Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders

Raja A. S. Mukherjee
Neil Aiton
Editors

Springer

Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders

Raja A. S. Mukherjee • Neil Aiton
Editors

Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders

Springer

Editors
Raja A. S. Mukherjee
National FASD Specialist Behaviour Clinic
Surrey
UK

Neil Aiton
The One Stop Clinic
Royal Sussex County Hospital
Brighton
UK

ISBN 978-3-030-73965-2 ISBN 978-3-030-73966-9 (eBook)
https://doi.org/10.1007/978-3-030-73966-9

This Springer imprint is published by the registered company Springer Nature Switzerland AG
The registered company address is: Gewerbestrasse 11, 6330 Cham, Switzerland

We would both like to dedicate this book to those who have been affected by prenatal alcohol exposure who have taught us so much along the way.

Grateful thanks to all the support given to me over years by my parents, Ananda and Sarbani Mukherjee, and to my Children, Joshua and Zac, and most importantly my Wife, Cheryl, for supporting me at every step of this FASD journey.
Raja A. S. Mukherjee

With grateful thanks to Fiona, who not only puts up with me with much grace and forebearance, but also the time ***it has taken*** *to edit this book.*
Neil Aiton

Foreword

Raja Mukherjee and Neil Aiton should be congratulated—a book to support the prevention, diagnosis and management of FASD is long overdue. I remember the day I asked Dr. Mukherjee to write an article on FASD for a new teaching website that I had just launched with the title 'Intellectual Disability and Health' (www.intellectualdisability.info). Little did I know that it would spark a clinical and research interest that would change the direction of his career!

In a medical textbook written by senior clinicians the first chapter is unusual but very refreshing, focusing as it does on the importance of people's stories. Being something of a storyteller myself, albeit in the long tradition of wordless stories [1], I do not need to be persuaded of their value. Indeed I suggest that listening is one of the most important skills for any clinician to learn, alongside the art of observing and patiently recording everything you have noticed. It is this kind of careful listening and observing over many years which has led to the co-creation of this informative and readable guide, so meaningfully informed by people directly affected by FASD.

I am reminded of the pioneering work of Dr. Byng-Hall who introduced Family Therapy to the Tavistock clinic in London in the 1970s. I remember him telling me about the way in which our family stories can influence our whole lives and that sometimes other people both within and without the family may have a version which an individual member does not recognise as their own authentic story. And as health and social care practitioners, we may be too quick to think we know a person's life history without taking time to really listen.

Byng-Hall [2] insisted that 'the ending of stories isn't necessarily already determined at the beginning but has the potential to be changed along the way'…an idea that is so important in the world of someone with FASD where there is no place for therapeutic nihilism. For children who were exposed to alcohol in utero this is a story that needs to be explored carefully so that everyone can come to terms with what happened and its implications. These issues are sensitively dealt with in some detail.

It goes without saying that early diagnosis is important so that the very best educational and social understanding and support is in place to meet each child and

young person's individual needs and to pave the way for their adult lives. There are many ideas to inspire a positive attitude in anyone who is learning about FASD and its effects for the first time and to renew and extend the knowledge and skills of more experienced practitioners.

I hope this book will instil confidence and lead to timely diagnoses and respectful and compassionate long-term support and management in the community. For such an under-diagnosed, commonly occurring and yet preventable condition, greater awareness should also reduce the number of babies harmed by alcohol in the future. I hope so.

Sheila the Baroness Hollins
Faculty of Health, Social Care and Education,
Kingston and St. George's (A joint enterprise of Kingston
University and St. George's, University of London)
London, UK

References

1. Hollins S, editor. Books beyond words. London; 1989–2021. https://www.booksbeyondwords.co.uk.
2. Byng-Hall J. The family script: a useful bridge between theory and practice. J Fam Ther. 1985;7:301–5.

Definition of Alcohol Units

The reader should be aware that the definition of a 'standard unit' of alcohol varies, and appropriate caution should be taken when referring to published literature. The different chapters in this book refer to a wide range of international sources which use different standards.

Most drinks show the concentration of alcohol as a percentage, often summarised as X.X% ABV (alcohol by volume)

UK	10 mL pure ethanol (8 g by weight)
	The labelling by law must show the number of units within the container
	In the UK, the number of units in a drink can be calculated as follows:
	Units = Volume of drink (in mL)/1000 mL × % alcohol
EU	Definition of a unit variable between member states:
	10 g ethanol most common, followed by 12 g (range 8–20)
	Labelling requirement: % alcohol by volume
	(member states may have additional requirements)
USA	A 'standard drink' contains 14 g ethanol, 0.6 oz (approx. 18 mL)
	[A standard drink is therefore 1.75× a UK standard unit]
Canada	A standard drink contains 13.6 ethanol (17.2 mL)
Australia	A standard drink contains 10g ethanol (12.5 mL)

Contents

Abbreviations

ABAS	Adaptive behaviour assessment system (e.g. ABAS-3)
ABI	Alcohol Brief Interventions (see also BI)
ACE	Adverse Childhood Experiences
Act	Act of Parliament-the method of making law in the UK
AD	Attachment Difficulty
ADD	Attention Deficit Disorder
ADH	Alcohol Dehydrogenase
ADHD	Attention Deficit Hyperactivity Disorder
ANS	Autonomic Nervous System
ARBD	Alcohol-Related Birth Defect
ARND	Alcohol-Related Neurodevelopmental Disorder
ASD	Autistic Spectrum Disorders
ATP	Adenosine triphosphate
AUDIT	Alcohol Use Disorders Identification Test (including AUDIT-C)
BI	Brief Interventions (see also ABI), also referred sometimes as S (screening) BI
BMA	British Medical Association
BMI	Body mass index
BRIEF	Behaviour Rating Inventory of Executive Function
CELF 4	Clinical Evaluation of Language Fundamentals, fourth UK Edition
CCG	Clinical Commissioning Group (UK) – groups which commission healthcare in each region
CGH array	Comparative Genomic Hybridisation (a form of genetic testing)
CIC	Children in Care
CNS	Central nervous system
CoP	Code of Practice
CYP	Child/young person (can also be plural)
DfBIS	Department for Business Industry and Skills
DfE	Department for Education (UK)
DfES	Department for Education and Skills
D-KEFS	Delis-Kaplan Executive Function System
DOH	Department of Health (UK)
DSM	Diagnostic and Statistical Manual (version often expressed in Roman numerals, e.g. DSM-V)

DWP	Department for Work and Pensions (UK)
EF	Executive function
EHCP	Education Health and Care plans
ELBW	Extremely Low Birth Weight
EUFASD	European FASD Alliance
FAE	Fetal Alcohol Effect
FAEE	Fatty Acid Ethyl Esters
FAS	Fetal Alcohol Syndrome (see also pFAS)
FASD	Fetal Alcohol Spectrum Disorders
FAST	Fast Alcohol Screening Test
FRI	Fluid Reasoning Index
FSIQ	Full Scale IQ
GABA	Gamma amino-butyric acid (a neurotransmitter)
GAI	General Ability Index
GGT	Gamma-glutamyl transferase (liver enzyme)
HC	Head Circumference (see also OFC)
ICA	Inter-country Adoption
ICD	International Classification of Diseases
IQ	Intelligence quotient
IUGR	Intra-Uterine Growth Restriction
LA	Local Authority (UK, responsible for local public services and facilities)
LAC	Looked After Children
LARC	Long-Acting Reversible Contraception
MI	Motivational Interviewing
MRI	Magnetic Resonance Imaging
NADH	Nicotinamide adenine dinucleotide
NDPAE	Neurobehavioural Disorder associated with Prenatal Alcohol Exposure
NHS	National Health Service (United Kingdom and devolved nations)
NICE	National Institute for Health and Care Excellence (UK)
NICU	Neonatal Intensive Care Unit
NOFAS (UK)	A Non-Governmental Charitable Organisation. Now called National FASD
NOFAS	National Organisation for Fetal Alcohol Syndrome (USA)
OFC	Occipito-frontal circumference (see also HC)
PAE	Prenatal Alcohol Exposure
PEth	Phosphatidylethanol
PF	Palpebral fissure
pFAS	Partial Fetal Alcohol Syndrome
PSI	Processing Speed Index
RCT	Randomised Controlled Trials
ROS	Reactive Oxygen Species
SD	Standard Deviation
SEN (SEND)	Special Educational Needs (and disabilities)

SENCo	Special Educational Needs Coordinator
SEN support	A system for identifying and meeting a child's needs if they need additional support for learning in school
SI	Sensory Integration
SIGN	Scottish Intercollegiate Guidelines Network
SPD	Sensory Processing Disorder
Statement of SEN	A statement of special educational needs sets out a child's needs and the help they should have. It is reviewed every year to make sure that any extra support given meets a child's needs
T-ACE	Alcohol screening test for pregnant women (Letters = mnemonic)
Tribunal	A collection of individuals responsible for handling appeals against local authority decisions regarding special educational needs
TWEAK	Alcohol screening test for pregnant women (Letters = mnemonic)
UN	United Nations
VSI	Visual Spatial Index
WAIS-IV	Wechsler Adult Intelligence Scales—Fourth Edition
WHO	World Health Organization
WISC-V	Wechsler Intelligence Scales for Children—Fifth Edition
WMI	Working Memory Index

Part I

It Starts at the Very Beginning

Section 1 Overview: The Importance of Stories

1

Neil Aiton and Raja A. S. Mukherjee

1.1 FASD: The Stories Which Need to Be Told …

History is who we are and how we are made. (David McCullough)

History is important—for individuals and for cultures, as well as for civilisations. Storytelling has become part of the innate way we, as humans, respond to and interact with each other. Stories by their nature have to be understood, remembered and shared. The storage and gradual accretion—as well as the shared understanding of those memories—are what build us into who we are as individuals and help define how we relate to others and to the world around us. The shared experiences re-lived in the telling of common stories of special or significant events help bind us together in society.

Books often contain stories, and in many ways, although this book is not a story book—as well as containing a collection of different stories—it does indeed tell its own powerful story: the story of FASD. Amongst the scientific references, the research studies and the prose lie glimpses of the real people whose stories need to be told. The centrepiece of that story is, of course, how these peoples' lives are continuing to be affected by the lifelong consequences of exposure to alcohol during fetal life. We are especially grateful to one individual, Lee, for contributing a chapter describing his own perspective. However, his story, although unique, can be seen in part, as representative of so many others. The individual chapters when

N. Aiton
The One Stop Clinic, Royal Sussex County Hospital, Brighton, UK
e-mail: neil.aiton@nhs.net

R. A. S. Mukherjee (✉)
National FASD Specialist Behaviour Clinic, Surrey, UK
e-mail: Raja.Mukherjee@sabp.nhs.uk

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_1

taken as a whole unveil the way that prenatal alcohol exposure impacts so many aspects of the way we function and interact as humans, and the challenges that arise—not only in assessment and diagnosis but also in understanding the unique nature of each individual who has been affected.

Then there is the story of the research itself and how that is progressing in so many different areas, so that we are able to find better ways to understand, prevent, diagnose and help those who are affected. Unfortunately, a book such as this only tells the story up to a certain point, but this story is continuing. Behind all of these research studies also lie the researchers themselves who have their own stories too, about how they first became involved and captivated by the importance of studying and researching this subject, as well as motivated to continue in the field.

Of course, the fundamental point of telling the story about FASD is to lead us to the position where we are able to prevent the problem from happening in the first place. Stories and experiences from other conditions where there are important adverse outcomes (death or lifetime disability) from a theoretically preventable problem such as leprosy, smallpox or TB have taken decades to progress. In a similar way, the societal change over the attitudes, prevalence and legal framework regarding smoking has taken decades to achieve since the severe adverse outcomes first became known in the 1960s and 1970s.

Chapter 2 begins with the story of how the historical understanding has developed regarding the effect of alcohol on the unborn child during pregnancy. Although most accounts about the historical aspect of FASD start with the classic paper by Jones et al. from 1973 [1] (and often ignoring a prior paper by Lemoine [2] because that story was told in a different language), it is likely that the dangers of alcohol consumption in pregnancy were not widely known earlier than this, just as even now this is not universal knowledge. There were concerns in the UK prior to the introduction of the gin laws in the 1750s, and also in Victorian times, although these were mostly around morality and social disorder. The story about the introduction of the gin laws and associated taxation has also largely been forgotten but this narrative can be compared very closely to modern attitudes and potential impacts with respect to the relatively recent arguments concerning the 'minimum unit pricing' taxation of alcohol [3, 4]. Of course, this is why *these* stories need to be told along with the ones in this book, so that we don't 'forget' again, and so that understanding and attitudes can eventually be changed.

Our own collective, cumulative, shared knowledge and experience in the field of FASD brings a moral imperative to strive to do anything we can to prevent this problem and overcome the barriers to change. Chapter 3 introduces the issues involved when considering the balance between the rights of the developing fetus and that of the mother, which have to be carefully considered as we attempt to address this problem: in particular to avoid the risk of unintentional harm to the fetus or the mother. Confusion over what the research shows and how this can be interpreted from different standpoints brings many challenges to how that conversation progresses. The importance of hearing others' perspective, as well as their stories, in the process of telling our own is highlighted—leading to a position of mutual understanding from which beneficial change can grow. If we wish to successfully

invoke change, we must make sure that despite any sense of frustration, we hold back from 'shouting' our stories too loudly and respect the positions of others. In telling our stories, we must also learn to frame them in ways which are mostly likely to be receptive to and gain traction with those whom we are addressing—in other words, pay more attention to the art of storytelling.

Sometimes, perhaps researchers are not particularly good at communicating their stories—it is after all a different skill. However, in competing against other priorities within the cacophony of all the other stories, it is important that the story presented is crafted well and able to stand out against the competition, whether applying for funding or in disseminating findings. There are others who are well-versed in the craft of 'storytelling' such as journalists who can help, but we must remember that they will be viewing the topic through a completely different lens, looking for the best angle, the most newsworthy detail or an interesting 'hook' to give a story greater character—and thus all the better when told and re-told. The party game when a message is passed around a circle in whispers (also called the 'telephone game') is well known but illustrates the point that after a story has been told, there is no control over what happens to it. For researchers who are struggling to discover and establish 'truth', particularly when findings are small and incremental, this apparent distortion of their normal paradigm can cause significant discomfort. It can also lead to miscommunication as in the example at the end of Chap. 3.

Consider, too, the far-reaching implications when there are gaps in our stories. Those who work within the field of 'children, looked after' within the care system and adoption will know this very well, along with those who may have been adopted themselves. As humans, we naturally search for meaning and context in our lives. The journey we are travelling on through life is one that has to have a beginning as well as an end: where did we come from and where are we going? What if we don't have people around us who are able to tell us our early stories, or even those stories that preceded them, to fill in the gaps before we developed our own conscious memories? Aside from the existential questions, it is clear that when that beginning is missing, there are powerful urges to try to fill in those gaps. Later chapters on children in care and adoption touch on some of these issues and the problems springing from lost information in trying to help these children in the best way as they face the challenges of growing up and establish their own new independent story.

Continuing the same theme, there has been increasing evidence that our early stories right at the start of lives are so important in shaping us, and that we can live out the consequences of these experiences for the rest of our lives [5]. These early events can cause changes in the nervous and endocrine systems which respond and adapt to those experiences way before the development of conscious memory, and possibly even during prenatal life as well [6]. Although our current state of knowledge in this area is in its infancy, it is extremely likely that prenatal alcohol exposure has a significant additional part to play in addition to the resulting neurobiology which has already been uncovered. The product of these changes means that those individuals face their future life with a nervous system which is 'wired differently' and therefore responds differently to external events compared with others, with additional long-term impacts on mental health through the lifespan. There is also a

chapter looking at how those early stories are intertwined with parenting and combine to influence the process of attachment which can also have far-reaching implications down to the next generation.

How we parent our children is the product of many complex factors, but those who work with pregnant women and in the fields of substance use cannot escape the observation that a great many of the women who are dependent on alcohol or use alcohol as a coping strategy in daily life themselves have complex and often traumatic early stories to tell. These stories really do need to be heard more widely in society, so that we can move away from a culture which very easily tends to blame and victimises 'women who choose to drink' towards one which is able to understand, support and encourage change. Statistics and figures about alcohol and deprivation (it is a common misapprehension that these two words are always associated) are important to collect, from a public health and political perspective because unless we understand the nature and size of problems, it is difficult to gauge progress when trying to effect change. The danger which can arise from just looking at statistics alone is that of de-personalisation and discrimination: here, individual stories can help with a wider understanding because they bring out the personal experiences and humanity of those managing to live, thrive and survive (or perhaps not?) despite significant adversity.

Of course, we don't have a right to hear people's private stories: we all own our own stories and can make choices about who to share them with. Often these stories are too intimate and private to be shared widely, and to share these type of stories takes trust. Many women who have used alcohol and/or other substances over long periods of time have experienced significant discrimination. This discrimination can be personal or institutional and intended or unintended. They have often learned to become suspicious and highly sensitive to what they perceive to be discrimination as well as judgemental attitudes. This leads to mistrust of professionals and institutions, and with that an understandable reluctance to share their story, or even part of their story. A common narrative for this type of experience is 'I've been let down before, why should this be any different?' There is a great need to understand and attempt to break down these barriers, but this is impossible unless there is provision for their stories to be heard.

The chapter on talking to pregnant women provides practical advice on trying to communicate, be 'real', show empathy and encourage women to 'tell their story', so that we can have a true and honest conversation with them about their pregnancy, and most importantly—for those individuals who may need it—how we can offer to help. It must also be remembered that because alcohol is so ubiquitous in our society, the knowledge that drinking alcohol in pregnancy can have profound and long-lasting effects is by no means universally known. Hence the importance of asking at initial pregnancy booking—not just about the current pattern but also about their knowledge and understanding, remembering that even women who do not drink alcohol regularly may drink significant amounts on special occasions or crises without the knowledge they could inadvertently be causing harm.

During pregnancy, how the fetus develops is itself a seemingly miraculous story about carefully choreographed patterns of cellular growth, any of which can be

potentially interrupted or altered by the presence of alcohol. It is those within the nervous system which have the greatest impact and long-standing consequences as told in many of the chapters in this book. Detailed knowledge of fetal development helps us understand the timing of impacts and give better understanding of the relationship of exposure to potential future outcome.

There is an irony that although the first descriptions about FAS were of newborns [1], it is currently extremely difficult to diagnose this problem in the new-born period using the currently available diagnostic frameworks. There is an urgent need for more detailed longer-term follow-up studies to investigate the relationship between exposure, early findings and biomarkers, which will help as useful predictors of future outcome. The encouragement to pursue this is the emerging evidence that earlier identification of FASD can help improve outcome. So the ending of stories isn't necessarily already determined at the beginning but has the potential to be changed along the way, rebutting a commonly held nihilistic fallacy (often used to justify inertia) that 'there's nothing thing that can be done anyway'.

Finally, we do like stories to have a 'happy ending'. The moral imperative to find better answers, to help those affected by alcohol in pregnancy as well as those affected by FASD in better ways, and to be better at preventing this problem in the first place means that this is an unfinished journey. A common journey on which all of us who have been impacted by our experience and understanding about FASD are bound together. We are privileged to build on the stories of those pioneers who first opened our eyes to this topic (too many to mention, and with the potential risk of accidental omission!). Looking back from the future to this present time, we too will be considered to have played our part in that journey—but will we have done enough? History will eventually have to make that judgement, but in the meantime, there is increasing hope, and still much to be done. The true happy ending to this story will be when (like polio) this problem has been virtually eliminated.

References

1. Jones KL, Smith DW. Recognition of the fetal alcohol syndrome in early infancy. Lancet. 1973;302(7836):999–1001. https://doi.org/10.1016/s0140-6736(73)91092-1. PMID: 4127281.
2. Lemoine P, et al. Les Enfants des parents alcholiques: anomalies observes a propos de 127 cas [The children of alcoholic parents: anomalies observed in 127 cases]. Quert Med. 1968;8:476–82.
3. Abel EL. The gin epidemic: much ado about what? Alcohol Alcohol. 2001;36(5):401–5. https://doi.org/10.1093/alcalc/36.5.401.
4. Dillon P. Gin: the much lamented death of madam Geneva the Eighteenth Century gin craze. London: Justin, Charles & Co; 2004. p. 263. ISBN 1-932112-25-1.
5. Larkin H, Shields JJ, Anda RF. The Health and Social Consequences of Adverse Childhood Experiences (ACE) Across the lifespan: an introduction to prevention and intervention in the community. J Pre Interv Commun. 2012;40(4):263–70. https://doi.org/10.1080/10852352.2012.707439.
6. Brandon AR, Pitts S, Denton WH, Stringer CA, Evans HM. A history of the theory of prenatal attachment. J Prenat Perinat Psychol Health. 2009;23(4):201–22.

Overview of FASD: How Our Understanding of FASD Has Progressed

2

Alan D. Price

Chapter Highlights

- Provides an overview of the background and history of fetal alcohol spectrum disorder (FASD)
- Reviews the mechanisms by which prenatal alcohol exposure can lead to FASD
- Describes evidence on low-level exposure and social determinants of alcohol-exposed pregnancies

The contemporary investigation into the impact of prenatal alcohol exposure is most often traced back to a series of articles by Kenneth Jones and colleagues at the University of Washington in the early 1970s [1–3], and to a lesser extent, to an article published in French 5 years earlier [4]. However, these publications have been described as rediscoveries of the harmful effects of alcohol on pregnancy, and such knowledge may have existed for hundreds or even thousands of years.

The earliest known warning concerning alcohol and pregnancy appears in the Old Testament Book of Judges, which is thought to have been written during or before the sixth century BCE. The Angel of the Lord appears to the Wife of Manoah to inform her that she will become pregnant, and warns her, amongst other instructions, to '… *drink no wine, nor other fermented drink* …' [5]. The woman gives birth to Samson, who becomes renowned for his strength and intellect. Whilst some have suggested that this represents knowledge of the harmful impact that alcohol may have on an unborn baby [6], others have argued that this may not be the case, and that the instructions were most probably given to ensure that proper religious rites were followed [5, 7].

A. D. Price (✉)
School of Health and Society, University of Salford, Salford, England
e-mail: A.D.Price2@salford.ac.uk

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_2

Similarly, the following quote has been attributed to Aristotle (384–322 BCE): '*Foolish, drunken, or haire brain women most often bring forth children like unto themselves, morose and feeble*' [8], and has often been held as evidence that the Ancient Greeks had at least a rudimentary knowledge of the ill effects of alcohol consumed during pregnancy [4, 9]. However, it later became apparent that renaissance scholar Robert Burton fabricated the quote, along with several others, in *The Anatomy of Melancholy* (1621). Aristotle did in fact discuss the influence of alcohol on conception in *Problemata* (reprinted 1927), but only in terms of its effect on the male libido, and that as a result of cooling the male body prior to intercourse, a couple might have the 'misfortune' to conceive a girl [7, 8].

There is little evidence of attention to the issue of prenatal alcohol exposure between these ancient clues and the end of the 1600s, when the abolishment of a monopoly on distillation in England coincided with a levy on the import of French brandy. By the early 1720s, the price of domestically produced gin had fallen so sharply that the working classes could afford to consume several pints each of the strong liquor per year [9, 10]. During the next 30 years, the widely documented public drunkenness on the streets of the English capital became known as the 'London gin epidemic'. Several observations [9] have been documented from that era, which appear to show that physicians were well aware of the harm that alcohol was having on unborn babies:

> *Half the train of chronical diseases with which we see children afflicted are only the secondary sighs and groanings … of parentive ill-spent life. These consequences … will be brought on infants by the debauchery of the Mother*. [11]

> *What must become an infant, who is conceived in gin? With the poisonous distillations of which it is nourished, both in the womb and at the breast*. [12]

> *… the enormous use of spirituous liquors … renders such infants as are born meagre and sickly, and unable to pass through the first stages of life*. [13]

Such quotes have generally been held as fairly strong evidence that the dangers of prenatal exposure to alcohol, or at least to gin, were well known during the early to mid-1700s [9, 14, 15]. Others have noted that these complaints about drunkenness only applied to the 'inferior classes' [16, 17]. Many social and economic forces influenced the lives of the working classes, the health of their children, and how they were perceived by upper-class physicians and politicians. It is also interesting to note that cheap, home-made gin was widely available on the black market, especially after a tax rise in 1736, which often contained additives such as sulphuric acid, lime oil and turpentine [18], and that prenatal exposure to turpentine has recently been shown to damage neurobehavioural functions in rats [19]. Exposure to turpentine, or other chemicals in bootleg gin, during the eighteenth century could, therefore, go some way to explain the problems seen in working-class children born during this time.

During the nineteenth century, public attitude towards alcohol in Europe and North America became more negative, fuelled in part by the temperance movement.

The first epidemiological studies on alcohol's harmful effects were published, such as Sullivan's [20] study of 120 pregnant inmates at a Liverpool women's prison. Maternal alcoholism was found to be associated with a higher rate of stillbirth or infant mortality than in cases where mothers had been denied alcohol. Sullivan also noted that paternal and grandparental alcoholism were less damaging than maternal exposure [7]. Such research was supported by, and conducted in support of, the temperance movement, whose attitude towards alcohol was based primarily on religious teachings [7]. Prohibition of alcohol in the United States was accompanied by often sensational claims about the harms of alcohol and dependency, so much so that following the end of prohibition in 1933, warnings about the harmful effects of alcohol were largely denounced as propaganda. Any previous research that supported the temperance view of alcohol was effectively discredited, and more recent findings pertaining to prenatal alcohol exposure were met with some incredulity by clinicians [7, 10].

In the second half of the twentieth century, a doctoral thesis and a related journal article, both written in French [4, 21], described physical and behavioural deficits in over 200 babies born to alcoholic mothers. The authors noted the impact of prenatal exposure to alcohol on the developing foetuses, but neither article managed to make an impact [6, 22]. The issue remained hidden until 1970, when Christie Ulleland, a junior paediatrician at the University of Washington School of Medicine, noticed that maternal alcoholism was a factor in a significant number of infants suffering with failure to thrive. She wrote that:

> *These observations indicate that infants of alcoholic mothers are at high risk for pre- and post-natal growth and developmental failure, and suggest that greater attention should be given to alcoholic women during the child bearing years.* [23]

This brief statement is arguably the true source of the modern investigation into the effects of prenatal exposure to alcohol. Eight of those infants from alcoholic mothers suffered from microcephaly, stunted growth, developmental delay, short palpebral fissures, joint anomalies and small jaws. This collection of defects was given the name Fetal Alcohol Syndrome and received global recognition in a series of articles written by Ulleland's colleagues Kenneth Jones and David Smith, whose names have become synonymous with the first descriptions of Fetal Alcohol Syndrome [1–3].

There was initial scepticism that such a ubiquitous substance as alcohol could do so much damage without having come to the attention of medical professionals. Since alcoholism frequently coincides with malnutrition, poverty and a chaotic home environment, many suggested that these environmental risks could explain a range of birth defects just as easily as alcohol [2, 10]. A steady increase in research attention mostly diminished these concerns, although the interaction between prenatal alcohol exposure and early environmental risk is still not fully understood [24]. Animal models provided experimental evidence of the harmful effects of alcohol [25, 26], even finding the same distinctive facial anomalies in mouse pups as those described by Jones and colleagues in human infants. Meanwhile, human

epidemiological studies [27–29] provided evidence of prevalence, range of outcomes, and began to demonstrate a link between the level of consumption and the severity of outcome.

In the time since this increase in attention to the issue, the harmful effects of alcohol used during pregnancy have become better understood and widely known to the general public, at least in societies where alcohol is widely used. Government warnings to reduce or avoid alcohol consumption during pregnancy in the United States in the 1970s and 1980s were followed by similar warnings in other Western countries, but the extent to which these warnings are having the desired effect is the subject of some debate [10, 30].

Alcohol is a teratogen—an agent that causes malformation to an embryo or fetus. There are several mechanisms by which alcohol can impact fetal development, as well as an increasing number of techniques that may be used to investigate these effects. Prenatal alcohol exposure (PAE) can impact the development of the whole fetus, and exposed individuals can present with structural damage to such areas as the digestive system [31], skeleton [32], heart [33], eyes [34] and the immune and endocrine systems [35].

Alcohol consumed during pregnancy enters the mother's bloodstream, passes freely though the placenta, and into the fetus and amniotic fluid [36]. In adults, alcohol is metabolised by a pair of enzymes called cytosolic alcohol dehydrogenase (ADH) and hepatic CYP2E1. These enzymes begin to appear in the individual during gestation, but do not reach maximum efficacy until well into childhood [37]. Instead, the main elimination method available to the fetus is transfer of alcohol back into the maternal bloodstream, but this is a complex process which can be impeded by alcohol-related constriction of blood vessels [36]. For these reasons, elimination of alcohol from the fetal compartment only occurs at around 3–4% of the maternal rate, which leads to prolonged fetal exposure [38].

Whilst in the fetal compartment, alcohol can disrupt the development of the fetus via a number of mechanisms. Apoptosis is the process of programmed cell death, which is usually beneficial and necessary in organic growth, but this process can be inappropriately initiated or supressed by some diseases or exposures to exogenous substances including alcohol [39, 40]. Apoptotic cell death due to alcohol exposure has been demonstrated in animal fetus models [41, 42], animal infant models [43, 44] and in vitro cell culture experiments [45, 46]. This process is seen in the adult human liver following alcohol consumption [47], but may be particularly harmful during fetal development since alcohol here is metabolised more slowly and since damage to cells during organogenesis can lead to improper development of those organs [36].

Alcohol can induce apoptosis by promoting the generation or inhibiting the elimination of reactive oxygen species (ROS)—unstable molecules formed during metabolic processes, which can react with many other molecules including those involved with cellular processes such as DNA and proteins [47]. Moreover, alcohol exposure can reduce the number of antioxidant cells, whose functions include elimination of ROS [47]. Raised levels of ROS can lead to uncontrolled apoptosis, and animal models have suggested this as a mechanism for the characteristic craniofacial and

neurodevelopmental abnormalities seen in FAS [48, 49]. Linked to this is the effect of alcohol on mitochondria, which are organelles present in all cells and are crucial to oxygen and energy production within the cell. There is evidence from rodent models that prenatal alcohol exposure can lead to apoptosis via mitochondria damage [50].

Besides direct impact on cellular processes, PAE may affect gene expression. DNA molecules are present in every type of cell, and contain essentially the same information, most of which will be unnecessary to the functioning of a given cell. Epigenetic markers are molecules that develop during gestation and attach to DNA molecules, and whose role is to activate or inhibit certain genes or groups of genes, ultimately controlling gene activity [51]. Various environmental factors can influence epigenetic markers and gene expression, sometimes resulting in adverse outcomes such as cancers [51]. There is emerging evidence from animal fetus and cell culture studies that PAE can alter epigenetic markers, resulting in widespread changes in gene expression [52]. Moreover, recent human clinical studies have shown expected epigenetic differences in children with fetal alcohol spectrum disorder (FASD), which support the role of epigenetics as a mechanism for PAE-related harm in humans [53, 54].

Alcohol-related damage to cells in the developing fetus is especially harmful for the developing brain, which develops throughout gestation and is therefore vulnerable to alcohol exposure during the entire pregnancy. Within the brain, however, different structures may be especially at risk during different periods, and this may explain some differences in FASD presentation. The rudimentary structures of the brain and nervous system begin to develop during the embryonic period, which lasts from conception to about 8 weeks gestation. The primitive brain provides the basic plan for development but continues to develop across the whole pregnancy and into the postnatal period. The brain first begins to form as a tube, which forms into sections that become the forebrain, midbrain and hindbrain. At around 4–8 weeks gestation, the first fissure of the brain starts to form, and this separation produces the two hemispheres of the cortex. The further sulci and gyri (grooves and ridges) of the cortex continue to develop until the late stages of pregnancy, at Weeks 23–35 [55]. The brain areas whose development is more active during the first trimester, such as the midbrain and the limbic system, which are associated with emotional and instinctive processes, may be more susceptible to harm during this time, whereas the cortical areas, which undergo more development later on, may be more at risk during the second or third trimesters. Some evidence for this comes from studies of the timing and pattern of alcohol exposure in humans. Alcohol consumption during the first, rather than second or third trimesters, has been shown to be associated with increased emotional difficulties [56, 57]. Craniofacial effects such as the thin vermillion border, smooth philtrum and microcephaly have been shown to be dependent on specific exposure times, such as gestational days 19 and 20 in non-human primates [58] and in the second 6 weeks of gestation in humans [59].

Children prenatally exposed to alcohol can present with a wide variety of physical, cognitive and behavioural problems, and to varying degrees. The relative

breadth and severity of deficits are associated with a number of risk and protective factors which contribute to the impact of alcohol on a developing fetus. The most influential of these is the drinking behaviour of the expectant mother. Consistently high levels of alcohol consumption throughout pregnancy and/or episodes of binge-drinking (more than 6 units per session for women [60]) are associated with severe cognitive deficits [61], behavioural disorders [62] and structural cortical abnormalities [63] in humans. Animal models have shown that high-dose alcohol exposure causes severe neurodevelopmental deficits in primates and rodents [64]. Results from studies into the impact of mild to moderate PAE in humans are less consistent, with some studies failing to find a significant detrimental effect on pregnancy outcome [65] or cognitive and behavioural development [66]. However, a recent meta-analysis found a small association between moderate PAE and behavioural issues in children [61], and a meta-analysis of high-quality studies showed an effect on cognitive outcomes and birthweight [67].

The mixed results of studies into low to moderate PAE may be partly due to the influence of confounding variables. True experiments in humans, where pregnant women are randomly assigned to consume alcohol, would be deeply unethical. Therefore, research is limited to observational designs such as case-control or cohort studies, which are susceptible to confounding variables [68]. For example, women who drink low to moderate amounts of alcohol during pregnancy tend to be more affluent than women who never drink and women who drink more heavily, and affluence is associated with advantages in offspring such as higher levels of intelligence and academic achievement [69]. Factors such as affluence could, therefore, partly explain the mixed results of studies into low to moderate alcohol exposure on cognitive functioning in school-age children. One technique that can bridge the gap between observational studies and experimental studies in humans is Mendelian randomisation. Named after early geneticist Gregor Mendel, this technique relies on the random distribution of genes from each parent to their offspring. This leads to random variation within the population of genetic determinants of behaviour, including alcohol consumption. Variations of a gene that controls the production of alcohol dehydrogenase—rs1229984 (ADH1B)—have been shown to predict alcohol consumption before and during pregnancy [70] and are considered the most useful tool to conduct quasi-randomisation to conditions of varying PAE in humans [71]. Studies that have employed this technique have demonstrated significant incremental differences between moderate, low and zero alcohol exposure in pregnancy, where higher exposures were associated with deficits in intelligence and academic achievement [71], early-onset persistent conduct problems [72] and atopic conditions such as asthma, hay fever and eczema [73].

There is some evidence of a genetic component in the teratogenic effects of alcohol. For example, PAE has been experimentally shown to have differing effects on five distinct inbred strains of mice [74]. One strain showed severe global physical defects following maternal prenatal intubation of alcohol, whereas another strain showed no teratogenesis and the remaining three strains showed intermediate effects. A mechanism for these kinds of results may be the variation in the rate of maternal or fetal metabolism of alcohol; the efficacy of enzymes such as those

mentioned above—ADH and CYP2E1—may be largely the result of genetic variation in humans as well as animals [75]. At least one human twin study has supported the role of genetics, with monozygotic alcohol-exposed twin pairs showing significantly greater concordance in terms of IQ and diagnosis than between dizygotic pairs [76], but these kinds of studies are scarce.

Increased or decreased exposure to other substances can also negatively impact fetal development. Risks relating to exposure to exogenous substances such as tobacco and street drugs are well known [77, 78], and there is some evidence that prenatal polydrug use, combined with alcohol exposure, may be more harmful than alcohol exposure alone [79]. Many women who consume alcohol during pregnancy, especially those who struggle with addiction, can be further affected by polydrug exposure [80], as well as other related issues such as malnutrition. Maternal diet or nutrient deficiency may also intensify the impact of PAE—low levels of iron [81] and zinc [82] in maternal blood have been shown to exacerbate the teratogenic effects of alcohol in animal models. Choline supplementation may be useful as an intervention during pregnancy and early childhood as this has been shown to moderate the impact of PAE in rats [83] and humans [84], and multivitamin supplements have shown a similar effect in humans [85].

Much of the research into the specific dangers and mechanisms of alcohol-related harm to a developing fetus is necessarily conducted using either in vitro tissue samples or animal model research methods. True experiments using human participants would clearly be harmful and unethical, so it is not possible to design a PAE study in humans where all relevant variables—genetics, drug exposure, pattern of alcohol exposure, environmental influences and so on—can be effectively controlled. Studies using tissue samples are useful under particular circumstances, and this method can be tightly controlled, uncomplicated and cost-effective, which can allow for multiple internally valid experiments [86]. However, tissue studies are limited in terms of their ability to demonstrate the impact of PAE, which necessarily involves intact animals during pregnancy, with a digestive system and many bodily influences on alcohol consumed and its eventual impact on the fetus [86]. Studies using live animals bridge this gap, and although no animal is a perfect surrogate for the human body, different animals can provide useful models for different research questions [87]. Smaller animals such as rodents are suitable for neurobiological or genetic studies, and their shorter gestation period allows for a greater turnover of results [88]. Larger animals such as sheep and pigs provide a gestation period and some behaviours similar to humans, whereas non-human primates offer the most similar model to humans in terms of genetics, neurological and cognitive development and social behaviours [87, 89]. There are, of course, ethical issues inherent in the use of animals in this kind of research, but the impact that these studies can have on our understanding of PAE is substantial [90]. Where observational studies of humans have provided tentative findings, the use of various animal models has been invaluable in the process of supporting evidence in a manner which would otherwise be unrealistic [86].

Early evidence of the physical impact of PAE on the human brain came entirely from post-mortem investigations of children born to alcoholic mothers following

heavy PAE [91]. By the late 1970s, alcohol had become widely accepted as a teratogen responsible for growth deficiency, craniofacial abnormalities, joint or skeletal defects, microcephaly, cardiac problems and mental deficiency [92]. Further studies during this time began to produce more specific neurological findings including hydrocephalus (excess fluid in the brain), extensive neuroanatomical disorganisation and leptomeningeal neuroglial heterotopia (an abnormal sheet of neural or glial tissue) around the meninges, cerebellum and brainstem [93]. Such studies highlight the extent to which heavy PAE can impact brain development, often leading to stillbirth or infant mortality, but these findings may not be representative of the full range of deficits, especially those seen following moderate alcohol exposure [59]. More recently, advances in brain imaging technologies have allowed the assessment of brain structure and function in live human patients. Magnetic resonance imaging (MRI) has shown that reductions in overall brain volume and surface area as well as malformations are common, but that some brain structures are more often damaged than others. These include the cerebellum, basal ganglia, hippocampus, the frontal and parietal lobes of the cortex, and the corpus callosum [94, 95]. The corpus callosum, the central bundle of neural fibres connecting the two hemispheres of the brain, which is responsible for integration of cognitive, sensory and motor functioning, appears to be particularly vulnerable to prenatal alcohol exposure. There is also an association between corpus callosum dysmorphology and the characteristic facial anomalies seen in FAS, which could potentially be used by clinicians to determine the extent of neurostructural damage [96]. Related to this neuroanatomical damage is the impact of PAE on the endocrine system. Animal models and human observational studies have demonstrated a link between PAE, hormone imbalance and an increased stress-response [97, 98], which implicates the hypothalamic-pituitary-adrenal (HPA) axis [99]. The HPA axis is formed of the hypothalamus and pituitary gland in the brain, and the adrenal glands which are located next to each kidney. The stress response begins with activation of the hypothalamus, and results in the release of cortisol, epinephrine and norepinephrine into the bloodstream. These hormones increase heart rate and glucose levels, which may be needed during the stressful situation [100]. PAE can increase activation of the HPA axis in humans [97], which means that individuals exposed to alcohol prenatally may be more susceptible to damage caused by stressful events during childhood or adulthood. Little research has been conducted on the relationship between prenatal alcohol and postnatal trauma, but an emerging pattern is that children with both of these exposures are more similar in terms of cognitive and behavioural functioning to children with just prenatal alcohol, than they are to children with just postnatal trauma [101]. This suggests that, rather than a synergistic effect of dual exposure, prenatal alcohol appears to be the driving force behind developmental difficulties in children with both exposures.

Fathers' drinking behaviour can also affect their unborn offspring, through both biological and environmental means. Alcohol consumption by biological fathers in the week leading up to IVF sperm collection has been found to predict failure to achieve live birth and spontaneous miscarriage, possibly due to the impact of alcohol on sperm count or quality [102]. In a mouse model, males who were intubated with alcohol prior to mating were more likely than non-exposed controls to produce

pups with teratogenic and developmental deficits [103]. Sperm quality was also suggested as an explanation for these effects, but the authors report that such mechanisms are poorly understood. Transgenerational epigenetic inheritance—the biological inheritance of acquired characteristics from the previous generation via epigenetic means—has been suggested as a mechanism for passing on drug-seeking behaviours from male mice to their offspring [104]. Epigenetic inheritance is also seen in plants and smaller animals such as nematodes, but the extent to which it may occur in mammals is poorly understood [105, 106]. Studies in humans have found associations between preconception paternal alcohol consumption and low birthweight [107], cognitive deficit [108], congenital heart defect [109] and leukaemia [110], but sperm quality is currently a better explanation for such effects than transgenerational epigenetic inheritance.

Aside from biological mechanisms, there appears to be a significant social effect on alcohol consumption in pregnancy, and particularly from the impact of male partners. A recent cross-cultural study of English and Swedish mothers [111] found that Swedish participants viewed women who drink during pregnancy as unfit mothers and were less likely to drink during pregnancy themselves, whereas English women emphasised alcohol as a social device and felt that drinking small amounts on special occasions was acceptable. The same study found that partners often reduced or abstained from alcohol during their partner's pregnancy, and this appeared to facilitate reduction or abstinence in the pregnant women. Women who drink during pregnancy have been shown to be more likely to live with a male partner who consumed alcohol [102], and women whose male partner was a heavy drinker were more likely to continue to drink during pregnancy [112]. The quality of a woman's relationship with her male partner has also been shown to influence her alcohol behaviour during pregnancy; women who reported higher relationship satisfaction were less likely to consume alcohol during pregnancy [112], and when fathers were involved in pregnancy care protocols, their female partners were less likely to drink during pregnancy [102]. Unfortunately, many of the findings relating to male partners' impact on FASD are taken from only a small number of high-quality studies, and there appear to have been no studies into the prenatal influence of female partners on maternal alcohol consumption [113]. The focus of investigations into the mechanisms of FASD has understandably been on in utero biological factors which necessarily implicate the biological mother, but there has been some concern that women have been unfairly villainised here, while the role of fathers, partners and wider society has been overlooked [113, 114].

There has been a huge increase during the last half century in the understanding of the impact of alcohol on many different biological mechanisms in the developing fetus. We have come to realise many different ways that the fetus can be affected by alcohol and how this might impact future growth and development. Although there is much more we need to learn, evidence from human, animal and cell tissue studies converges to provide strong and detailed evidence of the harmful impact of prenatal alcohol exposure. The infant can emerge from the uterus with biological and behavioural changes which can significantly affect adaptation to the postnatal environment.

References

1. Jones K, Smith D. Recognition of the fetal alcohol syndrome in early infancy. Lancet. 1973;302(7836):999–1001.
2. Jones K, Smith D, Streissguth A, Myrianthopoulos N. Outcome in offspring of chronic alcoholic women. Lancet. 1974;303(7866):1076–8.
3. Jones KL, Smith DW. The fetal alcohol syndrome. Teratology. 1975;12(1):1–10.
4. Lemione P, Harasseau H, Borteryu J, Menuet J. Les enfants de parents alcooliques: anomalies observées à propos de 127 cas [The children of alcoholic parents: anomalies observed in 127 cases]. Quest Med. 1968;21:476.
5. Abel EL. Was the fetal alcohol syndrome recognized in the ancient Near East? Alcohol Alcohol. 1997;32(1):3–7.
6. Calhoun F, Warren K. Fetal alcohol syndrome: historical perspectives. Neurosci Biobehav Rev. 2007;31(2):168–71.
7. Sanders JL. Were our forebears aware of prenatal alcohol exposure and its effects? A review of the history of fetal alcohol spectrum disorder. J Pop Ther Clin Pharmacol. 2009;16(2):e288.
8. Abel EL. Was the fetal alcohol syndrome recognized by the Greeks and Romans? Alcohol Alcohol. 1999;34(6):868–72.
9. Warner RH, Rosett HL. The effects of drinking on offspring: an historical survey of the American and British literature. J Stud Alcohol. 1975;36(11):1395–420.
10. Warren KR, Hewitt BG. Fetal alcohol spectrum disorders: when science, medicine, public policy, and laws collide. Dev Disab Res Rev. 2009;15(3):170–5.
11. Sedgwick J. A new treatise on liquors: wherein the use and abuse of wine, malt-drinks, water, etc. are particularly consider'd in many diseases, constitutions, and ages. London: Charles Rivington; 1725.
12. Fielding HA. Enquiry into the causes of the late increase of robberies, etc. with some proposals for remedying this growing evil in which the present reigning vices are impartially disposed. L. 1751.
13. Morris C. Observations on the past growth and present state of the City of London. To which are annexed a complete table of the Christnings and Burials within this City from 1601 to 1750; … together with a table of the numbers which have annually died of each disease from 1675 to the present time, etc. 1751.
14. Coffey TG. Beer street: gin lane. Some views of 18th-century drinking. Quarterly J Stud Alc. 1966;27(4):669–92.
15. Rodin AE. Infants and gin mania in 18th-century London. JAMA. 1981;245(12):1237–9.
16. Abel EL. The gin epidemic: much ado about what? Alcohol Alcohol. 2001;36(5):401–5.
17. Abel EL. Gin Lane: did Hogarth know about fetal alcohol syndrome? Alcohol Alcohol. 2001;36(2):131–4.
18. Solmonson LJ. Gin: a global history. London: Reaktion Books; 2012.
19. Fortier M-E, Luheshi GN, Boksa P. Effects of prenatal infection on prepulse inhibition in the rat depend on the nature of the infectious agent and the stage of pregnancy. Behav Brain Res. 2007;181(2):270–7.
20. Sullivan W. A note on the influence of maternal inebriety on the offspring. J Ment Sci. 1899;45(190):489–503.
21. Rouquette J. Influences of the parental alcoholic intoxication on the physical and psychological development of young children. Doctoral dissertation. Paris; 1957.
22. Lemoine P. The history of alcoholic fetopathies. J Pop Ther Clin Pharmacol. 2012;19(2):e224.
23. Ulleland C, Wennberg RP, Igo RP, Smith NJ. The offspring of alcoholic mothers. Pediatr Res. 1970;4(5):474.
24. Price A, Cook PA, Norgate S, Mukherjee R. Prenatal alcohol exposure and traumatic childhood experiences: a systematic review. Neurosci Biobehav Rev. 2017;80:89–98.
25. Chernoff GF. The fetal alcohol syndrome in mice: an animal model. Teratology. 1977;15(3):223–9.

26. Sulik KK, Johnston MC. Sequence of developmental alterations following acute ethanol exposure in mice: craniofacial features of the fetal alcohol syndrome. Am J Anat. 1983;166(3):257–69.
27. Harlap S, Shiono P. Alcohol, smoking, and incidence of spontaneous abortions in the first and second trimester. Lancet. 1980;316(8187):173–6.
28. May PA, Hymbaugh KJ, Aase JM, Samet JM. Epidemiology of fetal alcohol syndrome among American Indians of the Southwest. Soc Biol. 1983;30(4):374–87.
29. Sokol RJ, Miller SI, Reed G. Alcohol abuse during pregnancy: an epidemiologic study. Alcohol Clin Exp Res. 1980;4(2):135–45.
30. Burton A. Message on a bottle. Lancet Neurol. 2015;14(4):354–5.
31. Hofer R, Burd L. Review of published studies of kidney, liver, and gastrointestinal birth defects in fetal alcohol spectrum disorders. Birth Def Res A Cli Mol Teratol. 2009;85(3):179–83.
32. Simpson M, Duggal S, Keiver K. Prenatal ethanol exposure has differential effects on fetal growth and skeletal ossification. Bone. 2005;36(3):521–32.
33. Burd L, Deal E, Rios R, Adickes E, Wynne J, Klug MG. Congenital heart defects and fetal alcohol spectrum disorders. Congenit Heart Dis. 2007;2(4):250–5.
34. Strömland K, Pinazo-Durán MD. Ophthalmic involvement in the fetal alcohol syndrome: clinical and animal model studies. Alcohol Alcohol. 2002;37(1):2–8.
35. Zhang X, Sliwowska JH, Weinberg J. Prenatal alcohol exposure and fetal programming: effects on neuroendocrine and immune function. Exp Biol Med. 2005;230(6):376–88.
36. Gupta KK, Gupta VK, Shirasaka T. An update on fetal alcohol syndrome-pathogenesis, risks, and treatment. Alcohol Clin Exp Res. 2016;40(8):1594–602.
37. Zelner I, Koren G. Pharmacokinetics of ethanol in the maternal-fetal unit. J Pop Ther Clin Pharmacol. 2013;20(3):e259.
38. Heller M, Burd L. Review of ethanol dispersion, distribution, and elimination from the fetal compartment. Birth Def Res A Cli Mol Teratol. 2014;100(4):277–83.
39. Elmore S. Apoptosis: a review of programmed cell death. Toxicol Pathol. 2007;35(4):495–516.
40. Goodlett CR, Horn KH. Mechanisms of alcohol-induced damage to the developing nervous system. Alcohol Res Health. 2001;25(3):175.
41. Cartwright MM, Tessmer LL, Smith SM. Ethanol-induced neural crest apoptosis is coincident with their endogenous death, but is mechanistically distinct. Alcohol Clin Exp Res. 1998;22(1):142–9.
42. Ramachandran V, Perez A, Chen J, Senthil D, Schenker S, Henderson GI. In utero ethanol exposure causes mitochondrial dysfunction, which can result in apoptotic cell death in fetal brain: a potential role for 4-hydroxynonenal. Alcohol Clin Exp Res. 2001;25(6):862–71.
43. Ikonomidou C, Bittigau P, Ishimaru MJ, Wozniak DF, Koch C, Genz K, et al. Ethanol-induced apoptotic neurodegeneration and fetal alcohol syndrome. Science. 2000;287(5455):1056–60.
44. Young C, Olney JW. Neuroapoptosis in the infant mouse brain triggered by a transient small increase in blood alcohol concentration. Neurobiol Dis. 2006;22(3):548–54.
45. Chen CP, Kuhn P, Chaturvedi K, Boyadjieva N, Sarkar DK. Ethanol induces apoptotic death of developing β-endorphin neurons via suppression of cyclic adenosine monophosphate production and activation of transforming growth factor-β1-linked apoptotic signaling. Mol Pharmacol. 2006;69(3):706–17.
46. Katz GG, Shear NH, Malkiewicz IM, Valentino K, Neuman MG. Signaling for ethanol-induced apoptosis and repair in vitro. Clin Biochem. 2001;34(3):219–27.
47. Wu D, Cederbaum AI. Alcohol, oxidative stress, and free radical damage. Alcohol Res Health. 2003;27(4):277.
48. Cartwright MM, Smith SM. Increased cell death and reduced neural crest cell numbers in ethanol-exposed embryos: partial basis for the fetal alcohol syndrome phenotype. Alcohol Clin Exp Res. 1995;19(2):378–86.
49. Dunty WC Jr, Chen S, Zucker RM, Dehart DB, Sulik KK. Selective vulnerability of embryonic cell populations to ethanol-induced apoptosis: implications for alcohol-related birth defects and neurodevelopmental disorder. Alcohol Clin Exp Res. 2001;25(10):1523–35.

50. Manzo-Avalos S, Saavedra-Molina A. Cellular and mitochondrial effects of alcohol consumption. Int J Environ Res Public Health. 2010;7(12):4281–304.
51. Mazzio EA, Soliman KF. Basic concepts of epigenetics: impact of environmental signals on gene expression. Epigenetics. 2012;7(2):119–30.
52. Lussier AA, Weinberg J, Kobor MS. Epigenetics studies of fetal alcohol spectrum disorder: where are we now? Epigenomics. 2017;9(3):291–311.
53. Laufer BI, Kapalanga J, Castellani CA, Diehl EJ, Yan L, Singh SM. Associative DNA methylation changes in children with prenatal alcohol exposure. Epigenomics. 2015;7(8):1259–74.
54. Portales-Casamar E, Lussier AA, Jones MJ, MacIsaac JL, Edgar RD, Mah SM, et al. DNA methylation signature of human fetal alcohol spectrum disorder. Epigenetics Chromatin. 2016;9:25.
55. Stiles J, Jernigan TL. The basics of brain development. Neuropsychol Rev. 2010;20(4):327–48.
56. O'Leary CM, Nassar N, Zubrick SR, Kurinczuk JJ, Stanley F, Bower C. Evidence of a complex association between dose, pattern and timing of prenatal alcohol exposure and child behaviour problems. Addiction. 2010;105(1):74–86.
57. O'Leary CM, Bower C, Zubrick SR, Geelhoed E, Kurinczuk JJ, Nassar N. A new method of prenatal alcohol classification accounting for dose, pattern and timing of exposure: improving our ability to examine fetal effects from low to moderate alcohol. J Epidemiol Community Health. 2010;64(11):956–62.
58. Astley SJ, Magnuson SI, Omnell LM, Clarren SK. Fetal alcohol syndrome: changes in craniofacial form with age, cognition, and timing of ethanol exposure in the macaque. Teratology. 1999;59(3):163–72.
59. Feldman HS, Jones KL, Lindsay S, Slymen D, Klonoff-Cohen H, Kao K, et al. Prenatal alcohol exposure patterns and alcohol-related birth defects and growth deficiencies: a prospective study. Alcohol Clin Exp Res. 2012;36(4):670–6.
60. NHS. Binge drinking. https://www.nhs.uk/live-well/alcohol-support/binge-drinking-effects/.
61. Flak AL, Su S, Bertrand J, Denny CH, Kesmodel US, Cogswell ME. The association of mild, moderate, and binge prenatal alcohol exposure and child neuropsychological outcomes: a meta-analysis. Alcohol Clin Exp Res. 2014;38(1):214–26.
62. Fryer SL, Tapert SF, Mattson SN, Paulus MP, Spadoni AD, Riley EP. Prenatal alcohol exposure affects frontal-striatal BOLD response during inhibitory control. Alcohol Clin Exp Res. 2007;31(8):1415–24.
63. Sowell ER, Mattson SN, Kan E, Thompson PM, Riley EP, Toga AW. Abnormal cortical thickness and brain–behavior correlation patterns in individuals with heavy prenatal alcohol exposure. Cereb Cortex. 2008;18(1):136–44.
64. Maier SE, West JR. Drinking patterns and alcohol-related birth defects. Alcohol Res Health. 2001;25(3):168.
65. Henderson J, Gray R, Brocklehurst P. Systematic review of effects of low-moderate prenatal alcohol exposure on pregnancy outcome. BJOG. 2007;114(3):243–52.
66. Kelly Y, Iacovou M, Quigley MA, Gray R, Wolke D, Kelly J, et al. Light drinking versus abstinence in pregnancy - behavioural and cognitive outcomes in 7-year-old children: a longitudinal cohort study. BJOG. 2013;120(11):1340–7.
67. Mamluk L, Jones T, Ijaz S, Edwards H, Savovic J, Leach V, et al. Evidence of detrimental effects of prenatal alcohol exposure on offspring birthweight and neurodevelopment from a systematic review of quasi-experimental studies. Int J Epidemiol. 2020:dyz272.
68. Gray R, Mukherjee RA, Rutter M. Alcohol consumption during pregnancy and its effects on neurodevelopment: what is known and what remains uncertain. Addiction. 2009;104(8):1270–3.
69. O'Leary CM, Bower C. Guidelines for pregnancy: what's an acceptable risk, and how is the evidence (finally) shaping up? Drug Alc Rev. 2012;31(2):170–83.
70. Zuccolo L, Fitz-Simon N, Gray R, Ring SM, Sayal K, Smith GD, et al. A non-synonymous variant in ADH1B is strongly associated with prenatal alcohol use in a European sample of pregnant women. Hum Mol Genet. 2009;18(22):4457–66.

71. Zuccolo L, Lewis SJ, Smith GD, Sayal K, Draper ES, Fraser R, et al. Prenatal alcohol exposure and offspring cognition and school performance. A 'Mendelian randomization' natural experiment. Int J Epidemiol. 2013;42(5):1358–70.
72. Murray J, Burgess S, Zuccolo L, Hickman M, Gray R, Lewis SJ. Moderate alcohol drinking in pregnancy increases risk for children's persistent conduct problems: causal effects in a Mendelian randomisation study. J Child Psychol Psychiatry. 2016;57(5):575–84.
73. Shaheen SO, Rutterford C, Zuccolo L, Ring SM, Davey Smith G, Holloway JW, et al. Prenatal alcohol exposure and childhood atopic disease: a Mendelian randomization approach. J Allergy Clin Immunol. 2014;133(1):225–32.e1-5.
74. Downing C, Balderrama-Durbin C, Broncucia H, Gilliam D, Johnson TE. Ethanol teratogenesis in five inbred strains of mice. Alcohol Clin Exp Res. 2009;33(7):1238–45.
75. Gemma S, Vichi S, Testai E. Metabolic and genetic factors contributing to alcohol induced effects and fetal alcohol syndrome. Neurosci Biobehav Rev. 2007;31(2):221–9.
76. Streissguth AP, Dehaene P. Fetal alcohol syndrome in twins of alcoholic mothers: concordance of diagnosis and IQ. Am J Med Genet. 1993;47(6):857–61.
77. Cornelius MD, Day NL. Developmental consequences of prenatal tobacco exposure. Curr Opin Neurol. 2009;22(2):121.
78. Smith LM, LaGasse LL, Derauf C, Grant P, Shah R, Arria A, et al. The infant development, environment, and lifestyle study: effects of prenatal methamphetamine exposure, polydrug exposure, and poverty on intrauterine growth. Pediatrics. 2006;118(3):1149–56.
79. Rivkin MJ, Davis PE, Lemaster JL, Cabral HJ, Warfield SK, Mulkern RV, et al. Volumetric MRI study of brain in children with intrauterine exposure to cocaine, alcohol, tobacco, and marijuana. Pediatrics. 2008;121(4):741–50.
80. Harrison PA, Sidebottom AC. Alcohol and drug use before and during pregnancy: an examination of use patterns and predictors of cessation. Matern Child Health J. 2009;13(3):386.
81. Rufer ES, Tran TD, Attridge MM, Andrzejewski ME, Flentke GR, Smith SM. Adequacy of maternal iron status protects against behavioral, neuroanatomical, and growth deficits in fetal alcohol spectrum disorders. PLoS One. 2012;7(10):e47499.
82. Keen CL, Uriu-Adams JY, Skalny A, Grabeklis A, Grabeklis S, Green K, et al. The plausibility of maternal nutritional status being a contributing factor to the risk for fetal alcohol spectrum disorders: the potential influence of zinc status as an example. Biofactors. 2010;36(2):125–35.
83. Thomas JD, Abou EJ, Dominguez HD. Prenatal choline supplementation mitigates the adverse effects of prenatal alcohol exposure on development in rats. Neurotoxicol Teratol. 2009;31(5):303–11.
84. Wozniak JR, Fink BA, Fuglestad AJ, Eckerle JK, Boys CJ, Sandness KE, et al. Four-year follow-up of a randomized controlled trial of choline for neurodevelopment in fetal alcohol spectrum disorder. J Neurodev Disord. 2020;12(1):1–13.
85. Coles CD, Kable JA, Keen CL, Jones KL, Wertelecki W, Granovska IV, et al. Dose and timing of prenatal alcohol exposure and maternal nutritional supplements: developmental effects on 6-month-old infants. Matern Child Health J. 2015;19(12):2605–14.
86. Wilson SE, Cudd TA. Focus on: the use of animal models for the study of fetal alcohol spectrum disorders. Alcohol Res Health. 2011;34(1):92.
87. Cudd TA. Animal model systems for the study of alcohol teratology. Exp Biol Med. 2005;230(6):389–93.
88. Schneider ML, Moore CF, Adkins MM. The effects of prenatal alcohol exposure on behavior: rodent and primate studies. Neuropsychol Rev. 2011;21(2):186–203.
89. Kelly SJ, Goodlett CR, Hannigan JH. Animal models of fetal alcohol spectrum disorders: impact of the social environment. Dev Disab Res Rev. 2009;15(3):200–8.
90. Beydoun H, Saftlas AF. Physical and mental health outcomes of prenatal maternal stress in human and animal studies: a review of recent evidence. Paediatr Perinat Epidemiol. 2008;22(5):438–66.
91. Guerri C, Bazinet A, Riley EP. Foetal alcohol spectrum disorders and alterations in brain and behaviour. Alcohol Alcohol. 2009;44(2):108–14.

92. Clarren SK. Recognition of fetal alcohol syndrome. JAMA. 1981;245(23):2436–9.
93. Clarren SK, Alvord EC, Sumi SM, Streissguth AP, Smith DW. Brain malformations related to prenatal exposure to ethanol. J Pediatr. 1978;92(1):64–7.
94. Donald KA, Eastman E, Howells FM, Adnams C, Riley EP, Woods RP, et al. Neuroimaging effects of prenatal alcohol exposure on the developing human brain: a magnetic resonance imaging review. Acta Neuropsychiatr. 2015;27(5):251–69.
95. Lebel C, Roussotte F, Sowell ER. Imaging the impact of prenatal alcohol exposure on the structure of the developing human brain. Neuropsychol Rev. 2011;21(2):102–18.
96. Suttie M, Wozniak JR, Parnell SE, Wetherill L, Mattson SN, Sowell ER, et al. Combined face–brain morphology and associated neurocognitive correlates in fetal alcohol spectrum disorders. Alcohol Clin Exp Res. 2018;42(9):1769–82.
97. Haley DW, Handmaker NS, Lowe J. Infant stress reactivity and prenatal alcohol exposure. Alcohol Clin Exp Res. 2006;30(12):2055–64.
98. Hellemans KG, Verma P, Yoon E, Yu W, Weinberg J. Prenatal alcohol exposure increases vulnerability to stress and anxiety-like disorders in adulthood. Ann N Y Acad Sci. 2008;1144:154.
99. Weinberg J, Sliwowska JH, Lan N, Hellemans KG. Prenatal alcohol exposure: foetal programming, the hypothalamic-pituitary-adrenal axis and sex differences in outcome. J Neuroendocrinol. 2008;20(4):470–88.
100. Garrett B. Brain and behavior: an introduction to biological psychology. Philadelphia, PA: Sage Publications, Inc; 2009.
101. Price AD. The impact of traumatic childhood experiences on cognitive and behavioural functioning in children with foetal alcohol spectrum disorders. Salford: University of Salford; 2019.
102. Czeizel AE, Czeizel B, Vereczkey A. The participation of prospective fathers in preconception care. Clin Med Insights Reprod Health. 2013;7:1.
103. Lee HJ, Ryu J-S, Choi NY, Park YS, Kim YI, Han DW, et al. Transgenerational effects of paternal alcohol exposure in mouse offspring. Anim Cell Syst. 2013;17(6):429–34.
104. Finegersh A, Rompala GR, Martin DI, Homanics GE. Drinking beyond a lifetime: new and emerging insights into paternal alcohol exposure on subsequent generations. Alcohol. 2015;49(5):461–70.
105. Heard E, Martienssen RA. Transgenerational epigenetic inheritance: myths and mechanisms. Cell. 2014;157(1):95–109.
106. van Otterdijk SD, Michels KB. Transgenerational epigenetic inheritance in mammals: how good is the evidence? FASEB J. 2016;30(7):2457–65.
107. Passaro KT, Little RE, Savitz DA, Noss J. Effect of paternal alcohol consumption before conception on infant birth weight. Teratology. 1998;57(6):294–301.
108. Roeleveld N, Vingerhoets E, Zielhuis GA, Gabreëls F. Mental retardation associated with parental smoking and alcohol consumption before, during, and after pregnancy. Prev Med. 1992;21(1):110–9.
109. Steinberger EK, Ferencz C, Loffredo CA. Infants with single ventricle: a population-based epidemiological study. Teratology. 2002;65(3):106–15.
110. Milne E, Greenop KR, Scott RJ, de Klerk NH, Bower C, Ashton LJ, et al. Parental alcohol consumption and risk of childhood acute lymphoblastic leukemia and brain tumors. Cancer Causes Control. 2013;24(2):391–402.
111. Scholin L, Hughes K, Bellis MA, Eriksson C, Porcellato L. Exploring practices and perceptions of alcohol use during pregnancy in England and Sweden through a cross-cultural lens. Eur J Pub Health. 2018;28(3):533–7.
112. Bakhireva LN, Wilsnack SC, Kristjanson A, Yevtushok L, Onishenko S, Wertelecki W, et al. Paternal drinking, intimate relationship quality, and alcohol consumption in pregnant Ukrainian women. J Stud Alc Drugs. 2011;72(4):536–44.
113. McBride N, Johnson S. Fathers' role in alcohol-exposed pregnancies: systematic review of human studies. Am J Prev Med. 2016;51(2):240–8.
114. Gearing RE, McNeill T, Lozier FA. Father involvement and fetal alcohol spectrum disorder: developing best practices. J FAS Int. 2005;3(e14):1–11.

3 The Complex Issues Related to Alcohol and Pregnancy: Evidence, Ethics and the Law

Neil Aiton

Chapter Highlights

- An exploration of the complex issues involved with alcohol exposure in pregnancy and why some of the evidence appears conflicting
- Ethical considerations around the rights of the fetus and the mother
- The responsibilities of society, public bodies and individuals with respect to alcohol consumption in pregnancy

There are many factors which interplay to influence the approach people can take towards drinking alcohol in pregnancy. These can arise from personal experience, the attitude of friends and family, underlying culture, prevailing public perceptions and attitudes. Into that mixture comes the interpretation of the evidence, which sometimes can appear to conflict. Underlying these issues are often deeply held beliefs that individuals hold about autonomy, and the rights of the fetus versus the rights of the mother.

Some of the reasons for apparent conflicts in evidence on this subject are considered, along with an example of how evidence can be misinterpreted. There has also possibly been a mistrust of absolutist standpoints when that could not be robustly supported by evidence (e.g. the evidence of harm at very low levels of exposure). As well as these issues, this chapter explores some of the potentially conflicting legal, ethical and moral issues concerning maternal rights, and the rights of the fetus within the context of alcohol and pregnancy. Also, how understanding some of these perspectives can be important in ongoing conversations.

People approach the subject of alcohol in relation to pregnancy through the lens of a number of different fundamental frameworks: experiential, factual, ethical, moral, legal and professional paradigms. These different approaches can lead to different understandings regarding the relative importance of the different issues which

N. Aiton (✉)
The One Stop Clinic, Royal Sussex County Hospital, Brighton, UK
e-mail: neil.aiton@nhs.net

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_3

are presented when considering this topic. It can also lead to different interpretations when approaching the available data.

Understanding the paradigm through which individuals approach the subject can be helpful for professionals, because this might determine the approach taken in any ongoing conversation (see Chap. 5). Furthermore, it can help one to avoid approaches which may be either unhelpful, ineffective or potentially even counter-productive for any particular individual and therefore help to maintain professional/client relationships based on mutual understanding respect and acceptance of each other's standpoint.

3.1 Views About Science, Research and Accessibility

A quick internet search on the topic of drinking alcohol in pregnancy (e.g. using the keywords 'pregnancy, alcohol') brings up a plethora of results with links to widely disparate 'findings' in answer to that search. There might be links to seemingly authoritative bodies with appropriate guidelines and support (e.g. in the United Kingdom: the NHS, UK Government/Chief Medical Officer guidelines). There might also be links to professional bodies (Royal Colleges, health or pregnancy-related organisations). There will also be links to journalistic articles quoting research—with varying degrees of accuracy—indicating harms or lack of harms. Of course, journalists will most likely be highlighting a particular 'strap-line' or direct quote which may or may not challenge preconceptions or perceived orthodoxy about the subject, and perhaps presents a new angle. True research publications may perhaps be found several pages down and are often much harder to find, let alone access. So how does average person trying to find out what to do make sense of all this?

If one trusts independent non-partisan bodies (and the NHS (National Health Service) in the United Kingdom holds a high level of trust), then it's easy to accept the recommendations provided by such organisations and believe that these might be in your best interests. But inherent suspicion of central bureaucracy, central government or strongly held views about fetal or maternal rights might lead one to different sources, opinions and conclusions.

3.2 A Factual or 'Evidence-Based' Approach

A Factual or 'evidence-based' approach is based on an understanding that sifting and weighing-up the evidence can help one to come to a balanced understanding and conclusion. Inherent within this approach are contained beliefs about the 'truth' (or otherwise) of science itself as well as the 'system' which produces research. The quantity of evidence itself is growing significantly and seems overwhelming, with the risk that attention is drawn to the most recent studies, or ones which can support previously held preconceptions, or what is most easily accessible. How much weight does one attach to particular pieces of evidence, and how is it possible to reconcile apparent contradictory findings? These problems have been recognised for a long time and hence the growth of the science of meta-analysis, along with developments such as the

Cochrane Library (www.cochranelibrary.com) which attempt to provide a library of high quality authoritative and reliable healthcare information as well as resources to help synthesise evidence comprehensively, as objectively as possible and limit the introduction of bias into that process. The amount of evidence can sometimes be overwhelming, bring the problems of how to resolve prioritisation and weighting. Disparity in research findings can be reduced partly by attempts to refine the question that is actually being asked more carefully through the use of carefully framed questions [1], perhaps with the assistance of trained librarians. There are also particular research groups which also specialise in the particular area of evidence synthesis.

People who look at this subject through an evidence-based approach are likely to be amenable to support and help with discussions around appropriate evidence and meta-analyses, as well as acknowledgement of the areas of uncertainty - what don't we know the answer to, and where is the level of evidence poor, making it difficult to reach a conclusion?

Professionals themselves remain an important source of advice and support for those seeking healthcare, but perhaps this might be changing as people feel that they can access information for themselves in age where search engines and internet access is becoming more universally available. Interestingly there is some evidence that a degree-level education can be associated with a higher alcohol consumption in pregnancy compared to those without higher education. There may be several reasons for this, but one might be the possibility that the increased 'agency' that higher education brings might encourage individuals to search for evidence and reach their own conclusions [2].

Perhaps there is also a changing response in society (and even occasionally perhaps a suspicion?) about the role of the 'expert'? Previously, before the growth of the internet, the professions were a source of access to 'knowledge' which was held in books and specialist journals in libraries of esteemed institutions, and not available to access for most people. Now this knowledge is more widely available as well as accessible and the volume of it is increasing rapidly and exponentially. In fact, the problem is perhaps one of too much knowledge, and how to make sense of what there is.

Although professionals no longer control access to information, they can still have a really important role to help provide experience of similar situations, synthesis, weight and context in helping to make sense of the vast amounts of literature and evidence. They can help in the understanding of which evidence might apply in particular situations and be aware of the relative reliability (or otherwise) of different sources of evidence on which people might rely. Sensitivity, openness, honest concern and a non-judgemental attitude are likely to be important factors which help conversations to open up, allowing exploration of the relevant history to decide how help can best be given to those who are seeking it.

Sometimes, research is more likely to be quoted and read simply because it is more accessible (read and downloaded), or because attention was drawn to it for some reason, rather than because the points made are more important or more relevant to the question under consideration. Part of the problem is because of accessibility: the access to much published scientific research lies at the behest of publishing companies who retain the copyright and produce the journals within which much research is published—and make considerable sums of money by controlling access

for payment at institutional and personal level. So where access is difficult the abstract only might be viewed, rather than the whole paper, thereby missing potentially important information. More recently there has been a move to increase the amount of research which is available under 'open access' arrangements [3], and particularly so for research which is publicly funded. This debate is ongoing, but this particular issue—of alcohol and pregnancy—underlines the importance of being able to access wide-ranging research studies in a way that truly maintains balance according to merit and findings (rather than access arrangements) and demonstrates that specialist skills as well as institutional support are required to achieve that.

3.3 Reasons for Apparent Conflicts in Research Findings

So far, the problems of accessing data and finding balance without bias have been considered. But there are good reasons why in some situations the research evidence in this area can appear to be in conflict and contribute to confusion. When looking at research findings and seeking the answers to questions, it is important to pay close attention to the question which is being asked. Systematic methods exist which help to formulate the search question more rigorously [1] and this can help to narrow down the focus as far as the population studied, the type of study, the intervention or method of assessment used, and the way the outcome was measured. However, a quick internet search will not have the same degree of rigour and will most likely result in confusing multiplicity of results. As an example, just typing '*light drinking in pregnancy*' into a search engine brings up over 200,000 results, for which the headlines for each result range from: 'significant risks of adverse behavioural and learning outcomes', to 'no problems' or 'safe', that light drinking is 'dangerous', or conversely that is 'safe'. Underlying the disparity found in these results are the opinions of those writing the 'headlines' for each article. Different studies use different methods of assessment of alcohol consumption and what constitutes 'light drinking', different populations, and different ways of assessing outcome, as well as important differences which exist between countries at to what constitutes a 'unit' (see the introduction to this book for different definitions). It can take significant resources, multi-professional teams and specialist skills to try to come up with a carefully considered answer to this seemingly simple, but in actual fact quite complex question about 'light drinking' in pregnancy [14]—and even then, the results can be widely misrepresented (see the example given at the end of this chapter).

Apparent conflicts (or differences in research findings) may well be biological in origin: It has been known for a long time that the long-term outcome (i.e. defining the behavioural phenotype) resulting from particular levels of prenatal alcohol exposure is extremely variable and difficult to predict. Of course, the timing and level of exposure can explain some of this variability. However, there are clearly other factors involved, such as:

- the time period over which alcohol is consumed
- whether alcohol is taken with food
- genetic variations in metabolism

- the effect of induced metabolism of alcohol
- impairment of liver function which might affect alcohol metabolism and prolong exposure
- the quality of overall diet—affecting metabolic co-factors (e.g. choline, anti-oxidant function)
- presence of dietary deficiencies
- unknown genetic factors which affect metabolic and neurological resilience
- co-morbid factors—for example, smoking
- body composition (e.g. BMI)
- Epigenetics (level of DNA methylation)

So, the likely impact of any level of prenatal alcohol exposure on the nervous system (and hence future development) is likely to be a balance between a multiplicity of factors, some of which will serve to increase that impact and other factors which would be likely to be protective against damage being caused.

3.4 Ethical Approach to Understanding Maternal and Fetal Rights

There are a variety of ways that it is possible to approach ethical problems: from underlying principles, guided by the outcomes or consequences, or we can look at decision-making and the decision-maker. The four commonly held principles of medical (see Box 3.1) provide a widely acceptable basis with which to guide our approach [4] (although it does not provide a framework to help us when those principles might conflict).

Box 3.1 Four Principles of Medical Ethics [4]
- Beneficence (to bring about 'good')
- Non-maleficence (to avoid 'harm')
- Autonomy (respect for decisions of others about themselves)
- Justice (concepts of fairness, impartiality, equality)

Within the context of alcohol and pregnancy, it is possible to examine the issues presented through these different lenses. In terms of the first principle of beneficence, it would be easy to propose that we wish to reduce the number of babies affected by FASD—and no doubt that would universally be seen as a beneficial outcome. To achieve this objective, it is necessary to deal with the underlying causation of that problem: prenatal alcohol exposure, thus aim to prevent the consumption of alcohol in pregnancy. This would be good in terms of healthcare outcome for women themselves as well as the fetus and follows the first two principles. However, there are a range of choices to make about how one brings about that desirable outcome which can range from: passive recommendation, active encouragement,

provision of support or with increased severity, to legislation and associated criminality, even resulting in forced coercion though internment. Promoting the rights of the developing fetus necessarily thus imposes restrictions on the rights of the pregnant women to be able to drink alcohol even though she may be pregnant. A relatively liberal society would stop short of enforcing these restrictions, but prevailing politics and attitudes around healthcare and public health will affect how passive or active an approach is taken towards encouragement.

In 2000, the United Kingdom incorporated the rights set out in the European Convention on Human Rights into the Human Rights Act 1998. These rights become relevant for a baby from the moment of birth, but do not apply to the fetus which does not have the status of a legal person.

Variations in ethical viewpoints range from understanding the fetus as an autonomous individual with rights from the moment of conception, to the opposite view that the fetus does not have any rights until after birth and becomes a separate autonomous individual. Although even a newborn baby cannot be considered truly autonomous—and from an anthropological perspective the human infant is dependent upon its parents (albeit with decreasing importance) for many years. An intermediate viewpoint would be the consideration that the rights of the fetus take on a gradually increasing importance as gestation increases. However, while it is possible to consider this gradual increase in rights from a moral, ethical and philosophical standpoint, it has proved difficult to encapsulate this gradual change easily within a legislative framework, in a way which gives credence to a diversity of views and does not lead to unintended outcomes.

Within this context, sometimes the attempt to legislate for good (beneficence), while well-intended, can result in adverse consequences which might cause greater harm (breaking the principle of non-maleficence) and which outweigh the intended benefit. For example, making the consumption of alcohol in pregnancy a criminal offence. This approach has been tried previously [5, 6]. Due to risk of criminal penalty, the likely result is that those who continued to drink alcohol in pregnancy, for whatever reason, would become very unwilling to admit it. This might improve the apparent statistics recorded for alcohol consumption in pregnancy, but at the potential cost of hiding the true picture, and also reducing the likelihood of people seeking help for this problem during pregnancy, along with their expectation of being treated with sympathy and understanding as well as being offered appropriate help and support.

The issue of balance between the rights of the fetus versus the rights of the mother has been most widely discussed in society with respect to changes in laws concerning abortion, and this topic is highly relevant to take as a starting point when considering the same balance of rights with respect to alcohol consumption in pregnancy. Abortion was illegal in the United Kingdom prior to 1968 and prior to 2019 in Northern Ireland (Poland and Malta are the only European countries which have maintained highly restrictive laws prohibiting abortion.) Aside from religious justification, prevention of abortion might be considered to have the beneficial outcome of reducing loss of life (by reducing the number of foetuses where pregnancy would otherwise be terminated); however, the unintended consequence is invariably the loss of women's lives and the potential long-term health impacts on women who felt

driven to induce or seek abortions—for whatever reason, because statutory or voluntary services are not readily available. This very real human problem constituted a major driver for changes in the law.

The potential legal problem of an 'offence' (however that may be encapsulated in law) against the fetus, together with the consequences which would spring from that, does not exist if the developing fetus is not a 'person' and therefore has no 'rights' as far as the law is concerned until the moment of birth—and indeed this is the current situation in the United Kingdom. However, these commonly held views about the increasing rights of the fetus have been recognised in some countries and have influenced abortion legislation, placing limits on the ability to obtain abortion as gestation increases, often with checks and balances which include serious risk to the life or well-being of the fetus or the mother.

Contrasted with the rights of the fetus are the rights of the mother, her own autonomy and her right to make choices. It is only when maternal choices come into potential conflict with what might be considered to be the 'best interests' of the developing fetus that a potential ethical issue arises. In UK case law on this issue, the Court of Appeal upheld that while pregnancy increased the responsibilities of a woman, it did not diminish her entitlement to decide whether or not to accept medical treatment [7]. As in the arguments over abortion—these positions can become highly polarised: a women's 'right to choose' (principle 3: respect for autonomy) versus fetal rights (principles 2 and 4: non-maleficence and justice). This polarisation is often accompanied by considerable passion and sometimes sadly, a lack of respect for alternative views which may be held by others. The legal framework somehow has to balance these seemingly opposing views. It is important to acknowledge though that the legal framework may differ in important ways from one's own moral and ethical standpoint in the attempt to incorporate and hold together a broad and diverse range of opinion. In the United Kingdom this polarisation has also encroached on issues where some take the view that it is an 'infringement of liberty' when mothers are asked about their alcohol consumption at pregnancy booking, and whether that information should subsequently become part of the subsequent child health record [8]. Unfortunately journalistic attention to a 'story', which may be interesting, novel or divisive, doesn't always lead to balance in how these issues are presented, and propagates the continuing perception of conflict over these issues, rather than maturity of understanding with respect to alternative views and an atmosphere where resolution can be pursued. It also overlooks the benefits to the child of important information being available at a later time to aid with diagnosis (mentioned in several later chapters).

Additionally, the likelihood, if alcohol consumption in pregnancy were a criminal offence, is that those who were already under the attention of health and social care organisations for other reasons but known to have an alcohol dependency might be particularly singled-out. This would be in contrast to those with higher income and social status who would be able to continue to drink alcohol (again for whatever reason) but without the same level of supervision and consequences, and thus creating an inequitable and discriminative system. Depending on the legal expectations placed on professionals, this approach could also change how professionals involved in pregnancy were perceived and probably make an open and honest relationship more difficult.

The difficulty intrinsic to this approach to this problem: preventing the fetus being exposed to alcohol, potentially leads to a punitive approach towards the mother. Many professionals who work in this area will know that there are many reasons why mothers continue to drink in pregnancy (e.g. as a coping mechanism, helping with symptoms of anxiety, stress, relationship conflict, mental health problems), and understand that offering the right help and support is crucial to helping mothers to change. It is probably much more helpful to take a supportive approach towards the mother who may have many complex long-standing background problems which led to this issue in the first place. However, a society that takes a punitive approach and legislates against this problem is unlikely to provide effective services to help and support those who most need it - another resulting unintended consequence.

3.5 Societal Responsibilities

It is also necessary to set these issues in the context of what it means to live in a 'free society' where we have relative freedom of choice for our actions. There are certainly many benefits as well as responsibilities which come with our freedom of choice, but due to the fact that the majority of the human race lives together with others, most societies determine that 'freedom' has limitations—particularly where particular actions of individuals would be injurious to others. It could be considered that one 'cost' of this freedom is also perhaps an acknowledgement that a small minority will choose approaches which differ from a majority view. The exact balance of these freedoms and their limitations are for individual societies to determine and will vary over time as well as between societies (authoritarian compared with libertarian approach). Thus, in the United Kingdom, it is perhaps right that we have—and should continue to have—a framework where it is not illegal to drink alcohol during pregnancy, even though this is undesirable for the fetus, but this brings in the requirement for additional actions.

There are also responsibilities for society itself as a whole towards the individual: and here it might be considered that there are moral and ethical responsibilities on individuals in positions of responsibility and power, as well as organisations to make sure that there is a societal context present which enables and supports the individual to make the best choices for themselves. In this respect, the different organisations which exist at different levels: governmental, professional organisations as well as local service provision have responsibility to provide information, recommendations, as well as services to support and help the individual make their autonomous decisions, and perhaps, on occasions to help guide or 'nudge' individuals towards some choices rather than others [9]. In the wider context, and for the future this might begin with how we approach the education of our children—a point first made to the House of Commons in 1834 on this very subject? [10]. The acceptance of the implications of societal responsibility should lead to a re-evaluation of the pervasive and unhelpful negative attitudes about moral responsibility and blaming of pregnant women for their actions [11]—particularly in the context of lack of effort placed into education and public health messages.

3.6 Public and Cultural Attitudes and Public Health

Public attitudes and responses to drinking alcohol in pregnancy also play an important part in shaping the prevalence of alcohol consumption in pregnancy. With smoking, it took many years to effect large-scale public changes in attitudes. This began with the emergence of evidence about the adverse health effects of smoking but was several decades before a ban on smoking in public places was instituted in Scotland and then England. Subsequently smoking was banned when travelling in cars with young children (who were unable to make a choice). In between these times were campaigns against commercialism, arguments about individual rights, the role of sponsorship, advertising and an emergence of the dangers of 'second-hand smoke' on others who chose not to smoke. Tobacco company-funded research introduced bias into published scientific evidence with selective publication and in some case evidence suppression [12]. One hopes that we will be wary of falling into similar traps with respect to alcohol.

We do need to know and understand more about how to help inform the public and develop effective targeted health messages. It is likely that long-term changes in public attitudes and perception can be achieved to benefit families and reduce fetal alcohol exposure although so far there have been relatively few attempts from a research perspective to explore the best ways of doing this [13].

People who are viewed as have 'self-inflicted' illness are sometimes seen as less deserving of healthcare interventions, as they can be viewed as have been the architect of their own misfortunes ever since the Victorian Poor Laws and the concept of the 'deserving' and 'undeserving' poor. They can therefore be subject to discrimination at individual as well as at commissioning level [14]. However if we are better able to help those who require help to reduce and stop their alcohol use where this is harmful (either to themselves or because of pregnancy), then we need to hear more about the stories and reasons as well as backgrounds leading to these situations so that there can be understanding as well as more effective and earlier societal and health-related interventions.

3.7 Conclusion

Despite the apparent disagreements and different approaches, coupled with confusion over the evidence, there are several important conclusions and principles which can be drawn in our desire to improve the situation for the future, assuming that the underlying aim is to reduce the number of children growing up with FAS/FASD (Fetal Alcohol Syndrome/Fetal Alcohol Spectrum Disorders) who have been affected by exposure to alcohol during pregnancy.

There are a number of reasons why the process of change seems to progress so slowly (it took several decades with tobacco). It is likely that the persistence of conflict between those who approach this issue from different paradigms is an important factor. Debates about the rights of the mother versus the rights of the fetus are also important in continuing the appearance of unresolvable division through

polarisation of the debate. However, when moving forward, it is clear that those who (quite rightly) jealously guard the advances which have been made in maternal rights over the last few decades are reassured that progress in this area is not about framing guilt or victimising women. There are many aspects of lifestyle and attention to health which are worth addressing when considering pregnancy, or at the start of a pregnancy, and alcohol consumption is only one amongst many things which should be discussed.

There is of course the need to enquire, because this opens up the conversation, to provide appropriate information if necessary and make sure that where required the right type of help and support can be given to encourage and enable mothers to change in positive ways. The concerns raised about confidentiality and maternal information could, at least partly be addressed by anonymisation of maternal details where consent for transfer of information was not obtained, for whatever reason. This would help provide the important background information and context to feed into future diagnostic considerations if there are difficulties in child development at a later stage.

The importance of public health justifies the necessity to collect statistics regarding health to influence policy, health provision or consider interventions. Alcohol consumption at local, regional or national level is only one of a number of different health metrics when considering the health of populations and the ability to pick up adverse trends (e.g. the increasing incidence of liver failure in the female population) [15]. The opportunities for health intervention when individuals present to health services can be important in helping to inform mothers about their own longer-term risk and a chance to look at improving their own health outside of pregnancy when potentially harmful levels of drinking are involved. Unless we collect (unattributed) population data, we are unable to track changes and improvements over time, particularly in response to targeted campaigns.

From the newborn infant's perspective, unless we ask women about their alcohol consumption in pregnancy, we are unable to offer appropriate advice along with help and further support to encourage and help mothers reduce or stop their alcohol consumption at any point in pregnancy. That intervention can improve the outcome for that particular baby; however, there is also the possibility of secondary prevention of the same problem recurring in subsequent pregnancies.

Of course, there is an issue—and an accompanying responsibility—for us as individuals within our society, and there is clearly much more to do to improve how we approach this problem in pregnancy.

We all have different parts to play in this process in terms of highlighting issues, helping improve care and encouraging change. Despite the commonly repeated phrase that 'FASD is preventable', as long as alcohol is around in our society, children will continue to be affected by alcohol exposure during pregnancy. It remains incumbent on us to continue to explore the best ways of helping to reduce that adverse outcome (Box 3.2).

Box 3.2 This Example Demonstrates How Information from a Research Study About Alcohol in Pregnancy Can Be Misrepresented, and Misinterpreted

An article was printed on the front page of *The Times* date: 12/9/2017 about a meta-analysis published in the journal BMJ Open [16].

The headline was: '***Light drinking in pregnancy safe***', however although the title was a misrepresentation of the research, the actual article reported the research study fairly accurately.

The title of the research study was: '***Low alcohol consumption and pregnancy and childhood outcomes: time to change guidelines indicating apparently 'safe' levels of alcohol during pregnancy? A systematic review and meta-analysis***.'

The conclusion of the authors of the study was: Evidence of the effects of drinking ≤32 g/week in pregnancy is sparse. As there was some evidence that even light prenatal alcohol consumption is associated with IUGR (Intra-Uterine Growth Restriction) and preterm delivery, guidance could advise abstention as a precautionary principle but should explain the paucity of evidence.

I wrote to *The Times*: in response they changed the headline in the online version later that day and the letter was published the following day in the printed version, but of course, the effect of a small letter buried deep in the paper does not counteract a large headline on the front page.

Following correspondence with the Science correspondent of the paper, the Professor quoted in the article and the author of the research paper in question it became clear that:

The inappropriate headline which confused lack of evidence of an effect with lack of evidence about an effect was written by the copy editors, and not by the science correspondent who wrote the actual article. The Professor quoted in the article felt that he had been misquoted.

The author of the paper experienced considerable uncomfortable publicity due to her paper being misquoted as this was not the message that the findings of the research represented.

References

1. Schardt C, Adams MB, Owens T, et al. Utilization of the PICO framework to improve searching PubMed for clinical questions. BMC Med Inform Decis Mak. 2007;7:16. https://doi.org/10.1186/1472-6947-7-16.
2. Layte R, McCrory C. Growing up in Ireland: maternal health behaviours and child growth in infancy. Dublin: Department of Children and Youth Affairs, Stationary Office; 2014. ISBN 978-1-4064-2839-1 https://www.drugsandalcohol.ie/23285/1/Infant_Maternal_Health_4_web.pdf. Accessed 8 Dec 2020.

3. Day S, Rennie S, Luo D, et al. Open to the public: paywalls and the public rationale for open access medical research publishing. Res Involv Engag. 2020;6:8. https://doi.org/10.1186/s40900-020-0182-y.
4. Gillon R. Defending the four principles approach as a good basis for good medical practice and therefore for good medical ethics. J Med Ethics. 2015;41:111–6. https://jme.bmj.com/content/41/1/111.info.
5. Criminality and drinking in pregnancy. https://www.amnesty.org/en/documents/amr51/6203/2017/en/. Accessed 24 Sep 2020.
6. Söderström K, Skolbekken J-A. Pregnancy and substance use – the Norwegian z 10–3 solution.1 Ethical and clinical reflections related to incarceration of pregnant women to protect the foetus from harmful substances. Nordic Stud Alcohol Drugs. 2012;29(2):155–71. https://doi.org/10.2478/v10199-012-0011-1.
7. St. George's Healthcare NHS Trust v. S 1998 3 All ER 673, (1998) 2 CHRLD 323.
8. Williams Z. Bilge, booze and misogeny: why I'm outraged by a new idea to police pregnant women. Guardian. 2020. www.theguardian.com/lifeandstyle/2020/sep/18/bilge-booze-misogyny-outrage-police-pregnant-women. Accessed 21 Sep 2020.
9. Thaler R, Sunstein C. Nudge: improving decisions about health, wealth, and happiness. New Heaven, CT: Yale University Press; 2008. ISBN 978-0-14-311526-7.
10. Committee on drunkenness: House of Commons Select Committee Report. Hansard. 1834; 25:cc959-960. https://api.parliament.uk/historic-hansard/commons/1834/aug/05/committee-on-drunkenness.
11. Abadir AM, Ickowicz A. Fetal alcohol spectrum disorder: reconsidering blame. CMAJ. 2016;188(3):171–2. https://doi.org/10.1503/cmaj.151425.
12. Fiona G, Ruth M, Adam T, Catherine O, Andy B, Ian P, et al. Journal policy on research funded by the tobacco industry. BMJ. 2013;347:f5193.
13. Dumas A, Toutain S, Hill C, et al. Warning about drinking during pregnancy: lessons from the French experience. Reprod Health. 2018;15:20. https://doi.org/10.1186/s12978-018-0467-x.
14. Sharkey K, Gillam L. Should patients with self-inflicted illness receive lower priority in access to healthcare resources? Mapping out the debate. J Med Ethics. 2010;36:661–5.
15. Simpson RF, et al. Alcohol drinking patterns and liver cirrhosis risk: analysis of the prospective UK Million Women Study. Lancet Public Health. 2019;4(1):e41–8.
16. Mamluk L, Edwards HB, Savović J, et al. Low alcohol consumption and pregnancy and childhood outcomes: time to change guidelines indicating apparently 'safe' levels of alcohol during pregnancy? A systematic review and meta-analyses. BMJ Open. 2017;7:e015410. https://doi.org/10.1136/bmjopen-2016-015410.

How Does Alcohol Affect the Developing Fetus?

4

Neil Aiton

Chapter Highlights

- The complex ways in which prenatal alcohol exposure affects the developing fetus and in particular the fetal nervous system.
- How alcohol consumption in pregnancy can affect the fetus indirectly through effects on the mother and the pregnancy.
- How understanding of fetal development together with timing of exposure can inform the conversation with pregnant women and provide a framework for understanding future neurodevelopmental impairment.

Alcohol is described as a teratogen. A teratogen can be described as 'any agent that can cause a malformation in a fetus'. The science of teratology attempts to understand how something might intervene during fetal development to cause an abnormality. The complex nature and sequence of developmental processes means that there are multifarious mechanisms through which an exogenous agent such as alcohol might cause a problem during fetal development. In addition, it is important to consider the concept that the timing of exposure relative to the stage of development can be critical in this process: meaning that exposure at a particular point in fetal development might lead to a problem but not at other times [1].

When considering how alcohol might affect a developing fetus, it is common just to focus on the teratogenic effects of alcohol directly on the fetus. However, there are also important indirect effects. Although many of these actors are interrelated, it can be helpful to break them down into the following areas:

- **Mother**—Effects on the mother which can affect fetal growth and development: including effects on metabolism, diet, behaviour, relationships and

N. Aiton (✉)
The One Stop Clinic, Royal Sussex County Hospital, Brighton, UK
e-mail: neil.aiton@nhs.net

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_4

relationship conflict. Additional factors include: presence of any alcohol dependence and impacts on the mother due to long-term alcohol use. Maternal fetal alcohol exposure and early maternal childhood experiences can also have an influence.

- **Pregnancy**—Changes caused to the biology of pregnancy caused by alcohol: the maintenance of pregnancy itself, the hormonal environment associated with pregnancy, effects on the whole chorioamniotic/fetal unit, the placenta and placental function. Additional non-specific effects include an increased likelihood of low birth weight and prematurity.
- **Fetus**—Direct effects of alcohol on the developing fetus as it changes from a fertilised egg into the infant at term ready for postnatal life (teratogenicity).
- **Newborn**—Exposure in pregnancy which leads to effects on the infant in the early newborn period: intoxication, withdrawal, behaviour and neurology, including modification of maternal–infant interaction.

Preconceptual and Intergenerational Influences

Taking the timeline back much further, prior to pregnancy itself is the increasing evidence of the potential for trans-generational effects: due mainly to the role of epigenetic changes on parental gametes (ovarian follicles in the female and testicular germ cells in the male):

- **Paternal**: Through chronic effects of alcohol exposure on the primitive germ cells in the testes which give rise to spermatogenesis and also direct effects on spermatozoa during the process of spermatogenesis [2] (the formation of sperm)—a process which takes around 74 days. [So, fathers who might wish to optimise the preconceptual period without alcohol should allow a minimum 3-month period.]
- **Maternal**: The process of gamete formation is different from males and the oocytes (which later in life can turn into follicles and be released as eggs from the ovary during the process of ovulation) are already present in the fetal ovary by the beginning of the second trimester during fetal development. It is thought that the reserve of ovarian follicles then diminishes during the lifespan [3]. Thus, it is potentially possible, through epigenetic effects on these nascent oocytes, for factors which impact on the mother during her **fetal** life to influence the outcome of her children when she eventually conceives and gives birth in adulthood. Indeed, there are some early research findings that support this concept [4] with respect to maternal nutrition [5] alcohol exposure [6], and adverse childhood experiences (ACEs) [7] although the same effect has not been demonstrated in population-based cohort studies at the present time [8].

Although detailed discussion of some of these aspects is beyond the scope of this chapter, it is important to mention them for completeness, to emphasise the broad scope of mechanisms through which alcohol can have an influence on developing fetus besides direct teratological effects of exposure during pregnancy and because many have potentially far-reaching effects (Box 4.1).

Box 4.1 Epigenetics: Explanation and Further Information

The study of how genes are controlled and what factors influence gene regulation: turning genes 'off/on' (up- and down-regulation). This determines what effect an individual gene, or groups of genes will have (gene expression) in the cell, the organ or the body as a whole, depending on their function.

Factors related to the environment, our behaviour as well as diet can cause epigenetic changes.

Epigenetic changes are potentially reversible and do not affect the underlying DNA sequence but rather how this 'read' and activated.

For further reading see:

- www.whatisepigenetics.com/what-is-epigenetics/
- www.cdc.gov/genomics/disease/epigenetics.htm
- Epigenetics studies of fetal alcohol spectrum disorder: where are we now? Lussier AA, Weinberg J, Kobor MS. Epigenomics 2017; 9(3): 291–311. www.futuremedicine.com/doi/pdf/10.2217/epi-2016-0163

Much of the research into future outcomes associated with alcohol consumption in pregnancy is beset with difficulties of trying to account for confounding factors which themselves can also act as important additional determinants of fetal outcome. Trying to control for these confounding factors is often problematic and these issues are likely to have led to some difficulties in interpretation when relating fetal experience to later outcomes. Much useful data has been gleaned from long-term prospective cohort studies, where, although the recruitment and data collection are prospective, the research question and analysis are often retrospective [9] as opposed to prospective studies where these aspects can be designed into the study and controlled for in the best way.

The commonly perceived issues with studies which depend on recording and quantifying alcohol consumption include recall bias, minimisation of actual consumption, under-recording, difficulties in the methodology used. These explain some of the difficulties in trying to understand the relationship between prenatal alcohol exposure and future outcome. The following may also be contributory factors:

- Variability of exposure with respect to timing, amount and duration of gestation between individuals and between studies
- Variability in the definition and methodology of recording, quantification, description and categorisation of exposure
- Significant genetic variation in hepatic alcohol metabolism
- Induction of hepatic enzymes with regular alcohol consumption leading to more rapid metabolism (and increased tolerance) reducing exposure duration

- The variation in individual dietary intake associated with alcohol exposure
- The association with social factors which are known to affect outcome
- The effects on behaviour and behavioural interaction in early childhood
- The long-time interval between exposure and outcome
- The use of biomarkers as a proxy for future outcomes (e.g. findings on magnetic resonance imaging (MRI) scans)

The use of animal models means that when comparing outcomes between experimental groups, these additional factors can be controlled for more carefully in comparison to the element under investigation [10] but brings an additional problem of cross-species interpretation. When associations between a particular outcome and exposure is observed, it is important not to assume that this is causative, and to design further studies to test hypotheses generated in this way. Bradford-Hill's criteria and the GRADE framework can be helpful when attempting to establish an argument for causation of effects on the fetus [11] (Strength of association, consistency, temporality, biological gradient, specificity, biological plausibility, coherence, experimental evidence and reasoning by analogy). However, even this approach can still remain particularly difficult in the context of complex multiple associated confounding factors.

4.1 Effects on the Mother Which Indirectly Affect the Fetus

Moderate or high levels of alcohol consumption (in the UK, >14 units/week in the non-pregnant women) on a regular basis is associated with significant risk of a host of adverse health outcomes (e.g. various cancers, alcoholic liver disease, pancreatitis, cardiovascular disease, neurological degenerative disease). However, many of these do not affect women who are still young enough to bear children, and it is usually the effects of alcohol on diet, mental health, behaviour and relationships which can have the greatest impact on the developing fetus.

Chronic high level alcohol use comes with a high risk of impairment of liver function, cirrhosis, and eventual liver failure. Some mothers who conceive around the age of 40 may have been drinking heavily for up to 25 years. Additional coexistent chronic infection by hepatitis C can exacerbate impairment of liver function which will lead to slower alcohol elimination and prolonged fetal exposure, increasing the risk to the fetus particularly in those who have a long history of heavy alcohol use. With hepatitis C infection, there is the risk of higher maternal viral loads (measured on polymerase chain reaction (PCR) testing) thereby increasing the risk of maternal–fetal transmission of infection. Changes in body composition due to dietary differences and liver impairment can affect how alcohol distributes and equalises within body compartments after consumption further affecting distribution and the rate of metabolism and elimination.

A number of different studies have shown a relationship between level of alcohol intake, maternal diet and nutritional intake, with a reduction in healthy choices and

increase in processed food types thereby increasing the risk of malnutrition and dietary deficiencies [12]. The evidence from animal studies which suggested that prenatal dietary supplementation might help to protect against adverse effects of prenatal alcohol exposure was attractive and encouraging, particularly in view of the association with increased risk of free radical mediated oxidative damage. Potential early favourites were identified, for example, vitamins C [13], E [14] and choline [15]. Although initial results were disappointing [16], a further randomised controlled trial has shown some evidence of benefit [17]. One explanation may be that dietary supplementation is important for some subgroups, but that any detectable experimental difference is subsumed within the context of larger studies investigating effects on populations.

Alcohol also affects our behaviour, and there has been increasing evidence of the relationship between higher levels of alcohol consumption and intimate partner physical or sexual violence [18]. Evidence from the ACE study has shown the enduring effects of abuse and related adverse experiences in childhood into adult life [19]. It is also likely that 'difficult pregnancies' where there are frequent episodes of violence, conflict and stress result in changes in the fetal nervous system [20, 21]. There has also been a mushrooming of understanding over the last few decades regarding our understanding about post-natal attachment. However, there has been relatively little research looking into how the attachment process might be different in the context of potential disturbance of neurological function/disability (e.g. that which might be seen in children with fetal alcohol syndrome/fetal alcohol spectrum disorder [FAS/FASD]). Likewise, a greater understanding of the process of pre-natal attachment is needed [22], along with an understanding of how any interventions might improve outcomes. Future attention to pre-natal aspects offers the exciting potential of intervening to reduce or even prevent intergenerational propagation of adverse effects.

4.2 Effects of Alcohol Consumption on Pregnancy (Box 4.2)

One of the most important early factors relating to alcohol consumption on pregnancy is the significant increase in the likelihood of first trimester miscarriage, even at levels of alcohol consumption which would be considered relatively low level in

Box 4.2 The Risks of Alcohol on Pregnancy
– 5× risk of early miscarriage
– 3× risk of premature delivery
– increased risk of: placental dysfunction, hypertension, low birth weight

the non-pregnant population. In a large Danish population cohort study of over 40,000 deliveries, there was a background level of 1.4% for spontaneous miscarriage in mothers drinking <1 drink/week rising to over 5 times that figure: 8.8% in those drinking >5 drinks/week [23].

The effects of alcohol consumption on maternal and fetal hormones have not been extensively studied, but it is clear that there are effects even though the implication is uncertain [24] (Some of these may be related to factors like prematurity and low birth weight—see below).

Alcohol use is associated with placental dysfunction [25] causing decreased placental size, impaired blood flow and nutrient transport, umbilical constriction and risk of abruption, and more research is required to uncover the biology which lie behind these changes.

Most of the effects of alcohol consumption on a pregnancy itself (as opposed to maternal or fetal factors) are due to the non-specific increases in the risks of adverse pregnancy outcomes. There is three times the risk of premature delivery compared with those who do not drink within the same cohort [26]. Prematurity and low birth weight are then acting indirectly as additional factors contributing to future risk of neurodevelopmental problems. There is also an independent increased risk of low birth weight [27]. Alcohol consumption can also increase the likelihood of developing hypertension during pregnancy [28] which will independently affect placental circulation and increase risks of poor fetal growth.

4.3 Direct Effects of Alcohol on the Developing Fetus

There are numerous studies which demonstrate effects of prenatal alcohol exposure on many organs of the body. These effects can be subtle as well as pervasive, for example, affecting growth, facial features, immunity, the cardiovascular system, and have been explored in more detail using animal models [29]. However, it is the effects on the developing fetal nervous system which are most life-impacting in determining future outcome and hence are central to the diagnosis of FAS/FASD [30].

The wider dysmorphology related to prenatal alcohol exposure is discussed elsewhere (Chap. 10); however, the particular constellation of facial features which are affected—the triad of small palpebral fissures, smooth philtrum and thin upper lip, remains fairly specific to fetal alcohol exposure. Photography with computerised analysis has attempted to measure these aspects more objectively [31]. More recently, complex 3D photographic analysis may provide additional sensitivity as well highlighting other more subtle features which have not as yet been incorporated as part of the assessment such as mid-facial recession and mandibular hypoplasia [32]. There have been attempts to look at fetal facial features using ultrasound, although the place of this type of assessment is not clear. All fetal ultrasound datasets so far available are insufficiently controlled for fetala alochol exposure. Many children who are eventually found to have FASD do not exhibit the classical facial features, so although the presence of facial features can contribute significantly to diagnosis—particularly when there is no antenatal history available, absence of features does not exclude prenatal alcohol exposure or FASD. It is likely that timing of the window of exposure is critical: in a study on Macaque monkeys this was during Days 19–20 (total gestation 183 days) [33]. It is not clear exactly how this translates to human gestation, but rapid development of the head and facial maxillary prominences begins during the

5th week of fetal life (Week 7 of pregnancy) and continues over the next 2–3 weeks. The sonic hedgehog gene and signalling pathway are fundamental to orchestrating normal craniofacial development and are known to be affected by alcohol [34]. Therefore, a pattern of intermittent alcohol exposure (binge-drinking) could easily miss this relatively small critical window (Box 4.3).

Box 4.3 For Further Information on Embryology

Fetal development:

- The Developing Human. Clinically Orientated Embryology. Moore K, Persaud T, Torchia M. Saunders Elsevier Philadelphia 2019.
- Larsen's Human Embryology. Schoenwolf G, Bleyl S, Brauer P, Francis-West P. Churchill Livingstone Elsevier. 2014.

Abnormal embryogenesis of the brain with alcohol:

- Chapter 3 in Alcohol, Drugs and Medication in Pregnancy. Ed. Preece M, Riley E. 2011 MacKeith Press, London.

There are a number of different mechanisms which contribute to nervous system development during fetal life as the primitive nervous system evolves. Different genes become activated at different times creating complex intracellular signalling pathways which interact to cause cell proliferation in some areas combined with programmed cell death (apoptosis) in other areas. This is combined with the effect of cellular migration to create greater structural complexity. Neuronal migration is most clearly seen in the second and third trimesters where the fetal brain changes from having a smooth surface at 23–24 weeks gestation to a pattern of complex gyri. This process creates the multi-layered neuronal structure underlying the surface of the cerebral cortex, and the complex folding pattern has the function of increasing the surface are of the cortex (and is much less prominent in less advanced mammals). Both neuronal migration and gyral formation have been demonstrated to be abnormal in children with FAS/FASD [35, 36].

Simultaneously, the cells in the nervous system become more highly specialised, differentiating into supporting cells (glial cells) and nerve cells (neurones) of several different types. These nerve cells then develop numerous interconnections which begin to establish the 'wiring' of the brain pathways. This process also requires cell growth—outgrowths of the nerve cells towards each other to connect (dendrites) and make new connections alongside the continued process of 'pruning' of connections and nerve cells through apoptosis (programmed cell death), as the pathways refine themselves.

At the most simplistic level, fetal alcohol exposure can disrupt these processes through the following mechanisms [29, 37]:

- Alteration of normal pattern and level of gene activation through epigenetic effects—causing changes in embryological signalling pathways for nerve cells

- Reducing energy availability at cellular level through mitochondrial disruption (affecting cellular energy production)—reducing neuronal and glial cell growth and affecting nerve cell impulse activity—both of which are metabolically costly and could be affected by cellular energy availability [38]
- Increase in free radicles (ROS) and direct oxidative damage [39] to enzymes and cellular structures and processes
- Direct toxicity of alcohol and its metabolites (especially acetaldehyde) to enzymes and regulatory signalling molecules
- Increase in cell apoptosis seen following alcohol exposure
- Alteration of neurotransmitter levels repeatedly affecting the balances between inhibitory and excitatory pathways

The effects of alcohol on the mature adult central nervous system are mediated by actions on a variety of neurotransmitters, with a complex interplay between excitatory and inhibitory systems which varies with the concentration of alcohol and behaviour state. In particular, alcohol has been shown to affect the following neurotransmitters: dopamine, noradrenaline, endogenous opioids, GABA (gamma amino-butyric acid), glutamate and serotonin. In the developing fetal nervous system, it is likely that regular exposure to alcohol affects the development of the normal neural pathways as well as the balances between the inhibitory and excitatory neurotransmitters which are critical for normal neurological functioning, processing and behavioural responses. It is beyond the scope of this chapter to consider this in significant detail. However, a growing body of evidence has begun to highlight the widespread subtle structural and functional abnormalities present throughout different areas of the brain which result from prenatal alcohol exposure. Areas such as the cerebellum, the corpus callosum, the basal ganglia, hippocampus and also the cerebral cortex (particularly frontal lobes) can be particularly affected. These changes of course relate to the subsequent neurological and behavioural problems which can be identified in children with prenatal alcohol exposure. Of note, however is that although these differences have been described in the context of high levels of chronic exposure, conventional neurological imaging does not always easily distinguish differences in the brains of infants with milder levels of prenatal exposure (who may still develop later behavioural and neurological abnormalities) from those who are unexposed—even with functional pathway imaging [40]. One can hypothesise that differences which we are unable to detect with current technology are likely to relate to more subtle differences in the neural ‘circuitry’, that is the particular pathways and nerve connections which developed in different ways due to alcohol exposure. (For example, in children with FAS, one can observe difficulties in incorporating and translating learning into behaviour change, or difficulty laying down memory.)

The rapid cell growth and development of the fetus is dependent on both a good energy supply source and the ability to use that energy efficiently. Alcohol also interferes with energy metabolism and mitochondrial function in ways which are not yet fully understood. Mitochondrial function is essential for normal oxidative energy metabolism to provide the energy to support cellular function through the production of adenosine triphosphate (ATP). Mitochondria synthesise the majority of the ATP on

their cell membranes through the combination of a number of different closely related enzymes utilising the transfer of electrons—a process called the 'electron transport chain'. Mitochondria also produce reactive oxygen species (ROS) as a by-product of this process of oxidative phosphorylation and cellular energy production. These ROS are highly reactive and have the potential to damage intracellular proteins and structures. Therefore, there are cellular mechanisms to keep the ROS 'under check' and prevent mitochondrial and wider cellular damage, and this is the role of 'antioxidants' which include certain enzymes (e.g. superoxide dismutase) and the involvement of various co-factors (e.g. phenols, carotene) and vitamins (e.g. Vitamins C and E).

There is good evidence that chronic alcohol consumption increases oxidative stress [39] and that prolonged and frequent increases in ROS can contribute to ongoing cell damage [41]. Metabolism of ethanol through alcohol dehydrogenase to acetaldehyde, and the generation of NADH and the oxidation of acetaldehyde depend on the mitochondrial electron transport chain. Thus, alcohol metabolism creates competition for the mitochondrial mechanisms which is likely to create substantial acute metabolic challenges to the cell, reducing the energy available for normal processes. There is also an increase in free radical formation leading to enzyme and cellular protein damage and hence further impairment of energy production and possible cell death. All of these may contribute directly to impairment of function as well as neuronal damage.

Many children who have been subject to significant prenatal alcohol exposure who demonstrated evidence of intrauterine growth restriction continue to demonstrate poor growth for a prolonged period even though the alcohol exposure is no longer present and in the presence of an adequate nutritional intake. More research is needed into the biological mechanisms underpinning this observation. One possible mechanism is that epigenetic changes affect the expression, and hence activity, of the mitochondrial respiratory chain enzymes restricting the energy utilisation for a prolonged period along the lines of the Barker Hypothesis.

With respect to neurological function, it is the development of interconnections and pathways between different areas of the brain which allow us to develop complex levels of functioning as our nervous systems develop. Many of these pathways need to be able to interconnect and send messages back and forward very rapidly in response to multilateral input and processing. Hence these interconnecting pathways—which are early receivers of myelination during fetal life—are a process which helps to speed up neural conduction. It is the presence of this fatty myelin 'insulation' which gives rise to the term 'white matter'. A good example of one of these white matter tracts or pathways is the corpus callosum which connects a wide variety of brain regions between the two hemispheres. There is a large energy cost to maintaining high-speed nervous conduction because of the need to power cell membrane repolarisation and ion transfer required for nerve impulse transmission. If chronic alcohol exposure impairs mitochondrial function and energy production, it is reasonable to assume that these regions which are likely to be the most metabolically active would be more severely affected. Indeed abnormalities of the corpus callosum have been observed in association with fetal alcohol exposure, and the corpus callosum is smaller and a different shape in adults with FASD [42] as well as in infants (Box 4.4) [43].

Box 4.4 Pregnancy Dating and The Difference Between Fetal Age and Gestational Age

Embryological descriptions usually relate to the age since fertilisation. Pregnancy is usually dated from the first day of the last menstrual period (LMP) [additional correction may need to be made if the menstrual cycle is longer than normal].

Early dating ultrasound scans prior to 14 weeks have an accuracy of ±5–7 days.

Fetal age is always 2 weeks less than gestational age.

4.3.1 Fetal Brain Development and the Timing of Exposure

One of the most important questions to which a mother might wish to know the answer is 'What is chance of my baby having a problem in the future?'

There are a few important principles to remember when considering the answer to that question. If this conversation is taking place after delivery, then one can take a history, make a full assessment (see Chap. 7) according to recommended guidelines [30] and arrive either at a diagnosis or some broad estimate of possible future risk. If the conversation takes place during pregnancy, then there are some additional aims: It would be important to offer the mother help and support to stop drinking at this point, if she has not already done so, try to educate regarding the risks of continued alcohol use (which may provide motivation) and avoid misunderstandings such as 'it's too late' and 'pointless to stop'. The pregnancy calculator can be very helpful when drilling down into the detail of particular weekends or events which might be associated with high levels of potential exposure and occasionally can be useful to dismiss needless worry.

4.3.2 Clinical, Embryological and Exposure Correlation

Without going into detailed embryological explanations which can be found in embryology texts (and subject to possible criticism of oversimplicity) the following

gestational periods can be considered as important 'milestones' of brain development when considering any potential impact relating to exposure (Table 4.1):

Broadly speaking, although development is continuous and interconnected, once progression has taken place beyond the primitive streak and the notochord, the central nervous system development can be considered in three stages. The earliest part of fetal nervous system development (4th–8th week of pregnancy) which involves the development of the spinal cord and the outgrowth of the more 'primitive' central structures of the brain: the hind-brain, mid-brain and fore-brain. The cerebellum begins to form towards the end of this period. (The cerebellum is important in the coordination of movement.)

At 8–9 weeks of pregnancy (6–7 weeks fetal age) towards the end of this first 'stage' of development, there is evidence of the early presence of the thalamus, hypothalamus and the hippocampus, together with the beginning of the amygdaloid nucleus [44]. These structures form part of the limbic system which regulates emotions, behavioural responses and association of memory.

This next 'stage' (from 9 to 21 weeks of pregnancy) is the period when the cerebral hemispheres form and grow. It is also the time of neuronal differentiation, (especially in the subcortical grey matter) and the development of the interconnecting pathways between different areas within the brain (referred to as white matter as these pathways contain high levels of the insulating myelin which help to speed up conduction). These neural 'highways' (e.g. the corpus callosum) help to interconnect and pass messages rapidly between different parts of the brain to help coordinate function.

The final 'stage' is one of maturation, growth and increasing complexities, particularly of the cerebral hemispheres and interconnecting pathways. The outer layers of the cerebral cortex are where most of our important cognitive processes take place, as well as developing particular specialised areas for sensory information and movements.

In terms of risks relating to periods of alcohol exposure, we can be reasonably confident that there is a negligible risk of developmental problems prior to

Table 4.1 Summary of important points in fetal neurological development

	Fetal age since fertilisation	Pregnancy gestation (weeks)
Conception	Day 0	2
Implantation into the uterine (blastocyst)	Days 6–10	3+
Primitive central brain and spinal cord	Day 28	6
Thalamus, hypothalamus and hippocampus formed	6–7 weeks	8–9
Start of formation of cerebral hemispheres		
Primitive cerebral hemispheres now formed and start to increase in size	Days 49 (7 weeks)	9
Corpus callosum starts to form	10 weeks	12
Corpus callosum complete, smooth cerebral hemispheres	20 weeks	22
Increasing gyral formation and cortical complexity	Weeks 21 to term	Weeks 23 to term

implantation which occurs between Weeks 3 and 4 following the date of the last menstrual period.

Between Weeks 4 and 8, the period of 'primitive brain' development, the risk of subsequent problems, is low, but bearing in mind the central brain structures present and the early formation of some of the structures related to the limbic system, any potential future problems might be most likely to be focussed on emotional regulation and arousal, and possible memory involvement. The cerebral hemispheres are not yet developed, so significant cognitive processing deficits are unlikely.

The period around Weeks 7–9 is likely to be the most sensitive time for facial development.

The most important risk in this early phase during the first trimester is actually that of spontaneous miscarriage which may be as high as 10% (Fig. 4.1).

Exposure in the next stage of nervous system development (pregnancy Weeks 9–21) is more likely to affect neuronal migration, brain growth and the developing interconnecting pathways. So, the biggest effects are likely to be that of difficulties with learning, and difficulties in coordination of thought processes. The limbic system is more highly developed, so could still be impacted by further exposure. During this period, the cerebral hemispheres are beginning to form and grow, so there could be restriction in growth (future microcephaly) and increasing concern about cognitive impacts with increasing gestation.

The final stage (Weeks 23-term)—that of maturation and increasing complexity—means that any ongoing alcohol exposure is likely to raise increasing concern particularly about cognitive impact, in addition to further impacting the areas previously affected.

So, it is important for a mother to be aware that if it is possible to stop any alcohol exposure, even during the third trimester, this can still be important in helping to prevent further long-term impacts, particularly affecting cognitive outcome (Fig. 4.2).

4.4 Effects of Alcohol Exposure in Pregnancy on the Newborn Infant After Birth

Important effects on the newborn infant in the immediate postnatal period which can attributable to the direct effects of alcohol are usually only seen in mothers who continue drinking throughout pregnancy up until the time of delivery or who drink immediately prior to coming into hospital in labour. In these infants there is initially poor feeding and sedation from intoxication in the immediate post-birth period, followed 12–48 h later by irritability and unsettled behaviour together with risk of seizures. There is an additional risk of hypoglycaemia if there has been recent alcohol consumption during labour or preceding few days, with occasional evidence of metabolic acidosis due to interruption to normal metabolic pathways.

Seizures can be treated with the normal first-line treatment such as phenobarbitone which has a long history of use in the neonatal period, works well and often

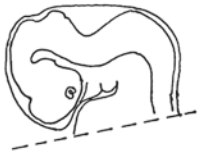
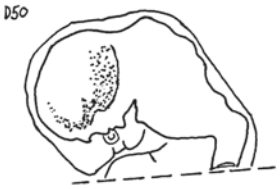
Fetal age (days) Weeks of pregnancy		Cross-section	Explanation
Day 21 Week 5			The notochord (precursor to the spinal cord) present in the neural groove
Day 28 Week 6		 (unfolded)	The notochord has grown to form the primitive hind brain and midbrain, and the midbrain flexure has formed.
Day 35 Week 7			The hind brain and midbrain developing. The Forebrain is now distinct. Towards the end of this period (around week 8) the forebrain begins to divide into two separate hemispheres. The cerebellum is forming.
Day 49 Week 9			The separate cerebral hemispheres are now visible, and continue to grow and enlarge.
Day 77 Week 13			The essential parts of the brain are now recognisable and continue to grow and develop. Nerve fibres begin to cross the midline from the areas at each end (commissures) to form the corpus callosum.

Fig. 4.1 The early development of the fetal nervous system (Fetal age in days since conception, gestational age of pregnancy in weeks) [derived from: K Moore, T Persaud. The Developing Human. Elsevier, Philadelphia]

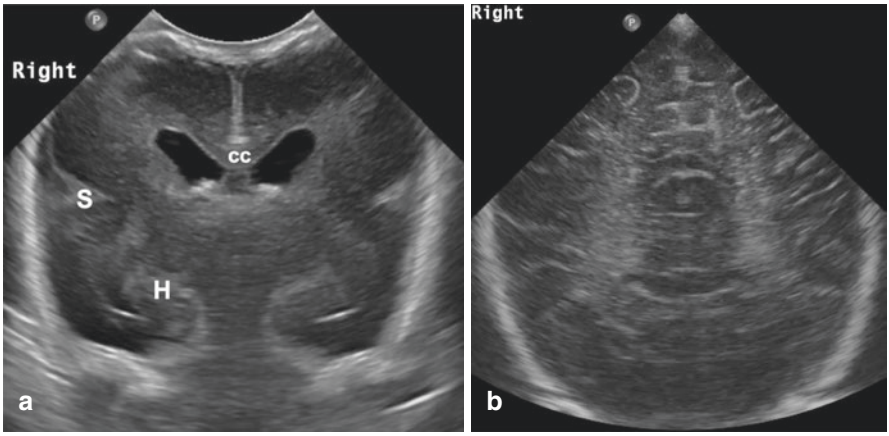


Fig. 4.2 Ultrasound images demonstrating difference in gyral formation between a 23 weeks premature infant and a full-term infant (40 weeks). (**a**) Mid-coronal transfontanelle ultrasound scan of 23 weeks infant showing primitive brain with smooth surface. The corpus callosum [cc] is fully formed. The lateral Sylvian fissures [S] are beginning to form, and the hippocampus [H] is also visible on each side. (The bright areas on each side are the skull bones). (**b**) Posterior coronal section through brain of term infant showing extensive gyral formation in the cerebral hemispheres. (In this example, the image is angled from the fontanelle towards the occiput, and the occipital skull sutures are visible as breaks in the bright skull outline). Some differences in echogenicity are showing evidence of the contrast between grey and white matter

only a single loading dose is required, although alternatives can be considered. Hypoglycaemia, if present should be corrected first.

Intrauterine growth restriction may be present and is an independent risk for hypoglycaemia which should be monitored and treated appropriately according to normal protocols. There is an additional risk of hypoglycaemia if there has been recent alcohol consumption during labour or preceding few days. Hypoglycaemia should be monitored and treated to prevent any further neurological injury. The potential risk of neurological injury in the presence of hypoglycaemia may be particularly high because of the suppression of free fatty acids as an alternative metabolic fuel by the alcohol. Consideration may need to be given to the possibility of using a lower threshold for determining hypoglycaemia in this context compared with the normal thresholds for this reason.

Even if alcohol exposure had ceased some time prior to the birth, some newborn infants with significant prenatal alcohol exposure can still exhibit subtle behavioural signs in the newborn period. These include difficulty in settling, disordered sleep, and dysfunctional sucking and feeding. Sometimes this can require nasogastric tube feeding and input from speech therapists with specialist experience in infant feeding can be helpful. Feeding dysfunction can also manifest itself at the time of weaning.

4.4.1 Protection from Damage in the Fetus (Resilience)

In many biological systems it is also important to consider mechanisms which help to protect us from ongoing damage. In the context of free radicle ROS damage there will be genetic as well as environmental factors which modify our ability to 'mop-up' these free radicles and thereby help to protect against cellular damage. A good term to use to summarise that concept is that of biological resilience. Genetic factors might affect the 'efficiency', and epigenetic factors affect the regulation of these protective enzymes.

Finally, those enzymes often require complicated dietary co-factors including minerals for efficient function, and hence the additional role played by the presence of essential micronutrients in the diet in this process. All of these factors interplay to result in the eventual outcome which we might eventually see in a developing child. Such a model also explains the lack of consistency between particular levels of exposure and predicting outcome. Likewise, this concept may explain the variability in response to studies involving micronutrient supplementation [16, 17].

In terms of predicting future outcome, there is considerable potential benefit in attempting to identify those at significant risk of future behavioural and developmental difficulties when many of those problems may lie several years ahead. In populations where there are problems with background levels of alcohol exposure in pregnancy where 5–20% of pregnancies might be affected, it is a challenge to pick out which children might be most at risk of future disability. If there was a biological marker which was related to the level of likely neuronal damage, this might be a better discriminator than a marker which was related to the level of alcohol exposure alone?

4.5 Conclusion

What happens to the fetus during pregnancy cannot be taken in isolation. There is a continuum from fetal to neonatal to infancy and beyond. The old-fashioned 'nature versus nurture' debate polarises the interpretation unnecessarily into whether the underlying biology or the subsequent experiences are more important—because of course the answer is 'both'. The human being is endlessly complex and adaptable, and our survival and success in so many different environments in the course of evolution is a testament to this. Unfortunately changes which may initially be beneficial in terms of an adaptive response to biological adversity may no longer prove to be advantageous in different, environments subsequently.

Trying to understand the principles of fetal development and the ways in which alcohol can cause harm can help in assessing the risk from a clinical perspective. For example, one might expect the greatest impact on cognitive outcome to be related to fetal exposure later in pregnancy, during the periods of gyral formation and neuronal maturation. Understanding these aspects can help to dismiss common myths ("all the damage is done in the first trimester, so theres no point changing")

and encourage active changes and support to mothers which can still make a difference even in leter pregnancy. However, it is important not to forget the wider ways in which the fetus might be affected indirectly through alcohol in addition to the direct teratogenic affects.

With significant maternal alcohol consumption, there is often an associated poor diet. So, there can be a double-effect of increased oxidative stress coupled with risk of nutritional deficiency. Of course, some of the differences in response seen to similar levels of exposure can be because of biological differences in resilience as well as response.

The Barker hypothesis [45] has contributed an understanding of how biological impacts of pregnancy can contribute to the development of chronic disease which presents much later in life. This grew from an understanding of how fetal nutritional status can be mediated either by maternal nutritional status or factors affecting fetal blood supply, such as pre-eclampsia and hypertension, leading to the concept of fetal programming. This knowledge combined with research on DNA methylation and acetylation contributed to our understanding of how the way our genes can be affected to varying degrees by these factors. Children with prenatal alcohol exposure may remain on the small side and exhibit poor growth and weight gain, even though the precipitating factor creating that situation has been removed. Furthermore, the frequent comorbidities which often co-exist alongside alcohol use can contribute additional adverse impacts as well as reduce resilience of the fetus to withstand the consequences of prenatal alcohol exposure.

Over the last decade there has been an explosion in our knowledge of how early postnatal experiences (ACEs) and even possibly antenatal experiences, also play their part. The infant with prenatal alcohol exposure who may have subtle underlying abnormalities of brain development may also be able to adapt less well to postnatal adversity.

The influence of alcohol on the fetus does not cease at the end of pregnancy. In terms of epigenetics there is evolving evidence of the transmission of these epigenetic factors not only during cell division over a lifetime, but also - more worryingly, causing transgenerational effects to future offspring. The clinical and societal consequence of this is the importance of recognising that we need to intervene to reduce these adverse experiences and prevent the potential propagation of adverse consequences through future generations.

References

1. Ali S, Champagne DL, Alia A, Richardson MK. Large-scale analysis of acute ethanol exposure in zebrafish development: a critical time window and resilience. PLoS One. 2011;6(5):e20037. https://doi.org/10.1371/journal.pone.0020037. PMID: 21625530; PMCID: PMC3098763.
2. Rompala GR, Homanics GE. Intergenerational effects of alcohol: a review of paternal preconception ethanol exposure studies and epigenetic mechanisms in the male germline. Alcohol Clin Exp Res. 2019;43(6):1032–45. https://doi.org/10.1111/acer.14029. PMID: 30908630; PMCID: PMC6551262.
3. Wallace WHB, Kelsey TW. Human ovarian reserve from conception to the menopause. PLoS One. 2010;5(1):e8772. https://doi.org/10.1371/journal.pone.0008772.

4. Buss C, et al. Intergenerational transmission of maternal childhood maltreatment exposure: implications for fetal brain development. J Am Acad Child Adolesc Psychiatry. 2017;56(5):373–82. https://doi.org/10.1016/j.jaac.2017.03.001. ISSN 0890-8567.
5. Gu L, Liu H, Gu X, et al. Metabolic control of oocyte development: linking maternal nutrition and reproductive outcomes. Cell Mol Life Sci. 2015;72:251–71. https://doi.org/10.1007/s00018-014-1739-4.
6. Kaminen-Ahola N, Ahola A, Maga M, Mallitt K-A, Fahey P, Cox TC, et al. Maternal ethanol consumption alters the epigenotype and the phenotype of offspring in a mouse model. PLoS Genet. 2010;6(1):e1000811. https://doi.org/10.1371/journal.pgen.1000811.
7. McDonnell CG, Valentino K. Intergenerational effects of childhood trauma: evaluating pathways among maternal ACEs, perinatal depressive symptoms, and infant outcomes. Child Maltreat. 2016;21(4):317–26. https://doi.org/10.1177/1077559516659556.
8. Sharp GC, et al. Maternal alcohol consumption and offspring DNA methylation: findings from six general population-based birth cohorts. Epigenomics. 2018;10(1):27–42.
9. McQuire C, Mukherjee R, Hurt L, Higgins A, Greene G, Farewell D, Kemp A, Paranjothy S. Screening prevalence of fetal alcohol spectrum disorders in a region of the United Kingdom: a population-based birth-cohort study. Prev Med. 2019;118:344–51. https://doi.org/10.1016/j.ypmed.2018.10.013. PMID: 30503408; PMCID: PMC6344226.
10. Kotch LE, Sulik KK. Experimental fetal alcohol syndrome: proposed pathogenic basis for a variety of associated facial and brain anomalies. Am J Med Genet. 1992;44:168–76. https://doi.org/10.1002/ajmg.1320440210.
11. Schünemann H, Hill S, Guyatt G, et al. The GRADE approach and Bradford Hill's criteria for causation. J Epidemiol Community Health. 2011;65:392–5.
12. Coathup V, Northstone K, Gray R, Wheeler S, Smith L. Dietary patterns and alcohol consumption during pregnancy: secondary analysis of Avon longitudinal study of parents and children. Alcohol Clin Exp Res. 2017;41:1120–8. https://doi.org/10.1111/acer.13379.
13. Naseer M, Ullah N, Ullah I, Koh P, Lee H, Park M, Kim M. Vitamin C protects against ethanol and PTZ-induced apoptotic neurodegeneration in prenatal rat hippocampal neurons. Synapse. 2011;65:562–71. https://doi.org/10.1002/syn.20875.
14. Marino MD, Aksenov M, Kelly SJ. Vitamin E protects against alcohol-induced cell loss and oxidative stress in the neonatal rat hippocampus. Int J Dev Neurosci. 2004;22(5–6):363–77. https://doi.org/10.1016/j.ijdevneu.2004.04.005. ISSN 0736-5748.
15. Thomas JD, et al. Neonatal choline supplementation ameliorates the effects of prenatal alcohol exposure on a discrimination learning task in rats. Neurotoxicol Teratol. 2000;22(5):703–11. https://doi.org/10.1016/S0892-0362(00)00097-0. ISSN 0892-0362.
16. Wozniak JR, et al. Choline supplementation in children with fetal alcohol spectrum disorders: a randomized, double-blind, placebo-controlled trial. Am J Clin Nutr. 2015;102(5):1113–25.
17. Jacobson SW, Carter RC, Molteno CD, Stanton ME, Herbert JS, Lindinger NM, Lewis CE, Dodge NC, Hoyme HE, Zeisel SH, Meintjes EM, Duggan CP, Jacobson JL. Efficacy of maternal choline supplementation during pregnancy in mitigating adverse effects of prenatal alcohol exposure on growth and cognitive function: a randomized, double-blind, placebo-controlled clinical trial. Alcohol Clin Exp Res. 2018;42:1327–41. https://doi.org/10.1111/acer.13769.
18. Devries KM, Child JC, Bacchus LJ, Mak J, Falder G, Graham K, Watts C, Heise L. Intimate partner violence victimization and alcohol consumption in women: a systematic review and meta-analysis. Addiction. 2014;109:379–91. https://doi.org/10.1111/add.12393.
19. Anda RF, Felitti VJ, Bremner JD, et al. The enduring effects of abuse and related adverse experiences in childhood: a convergence of evidence from neurobiology and epidemiology. Eur Arch Psychiatry Clin Neurosci. 2006;256:174–86. https://doi.org/10.1007/s00406-005-0624-4.
20. Rakers F, Rupprecht S, Dreiling M, et al. Transfer of maternal psychosocial stress to the fetus. Neurosci Biobehav Rev. 2020;117:185–97. https://doi.org/10.1016/j.neubiorev.2017.02.019. ISSN 0149-7634.
21. Miranda A, Sousa N. Maternal hormonal milieu influence on fetal brain development. Brain Behav. 2018;8:e00920. https://doi.org/10.1002/brb3.920.

22. Brandon AR, Pitts S, Denton WH, et al. A history of the theory of prenatal attachment. J Prenat Perinat Psychol Health. 2009;23(4):201–22.
23. Kesmodel U, Wisborg K, Olsen SF, Henriksen TB, Secher NJ. Moderate alcohol intake in pregnancy and the risk of spontaneous abortion. Alcohol Alcohol. 2002;37(1):87–92. https://doi.org/10.1093/alcalc/37.1.87.
24. Gabriel K, Hofmann C, Glavas M, Weinberg J. The hormonal effects of alcohol use on the mother and fetus. Alcohol Health Res World. 1998;22(3):170–7.
25. Burd L, Roberts D, Olson M, Odendaal H. Ethanol and the placenta: a review. J Matern Fetal Neonatal Med. 2007;20(5):361–75. https://doi.org/10.1080/14767050701298365. PMID: 17674239.
26. Albertsen K, Andersen A-MN, Olsen J, Grønbæk M. Alcohol consumption during pregnancy and the risk of preterm delivery. Am J Epidemiol. 2004;159(2):155–61. https://doi.org/10.1093/aje/kwh034.
27. Wright JT, Barrison IG, Lewis IG, Macrae KD, Waterson EJ, Toplis PJ, Gordon MG, Morris NF, Murray-Lyon IM. Alcohol consumption, pregnancy and low birth weight. Lancet. 1983;321:663–5. https://doi.org/10.1016/S0140-6736(83)91964-5. ISSN 0140-6736.
28. Iwama N, Metoki H, Nishigori H, et al. Association between alcohol consumption during pregnancy and hypertensive disorders of pregnancy in Japan: the Japan Environment and Children's Study. Hypertens Res. 2019;42:85–94. https://doi.org/10.1038/s41440-018-0124-3.
29. Fernandes Y, Buckley DM, Eberhart DK. Diving into the world of alcohol teratogenesis: a review of zebrafish models of fetal alcohol spectrum disorder. Biochem Cell Biol. 2018;96(2):88–97. https://doi.org/10.1139/bcb-2017-0122.
30. Cook JL, et al. Fetal alcohol spectrum disorder: a guideline for diagnosis across the lifespan. CMAJ. 2016;188(3):191–7. https://doi.org/10.1503/cmaj.141593.
31. Astley SJ, Clarren SK. A case definition and photographic screening tool for the facial phenotype of fetal alcohol syndrome. J Pediatr. 1996;129(1):33–41. https://doi.org/10.1016/S0022-3476(96)70187-7. ISSN 0022-3476. Further information: https://depts.washington.edu/fasdpn/htmls/literature.htm.
32. Suttie M, Foroud T, Wetherill L, Jacobson JL, Molteno CD, Meintjes EM, Eugene Hoyme H, Khaole N, Robinson LK, Riley EP, Jacobson SW, Hammond P. Pediatrics. 2013;131(3):e779–88. https://doi.org/10.1542/peds.2012-1371.
33. Astley SJ, et al. Fetal alcohol syndrome: changes in craniofacial form with age, cognition and timing of ethanol exposure in the macaque. Teratology. 1999;59:163–72.
34. Xavier GM, Seppala M, Barrell W, et al. Hedgehog receptor function during craniofacial development. Dev Biol. 2016;415(2):198–215. https://doi.org/10.1016/j.ydbio.2016.02.009. ISSN 0012-1606. http://www.sciencedirect.com/science/article/pii/S001216061630080X.
35. Hendrickson TJ, Mueller BA, Sowell E, et al. Cortical gyrification is abnormal in children with prenatal alcohol exposure. NeuroImage Clin. 2017;15:391–400. https://doi.org/10.1016/j.nicl.2017.05.015. ISSN 2213-1582.
36. de la Monte SM, Tong M, Carlson RI, et al. Ethanol inhibition of aspartyl-asparaginyl-β-hydroxylase in fetal alcohol spectrum disorder: potential link to the impairments in central nervous system neuronal migration. Alcohol. 2009;43(3):225–40. https://doi.org/10.1016/j.alcohol.2008.09.009. ISSN 0741-8329.
37. Guerri C. Neuroanatomical and neurophysiological mechanisms involved in central nervous system dysfunctions induced by prenatal alcohol exposure. Alcohol Clin Exp Res. 1998;22:304–12. https://doi.org/10.1111/j.1530-0277.1998.tb03653.x.
38. Crotty P, Sangrey T, Levy WB. Metabolic energy cost of action potential velocity. J Neurophysiol. 2006;96(3):1237–46.
39. Bailey SM, Cunningham CC. Contribution of mitochondria to oxidative stress associated with alcoholic liver disease. Free Radic Biol Med. 2002;32(1):11–6. https://doi.org/10.1016/s0891-5849(01)00769-9. PMID: 11755312.
40. Wozniak JR, Mueller BA, Mattson SN, et al. Functional connectivity abnormalities and associated cognitive deficits in fetal alcohol Spectrum disorders (FASD). Brain Imag Behav. 2017;11:1432–45. https://doi.org/10.1007/s11682-016-9624-4.

41. Nassir F, Ibdah JA. Role of mitochondria in alcoholic liver disease. World J Gastroenterol. 2014;20(9):2136–42. https://doi.org/10.3748/wjg.v20.i9.2136. PMID: 24605012; PMCID: PMC3942818.
42. Sowell ER, Mattson SN, Thompson PM, Jernigan TL, Riley EP, Toga AW. Mapping callosal morphology and cognitive correlates: effects of heavy prenatal alcohol exposure. Neurology. 2001;57(2):235–44. https://doi.org/10.1212/wnl.57.2.235. PMID: 11468307.
43. Jacobson SW, et al. Heavy prenatal alcohol exposure is related to smaller corpus callosum in newborn MRI scans. Alcohol Clin Exp Res. 2017;41:965–75. https://doi.org/10.1111/acer.13363.
44. Sulik KK, O'Leary-Moore K, Godin EA, Parnell SE. Alcohol, drugs and medication in pregnancy. In: Preece M, Riley E, editors. Clinics in Developmental Medicine, vol. 188. London: Mac Keith Press; 2011. p. P30.
45. Barker D, Osmond C. Infant mortality, childhood nutrition, and ischaemic heart disease in England and Wales. Lancet. 1986;327(8489):1077–81. https://doi.org/10.1016/s0140-6736(86)91340-1.

Conversations About Alcohol Use in Pregnancy

5

Anna Ferguson

Chapter Highlights

- How to start and continue meaningful conversations about alcohol use in pregnancy
- Practical approaches to asking difficult questions
- How to use motivational interviewing and deliver brief interventions effectively
- Delivering ongoing care

5.1 Introduction

Conversations about alcohol use in pregnancy can be challenging. Healthcare professionals can find it hard to raise the question and supporting behaviour change can be difficult.

Midwives and doctors may feel they do not have the time, the skills or even the inclination to shoehorn 'another thing' into an already packed schedule of antenatal care. At the same time, it is clear that these conversations are vital. As the evidence shows, and as has been illustrated in previous chapters, it is essential that alcohol use in pregnancy is comprehensively addressed as part of routine antenatal care. It is widely accepted that for many people pregnancy provides the motivation to stop drinking. However, recent studies suggest that some level of alcohol is consumed in up to 80% of pregnancies in the UK [1]. This chapter focuses on straightforward methods to communicate effectively about alcohol, in ways that are not time-consuming or complicated, and do not require specialist skills or training.

This chapter is not a 'how to' guide for behaviour change or brief interventions, neither is it a checklist for motivational interviewing nor a blueprint for the 'right' way to speak to pregnant women and people about alcohol use. What has already been

A. Ferguson (✉)
Specialist Midwife Substance Use, One Stop Clinic, University Hospitals Sussex NHS Foundation Trust, Brighton, UK
e-mail: anna.ferguson1@nhs.net

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_5

Box 5.1 Example from Practice
Back in 2004 I was a student midwife and coming to the end of my first community placement. I was making my first attempts at talking to clients and formulating my own 'patter'. On one particular day I was working with a new mentor. I was in the habit of arriving at a visit and scanning the room for the maternity notes—if I could just get my hands on them, I would have my safety blanket of questions and prompts. The woman welcomed us into her front room. My mentor knelt down on the carpet. She seemed to sink a little further into herself as she tucked her hands beneath her shins. 'So,' she said, 'what can we tell you?' I would so love to return to that moment and see my reaction. It seems odd now, reflecting on this, that those few words made such an impact, but they did. It was my first experience of a consultation as a joint undertaking: a real exchange of information, where listening, rather than telling, was the priority.

written about theoretical frameworks and practical guides exceeds anything replicated here. Instead, this chapter is a practical look at beginning, and where necessary continuing, conversations around alcohol use. The focus is that it is the *process*, rather than any formula, checklist or training, that is the key to achieving meaningful interactions about alcohol use. The simplicity of a 'what can I tell you?' approach, combined with one or two simple, straightforward questions and a commitment to actively listening to the person's responses can form the basis for an honest exchange of information, and where appropriate, the framework for behaviour change to take place. Within this dialogue, the factual information about alcohol use in pregnancy is included in such a way that it is tailored to the person's existing knowledge, and as part of a mutual conversation. This approach works on two levels: firstly, it creates an atmosphere where a client is able to give an honest account of their alcohol use, and secondly, where needed, it forms the basis for an on-going conversation.

The chapter is divided into sections in order to correspond with the main instances where such conversations may take place. Firstly, asking the question: this could be at the booking appointment, subsequent antenatal appointments or at a new, unplanned contact, which may, for example, be in a day assessment unit, or triage or labour ward. The second is continuing the conversation: this focuses on cases where clients are continuing to use alcohol during pregnancy, and a brief intervention and an on-going dialogue are indicated. Examples from practice are included in the hope that they will provide useful examples of possible ways of opening up channels of communication and/or continuing a dialogue (Box 5.1).

5.2 'Making It Better'

Our healthcare system places a huge emphasis on identifying the 'problem' and getting the information across to solve 'it'. For clinicians, the desire to help and to 'do something' can be very strong. When it comes to alcohol use in pregnancy, there is an added sense of urgency due to the finite 9-month period and the possible risks to the fetus. This, of course, is valid and vital: pregnant women and people do need clear and

Box 5.2 Example from Practice
Prior to 2012 I worked largely on busy labour wards, in a high-risk antenatal day assessment unit and on a postnatal ward where I was used to asking questions, giving advice, planning care and getting things done. If you had asked me at this point in my career if I was a good listener, if I wanted the best for the families I cared for, and if I was understanding and empathetic, and aimed to empower the people in my care, I would have certainly replied yes. I identified with these statements and agreed with the sentiments behind them. But I also was focussed on giving information—on making sure clients had everything they needed to make the right choices, to do the right thing, to be well, and crucially to minimise any possible risk for themselves and their babies. What I have come to realise is that this way of working, whilst appropriate sometimes, is more often than not unhelpful when it is applied to behaviour change.

timely information. At the same time, it is also generally accepted that behaviour change is not achieved by telling people what to do [2]. However explicit the facts may appear, however clearly the information is given and however much the practitioner may want to help, human beings make decisions based on their perception of the facts via complex and deeply personal processes and their own beliefs and feelings.

Recognising this dissonance is vital when it comes to talking to clients about alcohol use: supporting behaviour change requires that a practitioner step away from her role as information giver, and instead adopt a collaborative stance, where decision-making becomes a joint process (Box 5.2).

5.3 Asking the Question

Asking difficult questions can be difficult. That sounds trite and simplistic, but it needs to be acknowledged. Conflicting factors are at play: the need to find out specific, detailed information whilst trying to build or maintain a relationship; the aim of being alongside the client, whilst very obviously being the professional in the interaction; the focus on trying to be client-led, but also needing to give specific, sometimes challenging, information and the goal of providing sensitive care, whilst often working under time constraints and in a physical environment that may not be ideal. Practitioners may also be juggling their own feelings: alcohol is an emotive subject, and they may feel strongly about its use in pregnancy. Much has been written about the importance of non-judgemental, empathetic care, which is both supportive and informative, but how is this actually achieved?

Motivational interviewing (MI) can provide a workable tool to help address some of these issues. The spirit of MI is that communication is a collaborative process that respects individual autonomy and is based on knowledge the client already has. Therefore, it is by its nature client-led and, as the client takes a more central role, the practitioner naturally becomes less dominant in the process. The use of open questions places the client at the centre of the conversation and aims to make existing knowledge about alcohol use in pregnancy the starting point of the dialogue. This is positive in two ways: firstly, using open questions encourages a longer

and more considered response and the client is therefore more likely to be actively involved in the conversation, and secondly open questions signal a willingness to establish a relationship. Both these factors are far more likely to establish a collaborative relationship between client and practitioner.

As one of a raft of health and social care questions at the booking appointment, a closed question about alcohol use may provide the perfect opportunity for the subject to be skimmed quickly over. Gaining, and documenting, this information is vital, but conditions need to be created so that the question can actually be answered. This is especially important, as a person may be feeling uncomfortable, embarrassed or even ashamed about their alcohol use. On the other hand, they may feel defensive, annoyed or judged. They may well feel that a quick 'No' is the simplest response to the question

- 'Do you drink?'

In contrast, questions such as

- *'Can you tell me what you know about alcohol use in pregnancy?'*
- *'Is there anything I can tell you about drinking in pregnancy?'*
- *'Is there anything you would like to know about alcohol and its effects in pregnancy?'*

may provide the opportunity for the person to consider the issue more closely and carefully, and subsequently begin a conversation that may elicit richer and more comprehensive information. It also provides the opportunity to listen rather than talk—to let the person establish the agenda, and then focus in on particular elements that appear important to them or explore issues that may need further discussion or clarification. It may not be that the subject is resolved in this first appointment. Acknowledging and accepting a continuing conversation is a positive step: it signifies honesty, and recognition of the importance of the issues involved. It also reinforces the message for clinicians that they may not 'solve' the alcohol issue at the first appointment, and emphasises that the issue is not, in fact, the practitioner's issue to 'solve'.

If it transpires that a client has been, or is currently, using alcohol in pregnancy then it is essential that the conversation continues. Within this it is vital that a careful and thorough alcohol history is taken. This should not be the cue to commence a list of questions about how much and how often, although accurate information is of course very important. If a person says that they have been, or are, drinking alcohol then it is all the more essential that further information is sought in a way that encourages and enables them to share it openly and honestly. This also means that the interaction will continue as a two-way street, not an interrogation, which will hopefully ensure that the relationship between the practitioner and the client will also evolve and develop positively.

Examples of follow-up questions could be:

- *'Would you be able to describe what you might drink on a night out?'*
- *'What does a typical weekend look like for you?'*
- *'Can you describe how drinking fits in to your week? Might you drink differently at the weekend than on a weeknight?'*
- *'How many drinks might you have on a night out if you were counting?'*
- *'How about on a special occasion?'*

This type of open questioning helps obtain a more complete picture of a person's alcohol use—when they drink, why they drink and how alcohol fits in to their life.

Box 5.3 Example from Practice

In 2012 I began working as part of a team providing care to clients with substance-use issues in pregnancy. Part of my role was providing cannabis reduction and cessation sessions in pregnancy, and in preparation for this I attended a short brief intervention training session. I read some literature about motivational interviewing, and by the time I was ready to see my first client I was more than slightly apprehensive about how the session would progress. My first session, with a young woman who was 18 weeks into her pregnancy and smoking cannabis fairly heavily on a daily basis, was a revelation. I tentatively used the motivational interviewing skills I had read about, and the consultation opened up in a way that I had not experienced before. The initial relief I felt from the woman, when I said 'I'm not here to lecture you about cannabis in pregnancy, I bet there is nothing I can tell you that you don't already know', was tangible. By the time I introduced the commitment and readiness/confidence 'rulers' (more of those later!) I felt that we were truly engaged in a meaningful dialogue. When, mid-way through, she looked up and said 'I think I could reduce my use from Days 7 to 6, and maybe think about losing the morning joint too' it felt like a watershed moment.

As part of this dialogue the vital answers about what and how much should evolve, but in a way that fuels connection, both between practitioner/client and the client and their alcohol choices.

MI is often criticised and under-utilised by health professionals as it is seen as requiring a high level of skill and extensive training [3], but as is shown by the above examples there is no complicated formula, just a commitment to asking broader questions, listening to the answers and allowing a client-led conversation to develop. Indeed, William R. Miller, who introduced the concept of MI in 1983, has stated that there is no magic bullet, training course or tool kit for MI [4]. Rather, he suggests practitioners need to focus on asking simple open questions, listening carefully, remaining curious and guiding gently. He also stresses that MI is something that is learnt by doing, and with practice and commitment the process evolves and develops (Box 5.3).

5.4 Alcohol Brief Interventions

Where alcohol use is identified, a brief intervention (BI) can be used alongside MI to continue to communicate effectively with clients about alcohol. Although it is evident that more research is needed regarding their efficacy in pregnancy, there is substantial evidence that BIs are effective in reducing harmful drinking [5]. Even in light of the low-quality evidence that does exist, the 2014 World Health Organisation (WHO) guidelines on BIs strongly recommended that all pregnant women and people who are using alcohol should be offered a brief intervention [6].

An alcohol brief intervention (ABI) can be described as a short, personalised interaction, which takes place on one or more occasions. It is designed to give specific information on the consequences of alcohol consumption, and in doing so encourage behaviour change.

Box 5.4 What Are the Key Principles of Motivational Interviewing?

Express empathy: do not try to resolve a problem or sympathise about it. Empathy is best seen as **connection** between people, where one person takes the perspective of the other and recognises the vulnerability of their position.
Understand discrepancy/ambivalence: acknowledge the conflicting feelings that the client may have about their alcohol use. Accept that the issue may not be clear-cut for the client—that they may have simultaneous and contradictory feelings.
Roll with resistance and avoid arguing: accept that the client needs to present their own reasons for change.
Support hope and self-determination: ask clients what their reasons are for change—don't tell them why you think they should change.

Suggestions for further reading
Motivational Interviewing in Health Care. Helping Patients Change Behavior. By S Rollnick, WR Miller and CC Butler. Guilford Press 2008.
Motivational Interviewing, Third Edition: Helping People Change (Applications of Motivational Interviewing) By WR Miller and S Rollnick. Guilford Press 2012.

It is important at this point to recognise that an ABI should be seen as part of the on-going conversation, and as such motivational interviewing should be an intrinsic part of the interaction. In practical terms this again means using open-ended questions, actively listening to responses and trying to encourage a conversation about behaviour change rather than seeing the goal as providing as much information, as quickly as possible (Box 5.4).

Rollnick, Miller and Butler rightly state that informing is a crucial part of healthcare, but they also note that providing information is often done in a way that fails to evoke behaviour change. They argue that people make choices rather than doing what they are told and suggest incorporating MI into information-giving by asking permission as part of the ABI [3]. So, a good starting point could be questions such as:

- *'I have a concern about your alcohol use. I don't know if you are concerned about it too, but would it be ok if I tell you what I think, or is there anything else that you would like to ask about, or tell me, first?'*
- *'Can I tell you what I know about that?'*
- *'There is something I need to tell you here, is that ok?'*

As clients may already feel uncomfortable talking about their alcohol use, asking permission is useful in that it is inclusive and non-threatening. It includes the person in setting the agenda for the ABI, and in a subtle but very important way it places them in control of the conversation. The ABI will obviously need to include clear and concise information about alcohol use in pregnancy, but this may be more accessible and assimilated when it is given in a spirit of collaboration and respect.

5.5 Continuing the Conversation

When alcohol use is disclosed an on-going dialogue and appropriate response to different levels of use is important. Firstly, where there is alcohol dependence—that is, where withdrawal symptoms are described when use is reduced or stopped—an

in-patient programme in a specialist unit is indicated. Abrupt cessation of alcohol use where there is physical dependence may lead to miscarriage or pre-term labour, and it is important that reduction and cessation of use are managed appropriately. Secondly, where moderate/heavy alcohol use is described, or where low use is continuing, practitioners may feel that referral to a specialist service is required. This could be in the form of a specialist substance-use clinic within maternity services, or if this is not available, a local drug and alcohol service. Finally, it may be that a specialist service is not indicated, or not available, or the person declines referral to it.

In reality, getting down to the basics of what, how much and how often is not always a straightforward or linear process. Clients may not initially disclose their alcohol use, the history they give may change, or they may not feel able or want to describe their alcohol intake. Fear of disapproval may mean that clients deny or under-report their alcohol use. Hints may come from other sources, for example from partners or other agencies, and people themselves may not have a clear idea of how much they are drinking due to the fairly complicated system of units that is used to quantify alcohol use.

The following examples are taken from practice (Boxes 5.5, 5.6, and 5.7). They are intended as a tool to guide assessment and care, rather than a comprehensive guideline. It is suggested that they are read alongside the previous sections on motivational interviewing and brief interventions. Full information on issues such as FAS/FASD/fetal brain development are mentioned but not detailed here, as these are covered in full in Chapter 3.

Box 5.5 Scenario: *Sam is 21 years old and booking at 9/40 weeks gestation in her first pregnancy. She tells you that she has been drinking 'loads' of alcohol at weekends, and smokes 20 cigarettes a day. She says 'I am stopping' and 'will my baby be ok?'*

How do you respond?
• Use statements to accentuate the positive *'It sounds like you're doing really well'* *'I can hear that you want your baby to have the best start in life'* *'It seems like you know about alcohol use in pregnancy. Is there anything more I can tell you?'* *'The possibility of risk is hard to predict, but stopping drinking at any stage decreases the harm'*
What initial information do you need? How can you find it out?
• How much alcohol was used? How often? What type? Until what gestation? Is use on-going? What is she stopping—alcohol or cigarettes? • Consider using a 'typical night out' question: *'Could you talk me through what a typical night out looks like for you?'* • Limit additional questions—let the story evolve • Ask permission when clarification is needed *'Would it be ok if I asked you about that in a bit more detail?'*
Are there specific risks in this case? Information for an ABI:
• FAS/FASD • Fivefold increase in risk of first trimester miscarriage with >5 drinks/week [7] • Increased risk of poor maternal and foetal nutrition [8, 9]

Talking not telling
'Thank you for talking to me about this today' *'It's not always easy to talk about things like this'* *'That sounds like a really good plan'* *'May I make a suggestion?'*
What next?: Clinical care and follow-up
• Document alcohol history • Offer information to take away: link to online or leaflet • Offer appointment with obstetrician if not already in place • Offer 30/40 foetal ultrasound scan if alcohol use continues • Offer cranial ultrasound scan and assessment following birth • Suggest follow-up appointment in 2 weeks to assess and review and continue the conversation

Box 5.6 Scenario*: Polly is 34 years old and 28/40 weeks pregnant with her second baby. She is seen in triage with raised blood pressure. As part of the consultation, she tells you that she is continuing to drink alcohol occasionally. She says that she did this in her first pregnancy and her baby was fine. She says that she has looked at the evidence and decided that the 'risks are over-exaggerated'*

How do you respond?
• Aim for a collaborative, non-confrontational dialogue • Acknowledge ambivalence *'It sounds like you've done quite a lot of reading around this. Is there anything more I can tell you?'* *'Are there any issues you would like further information on?'* *'I can see that you've made a plan that's working for you at the moment'*
What initial information do you need? How can you find it out?
• How much alcohol is being used? And how often? Is there a plan to stop at any point? *'I'd like to talk to you in a bit more detail about your alcohol use. Is that OK?'* *'How does having an occasional drink fit in to your week?'* *'Can you describe the things you enjoy about the drinks that you have?'*
Are there specific risks in this case? Information for an ABI:
• Evidence around low-level drinking: inconclusive regarding at what level and in which cases harm occurs • There is no proven safe time, or amount, to drink alcohol in pregnancy • No alcohol in pregnancy is the safest choice • Foetal brain development from 28/40 onwards
Talking not telling
'I agree there is a lot of evidence out there, it can be confusing can't it!' *'Is there anything that worries you about your alcohol use?'* *'I have a concern about your plan that you may or may not share, but I feel like I need to express it. Would it be alright if I explained it now, or is there something else that you want to talk about first?'* *'Thank you for discussing this with me with me today'*
What next?: Clinical care and follow-up
• Document alcohol history • Offer information to take away: link to online or leaflet • Suggest follow-up with community midwife in 2–4 weeks to continue the conversation

Box 5.7 Scenario: *Clare is 26 years old and pregnant with her first baby. When you see her at her 24/40 antenatal appointment, she tells you that she went to 'a couple of festivals' in the first trimester where she used recreational drugs and 'got drunk quite a lot'. She tells you that she still smokes cannabis and seems concerned about her partner's cannabis use*

How do you respond?
• Aim for an open, non-judgemental response *'How do you feel about your alcohol/substance use?'* *'How do you feel things are going for you?'* *'Is there anything that worries you about your alcohol/substance use?'* *'What do you know about alcohol use in pregnancy? Is there anything I can tell you more about?'*
What initial information do you need? How can you find it out?
• How much alcohol was used? How often? What type? Until what gestation? Is there on-going use? • What other substances were used? How much? How often? Is there on-going use? *'Can you talk me through what a weekend at a festival looks like for you?'* *'Can you tell me a bit more about your substance use prior to pregnancy?'* *'I'd like to ask you about your drinking in a bit more detail'*
Are there specific risks in this case? Information for an ABI:
• Cocaine use: risk of vasoconstrictive effect and stroke, neuro-behavioural consequences of an ADHD-type • Other substances: evidence inconclusive, but possible risk of harm
Talking not telling
'Thank you for telling me this today' *'I really value your openness'* *'What might be some of the benefits of stopping drinking do you think?'* *'Can I help you look at this in a bit more detail?'* *'Would your partner be interested in coming in with you next time?'*
What next?: Clinical care and follow-up
• Document alcohol history • Offer information to take away: link to online or leaflet • Offer appointment with obstetrician if not already in place • Offer 30/40 growth and amniotic fluid index ultrasound scan if alcohol/substance use continues • Offer cranial ultrasound scan and assessment following birth if sustained alcohol history disclosed • Suggest follow-up appointment in 2 weeks to assess and review and continue the conversation

5.6 Continuing the Conversation When It's Challenging

In all these cases the goal is to continue the conversation. There will of course be occasions when clients hear information that alters their perception, or understanding, and leads to immediate change. But there will also be occasions where people need to assimilate the new information gradually and make new decisions more slowly. There may also be times when clients are annoyed or irritated by the discussion. They may feel the information being presented is irrelevant or incorrect or that they are being patronised or criticised. In these cases, it is vital that confrontation is avoided. It must be remembered that

it is not an opportunity for the practitioner to tell the person what to do. Rather, it is an opportunity to listen, understand, offer information (if the person indicates that they are happy to hear it) and to aim to establish a relationship where a further dialogue is possible.

In some cases, a conversation that starts about alcohol use may bring up other issues. This may be other substance use, or relationship concerns or mental health issues. This can feel challenging, but it is likely that the issues are interlinked, and addressing them may help to unpick the issues around a person's alcohol use. It is helpful to remember again at this point that the practitioner is not there to immediately solve these issues or come up with an instant action plan or try to remedy multiple problems. Whilst it may be appropriate to discuss referrals to other services, the most important thing is to listen to the client and hear the full context of their story. Miller's comment 'It's not a goal until your client shares it' is useful. However, much the practitioner may want the client to change, the impetus to do so must come from the client. To go back to the basics of behaviour change and human behaviour, people need their own reasons to change. They need to identify those reasons and want to act on them.

5.7 Balancing Risk

Where alcohol is used alongside other recreational drugs it makes sense to focus any BI where there is the most potential for harm. In cases where there is polydrug use, addressing more than one issue is likely to be counterproductive. For example, when there is alcohol and cannabis use, it is likely that tackling the two substances at the same time is inappropriate. In these cases, it is important to balance the risks, and make a plan to address less potentially harmful substance use at a later date. As a healthcare professional there can be a tendency to feel every 'box has to be ticked' immediately. Whilst it is of course important to share information so that people can make evidence-based decisions, sharing too much too quickly may create the opposite effect to the intended one.

5.8 The Importance, Confidence and Readiness Rulers

In cases where clients show a positive response to reducing their alcohol/substance use, the importance, confidence and readiness rulers are a useful self-evaluation tool. No specific charts or equipment is needed—just a piece of paper and pen. The following scale is drawn, followed by the question:

On a scale of 1–10, if 1 is not confident at all and 10 is 100% confident, how confident do you feel in reducing (or stopping) your alcohol (or other substance) use?

1 2 3 4 5 6 7 8 9 10

The number chosen is marked on the scale. The initial question can then be followed up by further questions such as

- '*Why do you say a 3 not a 4?*'
- '*What would help you to move from a 5 to a 6?*'

Box 5.8 Example from Practice
Reaching a dead end in a conversation is hard. Silence is difficult too. There have been occasions when I have sat, mind racing, and not known what to say, or where or how I should try and guide the conversation. I don't have a magic formula for these times, but I do know that time and practice definitely helps, and that just doing it—listening carefully, speaking less and remembering that change is usually a process and not a one-off event, has helped in these situations.

The scale and question(s) are then repeated with the terms 'confident' and 'ready'. This creates the opportunity for clients to express in their own words what is important to them, identify their perceived barriers to change and most importantly recognise and voice their own reasons to change in ways that could work for them personally. The sheet can be given to the client at the end of the consultation. Within the usual fairly formal system of healthcare documentation, this act of giving a piece of a paper, that has been created together, can in itself be a powerful symbol of a joint endeavour, which has been led, and defined, by the client.

5.9 The Professional–Client Relationship: Acknowledging the Issues

There will be times when interactions do not go to plan. Listening, making suggestions and trying to resist the urge to correct and confront can be challenging. There will be occasions when establishing a dialogue is not possible. Professionals will also be juggling their own views about alcohol, which may affect the advice they give, and how they give it. Some studies have also shown that midwives are not convinced of the value of ABIs in antenatal care and feel that talking about alcohol may alienate clients and impact negatively on issues of trust and rapport. Practitioners also frequently report that they do not have the necessary training to use MI and ABIs effectively [10]. Recognising these issues is an important part of the process of improving the way alcohol is talked about in pregnancy: accepting that there will be issues, problems and challenges that may not be resolved quickly, or at all, is vital (Box 5.8).

5.10 Conclusion

Obtaining a clear and accurate maternal alcohol history is essential, both to provide appropriate care for pregnant women and people who are using alcohol and also, where there is significant alcohol use, to form part of a child's health record. It is clear that people do use alcohol in pregnancy, and also clear that it is sometimes difficult for them to speak openly about this use. Years of mixed messages about alcohol, conflicting information, media spin and personal attitudes to alcohol mean that clients, and professionals, can find talking about alcohol challenging. It is hoped that this chapter has outlined straightforward ways to talk to clients about alcohol, and, where necessary, to continue the conversation. Each contact during

pregnancy may provide the opportunity for people to disclose and discuss their use, and may ultimately make a difference to fetal well-being, the long-term health outcomes of the child and ultimately maternal health too. It is hoped that the simple strategies discussed in this chapter, which are not time-consuming and do not require specialist skills or training, will help practitioners embrace this opportunity.

Acknowledgements With grateful thanks to the following publications.

'Alcohol and pregnancy: health professionals making a difference' Telethon Institute for Child Health Research. Western Australia. 2007

'How many drinks would you have on a night out of you were counting them?'

'It isn't easy to talk about things like this'

'You want your child to have the best chance in life.'

'What are some of the good things about your alcohol use?'

'What worries you about your alcohol use?'

Rollnick S, Miller WR, Butler CC. Motivational Interviewing in Health Care. Helping Patients Change Behavior. New York: Guilford Press; 2008.

'You can't see a way forward with this one at the moment'

'Would it be ok if I tell you one concern I have about your plan?'

'May I make a suggestion?'

'I have a concern about your plan that you may or may not share, but I feel like I need to express it. Would it be alright if I explained it now, or is there something else that you want to talk about first?'

References

1. O'Keefe L, Kearney PM, McCarthy FP, Khashan AS, Greene RA, North RA, et al. Prevalence and predictors of alcohol use during pregnancy: findings from international multicentre cohort studies. BMJ Open. 2015;5:e006323. https://doi.org/10.1136/bmjopen-2014-006323.
2. Kelly MP, Barker M. Why is changing health-related behaviour so difficult? Public Health. 2016;136:109–16. https://doi.org/10.1016/j.puhe.2016.03.030.
3. Doi L, Jepson R, Cheyne H. Alcohol brief interventions in Scottish antenatal care: a qualitative study of midwives' attitudes and practices. BMC Preg Childbirth. 2014;14:170. https://doi.org/10.1186/1471-2393-14-170.
4. Rollnick S, Miller WR, Butler CC. Motivational interviewing in health care. Helping patients change behavior. New York, NY: Guilford Press; 2008.
5. Platt L, Melendez-Torres GJ, O'Donnell A, et al. How effective are brief interventions in reducing alcohol consumption: do the setting, practitioner group and content matter? Findings from a systematic review and metaregression analysis. BMJ Open. 2016;6:e011473. https://doi.org/10.1136/bmjopen-2016-011473.
6. Chang G. The 2014 WHO guidelines on brief interventions. Oxford: Blackwell Publishing Ltd; 2014.
7. Kesmodel U, Wisborg K, Olsen SF, et al. Moderate alcohol intake in pregnancy and the risk of spontaneous abortion. Alcohol Alcohol. 2002;37(1):87–92.
8. Weinberg J. Nutritional issues in perinatal alcohol exposure. Neurobehav Toxicol Teratol. 1984;6(4):261–9.
9. Phillips DK, Henderson GI, Schenker S. Pathogenesis of fetal alcohol syndrome: overview with emphasis on the possible role of nutrition. Alcohol Health Res World. 1989;13(3):219–27.
10. Miller R. Motivational interviewing in treating addictions. In: Arkowitz H, Miller WR, Rollnick S, editors. Motivational interviewing in the treatment of psychological problems. 2nd ed. New York, NY: The Guilford Press; 2015. p. 249–70.

Part II

The Difficulties of Making a Diagnosis of FASD

Section 2 Overview: Standardising the Approach to Assessment and Diagnosis

6

Raja A. S. Mukherjee and Neil Aiton

6.1 Diagnostic History

As highlighted in Part I, the first recognition of FASD, at that point only FAS, dates back to 1973. The fundamental features described at that point had not significantly changed. Over the subsequent decades, the focus has shifted from considering physical stigmata to more trying to understand the neurological presentation and the pathological mechanisms underlying these. A recognition has developed that there are significant overlaps with other conditions. However, to some extent the knowledge related to this remains in its infancy, despite nearly 50 years of research into this area. Clarity and consistency remain a challenge.

6.2 Areas of Focus: Physical Versus Neurological

The initial focus related to the impacts of prenatal alcohol exposure on the developing fetus was primarily physical. This meant identifying bespoke physical stigmata, such as small eyes, flattened philtrum and a thin upper lip. This, alongside other associated features such as a flat mid face, upturned nasal nares and small chin (micrognathia). Also, wider physical stigmata across the body were used to identify a whole range of features that were thought to be predominantly linked to prenatal alcohol exposure. In the early days, whilst the neurological deficits were seen and

R. A. S. Mukherjee (✉)
National FASD Specialist Behaviour Clinic, Surrey, UK
e-mail: Raja.Mukherjee@sabp.nhs.uk

N. Aiton
The One Stop Clinic, Royal Sussex County Hospital, Brighton, UK
e-mail: neil.aiton@nhs.net

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_6

considered necessary, less focus was placed upon this in terms of the diagnostic formulations.

Full fetal alcohol syndrome therefore required the core characteristic facial features alongside growth, cognitive deficits and alcohol exposure to be identified. If the former three were found of a sufficient nature and degree, then alcohol exposure could be surmised. This was due to the considered unique presentation that had been identified. It was never the case that the facial features alone or physical stigmata only could identify the syndrome. In fact, the opposite was true. The neurological deficits needed to be identified; however, as these were non-specific and often overlapped with other conditions, arguments have perpetuated over the number of deficits that are required and the level of abnormality that was required to meet diagnostic thresholds. Over the last decade, it is this latter area that has predominated diagnostic research.

Having said that, new and emerging techniques are again beginning to shed light on how physical stigmata may aid the diagnostic process. For example, whilst many of the features that continue to be identified were reported almost from the first identification in 1973, they were not necessarily quantifiable or easily measurable. New technology such as 3D imaging has now allowed this to be better measured. It will open up a new era of diagnostic tools. These are not yet used in clinical practice and remain a research area, therefore not discussed in extensive detail here.

6.3 The Development of Different Diagnostic Approaches

Due to the discrepancies and discussions around the level of deficit required and disagreements over the best approach to diagnosis, different diagnostic schema have been developed. The Institute of Medicine in 1996 was one of the first to identify a range of disorders linked to the wide spectrum. However, a lack of agreement over the broader neurocognitive presentation meant that by 2003, the four-digit code was published by the University of Washington. This approach covered the whole range of outcomes linked to prenatal alcohol exposure. Only some of the diagnostic categories related to those who had the most significant deficits and overlapped with the Institute of Medicine criterion. By 2005, Canada had summarised findings to create its own approach. This was updated in 2016. Since then other countries, including Scotland, have reviewed the literature to produce its own approaches to the diagnostic process. In 2013, DSMV included a diagnostic approach for individual clinicians to use, which addressed the need for a multidisciplinary team to assess the condition.

It is important to recognise that all the diagnostic criteria use the same four basic parameters. Any disagreement relates to sensitivity and specificity. When considering the different approaches, all consider facial characteristics, growth, neurocognitive deficits and prenatal alcohol exposure. It is only around thresholds that there is disagreement. This is not different to many other diagnostic conditions where there is a gradation. For example, between unaffected, exposed but without a functional deficit and those exposed with clinical deficits. It is by trying to identify these cut-offs that disagreements have arisen. This has caused confusion in those not

specialist in the area; however, if there is any single takeaway message, it is that there is general consensus and agreement around diagnostic validity. Also in that using one of the approaches, in a consistent manner, is better than ignoring the disorder or not using any standard approach.

6.4 Comorbidities

Confusing this is a fact that at least from the neurological process, there are overlaps with other presentations. As such, attributing these to prenatal alcohol exposure can be difficult. Especially, as there is often poor alcohol exposure recording in many areas. Reviews have suggested that the effects of alcohol are extensive and that there are many, up to 428, identified comorbid presentations with those diagnosed with FASD. As such it is important to recognise the relationship between these disorders and those factors that influence the wider presentations. The diagnosis is therefore one of exclusion of other causes of neurological deficit and inclusion of expected features. This is why also understanding the relationship between aetiology and outcome becomes important.

6.5 The So What Question

Ultimately, it is through diagnosis that an individual can access treatment. Whilst diagnosis tends to be a medical model, it allows several factors to be identified and initiated. Firstly, diagnosis leans toward an understanding of subsequent management and how management strategies need to be modified in the light of that diagnosis. It also allows prognosis to be presented to individuals and their families. This helps to develop the individual understanding around the levels of support and help that need to be put in place in order to improve their overall prognosis. This offers trajectories to the individual and those families living to guide their futures. Also, but less important, it allows clarity to be established that conditions such as FASD which remain questioned by some, actually exist. It also allows research to be conducted internationally on the subject because it is clear that the same groups are being studied. Without these, progress cannot be made.

Arguments around formulation versus diagnostic approaches are often raised. These are not exclusive situations however. Good practice would suggest that even where there is diagnosis, a formulation around underlying individual needs, especially in the light of the wide variability, remains an important part of the presentation. This does not diminish the need to undertake diagnostic profiling. As will be discussed in later chapters, varying degrees of resource means that not everybody will have full evaluations. Understanding a diagnosis guides the individual to areas to explore, which offers the greatest impact for the individual. It also recognises potential areas that have not yet been assessed, but may be important at a later stage. Formulation alone, unless everything is studied in every individual, does not offer this. It is therefore important that the two aspects are taken together.

Ultimately however, it is about changing the management and trajectory for individuals that is important. This will be discussed more in Part III.

6.6 Overview of the Following Chapters

The following chapters in Part II will build and explain more with regards to the changing pattern of this condition in children as they grow older. Both physical and neurological deficits will be described alongside new concepts in diagnosis as well as how to recognise children in different settings such as those adopted. Some of the subtleties of both the physical and psychological presentation will be presented. Further, how these presentation impact on wider relationships, such as attachment, will be considered. The section will conclude with a consideration around how services can be developed and set up in order to support the diagnostic and management process.

7 Is My Baby Affected? Assessment and Diagnosis at Birth and During Infancy

Neil Aiton

Chapter Highlights

- Identifying newborn infants who might be at risk of later problems (fetal alcohol spectrum disorder [FASD]).
- The problems of trying to make a diagnosis of FAS or FASD in the newborn period.
- Structured assessment of the newborn infant who may have been exposed to alcohol in pregnancy.

The first published papers describing fetal alcohol syndrome which reached widespread attention concerned the description of 11 young children including newborn infants in 1973 [1].

Current understanding regarding the cardinal features of growth deficiency, characteristic facial phenotype and evidence of neurological dysfunction has not changed significantly since that first description, although the full extent of the complexities of the effect on the nervous system took considerable time to be fully appreciated. Despite recognition of the clear impact of prenatal alcohol exposure at such an early stage in these early papers, nearly 50 years later it is extraordinarily difficult to make a diagnosis of fetal alcohol syndrome or fetal alcohol spectrum disorders in the newborn period. There are a number of important reasons why this is so:

- In many places the single most important reason is due to poor identification of women who might be drinking alcohol in pregnancy (see Chaps. 5 and 32).

N. Aiton (✉)
The One Stop Clinic, Royal Sussex County Hospital, Brighton, UK
e-mail: neil.aiton@nhs.net

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_7

- Many children who may develop significant problems meriting a diagnosis of FASD much later in childhood do not demonstrate any observable clinical abnormality at birth.
- Systematic approach to assessment of infants in the newborn period who might be affected by prenatal alcohol exposure is rare in the UK and elsewhere at the current time.

From the population of mothers who were drinking alcohol in pregnancy there will be babies who will show clinical evidence of prenatal alcohol exposure at the time of birth, and almost certainly, a larger group who will not. We already know that there is an inconsistent relationship between the amount of alcohol exposure during pregnancy and future outcome. Therefore, because of the lack of a direct relationship, knowledge of the amount and frequency of alcohol exposure in pregnancy can only predict *risk* of a future outcome within extremely broad parameters. The task in the neonatal period in an ideal world is to identify:

- Infants who demonstrate clinical evidence which relates to prenatal alcohol exposure
- Infants who might be most at risk of future problems

If it is thought that an individual infant might be at significant risk of developing problems related to FAS/FASD in the future because of the knowledge about exposure during pregnancy, and it is not possible to make a diagnosis in the newborn period, there are still substantial impediments to achieving a later diagnosis for the following reasons:

- Even where a significant risk of developing FAS/FASD has been recognised, many children's developmental services are not commissioned to follow up children who do not yet have an identified problem. This runs counter to the principle of following up infants who are *at risk of future neurodevelopmental impairment* (which is so deeply enshrined within the field of neonatal medicine for follow-up of preterm infants) and means that opportunities for early recognition of problems and diagnosis can be missed.
- At the time of writing, with a minority of exceptions, in the UK there is a lack of clearly developed neurodevelopmental diagnostic pathways commissioned within childhood developmental services for referral of children who might have FAS/FASD [2].
- Where pathways do exist for diagnosis of FAS some children, who may have quite pervasive life-impacting problems, may not actually reach threshold criteria for diagnosis for either ADD/ADHD (Attention Deficit Disorder/Attention Deficit Hyperactivity Disorder) or ASD (Autistic Spectrum Disorder), and can end up without the true nature of their problems being recognised.
- Many children end up presenting at school age with educational or behavioural problems, and may not get referred for a full medical assessment, because there are no obvious medical or developmental issues (even if the pre-birth history had been known at a much earlier stage).

- The perinatal history may no longer be available—particularly for adoptive children.
- Even assuming that children's community developmental services are notified about those who might be 'at risk', this information may not end up being amalgamated with educational records thus the opportunity of providing the potential context for any future behavioural or educational difficulties, prompting medical referral can be missed. [Hopefully, this is becoming less common since the introduction of shared joint educational and healthcare plans (EHCP) in the UK.]
- Professional barriers: including training, resistance to asking about previous history
- Parental barriers: including guilt, minimisation of alcohol use, concern over safeguarding procedures

The gold standard should be that the possibility of the underlying diagnosis of FAS/FASD should be included as part of the diagnostic consideration in all children who present with neurodevelopmental problems, and be part of the diagnostic consideration in pathways for diagnosis of ASD and ADD/ADHD.

There are many good reasons for trying to make a diagnosis of FAS/FASD at the earliest opportunity. Correct diagnosis leads to the appropriate use of precious (and expensive) healthcare resources by making sure that appropriate treatments and interventions are utilised, and conversely resources are not wasted on inappropriate interventions which may not work. There is also some evidence that earlier diagnosis leads to better outcomes [3]. Probably because the correct help and support given earlier allows the opportunity to minimise the adverse outcomes—particularly those concerning long-term mental health—which can accrue over a prolonged period when this is lacking and management is sub-optimal.

The majority of women of child-bearing age in Western European Cultures drink alcohol on a reasonably regular basis. The actual proportion will vary according to differing cultural and regional factors: in the UK, from 29% described as binge-drinking in Bradford to 74% >2 units/week in Leeds, and even up to 92% (with 40% drinking every week) in Scotland [4–6]. In one survey, 20% of women aged 18–44 drank more than 14 units/week [4]. Over time, this level of alcohol consumption places their own health at risk irrespective of any pregnancy. There are various estimates of the number of pregnancies which are 'unplanned', but some estimates have placed this as high as 50%. Therefore, alcohol exposure within the first few weeks of pregnancy is relatively common: 9% during first trimester in Bradford dropping to 3% in the second. It is therefore not surprising that current health messages aimed at those trying to conceive suggest the 'precautionary approach' advocating abstention [7] particularly in view of the poor quality and strength of evidence available regarding low levels of consumption [8]. From this local population cohort-based evidence, which is limited culturally and geographically but robust in quality, we know that peri-conceptual alcohol consumption is common within the UK population.

It is current recommended standard practice to screen for alcohol consumption at initial pregnancy booking and ideally this should be done using standardised tools [9]. At the same time, all women should be given advice regarding the risks of alcohol consumption during pregnancy. Although there has been much discussion about the

use of screening tools, which have nearly all been designed to screen for hazardous or harmful drinking in various populations, rather than the context of screening for alcohol consumption in pregnancy, the underlying question which needs addressing at pregnancy booking is a binary one: 'is there any ongoing alcohol consumption or not?' There is high-level evidence that brief interventions are effective, and should be offered to those women who are continuing to drink alcohol [10]. Women who are alcohol-dependent have a history of hazardous or harmful drinking, or have a high level of consumption prior to pregnancy should be referred to more specialised services.

However, even if a perfect screening and identification programme exists, there are still important questions to answer about what steps to take next, for which the evidence is far from clear:

- What is the threshold for exposure during pregnancy which should lead to formal assessment after birth for signs of FAS/FASD and on-going follow-up? (number of episodes, and level of consumption)
- What is the threshold for exposure during pregnancy which ought to merit referral after birth for ongoing neurodevelopmental follow-up even if there are no observable signs at birth?
- What is the gestational age threshold above which the risk is high enough to merit further referral after delivery even if there is no continuing alcohol use? (for example: the mother who didn't realise that she was pregnant, but stops immediately the pregnancy is confirmed)
- What is place for biological screening tests for alcohol use in pregnancy? (e.g. meconium, hair strand testing, PETH testing)
- At a clinical service level, how do you balance the ideal situation of providing early detection of neurodevelopmental problems in infancy or childhood without excessive burden in areas with high levels of prenatal alcohol exposure.

Part of the reason these questions are difficult to answer is because not all of the children who have been subject to some degree of prenatal alcohol exposure, will subsequently end up with a diagnosis of FAS/FASD: demonstrating the difference between exposure and the effect or impact of exposure. From a pragmatic perspective, those babies where there is continuing exposure throughout pregnancy and those babies where there is exposure continuing into the second trimester (whether intermittent or regular) can be considered to be at the highest risk and should certainly be identified for assessment and follow-up. So far there are very few prospective cohort studies which have assessed the level of exposure during pregnancy and then go on to investigate the proportion of children who subsequently develop FAS/FASD on rigorous assessment—particular with low to moderate prenatal alcohol exposure during the first trimester. The AQUA study, which is still in progress, is good example of a study which should help to address this question when the results of longer-term follow-up are published [11]. The Safe Passage Study may also contribute useful information [12].

A further important reason is that the majority of the research concerning the diagnosis of FASD has sprung from the perspective of children who present with a

number of different problems (usually neurodevelopmental or behavioural) and the implicit question arising is whether this is related to prenatal alcohol exposure. The use of screening tools has been proposed [13]; however, these types of tools are often based on cohorts of children who have a diagnosis of FAS/FASD and examining the retrospective relationship between exposure and different outcomes, which is a different epidemiological question to looking at that relationship 'the other way round': the newborn infants who are born to a population of mothers who may have drunk alcohol in pregnancy who present later with developmental problems. There is therefore a potential problem with applying these data from screening tests which have been developed in a different population to newborn infants with prenatal alcohol exposure prospectively, but given the imperfect nature of the prospective follow-up data at the present time, this perhaps is the best evidence available, and so not without merit. A recent publication concerning twins demonstrates the challenges involved in predicting outcome even with the same level of exposure and illustrates that differences in genotype have important modifying effects [14].

Biochemical screening tests which have been suggested include measuring Phosphatidylethanol (PEth) and meconium for Fatty Acid Ethyl Esters (FAEE); however, the evidence is so far insufficient to support the reliable relationship between these markers and prenatal alcohol exposure, particularly at low levels of exposure [15, 16]. The use of meconium FAEE certainly adds additional information confirming prenatal alcohol exposure which has occurred in the second trimester when meconium starts to be produced, and therefore can be considered to have identified a higher-risk group. Hair strand testing has been used particularly with respect to safeguarding procedures, but there are insufficient data particularly relating to the changed physiology of pregnancy to reliably relate exposure with measurements made, and the implications of any particular values. At the current time it can be thought of as 'adding to the body of evidence' concerning the context of antenatal exposure for an individual child and may contribute additional information to guide safeguarding considerations when considering maternal aspects.

It is impossible to discuss the topic of early identification and assessment without, sadly, consideration of the topic of child-safeguarding and potential adoption. Included in these processes is the need to try and attempt to answer important questions such as:

- 'What has been the impact on the baby from what happened in pregnancy?'
- 'What will the future outcome be?'
- 'What type of support will the baby require?'

Due to the uncertainty which inevitably exists around predicting future development, there is also the importance of avoiding the opposing dangers of wanting to ensure that an appropriate environment, support and future monitoring can be provided by warning of potential issues ahead and yet not 'imprisoning' the child's future potential by limiting future expectation. The adoption of children who may have had prenatal alcohol exposure is discussed elsewhere in this book, and also in that context the difficulty, or sometimes impossibility of trying to obtain a prenatal history. It is therefore important that—along with all other determinants of future

health (e.g. family history, pregnancy, birth and early neonatal history, presence of viral infection) that any history of prenatal alcohol exposure is recorded in the child's records. Issues relating to maternal confidentiality may mean that this information should be recorded anonymously, without traceable demographic details, and this should probably be the ideal standard for all children and not just those being adopted.

Despite the limitations in making an assessment for FASD in the newborn infant, some things can be assessed: including documenting the antenatal exposure, growth parameters (particularly head circumference measurement looking for microcephaly), the presence or absence of facial features, assessment of neurology and brain imaging (Table 7.1).

Table 7.1 Structured assessment of infants with prenatal exposure in the neonatal period

Assessing and recording the history:
Pattern of alcohol exposure during pregnancy? Amount, frequency Special occasions Crises? • Sources of evidence: – Maternal history – Pregnancy—pregnancy booking and on-going records – Biochemical testing: urine—toxicology or dipstick records, hair strand testing – External assessments: Police reports (including episodes triggered by reported domestic violence), A&E attendance, GP general practitioner, Social services, substance misuse or alcohol services. (Include reliably documented episodes of observed intoxication by third parties) • Important pointers that should raise particular concern: – Self-reporting concern – Previous child with FAS/FASD – Reports of intoxication from other sources, e.g. police, domestic violence reports, hospital emergency departments, safeguarding reports (if other children involved) – High level and frequency of alcohol consumption prior to pregnancy – Reports from other services who may be involved (perinatal mental health, substance misuse or alcohol support services) – Evidence of intra-uterine growth restriction
Examination and physical assessment:
• Plot length, weight and head circumference on standard centile charts (microcephaly defined as head circumference <2 SD below the mean) • Assessment of dysmorphic features (see Chap. 10) • Assessment of sentinel facial features: – Palpebral fissure length <2 SD below mean – Philtrum rated 4 or 5 on University of Washington Lip-Philtrum scale – Upper lip rated 4 or 5 on University of Washington Lip-Philtrum scale • General and neurological examination • Clinical photography: for medical record, Computerised FASD assessment
Further investigations for consideration:
• Brain ultrasound examination • MRI brain—if neurological abnormality demonstrated (including dysfunctional feeding) or abnormality on ultrasound • Other causes of in-utero growth restriction: – TORCH screen (serology for: Toxoplasma, Rubella, Cytomegalovirus, Herpes Simplex) – Urine for Cytomegalovirus excretion • Dysmorphology: Blood test for chromosomes, CGH array

Table 7.1 (continued)

Possible outcomes following assessment: The following four points should probably be better arranged as follows:
1. ***Diagnosis: FASD with sentinal facial features***—facial features present with microcephaly 2. ***Diagnosis: FASD without sentinel facial features*** (extremely rare in neonatal period)—facial features present without microcephaly but with deficits in three or more domains 3. ***Confirmed prenatal alcohol exposure with prenatal features but no other features: neurodevelopmental follow up recommended*** 4. ***At risk for neurodevelopmental disorder and FASD, associated with prenatal alcohol exposure***
Further referral?
Genetic referral: Evidence of dysmorphic features may warrant further genetic advice. (Photographic record can be helpful and provides opportunity for repeat assessment at a later time.) *Ongoing follow-up*: Community paediatric services depending on local pathways *No follow-up* (in absence of any abnormal physical/developmental findings): documentation of exposure and copies of summary to GP, Health Visitor and Local Community Paediatric service for information in case of future referral

7.1 Diagnostic Criteria

One of the most comprehensive and easy to follow guidelines for the diagnosis of FAS/FASD remains the Canadian algorithm, which was revised in 2016 [17] and recommended with slight modification in the SIGN guideline [9].

It is very rare to be able to demonstrate significant central nervous system dysfunction in the newborn period in three or more domains of development as described in the diagnostic algorithm; however, recognition of significant nervous system dysfunction is potentially possible for the following domains: neuroanatomy/neurophysiology, attention, motor skills. For example, the presence of microcephaly (head circumference <2 SD below mean), an abnormal MRI demonstrating anatomical changes consistent with prenatal alcohol exposure, seizures (not including those related to immediate withdrawal, and with other causes excluded), abnormal oromotor skills demonstrated by dysfunctional feeding, that is, abnormal sucking/swallowing. There is not enough research information at the present time to understand aspects related to impulse control and self-regulation, abnormal visual function and social interaction. These might be predictive of future pervasive neurological abnormalities, but more research is needed in this area with long-term follow-up. Assessments therefore usually remain inconclusive for children under 6 years of age.

It remains to be seen whether routine imaging of the central nervous system may play a part in the standardised assessment of infants with documented exposure at some point in the future. However, further research is required to determine the part this might play and the relationship between particular findings at birth and future neurodevelopmental outcome. The attractiveness of using neurological 'biomarkers' of neuronal injury is that this is more likely to represent the impact of exposure, as opposed to the only other information available which is the record of exposure. MRI examination is resource-intensive and may require sedation and it is possible that other ways of assessing neuronal injury, which are easier to perform such as the use

of ultrasound, may play an increasing part. Trans-fontanelle ultrasound in particular in the neonatal period can provide good views of central brain structures such as the corpus callosum. Changes in the corpus callosum have been documented in infants and adults with FASD on MRI [18, 19], so these measurements may form a part of future assessments once the relationship between changes at birth and future outcome has been established. It is possible that 3D assessment of facial features may also play a part because of the ability to detect and measure subtle changes in facial features which cannot be detected by standardised dysmorphology assessment, and because of the evidence of relationship between facial morphology and brain development [20].

Although growth parameters do not form part of the diagnostic assessment for FASD in the latest Canadian guidelines, growth impairment should be carefully documented at birth, and may well be strongly related to prenatal alcohol exposure. Neonatal doctors and paediatricians are used to assessing the perinatal factors which may also be involved in contributing to causes of in-utero growth restriction. In that respect, the documentation of the presence, and also the *absence* of additional factors such as hypertension, smoking, maternal illness, can be important in understanding the degree to which prenatal alcohol exposure played a contributory part, with absence of these additional factors increasing the relative likelihood of the contribution prenatal alcohol exposure towards growth restriction. Whereas once many of the pregnancy and maternally related causes of intra-uterine growth restriction are removed following delivery (e.g. placental failure), there can be a period of catch-up growth in many infants over the next few months, this is often not seen in children with prenatal alcohol exposure. There are good reasons to include routine consideration of fetal alcohol exposure in the pathway as a diagnostic possibility in babies who are identified after birth with IUGR. (Prenatal alcohol exposure is likely to be at least ten times more common than the likelihood of CMV infection for example.)

Although routine genetic referral is not essential in the diagnostic process for children referred with suspected FASD [9], formal genetic testing (array CGH) can help to exclude alternative diagnoses or provide additional information to aid the process of diagnosis. In particular, genetic abnormalities may be identified which can be associated with facial dysmorphism and/or neuro-developmental abnormalities. Genetic abnormalities were identified in 3.6% of one recent retrospective case series review in the UK [21]. If a particular abnormality is identified, the findings can sometimes be difficult to interpret along with any associated implication, and the advice of a geneticist will be invaluable.

Computerised assessment of the facial features can be helpful (for example: using the Washington University software www.fasdpn.org) in providing a standardised assessment, with the aim of reducing subjectivity. Unfortunately lip-philtrum charts do not exist which relate particularly to the new born period, although palpebral fissure normal values are available. Even so, standardised photographs in the medical record can still be helpful, so that others can make their own conclusion, and these can be referred to in retrospect some years later.

7.2 Conclusion

Fundamentally, the most important long-term impact of prenatal alcohol exposure is that of neurodevelopmental abnormality. This is not possible to assess comprehensively so early in life in the newborn period due to immaturity of the nervous system. Therefore, it is difficult to make a diagnosis of FASD in the neonatal period and full diagnosis may need to wait until later in childhood. The optimal approach is to identify those infants who have evidence of prenatal alcohol exposure at the time of birth, or who are at significant risk of developing FASD.

There is not a direct relationship between the level of exposure and outcome for a number of reasons. Without a complete assessment as possible, the need for follow-up can only be determined by information on fetal exposure during pregnancy.

At the present time, there are also no early biomarkers present at birth which possess the ability to predict future neurodevelopmental outcome.

Early assessment is important in identifying infants who might be at risk, although there are difficulties in attempting to stratify that risk. More research is needed on the gestational age below which the likelihood of any significant impact is extremely low.

In the perinatal period, there is a vital role for professionals in documentation of exposure in pregnancy and transfer of information to baby's records. Potential conflicts exist between maternal rights and the rights of the child in transferring information to child records, particularly in the context of adoption, but it is reasonable to manage that by making the data factual and non-attributable. Standardisation in the way the data are recorded might help in the future?

In the neonatal period, a systematic and thorough assessment can be undertaken although there are limitations to the ways the nervous system can be assessed at such a young age. The most severely affected infants may reach threshold criteria for diagnosis of FASD.

Ideally follow-up should be instigated for those most at risk of future problems, although there are problems in identifying which babies should receive follow-up. There are also potential issues with childhood neurodevelopmental services being able to follow up children who do not (yet?) have a problem.

In the future, it may be that biomarkers which are better indicators of future impairment (rather than just indicators of exposure) such as brain scans. 3D photo assessment may provide a greater ability to stratify and predict risk.

References

1. Jones KL, Smith DW. Recognition of the fetal alcohol syndrome in early infancy. Lancet. 1973;302(7836):999–1001. https://doi.org/10.1016/s0140-6736(73)91092-1. PMID: 4127281.
2. A crisis of commissioning: CCGs are failing Government policy on FASD. National Organisation for FASD, London. 2019. https://nationalfasd.org.uk/documents/A%20Crisis%20of%20Commissioning%20report%20FINAL.pdf. Accessed 5 Feb 2021.
3. Streissguth AP, Bookstein FL, Barr HM, Sampson PD, O'Malley K, Young JK. Risk factors for adverse life outcomes for fetal alcohol syndrome and fetal alcohol effects. J Dev Behav Pediatr. 2004;25(4):228–38.

4. Cooper DL, Petherick ES, Wright J. J The association between binge drinking and birth outcomes: results from the Bradford cohort study. Epidemiol Commun Health. 2013;67:821–8.
5. Nykjaer C, Alwan NA, Greenwood DC, et al. Maternal alcohol intake prior to and during pregnancy and risk of adverse birth outcomes: evidence from a British cohort. J Epidemiol Community Health. 2014;68:542–9.
6. Symon A, Rankin J, Sinclair H, et al. Peri-conceptual and mid-pregnancy drinking: across-sectional assessment in two Scottish health board areas using a 7-day retrospective diary. J Adv Nurs. 2017;73(2):375–85. https://doi.org/10.1111/jan.13112.
7. UK Chief Medical Officer's low risk drinking guideline. 2016. https://www.gov.uk/government/publications/alcohol-consumption-advice-on-low-risk-drinking.
8. Mamluk L, Edwards HB, Savović J, et al. Low alcohol consumption and pregnancy and childhood outcomes: time to change guidelines indicating apparently 'safe' levels of alcohol during pregnancy? A systematic review and meta-analyses. BMJ Open. 2017;7:e015410. https://doi.org/10.1136/bmjopen-2016-015410.NHSguidelines.
9. Children and young people exposed prenatally to alcohol: a national clinical guideline. Scottish Intercollegiate Guidelines Network, Edinburgh. 2019. https://www.sign.ac.uk/media/1092/sign156.pdf.
10. Chang G, McNamara TK, Orav EJ, et al. Brief intervention for prenatal alcohol use: a randomized trial. Obstet Gynecol. 2005;105(5 Pt 1):991–8. https://doi.org/10.1097/01.AOG.0000157109.05453.84.
11. Muggli E, O'Leary C, Forster D, et al. Study protocol: Asking QUestions about Alcohol in pregnancy (AQUA): a longitudinal cohort study of fetal effects of low to moderate alcohol exposure. BMC Preg Childbirth. 2014;14:302. https://doi.org/10.1186/1471-2393-14-302.
12. Dukes KA, Burd L, Elliott AJ, Fifer WP, Folkerth RD, Hankins GD, Hereld D, Hoffman HJ, Myers MM, Odendaal HJ, Signore C, Sullivan LM, Willinger M, Wright C, Kinney HC. Safe passage study. Paediatr Perinat Epidemiol. 2014;28:455–65. https://doi.org/10.1111/ppe.12136.
13. FASD. Screening toolkit. Ottawa, ON: Childrens' Healthcare Canada. https://ken.childrenshealthcarecanada.ca/xwiki/bin/view/FASDScreeningToolkit/National+Screening+Tool+Kit+for+Children+and+Youth+Identified+and+Potentially+Affected+by+FASD. Accessed 14 Dec 2020.
14. Hemingway SJA, Bledsoe JM, Davies JK, Brooks A, Jirikowic T, Olson EM, et al. Twin study confirms virtually identical prenatal alcohol exposures can lead to markedly different fetal alcohol spectrum disorder outcomes-fetal genetics influences fetal vulnerability. Adv Pediatr Res. 2018;5:23. https://doi.org/10.24105/apr.2019.5.23.
15. Howlett H, Abernethy S, Brown NW, Rankin J, Gray WK. How strong is the evidence for using blood biomarkers alone to screen for alcohol consumption during pregnancy? A systematic review. Eur J Obstet Gynecol Reprod Biol. 2017;213:45–52. https://doi.org/10.1016/j.ejogrb.2017.04.005. PMID: 28426943.
16. McQuire C, Paranjothy S, Hurt L, et al. Objective measures of prenatal alcohol exposure: a systematic review. Pediatrics. 2016;138(3):e20160517. https://doi.org/10.1542/peds.2016-0517.
17. Cook JL, Green CR, Lilley CM, Anderson SM, Baldwin ME, Chudley AE, Conry JL, LeBlanc N, Loock CA, Lutke J, Mallon BF, McFarlane AA, Temple VK, Rosales T, Canada Fetal Alcohol Spectrum Disorder Research Network. Fetal alcohol spectrum disorder: a guideline for diagnosis across the lifespan. CMAJ. 2016;188(3):191–7. https://doi.org/10.1503/cmaj.141593. PMID: 26668194; PMCID: PMC4754181.
18. Sowell ER, Johnson A, Kan E, et al. Mapping white matter integrity and neurobehavioral correlates in children with fetal alcohol spectrum disorders. J Neurosci. 2008;28(6):1313–9. https://doi.org/10.1523/JNEUROSCI.5067-07.2008Jacobsen.
19. Jacobson SW, Jacobson JL, Molteno CD, et al. Heavy prenatal alcohol exposure is related to smaller corpus callosum in newborn MRI scans. Alcohol Clin Exp Res. 2017;41(5):965–75. https://doi.org/10.1111/acer.13363.

20. Suttie M, Wozniak JR, Parnell SE, et al. Combined face-brain morphology and associated neurocognitive correlates in fetal alcohol spectrum disorders. Alcohol Clin Exp Res. 2018;42(9):1769–82. https://doi.org/10.1111/acer.13820.
21. Lam Z, Johnson K, Jewell R. Genetic testing in patients with possible fetal alcohol spectrum disorder. Arch Dis Child. 2020; https://doi.org/10.1136/archdischild-2020-319572.

How FASD Presents Across the Lifespan

8

Ana C. Hanlon-Dearman

Chapter Highlights

- Presentation of fetal alcohol spectrum disorder (FASD) across different ages
- Key assessment points for clinicians across different ages
- Summary of clinical anticipatory guidance and interventions for individuals with FASD across the lifespan

The importance of early recognition of fetal alcohol spectrum disorder (FASD) is critical to supporting early diagnosis, and facilitating referrals for appropriate interventions to promote optimal outcomes and reduce comorbidity at every age [1]. It is therefore important for the clinician to be able to recognize not only the facial and physical characteristics but also the clinical neurobehavioral symptoms of FASD across the age span. The purpose of this chapter is to review the presentation of FASD across infancy to adulthood and provides key practice points for clinical assessment.

8.1 Newborn and Infancy

Infants exposed to alcohol are often typically initially recognized by their facial characteristics. The classical or sentinel facial features seen in children with prenatal alcohol exposure include shortened palpebral fissures, a thin vermillion border of the upper lip, and a long smooth philtrum [2–7]. Other features that may be observed include midface hypoplasia and flattened nasal bridge, micrognathia, ear abnormalities such as a railroad track pattern to the helix, a high arched palate, ptosis

A. C. Hanlon-Dearman (✉)
Pediatrics and Child Health, Max Rady College of Medicine, Rady Faculty of Health Sciences, University of Manitoba, Winnipeg, MB, Canada

Manitoba FASD Centre and Provincial FASD Network, Rehabilitation Centre for Children, Specialized Services for Children and Youth, Winnipeg, MB, Canada
e-mail: ahdearman@hsc.mb.ca

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_8

of the eyelids, digital abnormalities such as incomplete extension, joint contractures, reduced supination/pronation at the elbows, abnormal palmar creases, short upturned nose, and limb abnormalities [8, 9]. The presence of these features should be noted as part of a comprehensive assessment but are not felt to be pathognomonic of FASD.

The behavioral characteristics attributable to prenatal alcohol exposure should be recognized in infants both with and without the classical physical features. One of the distinguishing behavioral characteristics of prenatal alcohol exposure (PAE) in infants is excessive arousal and persistent difficulties with state regulation [10–12]. These difficulties may include significant feeding difficulties, sleep difficulties, reflexive abnormalities (often hyperreflexive, but may be variable), tone abnormalities including both hypertonia and hypotonia, high pitched vocalizations, difficulties sustaining attention, and difficulties with autonomic regulation (e.g., temperature instability). The impact of PAE has even been noted in the fetal state, reported to include abnormal state regulation and delayed maturation of fetal startle responses reflecting alcohol's early impact on inhibitory pathways [13, 14]. Neonatal and infant sleep difficulties have been correlated with early changes in infant EEG [15–17]. Changes in infant stress reactivity have also been observed with changes to cortisol reactivity, heart rate, and infant effect [18]. Recent work using cardiac orienting responses as alcohol sensitive (though not specific) and developmentally predictive measure of an infant's response to environmental stimuli supports the negative impact of PAE on both physiologic and learning responses in infants with alcohol exposure [19]. Finally, a recent prospective study of mother–infant pairs enrolled in the Rhode Island Child Health Study between 2009 and 2013 ($N = 627$) using the NICU Network Neurobehavioral Scale described atypical infant behavioral profiles in those with PAE, characterized by low levels of self-regulation and attention, and high levels of arousal and excitability [20].

Developmental and behavioral delays have been described in infants with PAE including motor, language, and cognitive delays as well as delays in social behavior [11, 21–23]. Infants with PAE have shown significantly lower infant mental development using the Bayley Infant Mental Development Index [24]. Infants have also been shown to have more "difficult" behavior, including being more difficult to soothe, more labile and less responsive, and this so-called "difficultness" has been linked to future risk for behavior problems as well as parenting stress [25, 26].

While it is often thought that the history of prenatal alcohol exposure (PAE) would be most easily available to clinicians caring for infants, in fact it can be very difficult to confirm. Women may not be aware of the concerns around prenatal alcohol use and may feel very cautious about sharing this history if they feel there will be judgement of their parenting or if they feel their children may be taken from them. It is incumbent on the clinician to be skilled in approaching this subject and sensitive to the many issues that may contribute to the use of alcohol in pregnancy. Clear and evidence-informed information for clinicians, including short videos with trauma-informed language recommendations on talking with women about alcohol use in pregnancy, is available at the website of the Centre of Excellence for Women's Health at http://bccewh.bc.ca/webinars-and-media/webinars/. Chapter 5 also offers suggestions on how to approach and discuss these issues with pregnant women.

The infant with known PAE should be evaluated in the nursery to identify classically affected infants early and to refer for early intervention programs. Infants

without classical physical features but who show evidence of neonatal abstinence or withdrawal in the context of PAE should be closely followed with appropriate developmental surveillance, as well as medical and behavioral support. Families who care for these infants should be provided with anticipatory guidance on medical and developmental co-morbidity and management.

Practice Points: Infants

1. Assess neonates for abstinence and consider prenatal alcohol exposure and further assessment for FASD in those infants showing Neonatal Abstinence Syndrome.
2. Know how to sensitively and effectively elicit a history of prenatal alcohol use in order to talk with women about providing early intervention for them and their infants in a supportive environment, dealt with more in Chap. 5. Helpful videos including recommended conversational scripts are available at the website of the Centre of Excellence for Women's Health at http://bccewh.bc.ca/webinars-and-media/webinars/.
3. Assess neonates and infants with PAE using a standardized approach for classical facial dysmorphology (shortened palpebral fissures (PF), thin vermillion border of the upper lip, and long smooth philtrum) as well as for growth restriction and associated features. Measurements considered specific to FASD profiles include palpebral fissure lengths <2, standard deviations from the mean on standardized PF charts, a thin upper lip Rank 4 or 5, and smooth philtrum Rank 4 or 5 on the Washington Diagnostic Prevention Network (DPN) Lip Philtrum Guide (images and links to ordering Lip Philtrum Guide available at https://depts.washington.edu/fasdpn/htmls/fas-face.htm).
4. Provide developmental surveillance and assessment for infants with confirmed PAE who are at risk for FASD. Refer for early intervention and therapy including speech and language therapy as infants are at risk for language delays and occupational therapy to address motor delays and support state regulation (feeding, sleeping, and self-soothing). Infants with PAE benefit from early referrals to community infant and child development programs where available.
5. Ensure the family receives early and appropriate support including education on the effects of PAE, parenting strategies, and family supports such as respite and family support groups.

8.2 Preschool Years

Early research on the cognitive profile of FASD was based on the study of preschool children with heavy PAE described by researchers such as Ann Streissguth as early as 1976 [27]. This work was among the earliest to describe the effects of prenatal alcohol exposure on intellectual functioning, motor delays, neurological soft signs, hyperactivity, inattention, and self-stimulating behaviors [27]. Two years later,

Streissguth was able to describe the cognitive range of toddlers, 60% of whom had IQs two or more standard deviations below the mean [28]. Research since that time has continued to describe the persistent impact of PAE on developmental delays in preschoolers.

There are unique considerations in the assessment of preschoolers with PAE, but the primary considerations are understanding their developmental needs in the context of their family and providing early intervention to support optimal developmental progress. Early longitudinal follow-up developmental examinations of preschool children between birth and 4 years of age who have been prenatally exposed to variable amounts of alcohol have shown the persistence of statistically significant behavioral effects at 4 years even after adjustment for confounders such as maternal substance use, education, and family stress [29].

Referrals for assessment of preschoolers with PAE are often complicated by additional environmental considerations including the number of placements for children in care, quality of the home environment, attachment concerns, and readiness of birth families or kin for assessment. In the absence of classical dysmorphology and/or confirmation of alcohol exposure, these considerations may result in a deferred diagnosis until school age or later. However, they should not prevent the clinician from completing a comprehensive physical and developmental assessment focusing on the child's needs and abilities and initiating referrals for early intervention. Specific reasons for early assessment, therefore, include: activation of appropriate referrals, optimizing medical management through diagnosis and management of co-morbid conditions, communication of the child's needs and anticipatory guidance with families and other providers, promotion of developmental and behavioral strategies with home and community, prevention and mitigation of future disabilities, prevention of future alcohol-affected children, and the development of a circle of support around the child and family [30].

Neurobehavioral symptoms of PAE in preschoolers can include cognitive delays, hyperactivity, attention problems, language delays, motor incoordination, atypical sensory responsivity (e.g., delayed auditory processing, tactile sensitivities impacting behavior and managing dailies activities such as dressing, oral sensitivities impacting chewing behaviors, texture or taste sensitivities impacting feeding), weak memory, aggression, and delayed play and social skills [31]. Studies of the developmental performance of preschoolers with heavy PAE have been shown to be substantially lower than non-exposed preschoolers and their parenting and support needs, even with home visiting intervention, are substantially higher [32]. Communication impairments are often significant in the preschool period and contribute to behavioral challenges which often reflect frustration and difficulty coping with environmental demands [33]. Recent work profiling a cohort of preschool children in Alberta, Canada showed patterns of neurobehavioral deficits including deficits in intellectual functioning, executive functioning, language and adaptive functioning [34]. However, despite the predictable deficits in this preschool group and the availability of multidisciplinary assessment, the diagnosis was still deferred in 15% of the children with heavy

alcohol exposure, most of whom lacked classical dysmorphology to reinforce diagnosis and many of whom experienced high rates of adversity, such as multiple and unstable caregiving environments, further complicating assessment [34]. It is critical for clinicians to ensure that even young children are referred for diagnostic assessment to support optimal outcomes and availability of appropriate support throughout their lifespan.

The assessment of preschoolers can be complicated by the more limited availability of formal psychometric measures available in this age group. In fact, while diagnostic guidelines define neurobehavioral domains to be assessed and discuss assessment at various ages, not all domains are easily available for objective assessment at different ages (see Table 8.1). Cognitive functioning in the preschool age group can be assessed using the Weschler Preschool and Primary Scale of Intelligence-4th Edition (WPPSI-IV) in children from 2 years and 6 months to 7 years and 7 months; though in many clinics, psychologists may be less involved with the assessment of the younger preschool groups (more details on psychological testing can be found in Chap. 14 and the approach of a multidisciplinary team in Chap. 22). Questionnaires are frequently used to gain caregiver and teacher evaluations of attention, executive functioning, and adaptive functioning. Communication skills may also be assessed using both clinician-driven interview measures and formal assessments using tools such as the Clinical Examination of Language Fundamentals – Preschool (CELF-P2; more details on language testing can be found in Chap. 14). Domains such as memory, or academic achievement, are generally not directly measured in this age group. However, careful clinician interview will be able to assess caregiver and preschool observations of symptoms such as the need to repeat previously taught information as well as the ability of the child to retain information.

The complexity of needs evident even in the preschool period supports the need for early referral for multidisciplinary developmental assessments, and both functionally and diagnostically informed recommendations and early intervention.

Table 8.1 Neurocognitive domains available for assessment by age (using domains defined in Canadian guidelines for FASD diagnosis [2])

Domains of function	Birth–infancy	Preschool	School age	Adolescence/adult
Neuroanatomy/ Neurophysiology	++++	++++	++	++
Sensory processing	++++	+++	+	+
Motor skills	++++	++++	++	++
Cognition	+	++	++++	++++
Academic achievement	–	++	++++	+++
Language	+	++++	+++	++
Memory	–	+	+++	++++
Attention	+	++	++++	+++
Executive function	–	++	+++	++++
Affect regulation	+	+	++	++++
Adaptive behavior	–	+	+++	++++

Practice Points: Preschoolers

1. Preschoolers with PAE need an early referral for multidisciplinary developmental assessments. These should involve:
   (a) Occupational therapy for assessment of motor, sensory, self-regulation and functional play;
   (b) Speech and language therapy for assessment of communication skills including receptive, expressive, and social or pragmatic communication;
   (c) Developmental assessment by pediatricians and/or psychologists experienced in the developmental and medical assessment of young children;
   (d) Audiological assessment; and
   (e) Ophthalmologic assessment.
2. Advocacy is critical for an appropriate understanding of functional developmental limitations of young children and early intervention resources. Additional support and therapy resources should be in place for preschoolers who may be in daycares or other preschool environments. These additional supports may include child development support, inclusion support in preschool or daycare programs, and respite support for the family. Supports should be informed by an understanding of the impact of prenatal alcohol on the developmental needs and behaviors of young children.
3. Consistency in a medical home should be ensured for children with PAE. Children with PAE may be in foster care and unfortunately may change homes and addresses. Even children who are in their family's care may also change addresses frequently for various reasons. These children need a consistent physician who is knowledgeable in their health throughout their lifespan. Ongoing surveillance for developmental and medical issues should be ensured. Annual monitoring should occur including:
   (a) Growth
   (b) Developmental screening and assessment
   (c) Complete review of systems
   (d) Feeding
   (e) Sleeping
   (f) Seizures
   (g) Social skills, pervasive developmental disorders
   (h) Other behavioral concerns including anxiety, attachment disorders
4. Advocacy for families caring for preschoolers with PAE should include education and anticipatory guidance for their developmental needs, supports regarding the transition to school, and appropriate respite services.

8.3 School-Age Children

The symptom profiles of school-age children with FASD are among the best recognized and studied and form the basis of the brain domains described in most diagnostic guidelines [2, 7, 35]. These guidelines generally describe central nervous

system domains including both structural, such as microcephaly and functional, including both cognitive and behavioral domains [2, 7, 35]. These cognitive and behavioral domains include intellectual and academic functioning, memory, attention, executive functioning, communication, self-regulation including affect regulation and sensorimotor functioning (for descriptions of the various domains, see Table 8.2) [2, 7, 35]. Recognizing inefficient or atypical cognitive processing, neural activation and integration of information is often key to understanding the deficits produced by PAE [36]. Evidence of impairment (≤2 SD below the mean or 1–1.5 SD difference between major subdomains) in three or more of the domains as described in the Canadian Guidelines is required for diagnosis [2]. In the Diagnostic Statistical Manual 5 (DSM-5), these domains are further clustered into "neurocognitive functioning" (including evidence of global intellectual dysfunction, learning difficulty, memory impairment, impairment in executive functioning, and difficulties with visual-spatial reasoning), self-regulation (includes impairments in mood and behavior regulation, attention deficit, and impulse control) and adaptive functioning (communication or social communication impairment, impairments in motor skills, and impairments in completing daily living skills) [37]. In general, a comprehensive assessment of each of these areas is recommended as part of the diagnostic evaluation (DSM V criteria are discussed further in Chap. 10).

Table 8.2 Description of cognitive and behavioral domains

Sensory processing	Refers to the neurologically based processing of environmental information received through senses including touch, vision, hearing, and movement
Self-regulation	Broadly, the ability to adjust one's emotions, attention, and behavioral responses appropriately for the condition or environment
Cognition	Reflects typical areas of intellectual development and functioning, usually including major domain areas of verbal and non-verbal intelligence, fluid reasoning along with a number of subdomains
Memory	Complex concept defined in many different ways and may include concepts of visual memory, verbal memory, long- and short-term memory
Academic achievement	Reflects academic functioning in basic areas including reading, spelling, and mathematics, and is usually in keeping with cognitive abilities
Language	Refers to one's ability to understand others communication (receptive language) and express oneself using appropriate language structures and content (expressive language)
Executive functioning	Typically includes concepts of working memory, inhibition/impulse control, hyperactivity, planning and problem solving or shifting and cognitive flexibility; difficulties would reflect impairments in integrating knowledge and basic cognitive processes such as attention, memory, and perception, in order to successfully accomplish tasks
Attention	Described by the ability to sustain or selectively focus and resist distraction
Affect regulation	Affect regulation: a new concept in Canadian FASD Guidelines [2], intended to reflect the regulation of mood and emotion; dysregulation would be defined as diagnostic meeting criteria for anxiety or depression; severity and age need to be considered as this concept is intended to describe adolescence or adults
Adaptive behaviors	Typically reflect the application of day to day functioning in conceptual, social, and practical areas, often including functions such as managing personal care, social interactions, and financial and home management

Researchers have been studying the neurocognitive profiles of children with FASD for at least the last three decades with consistent results. In 1985, Streissguth published a follow-up study of the 11 children first diagnosed in the United States with fetal alcohol syndrome with results showing persistent intellectual and adaptive deficits [38]. Streissguth continued her examination of the neurobehavioral functioning of alcohol-affected children, describing patterns of attention and memory deficits, deficits in cognitive processing and integration, executive dysfunction, and difficulties with visual-spatial organization and mathematics, even at lower levels (so-called "social drinking") of alcohol exposure [39]. Later longitudinal study of school-age children with PAE demonstrated dose-dependent effects of measures of neurobehavioral functioning from birth to 14 years including difficulties with attention, processing speed, and learning problems especially in arithmetic [40]. A 2009 review of the neurocognitive profiles in children with FASD described borderline to low average cognitive functioning, slow information processing, executive dysfunction, deficits in memory, as well as language, and social communication impairments [41]. In 2010, the clinical school-age cohort of the Washington DPN also showed similar cognitive behavioral profiles including cognitive delays, executive dysfunction, difficulties with abstract thinking, memory and judgement, atypical sensory motor integration, and adaptive dysfunction [42]. Other described features of PAE that impact school performance include deficits in fine motor and visual motor abilities that impact functional handwriting [43–45]. Work examining the gross motor functioning and sensory integration of children with FASD has also shown significant impairments in sensory processing, balance, coordination, and ball skills [46–49].

The academic achievement of children with PAE is significantly impacted and is often the trigger for the assessment of children who have not previously been identified. A specific academic profile has been recently suggested with primary deficits in mathematical reasoning greater than spelling and word reading; however, this is not a universal pattern [50]. Spelling performance has also been identified as an area of impairment, with deficits in working memory contributing to these difficulties [51]. Deficits in math, along with reading and spelling, have been shown to persist into adolescence [52]. Teachers are often involved in school-age identification, particularly of children not already identified through special education funding streams based on the diagnosis of intellectual disability. Specific academic challenges have been described not only in mathematics, but also in reading, writing, and spelling [50, 51, 53–56]. The behavioral challenges resulting from impulsivity, poor executive functioning, poor judgement, and cognitive rigidity are seen in interactions with teachers and peers and comorbid mental health challenges of ADHD, mood disorders, and conduct disorders provide severe challenges to group educational programming [50].

Atypical communication and social behaviors of children with FASD have been described by several authors, including concerns regarding possible autistic overlap in the social profiles [57–60]. Communication deficits have been recognized for over two decades and children with FASD/PAE are commonly referred to speech and language pathologists for treatment [61, 62]. An interesting observational study of the social behavior of children with FASD not only noted more passive/disengaged and irrelevant behaviors compared to classroom peers, but also showed more but shorter instances of prosocial behaviors [63]. Other work has described typical social behavior in children with FASD, but fewer restricted and repetitive behaviors

distinguishing FASD from the typical presentation on the autism spectrum [58]. Sensory processing differences and executive dysfunction may further interact with social perception and interactional skills producing atypical social behaviors [64]. Further deficits in risk-taking and affecting regulation in combination with social dysmaturity also negatively impact social relatedness [65] (overlapping neurodevelopmental findings are also discussed in Chap. 12). Referrals of children with PAE often describe challenges in peer interactions and may include aggressive or antisocial behaviors, but careful clinical history taking often reveals vulnerabilities in social understanding and a desire for acceptance by peers that may be inconsistent with an autistic phenotype. Early diagnosis has been shown to support improved social integration over time [66].

Children with FASD benefit from additional support in schools addressing their cognitive, communication, and behavioral needs. Classroom adaptations may include: environmental modifications to reduce visual clutter and structure personal space, graphically simple and clear visual prompts, opportunities for physical breaks or short exercise programs distributed through the day, role play and supported social skills opportunities [67]. Behavior management strategies in the classroom may include the use of behavioral modification with immediate consequences for behaviors, social skills teaching and scripting, and specific teaching of skills to self-monitor and organize behavior [68]. Effective educational interventions are based on a complete understanding of the cognitive and behavioral needs of the child, which is informed by a thorough diagnostic assessment including the description of strengths and challenges in each brain domain [67, 68]. The physician's role includes referring for appropriate diagnostic evaluation, communication of the results of that assessment to the school and educational therapists, ongoing surveillance for medical and behavioral co-morbidity, and ongoing treatment as needed [30]. Children who will ultimately be responsible for their own care and treatment decisions will need to become aware of their diagnosis and understand their profile of strengths and challenges. The physician has a responsibility to work with the child, their family/guardians, and community team to facilitate this understanding and provide ongoing care and treatment [30, 69] (Chaps. 8 and 29, respectively, address recognizing and managing the FASD child within a classroom setting).

Practice Points: School-Age Children

1. School-age children are the commonest age group seen for diagnosis in FASD clinics; however, their symptoms have often been present in the preschool period. Consider reviewing prenatal exposure histories with parents of school-age children who are presenting with learning difficulties in combination with behavioral challenges; PAE history may have been missed or parents may have not disclosed this information earlier and may be more ready to disclose this history in the context of a trusted professional relationship when they are more aware of the school difficulties their child is experiencing, concerned regarding the etiology of these difficulties, and understand supports available.

2. Teachers are excellent sources of information on behavioral and cognitive functioning within the school environment. Schools may be able to provide additional information and assessments of academic functioning at the request of the clinician.
3. Comprehensive diagnostic assessment informs specific environmental modifications and individualized educational programming. The physician should use the details of the assessment to help schools and families understand patterns of strengths and weaknesses of the child, inform Individualized Educational Planning, and identify specific supports required.
4. Understanding the differences in environmental demands of home and school may help explain differences in the functioning of the child across different settings.
5. Interventions to support a child's strength are often overlooked and are often as important to supporting the child if not more so. Many children have found that involvement in sports, arts, music, caring for animals, cooking, or other activities have found new areas for social skill development and have also found areas for them to pursue in long-term career planning
6. Ongoing surveillance for medical and behavioral co-morbidity, with appropriate evaluation and treatment, is critical to supporting the success of the child. Medical surveillance should review systems annually including review for areas of head injuries or seizures, metabolic monitoring as needed (regular checks of blood pressure, blood sugar, and lipid profile), musculoskeletal problems especially for those in sports, chronic pain, ADHD, social communication deficits, mood disorder (anxiety or depression), other psychiatric disorder, feeding, sleeping, and disturbances of self-care such as toileting and dressing.
7. Providing and maintaining longitudinal care of the child within their family and through their transition to adolescence and adulthood is critical to providing appropriate anticipatory guidance, early intervention, and treatment. Families may benefit from specialized parenting supports or may feel stressed and benefit from family therapy or respite supports. Siblings may also feel stressed and benefit from additional support and monitoring for the need for family therapy.

8.4 Adolescents and Adults

As early as 1991, in an article in JAMA Streissguth elegantly observed that "Fetal alcohol syndrome is not just a childhood disorder; there is a predictable long term progression of the disorder into adulthood, in which maladaptive behaviours present the greatest challenge to management." [70]. Olson et al. [71] were among the earliest to study adolescent patients with FAS and compare their cognitive profiles to control adolescents without PAE, showing in their patients' persistent cognitive dysfunction in the regulation of attention, memory, learning, poor cognitive flexibility, poor visual spatial skills, and adaptive dysfunction including poorer social

competence [71]. Consistently, a German cohort later showed prominent emotional and behavioral difficulties persisting into adulthood including attentional problems, aggression, delinquency, independent of IQ and of formal diagnosis [72]. Prenatal alcohol exposure, even without FASD, has also been shown to affect cognitive functioning and academic achievement [52].

There are many symptoms that continue to be apparent in adolescence and adulthood and that continue to impact functioning. Some of these include attention deficits, working memory deficits, learning disability, impulsivity and distractibility [52, 73–75]. In addition, cognitive gaps in the thinking process include difficulty in forming associations and predicting, difficulties with executive functioning and abstract reasoning, cause and effect, and generalization [75–77]. Adults with FASD who do not meet IQ criteria for intellectual disability often struggle with educational and occupational functioning, out of keeping with apparent cognitive potential, and often do not meet threshold requirements for community supports. In a 1997 study on a group of young adults with IQ's in the borderline to the average range, testing clearly demonstrated greater deficits on neuropsychological measures of complex attention, verbal learning, and executive functioning, than would have been predicted based on their measured IQ [78]. More recent work has shown that PAE not only affects adaptive functioning, but also affects role transition into adult life including less school completion and more likely unemployment [79].

Characteristic difficulties, however, include the significance of adaptive and social dysfunction out of keeping with cognitive potential [77]. Severe communication gaps are often overlooked and deficits in social communication significantly impact functioning [75]. Specific challenges in social functioning may include difficulties understanding the rules of social interactions, poor social judgement, difficulties understanding social cues, and even understanding situations that may be embarrassing to others [75]. Difficulty with judgment in day-to-day functioning can encompass difficulty in managing time, money, and schedules; difficulty in evaluating situational risk; and difficulty in understanding safety and danger. Coupled with social dysmaturity, these difficulties can lead to a heightened risk of exploitation and victimization [79].

There is also substantial risk of mental health disorders in individuals with FASD [77, 80]. These include high rates of internalizing and externalizing disorders in adolescents and adults [81]. While these have been typically described as a "secondary disability," it is increasingly recognized that they reflect not only the collective experience of genetic predisposition and environmental influences, but more recently are being understood as a primary effect of PAE's dysregulating effect on the stress controlling hypothalamic–pituitary–adrenal (HPA) axis [81–84]. This may also explain the increased risk for substance use disorders frequently described in individuals with FASD [85]. Finally, it is also suggested that adults with PAE may experience accelerated cognitive decline with aging, in part related to lower cognitive reserve capacity [86].

Recent literature suggests that adults with FASD may be more susceptible to other health conditions as a consequence of the impact of alcohol on early fetal programming [86, 87]. Work on immune and neuroendocrine dysfunction related to prenatal alcohol exposure is suggesting an altered immune response to viral infections and an increased risk of chronic diseases such as hypertension, diabetes

mellitus, diabetes insipidus, arthritis, hypertriglyceridemia and metabolic syndrome [88–95]. Prenatal alcohol exposure is also being linked to an increased risk of development of certain types of cancer [96, 97]. While the impact of other environmental influences on fetal programming is acknowledged, more research is needed on the impact of PAE on adult disease outcomes and potential ameliorating strategies [98].

While adults with FASD are at substantially higher risk for significant life challenges, early diagnosis even in adulthood is associated with improved outcomes. In a Life History Interview study of a cohort of 415 adolescent and adult patients diagnosed with FASD, Streissguth and colleagues showed the presence of significant life challenges in the cohort, with a life span prevalence of 61% for disrupted school experiences, 60% for trouble with the law, 50% for confinement (in detention, jail, prison, or a psychiatric or alcohol/drug inpatient setting), 49% for inappropriate sexual behaviors on repeated occasions, and 35% for alcohol/drug problems [1]. The risk for the development of alcohol use problems in young adults with PAE was also demonstrated in a 21-year longitudinal analysis regressing measures of parental alcohol use against self-reports of alcohol use and a measure of alcohol-related problems and dependence [99]. However, recent work has shown that lower substance use among adults with FASD, along with early diagnosis and intervention, is associated with more positive outcomes [100].

Practice Points: Adolescents and Adults

1. Advocate for access to FASD diagnosis in adolescents and adults. The challenges may include accessing accurate prenatal alcohol histories and the complicating factors of trauma histories and environmental stressors. It is important, however, to still consider FASD in this age group, especially where adaptive dysfunction is out of keeping with cognitive functioning and has not been addressed with typical interventions.
2. Screen for substance use in adolescents and adults, particularly in child bearing years and pregnancy, and refer early for appropriate treatment and support.
3. Ensure regular medical follow-up and screening of chronic disease including immune and metabolic disorders.
4. Screen regularly for psychiatric disorders including mood disorders, substance abuse disorder, and early onset cognitive dysfunction.
5. Support independence through coaching and community integration. It is very important for clinicians to advocate for their patients and families living with FASD by providing accurate information on prenatal alcohol exposure and FASD and participating in the development of appropriate community support systems.
6. Clinicians can help reduce stigma by using person-first language (e.g., "Person with FASD" not "FASD person") and by using neutral and objective language in talking with families and writing reports (e.g., "Prenatal alcohol exposure is confirmed," not "woman abused alcohol in pregnancy"). A good source of recommendations for language use is the FASD Language Guide available at http://www.fasdcoalition.ca/looking-after-each-other-project/fasd-language-guide/.

8.5 Conclusions

Despite these difficulties, early diagnosis remains one of the strongest factors associated with improved long-term outcomes, including lower rates of disrupted school experiences, reduced inappropriate behavior, reduced legal involvement, reduced later substance abuse and lower rates of psychiatric comorbidity [1]. The comorbidity of FASD can be significant and contributes to the high costs associated with this disorder [80, 101]. Furthermore, the productivity losses in adults with FASD associated with lack of appropriate community programs and supports are also significant [102]. For these reasons, it is recommended that individuals with prenatal alcohol exposure and developmental concerns are seen for diagnostic assessment early to provide appropriate understanding and support for the individual with FASD, their family, and the community. The value of an FASD assessment and diagnosis has been demonstrated showing a high degree of satisfaction with the process of diagnosis and ability to access supports, particularly for younger children, and likely reflecting the more limited resources available for adults [66, 103]. It is critical, therefore, to recognize patterns of presentation of FASD at all ages from infancy to adulthood and refer for appropriate diagnostic assessment in order to advocate for individual, family, and community understanding of behavior and establish comprehensive and appropriate supports for the affected individual.

References

1. Streissguth AP, Bookstein FL, Barr HM, Sampson PD, O'Malley K, Young JK. Risk factors for adverse life outcomes in fetal alcohol syndrome and fetal alcohol effects. J Dev Behav Pediatr. 2004;25(4):228–38.
2. Cook JL, Green CR, Lilley CM, Anderson SM, Baldwin ME, Chudley AE, et al. Fetal alcohol spectrum disorder: a guideline for diagnosis across the lifespan. CMAJ. 2016;188(3):191–7.
3. Chudley AE, Conry J, Cook JL, Loock C, Rosales T, LeBlanc N, et al. Fetal alcohol spectrum disorder: Canadian guidelines for diagnosis. CMAJ. 2005;172(5 Suppl):S1–S21.
4. Astley SJ, Clarren SK. A fetal alcohol syndrome screening tool. Alcohol Clin Exp Res. 1995;19(6):1565–71.
5. Astley SJ, Clarren SK. Measuring the facial phenotype of individuals with prenatal alcohol exposure: correlations with brain dysfunction. Alcohol Alcohol. 2001;36(2):147–59.
6. Hoyme HE, May PA, Kalberg WO, Kodituwakku P, Gossage JP, Trujillo PM, et al. A practical clinical approach to diagnosis of fetal alcohol spectrum disorders: clarification of the 1996 institute of medicine criteria. Pediatrics. 2005;115(1):39–47.
7. Hoyme HE, Kalberg WO, Elliott AJ, Blankenship J, Buckley D, Marais AS, et al. Updated clinical guidelines for diagnosing fetal alcohol spectrum disorders. Pediatrics. 2016;138(2):e20154256.
8. Leibson T, Neuman G, Chudley AE, Koren G. The differential diagnosis of fetal alcohol spectrum disorder. J Popul Ther Clin Pharmacol. 2014;21(1):e1–e30.
9. Jones KL, Hoyme HE, Robinson LK, Del Campo M, Manning MA, Prewitt LM, et al. Fetal alcohol spectrum disorders: extending the range of structural defects. Am J Med Genet A. 2010;152A(11):2731–5.
10. Coles CD. Impact of prenatal alcohol exposure on the newborn and the child. Clin Obstet Gynecol. 1993;36(2):255–66.
11. Coles CD, Smith IE, Falek A. Prenatal alcohol exposure and infant behavior: immediate effects and implications for later development. Adv Alc Subst Abuse. 1987;6(4):87–104.

12. Streissguth AP, Martin DC, Martin JC, Barr HM. The Seattle longitudinal prospective study on alcohol and pregnancy. Neurobehav Toxicol Teratol. 1981;3(2):223–33.
13. Hepper PG, Dornan JC, Little JF. Maternal alcohol consumption during pregnancy may delay the development of spontaneous fetal startle behaviour. Physiol Behav. 2005;83(5):711–4.
14. Little JF, Hepper PG, Dornan JC. Maternal alcohol consumption during pregnancy and fetal startle behaviour. Physiol Behav. 2002;76(4–5):691–4.
15. Havlicek V, Childaeva R. E.E.G. component of fetal alcohol syndrome. Lancet. 1976;2(7983):477.
16. Havlicek V, Childiaeva R, Chernick V. EEG frequency spectrum characteristics of sleep states in infants of alcoholic mothers. Neuropadiatrie. 1977;8(4):360–73.
17. Chernick V, Childiaeva R, Ioffe S. Effects of maternal alcohol intake and smoking on neonatal electroencephalogram and anthropometric measurements. Am J Obstet Gynecol. 1983;146(1):41–7.
18. Haley DW, Handmaker NS, Lowe J. Infant stress reactivity and prenatal alcohol exposure. Alcohol Clin Exp Res. 2006;30(12):2055–64.
19. Mesa DA, Kable JA, Coles CD, Jones KL, Yevtushok L, Kulikovsky Y, et al. The use of cardiac orienting responses as an early and scalable biomarker of alcohol-related neurodevelopmental impairment. Alcohol Clin Exp Res. 2017;41(1):128–38.
20. Appleton AA, Murphy MA, Koestler DC, Lesseur C, Paquette AG, Padbury JF, et al. Prenatal programming of infant neurobehaviour in a healthy population. Paediatr Perinat Epidemiol. 2016;30(4):367–75.
21. Coles CD, Kable JA, Drews-Botsch C, Falek A. Early identification of risk for effects of prenatal alcohol exposure. J Stud Alcohol. 2000;61(4):607–16.
22. Jacobson JL, Jacobson SW, Sokol RJ, Martier SS, Ager JW, Kaplan-Estrin MG. Teratogenic effects of alcohol on infant development. Alcohol Clin Exp Res. 1993;17(1):174–83.
23. Golden NL, Sokol RJ, Kuhnert BR, Bottoms S. Maternal alcohol use and infant development. Pediatrics. 1982;70(6):931–4.
24. Testa M, Quigley BM, Eiden RD. The effects of prenatal alcohol exposure on infant mental development: a meta-analytical review. Alcohol Alcohol. 2003;38(4):295–304.
25. Bates JE, Freeland CA, Lounsbury ML. Measurement of infant difficultness. Child Dev. 1979;50(3):794–803.
26. Atella LD, DiPietro JA, Smith BA, St James-Roberts I. More than meets the eye: parental and infant contributors to maternal and paternal reports of early infant difficultness. Parenting. 2003;3(4):265–84.
27. Streissguth AP. Psychologic handicaps in children with the fetal alcohol syndrome. Ann N Y Acad Sci. 1976;273:140–5.
28. Streissguth AP, Herman CS, Smith DW. Intelligence, behavior, and dysmorphogenesis in the fetal alcohol syndrome: a report on 20 patients. J Pediatr. 1978;92(3):363–7.
29. Streissguth AP, Barr HM, Martin DC. Alcohol exposure in utero and functional deficits in children during the first four years of life. CIBA Found Symp. 1984;105:176–96.
30. Hanlon-Dearman A, Green CR, Andrew G, LeBlanc N, Cook JL. Anticipatory guidance for children and adolescents with Fetal Alcohol Spectrum Disorder (FASD): practice points for primary health care providers. J Popul Ther Clin Pharmacol. 2015;22(1):e27–56.
31. Streissguth AP, Sampson PD, Barr HM, Clarren SK, Martin DC. Studying alcohol teratogenesis from the perspective of the fetal alcohol syndrome: methodological and statistical issues. Ann N Y Acad Sci. 1986;477:63–86.
32. Kartin D, Grant TM, Streissguth AP, Sampson PD, Ernst CC. Three-year developmental outcomes in children with prenatal alcohol and drug exposure. Pediatr Phys Ther. 2002;14(3):145–53.
33. Proven S, Ens C, Beaudin PG. The language profile of school-aged children with fetal alcohol spectrum disorder (FASD). Can J Speech Lang Pathol Audiol. 2014;37(4):268–79.
34. McLachlan K, Andrew G, Pei J, Rasmussen C. Assessing FASD in young children: exploring clinical complexities and diagnostic challenges. J Popul Ther Clin Pharmacol. 2015;22(1):e108–e24.

35. Astley SJ. Diagnostic guide for fetal alcohol spectrum disorders: the 4-digit diagnostic code. Seattle, WA: University of Washington; 2004.
36. Moore EM, Migliorini R, Infante MA, Riley EP. Fetal alcohol spectrum disorders: recent neuroimaging findings. Curr Dev Disord Rep. 2014;1(3):161–72.
37. American Psychiatric Association. Diagnostic and statistical manual of mental disorders. 5th ed. Arlington, VA: American Psychiatric Publishing; 2013.
38. Streissguth AP, Clarren SK, Jones KL. Natural history of the fetal alcohol syndrome: a 10-year follow-up of eleven patients. Lancet. 1985;2(8446):85–91.
39. Streissguth AP, Bookstein FL, Sampson PD, Barr HM. Neurobehavioral effects of prenatal alcohol: Part III. PLS analyses of neuropsychologic tests. Neurotoxicol Teratol. 1989;11(5):493–507.
40. Streissguth AP, Barr HM, Sampson PD, Bookstein FL. Prenatal alcohol and offspring development: the first fourteen years. Drug Alcohol Depend. 1994;36(2):89–99.
41. Kodituwakku PW. Neurocognitive profile in children with fetal alcohol spectrum disorders. Dev Disab Res Rev. 2009;15(3):218–24.
42. Astley SJ. Profile of the first 1,400 patients receiving diagnostic evaluations for fetal alcohol spectrum disorder at the Washington State Fetal Alcohol Syndrome Diagnostic & Prevention Network. Can J Clin Pharmacol. 2010;17(1):e132–64.
43. Duval-White CJ, Jirikowic T, Rios D, Deitz J, Olson HC. Functional handwriting performance in school-age children with fetal alcohol spectrum disorders. Am J Occup Ther. 2013;67(5):534–42.
44. Doney R, Lucas BR, Jirikowic T, Tsang TW, Watkins RE, Sauer K, et al. Graphomotor skills in children with prenatal alcohol exposure and fetal alcohol spectrum disorder: a population-based study in remote Australia. Aust Occup Ther J. 2017;64(1):68–78.
45. Doney R, Lucas BR, Jones T, Howat P, Sauer K, Elliott EJ. Fine motor skills in children with prenatal alcohol exposure or fetal alcohol spectrum disorder. J Dev Behav Pediatr. 2014;35(9):598–609.
46. Lucas BR, Latimer J, Doney R, Watkins RE, Tsang TW, Hawkes G, et al. Gross motor performance in children prenatally exposed to alcohol and living in remote Australia. J Paediatr Child Health. 2016;52(8):814–24.
47. Jirikowic TL, McCoy SW, Lubetzky-Vilnai A, Price R, Ciol MA, Kartin D, et al. Sensory control of balance: a comparison of children with fetal alcohol spectrum disorders to children with typical development. J Popul Ther Clin Pharmacol. 2013;20(3):e212–28.
48. Jirikowic T, Olson HC, Kartin D. Sensory processing, school performance, and adaptive behavior of young school-age children with fetal alcohol spectrum disorders. Phys Occup Ther Pediatr. 2008;28(2):117–36.
49. Hansen KD, Jirikowic T. A comparison of the sensory profile and sensory processing measure home form for children with fetal alcohol spectrum disorders. Phys Occup Ther Pediatr. 2013;33(4):440–52.
50. Glass L, Moore EM, Akshoomoff N, Jones KL, Riley EP, Mattson SN. Academic difficulties in children with prenatal alcohol exposure: presence, profile, and neural correlates. Alcohol Clin Exp Res. 2017;41:1024.
51. Glass L, Graham DM, Akshoomoff N, Mattson SN. Cognitive factors contributing to spelling performance in children with prenatal alcohol exposure. Neuropsychology. 2015;29(6):817–28.
52. Howell KK, Lynch ME, Platzman KA, Smith GH, Coles CD. Prenatal alcohol exposure and ability, academic achievement, and school functioning in adolescence: a longitudinal follow-up. J Pediatr Psychol. 2006;31(1):116–26.
53. Coles CD, Kable JA, Taddeo E. Math performance and behavior problems in children affected by prenatal alcohol exposure: intervention and follow-up. J Dev Behav Pediatr. 2009;30(1):7–15.
54. Crocker N, Riley EP, Mattson SN. Visual-spatial abilities relate to mathematics achievement in children with heavy prenatal alcohol exposure. Neuropsychology. 2015;29(1):108–16.

55. O'Leary CM, Taylor C, Zubrick SR, Kurinczuk JJ, Bower C. Prenatal alcohol exposure and educational achievement in children aged 8-9 years. Pediatrics. 2013;132(2):e468–75.
56. Kopera-Frye K, Dehaene S, Streissguth AP. Impairments of number processing induced by prenatal alcohol exposure. Neuropsychologia. 1996;34(12):1187–96.
57. Jargin SV. Attention Deficit Hyperactivity (ADHD) and Autism Spectrum Disorder (ASD): on the role of alcohol and societal factors. Int J High Risk Behav Addict. 2013;1(4):194–5.
58. Stevens SA, Nash K, Koren G, Rovet J. Autism characteristics in children with fetal alcohol spectrum disorders. Child Neuropsychol. 2013;19(6):579–87.
59. Nanson JL. Autism in fetal alcohol syndrome: a report of six cases. Alcohol Clin Exp Res. 1992;16(3):558–65.
60. Fombonne E. Is exposure to alcohol during pregnancy a risk factor for autism? J Autism Dev Disord. 2002;32(3):243.
61. Church MW, Abel EL. Fetal alcohol syndrome. Hearing, speech, language, and vestibular disorders. Obstet Gynecol Clin N Am. 1998;25(1):85–97.
62. Popova S, Lange S, Burd L, Shield K, Rehm J. Cost of speech-language interventions for children and youth with foetal alcohol spectrum disorder in Canada. Int J Speech Lang Pathol. 2014;16(6):571–81.
63. Olswang LB, Svensson L, Astley S. Observation of classroom social communication: do children with fetal alcohol spectrum disorders spend their time differently than their typically developing peers? J Speech Lang Hear Res. 2010;53(6):1687–703.
64. Kully-Martens K, Denys K, Treit S, Tamana S, Rasmussen C. A review of social skills deficits in individuals with fetal alcohol spectrum disorders and prenatal alcohol exposure: profiles, mechanisms, and interventions. Alcohol Clin Exp Res. 2012;36(4):568–76.
65. Kully-Martens K, Treit S, Pei J, Rasmussen C. Affective decision-making on the Iowa gambling task in children and adolescents with fetal alcohol spectrum disorders. J Int Neuropsychol Soc. 2013;19(2):137–44.
66. Alex K, Feldmann R. Children and adolescents with fetal alcohol syndrome (FAS): better social and emotional integration after early diagnosis. Klin Padiatr. 2012;224(2):66–71.
67. Millar J, Thompson J, Schwab D, Hanlon-Dearman A, Goodman D, Koren G, Masotti P. Educating students with FASD: linking policy, research and practice. J Res Spec Educ Needs. 2014;17:3–17.
68. Green J. Fetal alcohol spectrum disorders: understanding the effects of prenatal alcohol exposure and supporting students. J Sch Health. 2007;77(3):103–8.
69. Todorow M, Paris K, Fantus E. Ethical considerations when communicating a diagnosis of a fetal alcohol spectrum disorder to a child. J Popul Ther Clin Pharmacol. 2012;19(3):e361–8.
70. Streissguth AP, Aase JM, Clarren SK, Randels SP, LaDue RA, Smith DF. Fetal alcohol syndrome in adolescents and adults. JAMA. 1991;265(15):1961–7.
71. Olson HC, Feldman JJ, Streissguth AP, Sampson PD, Bookstein FL. Neuropsychological deficits in adolescents with fetal alcohol syndrome: clinical findings. Alcohol Clin Exp Res. 1998;22(9):1998–2012.
72. Spohr HL, Willms J, Steinhausen HC. Fetal alcohol spectrum disorders in young adulthood. J Pediatr. 2007;150(2):175–9, 9.e1.
73. Louth EL, Bignell W, Taylor CL, Bailey CD. Developmental ethanol exposure leads to long-term deficits in attention and its underlying prefrontal circuitry. eNeuro. 2016;3(5):ENEURO.0267-16.2016.
74. Rangmar J, Hjern A, Vinnerljung B, Stromland K, Aronson M, Fahlke C. Psychosocial outcomes of fetal alcohol syndrome in adulthood. Pediatrics. 2015;135(1):e52–8.
75. Rangmar J, Sandberg AD, Aronson M, Fahlke C. Cognitive and executive functions, social cognition and sense of coherence in adults with fetal alcohol syndrome. Nord J Psychiatry. 2015;69(6):472–8.
76. Tamana S, Pei J, Massey D, Massey V, Rasmussen C. Neuropsychological impairments and age-related differences in children and adolescents with fetal alcohol spectrum disorders. J Popul Ther Clin Pharmacol. 2014;21(2):e167–80.

77. Chudley AE, Kilgour AR, Cranston M, Edwards M. Challenges of diagnosis in fetal alcohol syndrome and fetal alcohol spectrum disorder in the adult. Am J Med Genet C: Semin Med Genet. 2007;145C(3):261–72.
78. Kerns KA, Don A, Mateer CA, Streissguth AP. Cognitive deficits in nonretarded adults with fetal alcohol syndrome. J Learn Disabil. 1997;30(6):685–93.
79. Lynch ME, Kable JA, Coles CD. Prenatal alcohol exposure, adaptive function, and entry into adult roles in a prospective study of young adults. Neurotoxicol Teratol. 2015;51:52–60.
80. Popova S, Lange S, Shield K, Mihic A, Chudley AE, Mukherjee RA, et al. Comorbidity of fetal alcohol spectrum disorder: a systematic review and meta-analysis. Lancet. 2016;387(10022):978–87.
81. Pei J, Denys K, Hughes J, Rasmussen C. Mental health issues in fetal alcohol spectrum disorder. J Ment Health. 2011;20(5):438–48.
82. Famy C, Streissguth AP, Unis AS. Mental illness in adults with fetal alcohol syndrome or fetal alcohol effects. Am J Psychiatry. 1998;155(4):552–4.
83. Lan N, Chiu MP, Ellis L, Weinberg J. Prenatal alcohol exposure and prenatal stress differentially alter glucocorticoid signaling in the placenta and fetal brain. Neuroscience. 2017;342:167–79.
84. Sliwowska JH, Song HJ, Bodnar T, Weinberg J. Prenatal alcohol exposure results in long-term serotonin neuron deficits in female rats: modulatory role of ovarian steroids. Alcohol Clin Exp Res. 2014;38(1):152–60.
85. Uban KA, Comeau WL, Bodnar T, Yu WK, Weinberg J, Galea LA. Amphetamine sensitization and cross-sensitization with acute restraint stress: impact of prenatal alcohol exposure in male and female rats. Psychopharmacology. 2015;232(10):1705–16.
86. Moore EM, Riley EP. What happens when children with fetal alcohol spectrum disorders become adults? Curr Dev Disord Rep. 2015;2(3):219–27.
87. Barker DJ. Sir Richard Doll Lecture. Developmental origins of chronic disease. Public Health. 2012;126(3):185–9.
88. Zhang X, Lan N, Bach P, Nordstokke D, Yu W, Ellis L, et al. Prenatal alcohol exposure alters the course and severity of adjuvant-induced arthritis in female rats. Brain Behav Immun. 2012;26(3):439–50.
89. McGill J, Meyerholz DK, Edsen-Moore M, Young B, Coleman RA, Schlueter AJ, et al. Fetal exposure to ethanol has long-term effects on the severity of influenza virus infections. J Immunol. 2009;182(12):7803–8.
90. Chen L, Nyomba BL. Effects of prenatal alcohol exposure on glucose tolerance in the rat offspring. Metabolism. 2003;52(4):454–62.
91. Pennington JS, Shuvaeva TI, Pennington SN. Maternal dietary ethanol consumption is associated with hypertriglyceridemia in adult rat offspring. Alcohol Clin Exp Res. 2002;26(6):848–55.
92. Knee DS, Sato AK, Uyehara CF, Claybaugh JR. Prenatal exposure to ethanol causes partial diabetes insipidus in adult rats. Am J Phys Regul Integr Comp Phys. 2004;287(2):R277–83.
93. Gray SP, Kenna K, Bertram JF, Hoy WE, Yan EB, Bocking AD, et al. Repeated ethanol exposure during late gestation decreases nephron endowment in fetal sheep. Am J Phys Regul Integr Comp Phys. 2008;295(2):R568–74.
94. Xia LP, Shen L, Kou H, Zhang BJ, Zhang L, Wu Y, et al. Prenatal ethanol exposure enhances the susceptibility to metabolic syndrome in offspring rats by HPA axis-associated neuroendocrine metabolic programming. Toxicol Lett. 2014;226(1):98–105.
95. Vaiserman AM. Early-life exposure to substance abuse and risk of type 2 diabetes in adulthood. Curr Diab Rep. 2015;15(8):48.
96. Polanco TA, Crismale-Gann C, Reuhl KR, Sarkar DK, Cohick WS. Fetal alcohol exposure increases mammary tumor susceptibility and alters tumor phenotype in rats. Alcohol Clin Exp Res. 2010;34(11):1879–87.
97. Chiappelli F, Kung MA, Tio DL, Tritt SH, Yirmiya R, Taylor AN. Fetal alcohol exposure augments the blunting of tumor necrosis factor production in vitro resulting from in vivo priming with lipopolysaccharide in young adult male but not female rats. Alcohol Clin Exp Res. 1997;21(8):1542–6.

98. Lunde ER, Washburn SE, Golding MC, Bake S, Miranda RC, Ramadoss J. Alcohol-induced developmental origins of adult-onset diseases. Alcohol Clin Exp Res. 2016;40(7):1403–14.
99. Baer JS, Sampson PD, Barr HM, Connor PD, Streissguth AP. A 21-year longitudinal analysis of the effects of prenatal alcohol exposure on young adult drinking. Arch Gen Psychiatry. 2003;60(4):377–85.
100. Currie BA, Hoy J, Legge L, Temple VK, Tahir M. Adults with fetal alcohol spectrum disorder: factors associated with positive outcomes and contact with the criminal justice system. J Popul Ther Clin Pharmacol. 2016;23(1):e37–52.
101. Popova S, Lange S, Burd L, Rehm J. The economic burden of fetal alcohol spectrum disorder in Canada in 2013. Alcohol Alcohol. 2016;51(3):367–75.
102. Easton B, Burd L, Sarnocinska-Hart A, Rehm J, Popova S. The cost of lost productivity due to fetal alcohol spectrum disorder-related premature mortality. J Popul Ther Clin Pharmacol. 2015;22(1):e3–8.
103. Astley SJ. The value of a FASD diagnosis (2013). J Popul Ther Clin Pharmacol. 2014;21(1):e81–e105.

Identifying the Child with FASD in Educational Settings

9

Carolyn Blackburn

Chapter Highlights

- Recognising a child with FASD in an educational setting.
- Policy contexts to identifying FASD in the educational setting.
- Wider considerations and comorbidities seen in an educational setting.

Children with fetal alcohol spectrum disorder (FASD) represent an under-recognised group of children in education as professional knowledge amongst the teaching profession about FASD is low. In addition, not all children prenatally exposed to alcohol will be diagnosed with FASD but may be diagnosed with an alternative condition such as attention deficit hyperactivity disorder (ADHD) or autism. This presents challenges and potential barriers to children with FASD being able to access the curriculum in a way that is meaningful to them.

9.1 Policy Background

The UK Special Educational Needs and Disability (SEND) policy development has been influenced by international human rights agendas and the need to reduce the social cost of failing to provide sufficient support to children with SEND early enough to improve their future success and life chances.

The *Special Educational Needs and Disability Act* SENDA (HMT) [1] provides protection for children with SEND against discrimination, and the right to education in mainstream settings. The SEN Code of Practice [CoP] [2] for education settings placed emphasis on the role of educators to support families in identifying children's needs through observation and monitoring and required schools to appoint a

C. Blackburn (✉)
Birmingham City University, Birmingham, England
e-mail: Carolyn.Blackburn@bcu.ac.uk

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_9

Special Educational Needs Co-ordinator (SENCO) with overall responsibility for children with SEND. The landmark Children and Families Act [3] introduced a revised Special Educational Needs Code of Practice (CoP) [4].

9.1.1 The Children and Families Act 2014 (Part 3): Key Points

Definition of SEN: a child has a special educational need if they have a learning difficulty or disability that calls for special educational provision. A learning difficulty is a significantly greater difficulty in learning than the majority of children of the same age.

In this context, disability is a disability that prevents or hinders a child from taking advantage of the facilities generally available. Special educational provision is a provision that is additional to or different from that which is normally available in mainstream settings. For a child under the age of 2, special educational provision means the provision of any kind. A child under school age has SEN if he or she is likely to have SEN when they reach school age, or would do so if the special educational provision were not made for them. The principles of the Act recognise the importance of the wishes, views and feelings of children, young people and their parents and encourage educators to:

- Promote their participation in decision making;
- Recognise the importance of information and support; and
- Focus on the best possible outcomes for children and young people with SEND.

The Act:

- Requires a joint approach across education, health and social care to commission;
- Services and co-operate at a local level to meet children and young people's needs;
- Requires local authorities to publicise these services as a local offer so that parents and young people are clear what support is available locally in terms of SEN and disability;
- Establishes a framework from birth to 25 years, with Education Health and Care Plans replacing statements of SEN (although there will be a period of transition allowing Local Authorities until 2018 to transition children who have a Statement to a single EHC plan); and
- Sets out a new framework for SEN and a new SEN and Disability Code of Practice.

All educational settings must:

- Have regard to the SEN and Disability Code of Practice (discussed below). This means that they must take it into account whenever they make decisions about children;
- Ensure that they have the necessary expertise to support children and families; and
- Co-operate with the local authority in meeting its duties to children with SEN.

The type of support that children with SEND receive will vary. Two broad levels of support are legislated for:

- SEN support—given to a child in their pre-school or school. In pre-school children under the age of 5 will be assessed at the age of 2 and again in the summer term of the first year of primary school. Reasonable adjustments will be made for disabled children.
- Education Health and Care plans (EHC)—for children up to the age of 25 who need more support than is available through SEN support. They aim to provide more substantial support for children through a unified approach that integrates education, health care and social care needs.

Local authorities have a clear duty to assess a child or young person's education, health and care needs where they may have SEN (special educational needs) and they may need special educational provision to be made for them at a level or of a kind which requires an assessment of a child or young person's education, health and care needs. This is called an EHC needs assessment. It is also sometimes called a 'statutory assessment'—an assessment that a local authority is required to carry out in accordance with statute, in this case the Children and Families Act 2014. It has replaced the old form of statutory assessment under the Education Act 1996. If a parent, young person or a school/college asks the local authority (LA) to carry out an EHC needs assessment then the LA must respond to the request within 6 weeks saying if they will or will not carry out the assessment. If they refuse, the parent/young person must be informed and has the right to appeal to the Special Educational Needs and Disability Tribunal. Once an LA agrees to carry out an EHC needs assessment they must by law seek advice and information from a number of key professionals as part of the process. Based on the evidence they have gathered they must then decide whether they will issue an EHC plan for that child or young person.

9.1.2 Special Educational Needs and Disability Code of Practice: 0–25 years (2015)—Key Points

This is statutory guidance for organisations who work with and support children and young people with special educational needs and disabilities. The leaders of early years settings, schools and colleges should establish and maintain a culture of high expectations that expect those working with children and young people with SEN or disabilities to include them in all the opportunities available to other children and young people so they can achieve well. The SEN and Disability Code of Practice: 0–25 years, para 1.31 requires:

- Early identification and an early response to SEND;
- Identification of SEND with parents;
- A graduated approach to responding to SEND;

- A cycle of assess, plan, do, review; and
- The involvement of specialists where a child continues to make less than expected progress.

Four broad areas of special education needs give an overview of the range of needs that should be planned for, not to fit a child into a category. The four broad areas of need that can be planned for are:

- Communication and interaction
- Cognition and learning
- Social, emotional and mental health
- Sensory and/or physical needs

Obviously, children will have SEND that overlap these areas and educators must plan for children's diverse and complex needs to ensure that their needs are met.

9.2 What to Look for in Educational Settings

In educational settings, the delays and difficulties in development and behaviour can manifest in a number of different ways and change both over time and be variable from day to day. Broadly speaking, there are a number of challenges for educators, namely:

- Memory/Learning/Information processing difficulties, including inconsistent retrieval of learned information, being slow to learn new skills, inability to learn from past experiences, problems recognising consequences of actions and problems with information processing speed and accuracy.
- Planning/Temporal skills including needing considerable help organise daily tasks, inability to organise time, not understanding the concept of time, difficulty in carrying out multi-step tasks.
- Behaviour Regulation/Sensory motor integration difficulties including poor management of anger/tantrums, mood swings, impulsivity, compulsive behaviour, perseveration, inattention, inappropriately high or low activity level, lying/stealing and unusual (high or low) reaction to sound/touch/light.
- Abstract Thinking/Judgement difficulties including exercising poor judgement, requiring constant supervision, poor abstract thinking, and poor understanding of safety and danger.
- Spatial Skills/Spatial memory difficulties get lost easily, and have difficulty in navigating from one destination to another.
- Social Skills and adaptive behaviour including behaving at a level notably younger than their chronological age and poor social/adaptive skills.
- Motor/Oral motor control including poor/delayed motor skills, poor balance and difficulty in feeding (chewing, swallowing and sucking). A lack of clarity in

speech may be compounded by cleft palate or palatal dysfunction. This can affect children in the following ways:

Children with cleft palate and FASD may have specific needs regarding their speech and resonance. If they have a cleft palate or velopharyngeal dysfunction, that is, their palate is not functioning correctly. They should be under the care of a Regional Cleft Palate Centre. There are two aspects to assessing a child with cleft palate:

1. Resonance—this will be determined by how effectively their palate functions. There may be normal resonance or there may be signs of increased nasality—hypernasality or audible air escape
2. Articulation—the production of consonants and vowels.

If there are difficulties in either or both of these areas then this can impact on their intelligibility. This may lead to frustration if they are not understood resulting in inappropriate behaviour. Liaison with the Speech and Language Therapist at the Regional Cleft Palate Centre would be recommended to ensure that young people are supported in the best way possible. It may be that speech targets can be incorporated in their general language and social programme.

- Cognition/Academic achievement, including working at curriculum levels below peers, requiring constant repetition of instructions, rules and subject areas.
- Language/Social communication including lack of understanding of social cues, strong expressive language coupled with poor receptive language skills, and lack of empathy for others. Communication can also be affected by cleft palate (see above) and the following factors:
  - Hearing—conductive hearing loss as a result of chronic otitis media (glue ear) is common. If the young person is not understanding/responding then their hearing should be checked to eliminate the compounding factor of a hearing loss.
  - Hoarseness—It can be common and makes the voice sound distinctive [5].

9.3 Commonly Found Co-existing Problems

A number of authors describe the incidence of one birth of a child with FASD as community or societal failure or a matter of social justice and inequality [6]. This is because there are wider issues and concerns associated with FASD. These relate to the context in which it is acceptable and possible for women to drink alcohol in pregnancy, including price and availability of alcohol issues, but also because there are complex factors related to FASD. For example, in a study that was undertaken with aboriginal populations in Australia, the following characteristics of children with FASD were observed [7]:

- 35% are born preterm;
- 65% are born with low birth weight;
- Only 7% diagnosed at birth, the average age of diagnosis is 3.3 years;
- 53% have microcephaly (small head circumference associated with the underdeveloped brain);
- 24% have significant birth defects;
- 85% have behavioural problems;
- 40% live with the birth parent (therefore, 60% lived with foster/adopted parents); and
- 51% have a sibling with FASD.

As highlighted above points, there is an interaction between PAE and preterm birth, and it is worth noting that the World Health Organisation (WHO) defines preterm birth as babies born alive before 37 weeks of pregnancy are completed. There are sub-categories of preterm birth, based on gestational age:

- extremely preterm (less than 28 weeks)
- very preterm (28–32 weeks)
- moderate to late preterm (32–37 weeks)

Each year in England, around 10,000 children are born very preterm (at less than 32 weeks gestation) and a further 60,000 are born moderately preterm (at 32–36 weeks gestation). The number of preterm births has increased in the last two decades, and more preterm children are surviving due to improved neonatal care [8]. However, the prevalence of cognitive, behavioural and emotional problems in preterm populations has not changed. In particular, children born preterm have been found to experience specific learning problems including difficulties with mathematics, visual-spatial skills, memory and attention. There is still much we do not know about the nature and spectrum of these learning difficulties, their long-term consequences, and how to deal with them. In particular, there is controversy about whether moderately preterm children experience similar but milder learning problems than children born very preterm. This is compounded by any difficulties associated with PAE. Children born with extremely low birth weight (ELBW) of less than 1000 g have been found to require some form of special educational support, and experience particular difficulty with either numeracy or reading [9, 10]. Children with FASD are often born small and sometimes premature as well. Assessment is more difficult if the pregnancy dates are not known and an early pregnancy scan is not available.

It is thought that up to 80% of children with FASD enter foster or adoption placement [11]. Sometimes, children with FASD experience multiple foster placements and can difficult to place for adoption due to their challenging behaviour [12]. This means that may have difficulties developing and sustaining relationships with others. Furthermore, Gregory et al. [13] found a history of prenatal exposure has been

found in 55 out of 160 health assessments for looked after children (34%) and in 34 out of 45 medicals for adoption (75%) [13]. Elliott's findings also suggest that if there is already a child with FASD in the family, this should trigger more support for the family in future pregnancies.

Given the characteristics noted by Elliott [7] and the arguments noted above, the imperative for diagnosis can related to personal, psychological, educational and societal benefits. It could be argued that:

> Exposure to alcohol before birth is the most important preventable cause of brain damage in children today affecting substantial numbers of children. Its effects range from devastating physical and learning disabilities to subtle damage leading to poor behaviour, violence and predisposition to criminality. The human cost to affected infants and their families is huge let alone the economic impact and burden on our health, education and social care services, and on the family and criminal justice systems. [14]

What differences might I notice about children with FASD in an educational setting?

9.4 Strengths

Children with FASD have many strength and these should be used to identify opportunities for learning and development and interventions for children. These include:

- Often gregarious, fun loving, caring and affectionate
- Can be sensitive, loyal, kind and trusting in relationships
- Can succeed in structured situations
- Often enjoy repetitive work
- Strong practical skills and tend be good at drama, art and woodwork
- Strong sense of fairness
- Strong visual memories
- Good verbal fluency

9.5 Observable Difficulties

9.5.1 Some or All of the Following May Be Observed in Infants with FASD

- Often trembling and difficult to sooth, may cry a lot;
- Weak sucking reflex;
- Little interest in food, feeding difficulties (feeding can take hours);
- Difficulties adjusting to solid food because of disinterest and poor appetite;

- Weak muscle tone;
- High susceptibility to illness;
- Unpredictable sleep patterns/cycles;
- High sensitivity to sights, sounds and touch;
- Failure to thrive (may continue to lose weight longer than normal after delivery);
- Delayed developmental milestones (e.g. walking and talking);
- Problems with bonding (this may be exacerbated if mum is also an excessive drinker and/or is depressed or finding bonding difficult herself); and
- Small for age, underweight, may be have been born prematurely.

9.5.2 Some or All of the Following May Be Observed in Preschoolers with FASD

- Feeding and sleeping problems;
- Poor motor coordination, and poor fine and gross motor control;
- Short attention span, flitting from one activity/area to another, exhibiting butterfly-like movements;
- Demonstrates more interest in people than objects;
- Overly friendly and indiscriminate with relationships may seek affection constantly;
- Expressive language may be delayed or children may be overly talkative (but lack richness of speech, thought or grammar complexity);
- Receptive language often delayed; even if children are talkative, they may not understand much of what is said to them. May follow other children's lead often;
- Inability to understand the danger, often fearless;
- Low tolerance for frustration and prone to temper tantrums;
- Easily distracted or hyperactive;
- Difficulty with changes and transitions, prefer routines; and
- Difficulty integrating sensory information, such as sound, touch, light, smells, movement.

9.5.3 Some or All of the Following Might Be Observed in Children in Compulsory Education

Developmental Difficulties

- Significant delays in achieving developmental milestones such as toileting and hygiene skills, in some cases beyond the primary years.

Medical Difficulties

- Medical and health-related difficulties including organ damage, poor sleep patterns, eating and dietary difficulties, small stature, vision and hearing impairments.

> *I've always wanted somebody to come along and say, 'I'm going to look this child with FAS as a child with severe medical problems.' We've had so many medical professionals involved …. not many of them have known enough about FASD to say this is standard procedure with a child with this problem.* [15]

Learning Difficulties

- Understanding cause and effect.
- Speech language and communication delays/disorders including verbosity, poor understanding, poor social cognition and communication skills and a difficult using sophisticated language in social contexts.
- Cognitive difficulties, including poor short-term memory, and poor concentration.
- Difficulty in understanding mathematical concepts, such as time, understanding money.
- Frontal lobe damage to the brain, which is associated with FASD, results in impaired executive functioning leading to deficits, such as impaired ability to organise, plan, understand consequences, maintain and shift attention, and process and memorise data. This has an impact on independence in a range of situations. Executive functioning impacts on daily living skills.

Behavioural Difficulties

- Behavioural difficulties, including hyperactivity, inattention, aggression, obsessions with people and objects, agitation, can cause anxiety and frustration for children as well as parents and educators. These difficulties, whilst often seen as behavioural issues, can also be related to sensory processing disorders requiring occupational therapy input:

> *She's not always aware of the impact of her behaviours on others and that upsets her, because after the event when people explain to her, she's very apologetic. She doesn't like to be like that but at the same time, she really cannot control it. This is the paradox, she's aware that she can't control herself and that's frustrating for her. In Science we're hitting difficulties because of her impulsivity around behaviour, she's very drawn to the apparatus, she likes anything that's very pretty and sparkly and if you've got colourful flames and splints and nice things bubbling in jars that's a temptation for her, she wants to get in there with her hands.* [16]

Social Difficulties

- Difficulty acquiring appropriate social and emotional skills, which have an impact on relationships, friendships, and any activity which requires an understanding of the state of mind of others and predicting how this might affect their actions.
- Understanding boundaries: children can be frustrated by their own behaviour, but seemingly unable to control it, leading to challenges in self-esteem and peer relationships:

> *It's almost as if he peaked in year 4 and hasn't developed much since then. The others in his class are now just as verbal as him but their understanding of things is much higher. The other students are now almost anti-Collin now because he's so immature compared to them and what they're expecting of their peers he's not reaching, so they have shunned him, we have to have sessions to deal with this as they are now bullying him.* [17]

Emotional Difficulties

- The need to rely on external prompts from adults can result in low self-esteem and frustration
- Children begin to identify the differences between themselves and peers (and vice versa) even in special school settings, again resulting in low self-esteem.
- Secondary disabilities, such as mental health problems, disrupted school experience, trouble with the law, confinement, inappropriate sexual behaviour, problems with independent living and employment can result from a lack of identification/support when children are at primary age.

Transition

Parents and educators share concern over the social and emotional vulnerability as children with FASD move through the education system and into adult life due to their difficulties with relationships and change to routines, need for constant supervision and difficulties acquiring key life skills.

- Children with FASD will continue to need provision and support throughout their adult life, which ideally include:
  - Ongoing multi-disciplinary assessment leading to appropriate and sensitive support packages
  - A commitment to maximising appropriate levels of independence
  - The provision of supported/sheltered living accommodation with access to assistance with daily living skills
  - Supported work and leisure opportunities
- Schools can support families in finding an appropriate and suitable placement for children and providing information to future placement about how best to support the child.

The overall implications for learning are that there is a necessity for extrinsic motivation to learn skills or complete tasks such as life skills, hygiene routines, and school-based tasks, particularly in secondary aged children, requiring repetitive reminders and re-focussing from adults. In addition, social communication difficulties mean that inappropriate interactions with others can leave children with FASD vulnerable to bullying and other forms of abuse, or they may intimidate others with their over friendly or over powering behaviour.

These challenges can be compounded by other co-existing disorders, such as ASD and ADHD. Linked to this, many children will experience sensory processing disorders (SPD) and may present with sensory seeking behaviour such as inattention, hyperactivity and distractibility. Children's early family experiences may imply that attachment difficulties (AD) are also a consideration for educators to be aware of, particularly where children have been placed in foster or adoptive families, as is most often the case for children with FASD.

Sensory Processing Disorders

Sensory processing disorder (SPD) relates to the inability to use the information received through the senses in order to function smoothly in daily life. SPD is an umbrella term to cover a variety of neurological disabilities. Included in this are:

- **Sensory modulation problems** that pertain to how a child regulate his/her responses to sensations. This may result in a child being over responsive (hypersensitive), under responsive (hyposensitive) or sensory seeking and some children may fluctuate between these positions.
- **Sensory discrimination difficulties** pertain to children who may have difficulty in distinguishing one sensation from another. We each have eight senses, including five external senses, visual, auditory, olfactory, tactile and gustatory, and three internal senses proprioceptive, vestibular and organic. Each of these senses presents implications for the way we perceive and respond to our environment and perceive sensations such as pain, smell, taste, balance and sound.
- **Sensory-based motor problems** relate to children who may position their body in unusual ways and difficulty in conceiving of an action to do, planning how to organise and move their body and carrying out the plan [18].

SPD can result in a range of symptoms from mild to severe which can impact on interaction with others, functioning in daily life, and the ability to learn and general success throughout life. General strategies for including children and young people with SPD in an educational setting are included in the chapter on teaching and learning.

Attachment Difficulties

A solid and healthy attachment with a primary caregiver appears to be associated with a high probability of healthy relationships with others while poor attachment with the mother or primary caregiver appears to be associated with a host of emotional and behavioural problems later in life [19]. Studies have shown that children who have positive early attachment experiences do well as pre-school children [20, 21] achieving high scores on ego-resilience and self-esteem, less dependency on the educator and more positive effect towards the educator. Securely attached children demonstrate a capacity to adapt to school and respond to the demands of academic and social setting in which learning takes place [22] Fundamentally, appropriate attachment with a primary caregiver is viewed as a foundation for optimal language, cognitive and emotional development [23] highlighting the interconnectedness between attachment and children's holistic development.

Children's ability to adapt to a new caregiver is thought to be easier in the first 6 months of life than afterwards, and 6-month-old children are less rejecting and more accepting of new adoptive or foster parents than older infants. However, the more caregivers a CYP is required to interact with the ability to securely attach to others diminishes [24], which has implications for the attachment style of CYP who are exposed to multiple foster placements. In addition, there is a general feeling amongst clinicians working with children who are adopted that such children have

the same issues whether they were adopted at birth or as teenagers. These issues relate to separation and loss, trust, rejection, guilt and shame, identity, intimacy, loyalty and mastery, or power and control.

As infants, children develop a reflective sense of what others are likely to do in response to their own behaviour [25]. This allows them to build mental representations or internal working models of expectations based on past experiences, allowing them to regulate the negative emotions of fear, distress and anxiety when insecurity is felt. The formation of attachment relationships serves an important function as an emotional and physiological regulator for all humans as a social species [26]. Self-regulation in children is the developmental integration of emotion and cognition in early childhood [27] and has been found to account for variance in academic outcomes among 3- to 5-year-old children, indicating that success in self-regulation helps prepare children for being successful in school.

Thus, through the development of internal working models with sensitive attachment figures, children will begin to predict what might happen when feelings are expressed or needs displayed. A child's internal working model of their relationship with a caregiver will include concepts of:

- The self
- Others
- Expectations of the relationship and how the self and others are likely to behave and react

The consistent availability of a sensitive caregiver in the development of a CYP's internal working model, therefore, is important as children begin to organise their attachment behaviour to increase the availability, proximity and responsivity of their carers to meet their needs. Adverse experience of early attachment, such as the inconsistent, unpredictable or unavailability of a primary caregiver, which is not relieved by more positive relationships with others later on, is very likely to have negative implications for both behaviour and learning. The educator can act as the specific attachment person in schools, particularly for anxious children [28]. Educators can represent a 'secure base' in a school setting by providing sensitive, predictable responses to behaviour and learning needs for children with attachment difficulties.

Mental Health

Children with FASD are at risk of developing secondary disabilities if sensitive and appropriate support packages are not received in their early years and primary education, one aspect of which is mental health problems. Mental health problems affect 30–40% of all CYP at some time during childhood [29]. The Mental Health Foundation estimates that 20% of children up to the age of 16 years' experience a mental health problem at some point during their development and 10% present with a 'clinically recognisable' mental health disorder, including emotional disorders such as anxiety, phobias and depression, self-harm and suicide, conduct

disorders, hyperkinetic disorders, autistic spectrum disorders, psychotic disorders, eating disorders and substance, and drug abuse. Two percent of children are diagnosed with two or more of these disorders.

For individuals affected by FASD, the figure for the number affected by poor mental health rises to 87% of individuals with FASD experiencing poor mental health in adolescence and adult life, leading to the risk of suicide (23%) [30]. Mental health problems can have a significant effect on engagement with the curriculum and learning. If a child is depressed, for example, this affects their ability to pay attention and access memories, allocate sufficient resources to tasks because they are distracted by negative thoughts [31], and demonstrate enthusiasm for learning and socialising. Generally, children who are depressed engage less effectively than those who are not depressed [32] Anxious children on the other hand may display a range of attention problems, such as narrow attention span and distractibility. Anxiety and worry can have a negative impact on information processing, motivation and memory [33, 34]. Educators supporting children with FASD, therefore, will need to be aware of mental health issues, how these may manifest in children and how best to support a child with organic brain damage and compounding mental health difficulties.

Educators and the professionals who support children with FASD in the classroom will require an understanding of the compounding factors associated with the condition, and ensure continuing multidisciplinary assessment that is necessary to allow identification of secondary disabilities so proactive action can be taken to ameliorate the effects. The importance of working closely with families and putting families at the centre of their child's educational journey in these circumstances cannot be overestimated [35].

9.6 Conclusion

This chapter has discussed the policy context in which educational settings must provide inclusive practice children and young people with FASD and some of the ways in which FASD can be identified in educational settings. The overlap with attachment and relational issues as well as hyperactivity and inattention linked to sensory integration problems has been raised.

The chapter has discussed transitions as a particularly challenging time and this includes transition between activities during the school day and between key education phases such as nursery education to primary school and primary school to secondary school. These are times when underlying difficulties may be more obvious as the structure and routine needed to enable children to regulate their emotions and predict the environment are in a state of flux. Transitions will need careful management and communication between agencies and with families.

The effects of the recent changes to legislation for children with SEND remain to be seen especially in relation to children with FASD as there is no research or evaluation of this currently.

References

1. Her Majesty's Government. (2001). Special Educational Needs and Disability Act. London, UK: Her Majesty's Stationery Office.
2. Department for Education and Skills (DfES). Special Educational Needs Code of Practice. Nottingham: DfES Publications; 2001.
3. Department for Education (DfE), Department for Business, Innovation and Skills, Department for Work and Pensions (DWP), Department of Health (DOH). Ministry of Justice Children and Families Act http://www.legislation.gov.uk/ukpga/2014/6/contents/enacted. Accessed 31 Mar 2020.
4. Department for Education (DfE), Department of Health (DOH). Special Educational Needs (SEN) code of practice: statutory guidance for organisations who work with and support children and young people with SEN. 2015.
5. Astley SJ. The 4-digit diagnostic code, diagnostic guide for fetal alcohol spectrum disorders centre on human development and disability. Seattle, WA: University of Washington; 2004.
6. Karr-Morse R, Wiley MS. Ghosts from the nursery: tracing the roots of violence. New York, NY: Atlantic Monthly Press; 1997.
7. Elliott E. Australian perspectives on fetal alcohol spectrum disorders. In: Carpenter B, Blackburn C, Egerton J, editors. Interdisciplinary approaches to FASD. London: Routledge; 2013.
8. National Neonatal Audit Programme Annual Report on 2014 data. 2015. http://www.rcpch.ac.uk/improving-child-health/qualityimprovement-and-clinicalaudit/national-neonatal-audit-programme-nnap. Accessed 11 Apr 2016.
9. Bowen J, Gibson F, Hand P. Educational outcome at 8 years for children who were born extremely prematurely: a controlled study. J Paediatr Child Health. 2002;38(5):438–44.
10. Wolke D, Strauss VY-C, Johnson S, Gilmore C, Marlow N, Jaekel J. Universal gestational age effects on cognitive and basic mathematic processing: 2 cohorts in 2 countries. J Pediatr. 2015;166(6):1410–1416.e1412. https://doi.org/10.1016/j.jpeds.2015.02.065.
11. Streissguth A, Clarren S, Jones K. Natural history of the fetal alcohol syndrome: a 10 year follow-up of eleven patients. Lancet. 1985;2:85–91.
12. Blackburn C, Carpenter B, Egerton J. Educating children and young people with foetal alcohol spectrum disorders. London: Routledge; 2012.
13. Gregory G, Reddy V, Young C. Identifying children who are at risk of FASD in Peterborough: working in a community clinic without access to gold standard diagnosis. Adopt Foster. 2015;39(3):225–34.
14. Aynsley-Green A. Foreword in British Medical Association Alcohol and pregnancy Preventing and managing fetal alcohol spectrum disorders. London: British Medical Association; 2016. p. ix.
15. Blackburn, C., Facing the Challenge and Shaping the Future for Primary and Secondary aged Students with Fetal Alcohol Spectrum Disorders (FAS-eD Project), Project Toolkit including project report, literature review, case studies, podcasts and primary and secondary learning and teaching frameworks). 2012.
16. Blackburn C. Personal communication from Secondary Education Teacher.
17. Blackburn C. Personal Communication with Specialist teacher in a special school for children with Severe Learning Difficulties.
18. Kranowitz CS. The out of sync child: recognizing and coping with sensory processing disorder. London: Penguin Group; 2005.
19. Perry B. Childhood experience and the expression of genetic potential: what childhood neglect tells us about nature and nurture. Brain Mind. 2002;3:79–100.
20. Sroufe LA. Infant–caregiver attachment patterns of adaptation in pre-school: the roots of maladaptation and competence. In: Permutter M, editor. Minnesota Symposium of Child Psychology, vol. 16; 1983. p. 41–81.
21. Sroufe A. Appraisal: Bowlby's contribution to psychoanalytic theory and developmental psychology; attachment: separation: loss. J Child Psychol Psychiatry. 1986;27(6):841–9.

22. Geddes H. Attachment in the classroom: the links between children's early experience, emotional well-being and performance in school. London: Worth Publishing; 2006.
23. Blackburn C. Walking through a moonless night: FASD and early childhood intervention. In: Carpenter B, Blackburn C, Egerton J, editors. FASD and interdisciplinary perspectives. London: Routledge; 2013. p. 102–22.
24. Newton Verrier N. The primal wound understanding the adopted child. Baltimore, MD: Gateway Press; 1993.
25. Howe D. Child abuse and neglect: attachment, development and intervention. Hampshire: Palgrave Macmillan; 2005.
26. Chesney AR, Champion P. Understanding the dynamics between preterm infants and their families. Support Learn. 2008;23(3):144–51.
27. Blair C, Razza RP. Relating effortful control, executive function, and false belief understanding to emerging math and literacy ability in kindergarten. Child Dev. 2007;78:647–63.
28. Barrett M, Trevitt J. Attachment behaviour and the schoolchild. London: Routledge; 1991.
29. Carpenter B, Coughlan B, Fotheringham J. Mental health and emotional well-being: the new dimension in the curriculum for children and young people with special educational needs. Complex needs series, No.4. London: Specialist Schools and Academies Trust; 2010.
30. Streissguth A, Kanter J, editors. The challenge of fetal alcohol syndrome: overcoming secondary disabilities. Seattle, WA: University of Washington Press; 1997.
31. Ellis HC, Ashbrook PW. Resource allocation model of the effects of depressed mood states on memory. In: Fielder K, Forgas J, editors. Affect, cognition, and social behaviour. Toronto, ON: Hogrefe; 1998.
32. Cooper P, Tiknaz Y. Nurture groups in school and at home. Connecting with children with social, emotional and behavioural difficulties. London: Jessica Kingsley; 2007.
33. Eysenck MW, Calvo MG. Anxiety and performance: the processing efficiency theory. Cognit Emot. 1992;6:409–34.
34. Eysenck MW, Keane MT. Cognitive psychology: a student's handbook. East Sussex: Psychology Press; 1995.
35. Blackburn C. Developing inclusive practice for young children with fetal alcohol spectrum disorders: a framework of knowledge and understanding for the early childhood workforce. London: Routledge; 2017.

It Isn't All About the Facial Features

10

Miguel del Campo

Chapter Highlights

- What do fetal alcohol syndrome (FAS) and fetal alcohol spectrum disorder (FASD) look like.
- Methods to measure and assess.
- Wider considerations and pitfalls in the assessment process.

[Part of the content of this chapter including some of the images are reprinted from Del Campo M, Jones KL. A review of the physical features of the fetal alcohol spectrum disorders. Eur J Med Genet. 2017 Jan;60(1):55-64. https://doi.org/10.1016/j.ejmg.2016.10.004. Epub 2016 Oct 10. PMID: 27729236.]

10.1 Dysmorphology: Congenital Anomalies

Dysmorphology refers to the study of the differences in body structure resulting from abnormal development of the embryo and the fetus. The characterization of major and minor congenital anomalies has facilitated the recognition of multiple genetic and non-genetic "multiple malformation syndromes." Major anomalies represent structural differences with significant impact on the health or appearance of the individual, whereas minor anomalies are differences in morphology without such impacts. Minor anomalies are much more frequent than major anomalies, and their recognition is much more useful for the delineation of genetic and non-genetic conditions. Minor anomalies are variants in morphology that are present, by definition, in a small proportion of the general population (2–10% depending on the

M. del Campo (✉)
Department of Pediatrics, Division of Dysmorphology and Teratology, University of California, San Diego,
La Jolla, CA, USA
e-mail: midelcampo@ucsd.edu

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_10

anomaly and the population). Congenital anomalies can occur de-novo or can be inherited from a parent. Those minor anomalies that are inherited from a parent or are recurrently found in a family or ethnic group can be considered familial or ethnic variants, and their significance in a specific patient certainly has to be weighed in the context of the family and the population. If this structural variant is not associated in the family to an unhealthy phenotype, it will be considered irrelevant and not taken into account as one of the features of the phenotype in the patient. The same is true for continuous morphological traits and anthropometric measures. For example, the potential significance of short stature is very different in the context of a family with short stature. Also, the very common presence of epicanthal folds in some Asian populations makes its presence in a patient of Asian descent much less significant to the overall phenotype.

After the initial identification of the basic concepts of dysmorphology and teratology by David W Smith and others in 1966 [1], much work has been carried out in order to delineate precisely the concepts of morphologic anomalies, variants, malformations, and other terms, as well as the precise nomenclature to be used for each of these [2]. Major anomalies (birth defects) are those that impair the function in the individual and may require treatment or repair, such as a cleft palate or a congenital heart defect. Minor anomalies such as short palpebral fissures or abnormal hand creases are infrequent morphologic findings (generally less than 4% of the population) that may be markers of abnormal morphogenesis but do not cause any cosmetic or functional deficits for the individual.

Major and minor congenital anomalies can result from different processes in alteration of morphogenesis. The term malformation refers to a primary alteration in development, which can be caused by an abnormal genetic program, or the impact of a teratogen in early development of an organ or structure. In contrast, deformations represent the impact of external forces in a previously normally developed structure, and disruptions indicate the abrupt interruption of normal development or destruction of a previously well-developed structure. The anomalies observed in FASD are a combination of minor and major malformations.

10.2 The Pattern of Physical Features as a "Biomarker" of Prenatal Alcohol Exposure

No major or minor malformation in isolation is specific enough to provide certainty of a specific diagnosis. Only a pattern of multiple malformations may be specific enough to delineate a syndrome and indicate that a specific cause is responsible for the observed phenotype. Dysmorphology uses pattern recognition for syndrome identification.

Since its delineation, physical features have been the hallmark for the recognition of the fetal alcohol syndrome (FAS). The consequences of prenatal alcohol exposure (PAE) were initially defined by pediatrician dysmorphologists, trained to recognize minor anomalies and to associate them to specific etiologies. Paul Lemoine, a French pediatrician, had described in 1968 the potential effects of

alcohol exposure in pregnancy in 127 children born to alcoholic mothers or fathers and published the description of his findings in the French literature [3]. Lemoine pointed to a phenotype of microcephaly, a short anteverted nose, fine lips, and midface hypoplasia. Jones and Smith reported several children born to alcoholic mothers in 1973 [4]. The pattern of malformations included prenatal and postnatal growth deficiencies, microcephaly, short palpebral fissures, maxillary hypoplasia, epicanthal folds, joint limitations, altered palmar creases, and septal or ductal defects of the heart. These authors proposed maternal alcohol consumption during pregnancy was responsible and proposed pathogenetic mechanisms for the teratogenic origin of the defects. This publication led to widespread recognition of the condition and to a large number of studies to further delineate the clinical phenotype and understand its developmental pathogenesis. Both Jones and Lemoine recognized major ocular and cardiac malformations in these children. A broad range of less frequent anomalies were found in some of the cases.

Forty years of clinical, animal and cell model research has identified a broader and more precise physical phenotype for these children. The pattern of physical features of FAS is today considered specific enough that a diagnosis of FAS can be established in the absence of confirmation of PAE by the most widely used FASD diagnostic systems [5–8]. Unfortunately physical features are considered differently in different diagnostic systems, leading to significant discordance in the diagnosis of patients [9]. In the absence of a specific laboratory "biomarker" of exposure, still unidentified in spite of continuous efforts by many researchers, the physical features or stigmata of FAS have become the actual biomarkers of exposure. This pattern reflects the consequences of PAE on growth, abnormal development of the head and face, and other organs and structures. Understanding this specificity is particularly important because reliable assessment of PAE is often impossible, as biological mothers may not admit to use or abuse of alcoholic beverages during pregnancy, and many of these children are placed in orphanages, foster care or adoption homes, making prior information on exposure often unavailable. In the case of FAS, animal model research has allowed us to better understand the relationship between PAE and its multiple effects on brain and facial development, and has provided graphic explanation for the pathogenesis of the observable features of FAS. Animal research has allowed us to better understand the significance of the dysmorphic features and certainly confirms their specificity for PAE.

10.2.1 Key Embryologic Facts to Understand the Origin of the Physical Features of FASD

Studies in mice that began soon after the identification of FAS provided the first experimental evidence that both brain and craniofacial abnormalities result from PAE, and that the defects in the mice are similar to those seen in humans [10]. Striking changes appeared in the developing rostral neuroectoderm, particularly the anterior neural ridge (ANR) at the rostral boundary of the forebrain of the fetuses, after the pregnant mice were exposed to alcohol. Other cells such as in the neural

crest, epibranchial placodes, and otic placodes are also affected. These early embryonic tissues originate the frontal brain as well as the craniofacial structures. An underdevelopment of the medial nasal processes leads to the short nose, significant smoothness of the philtrum and underdevelopment of the upper lip in FAS [10, 11]. These facial dysmorphic features are stage dependent in mice. Exposure on gestational day (number of days since conception) (GD) 7 produces a constellation of dysmorphic facial features characteristic of human FAS (severe midfacial hypoplasia, shortening of the palpebral fissures, an elongated upper lip, and a deficient philtrum), whereas ethanol exposure on GD8.5 causes mild midfacial hypoplasia and palpebral fissure shortening, a shortened upper lip, and a preserved philtrum [12].

Studies on the human fetus from 10 to 16 weeks gestation indicate that the philtrum is smooth and the upper lip is thin and lacks the Cupid's bow configuration up until week 10 of gestation [13].

All the evidence points to the fact that whereas growth deficiency, microcephaly and neurobehavioral abnormalities can be a result of exposure at any time throughout pregnancy, the presence of characteristic dysmorphic features indicate exposure during the first trimester.

10.3 Physical Features Are Diagnostic of FAS and Partial FAS and Are also Frequent in Other Infants with FASD

Syndromes are often multidimensional in their manifestations and include, in addition to malformations, features related to growth, cognition, behavior, and overall health and function of the affected individuals.

The physical features of FAS are not the major problem for the affected children, although major birth defects and significant growth deficiencies can be relevant medical issues. Both cognitive impairment and behavioral manifestations including secondary disabilities are the major problems for the life of these children and their families. Most patients affected by PAE do not have the classic growth deficits, small head circumference (HC) and the three key facial features of FAS. Therefore, the physical features as biomarkers of exposure are only useful for a formal diagnosis of FAS in a small subset of prenatally exposed children. It is estimated that 5–10 children that suffer the consequences of PAE will not have the physical features of FAS for each child with FAS or pFAS [14, 15].

The term fetal alcohol spectrum disorders (FASD) is an umbrella term which includes the full spectrum of defects resulting from PAE, with or without recognizable physical features and growth deficits. This spectrum of anomalies and its different diagnostic categories were initially set forth by the Institute of Medicine of the National Academy of Scientists in 1996 to include all features seen in children affected by PAE. Years later, these diagnostic categories were clarified for clinical use by Hoyme et al. [16] and were recently updated by the same author [6]. Even though the diagnostic criteria within the IOM scheme and most of the other systems include two or three of the three classic facial features, microcephaly and growth

deficits, clinical epidemiologic research has confirmed that many other facial and non-facial features are specifically associated with FAS as well, and can often be present in other patients with FASD who do not fulfil the formal criteria for FAS. The prevalence of the three classic features included in the diagnostic criteria and these additional dysmorphic features has been determined in several studies and is certainly variable. The additional dysmorphic features not included in the diagnostic criteria are found significantly more often in FAS. Therefore, they are considered part of the dysmorphic pattern of FASD [17]. Moreover, several of these features and the total number of those in the scheme of a "Dysmorphology Score" have been proven to be related to more severe FASD, with more impact on cognition and functional disability [14, 15, 18].

Even in cases without the minimal features necessary to make a diagnosis of FAS, including growth deficit, microcephaly and two or three of the three key facial features, other associated dysmorphic features have been more frequent in these "FASD without FAS" cases than they are in non-PAE children in some studies. The knowledge and quantification of these physical features will be important for all patients with PAE, even for those that will not be diagnosed with FAS.

In this chapter, we will review the structural defects associated with PAE that will allow recognition of FAS, including dysmorphic features and growth deficits. We will describe the full spectrum of physical anomalies associated with PAE, and not only those included in the different diagnostic schemes. We will provide a definition of the most relevant features, discuss their embryologic origin, their importance for the diagnosis of FAS and their consideration within the currently used diagnostic schemes.

We will also review the technical aspects to achieve precision in the evaluation of these features and the limitations existing for their assessment.

10.4 Physical Features of FASD: Description, Assessment, and Significance (Figs. 10.1, 10.2, 10.3, and 10.4)

Physical features included in the diagnostic criteria.

There are multiple sets of diagnostic guidelines available for use, which host significant differences. In general, the term FAS refers to the combination of growth deficits, and a small head size, and two of the three key facial features (short palpebral fissures, smooth philtrum and thin vermillion border of the upper lip) or all three. In partial FAS (pFAS), the facial features are still necessary but only growth deficit or a small head circumference and not the two together are required. If there is confirmation of exposure and neurobehavioral deficit, the two facial features alone can lead to a pFAS diagnosis. For patients with PAE, and a consistent neurobehavioral phenotype, but normal growth, head size and not two or three of the facial features, the term alcohol-related neurodevelopmental disorder (ARND) has been coined. The fetal alcohol spectrum of disorders includes the three diagnoses FAS, pFAS and ARND, plus the diagnosis of alcohol-related birth defects (ARBD) when major malformations are present.

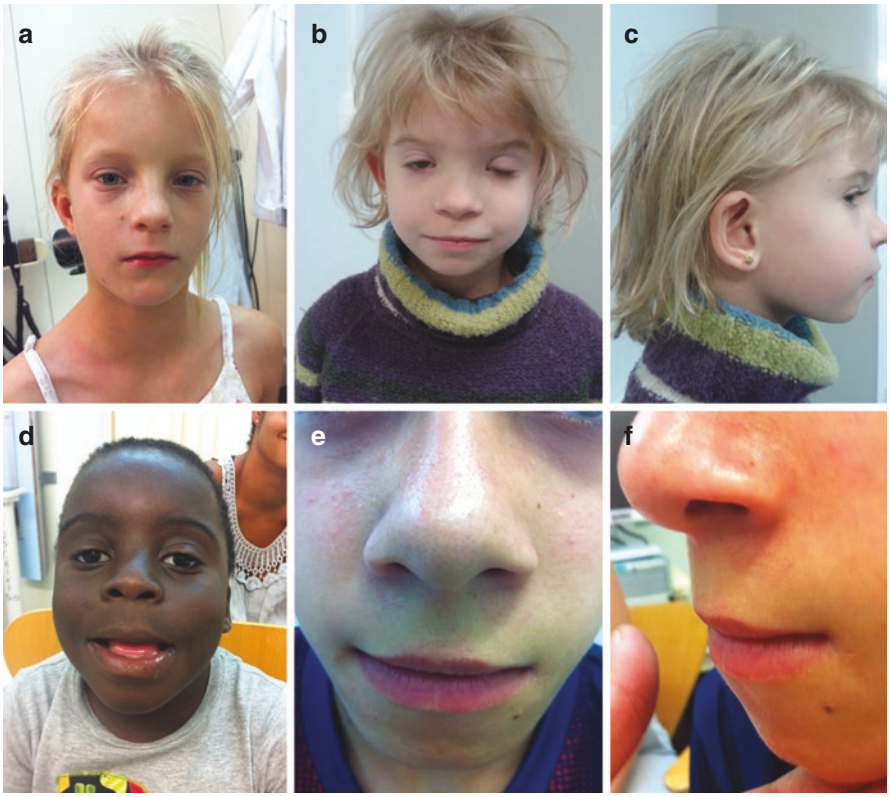


Fig. 10.1 The faces of FASD. (**a**) Short palpebral fissures, normal philtrum pillars, narrow and linear vermillion, midface hypoplasia. (**b**) Apparent hypertelorism with normal measurements due to the very short palpebral fissures. Prominent epicanthal folds, bilateral ptosis. (**c**) Midface hypoplasia and railroad track ears. (**d**) Mild ptosis, short anteverted nose with long smooth philtrum, fleshy lips. (**e**, **f**) Frontal and lateral view of the smooth philtrum and linear vermillion border of the upper lip that has lost the characteristic Cupid's bow configuration. Midface hypoplasia

Physical features for the diagnosis of FAS and pFAS (IOM)

- Weight or length ≤10%
- Head circumference ≤10%
- Palpebral fissures length ≤10%
- Smooth philtrum (scores 4–5) ≤10%
- Thin vermillion of upper lip (score 4–5) ≤10%

10.5 Growth Deficits

Growth deficits are frequent and were part of the initial definition of FAS. In our experience, significant growth deficits are seen in the most severely affected patients and many studies show more severe growth deficits are associated with more severe

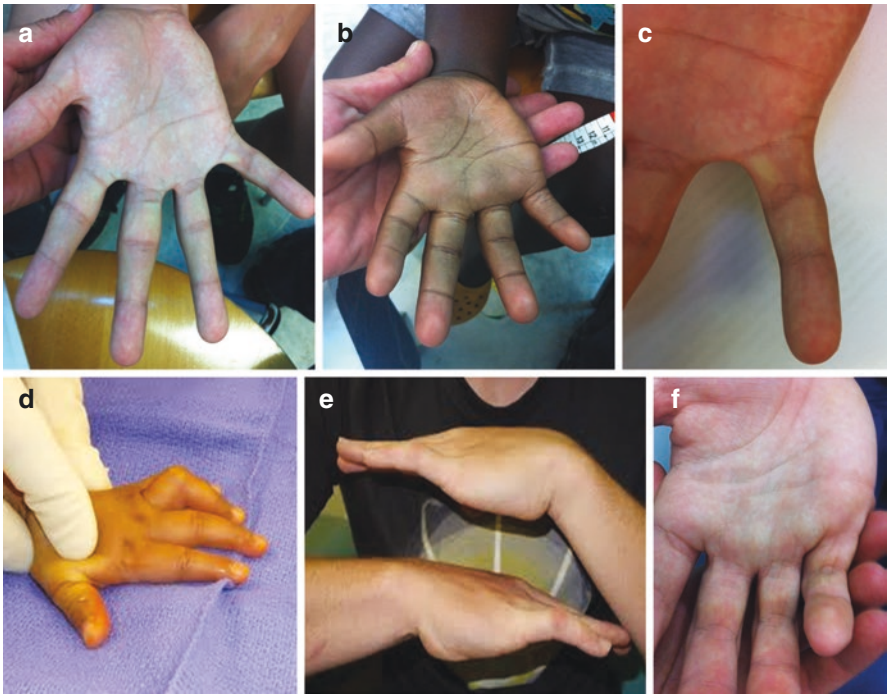


Fig. 10.2 The hands of FASD. (**a**) Hockey stick crease, absent proximal transverse palmar crease, normal thenar crease. (**b**) Hockey stick crease, absent proximal transverse palmar crease, absent proximal interphalangeal crease of the fifth finger, limitation to extension of fourth and fifth fingers indicating mild camptodactyly. (**c**) Clinodactyly of the fifth finger. (**d**) Camptodactyly of the fourth finger. (**e**) Bilateral camptodactyly of the fifth fingers and (**f**) Camptodactyly of fingers 3–4 and 5

intellectual deficits [18]. Growth deficits can be present at birth or become evident postnatally. Maternal health, prenatal nutrition and placental function are important determinants of variation in prenatal growth, and can modify the effects of PAE. Mothers with poor health and nutrition can have infants with significant prenatal growth deficit. In those cases, catch-up can occur after birth if these external factors have been determinant. When these additional prenatal factors are not present, growth parameters at birth can be normal, and a slow decline in centiles the growth chart can be seen over time, often manifest by two years of age. This type of postnatal onset growth deficit is common to many genetic conditions. Therefore, weight and length at birth should not be used to assess growth if evaluations at later ages are available.

Population-specific charts for objective assessment of weight and height should be used when available, but more specific curves have not been validated for all populations, and almost never for specific ethnic groups within a population. And, of course, ethnic mixes will not have their own charts. Finally, familial stature and weight should ideally be taken into account. When, in the absence of PAE, the parents and other family members show growth deficits or significantly tall stature, it can be difficult to interpret that the patient's growth status is determined by PAE,

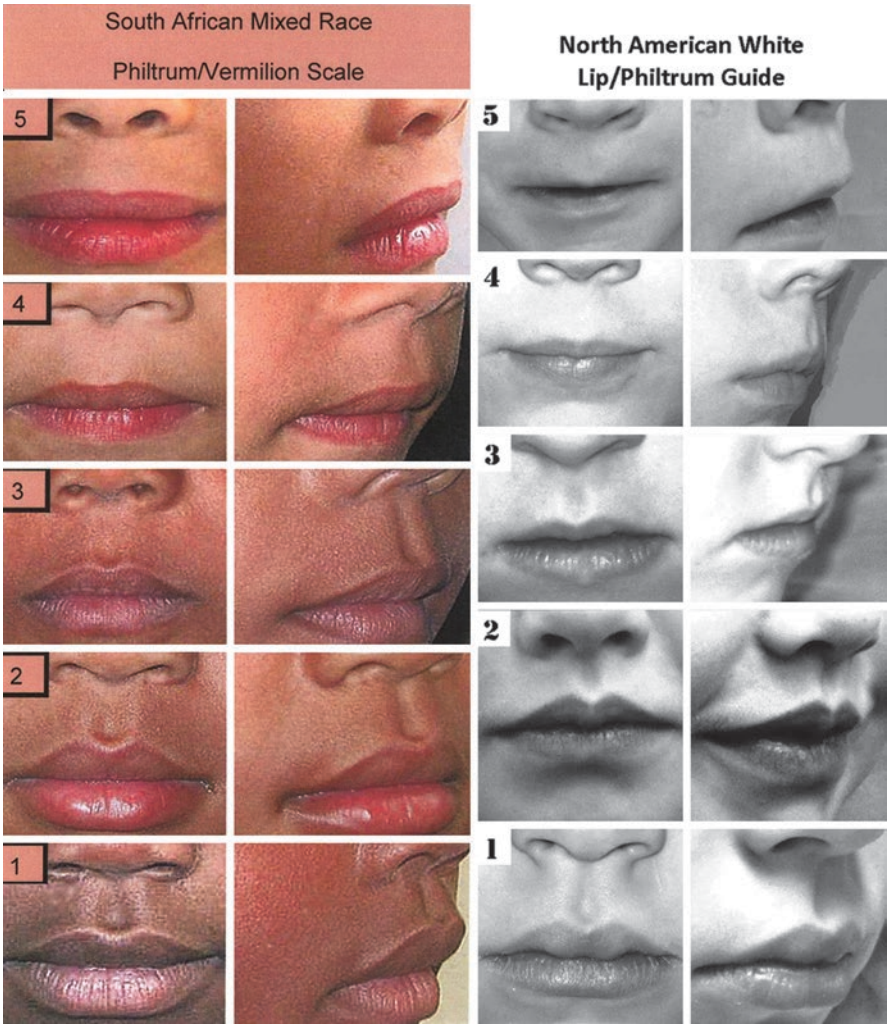


Fig. 10.3 Lip philtrum guide for the Black South African race and US Caucasians. In both guides, the prominence of the philtrum pillars is lost from 1–5, which can be best appreciated on a 45^0 view. Only scores 4 and 5 are used for the diagnosis of FAS. These represent a complete absence of the pillars (5) or very faint or incompletely visible pillars (4). For the vermillion border of the upper lip, the Cupid's bow shape is either lost (5) or very underdeveloped (4), and the vermillion border becomes linear. The upper lip is much thinner in Caucasians, a fact that should be taken into account with the use of race-specific lip-philtrum guides. Courtesy of Prof Eugene Hoyme, University of Arizona) (Hoyme et al. [6]; Hoyme et al. [7])

and not only by familial stature or body complexion. However, none of the diagnostic criteria for growth have yet taken into account these facts in an objective way.

Both height and weight can be abnormally decreased. Inability to gain weight in spite of normal nutrition, often reflected in a lean and apparently muscular

Fig. 10.4 Measurement of the palpebral fissures with a caliper or a hard ruler

build due to a low BMI with decreased fat content, is often observed. Hyperactivity or abnormal basal metabolic rates may also underlie this fact. This weight/height discordance is formally recognized by the fact that the Canadian criteria for FASD diagnosis include a weight for length ratio equal or below the tenth centile (10% of the population would fall below this line) as an additional indicator to deficits in weight or stature of abnormal growth in these patient [19]. Decreased cell mass and increased metabolic rates are most likely explanations for this body constitution and have been noted in animal models with growth deficits after PAE. Short stature in these patients is not commonly related to growth hormone deficiency and response to human growth hormone (HGH) treatment is not usually favorable [20, 21].

It is important to realize that all diagnostic criteria use the tenth centile as cut-off for both weight and height deficits, instead of -2SD (2–3%), which is commonly used for the definition of endocrine and genetic causes of impaired growth. All clinicians have observed that very severe or profound growth deficits caused by PAE leading to severe dwarfism/short stature are extremely infrequent in FASD.

10.6 Small Head Circumference (HC) and Structural CNS Defects

Similar to growth, HC ≤10% is considered in the diagnostic criteria for FAS, whereas the usual definition of microcephaly is HC ≤-2SD (2–3%). Microcephaly is caused by a global decrease in brain volume, well observed in animal studies of PAE. Structural anomalies of the CNS have been recurrently reported, most frequently agenesis, partial agenesis or dysgenesis of the corpus callosum, as well as posterior fossa anomalies such as a hypoplastic cerebellum and the Dandy Walker malformation [22, 23]. Other CNS anomalies have been reported, including reductions in brain volume in all areas but most significantly in certain regions such as the basal ganglia and the diencephalon [24]. The size of the head circumference is most related to cognitive function in FASD in many studies. Infants with smaller heads have more severe cognitive impairments. In addition, the common embryological origin of the brain and facial mesenchyme explains that children with more severe

microcephaly are often the most dysmorphic, particularly for the three key facial features of FASD, if they were exposed in the first trimester.

Ideally, HC of the parents should always be measured, since mild microcephaly may be a familial trait often not associated with cognitive or behavioral deficits. Similarly, a genetic background of increased head size can make the absence of a small HC in a child with PAE difficult to interpret. Also, ethnic variation in HC is frequent and specific charts for specific populations, if available, should be always used.

Head circumference measurement is in fact the measurement of the occipito-frontal circumference (OFC). A soft tape measurer should be used and circle the cranial vault 1 cm above the supraorbital processes in the frontal area and across the occipital prominence in the posterior aspect head, in order to get the larger possible measure (Fig. 10.1). When abundant hair is present, an effort should be made to get an accurate measurement by squeezing the hair. Similarly to growth, only severe effects on brain development will cause microcephaly at birth, and measures below the tenth centile may often become apparent after 2 years of age, indicating a reduced potential for head growth in some affected infants. Also measurement of the HC at birth may be modified by molding in utero and during delivery, and accurate and valid measurements will be more difficult to obtain. HC measurements at later ages will be more suitable for diagnosis than HC at birth. A small head is also a predictor of more severe intellectual disabilities after PAE, as is growth and having a diagnosis of FAS [18]. Both volume of exposure and timing in pregnancy will determine the physical consequences of PAE, and almost every study has identified lower IQ in patients with FAS in comparison to those without any physical features.

10.7 Short Palpebral Fissures

Palpebral fissures (PF) are considered abnormally short when their size is ≤tenth centile according to the IOM Hoyme criteria, but other systems use ≤-2SD as cut-off. When the latter is the case, this feature is only present in a small number of patients, those most severely affected. The size of the palpebral fissure is dependent on the size of the eye globe. The eyes are in fact invaginations of most rostral aspect of the developing brain and appear to be selectively affected by the teratogenic consequences of PAE. Major malformations of the eyes occur in FAS and include refraction errors, optic nerve hypoplasia and occasionally microphthalmia [25]. Reduced volume of the eye globes has been proven in animal studies and humans with ultrasound and other imaging techniques [26, 27]. Ethnic and familial variation in palpebral fissure size has been proven, but curves for different populations are rarely available. Correct measurement is of utmost importance, since 1 mm can make a difference in the overall diagnosis. Measuring the palpebral fissures close to the face and following the correct angle of the orbit with a rigid ruler is considered standard. A Vernier caliper can also be used, with much caution not to poke the eye. When using a ruler, one should approach the eye at an angle determined by the ruler being at equal distance from the inner-canthus (inner corner of the eye opening) and

the outer-canthus (outer corner). Failure to follow this angle, different for every subject, may lead to shorter measurements, due to parallax error. For precise measurement, the ruler should be as close to the eye as possible, but should not touch the lower eyelids, to avoid causing reflex closure of the eyelids. In order to avoid the fear of being poked, it is ideal to have the subject look up at the ceiling, for which sometimes a helping hand showing an object up high can be useful (Fig. 10.2).

Microcephaly is responsible for only 10% of the variation in the size of the palpebral fissures, indicating that the eye is selectively affected by PAE, independently of the HC [28]. In order to avoid the effect of the HC modifying the palpebral fissures, and to take into consideration familial and individual variations, Hoyme et al. have recently included the decreased ratio of the interpupillary distance (IPD) to the inner-canthal distance (ICD) as a feature used in the Dysmorphology score. The fact that the ICD is not modified in FAS, and the IPD reflects the medial aspect of the PF plus the ICD, would help override individual and familial variations of the size of the PF dependent on the overall size of the face. IPD/ICD ≤25% would, therefore, be another measure of the selective reduction in the size of the eye opening and the eye globe in FASD, but is not used routinely, has not been sufficiently validated and is more complicated to assess than the size of the PF themselves.

Decreased size of the palpebral fissures in FASD has been associated with lower IQ in many studies, highlighting their specificity as markers of severity of the impact of PAE.

10.8 **Smooth Philtrum** (Figs. 10.3 and 10.4)

A smooth philtrum is one of the three facial features of FAS. This is a very subjective feature and certainly difficult to assess. Individual, familial and ethnic variations are very important for the shape of the philtrum pillars and for the size and shape of the vermillion border of the upper lip. The philtrum pillars run from the base of the nose to the vermillion border of the upper lip. The pillars pull up the vermillion border at their places of lower insertion in the upper lip. Assessing the feature at an angle of 45° is necessary to evaluate the volume of the philtrum pillars as well as the groove created between them.

In order to assess this relevant dysmorphic feature, Astley and Clarren at the University of Washington developed for their evaluation the "lip-philtrum guide" for scoring the feature in a Likert scale 1–5 [29]. Only significant smoothness of the philtrum reflected in grades 4 and 5 are considered features of FAS (Fig. 10.3). A score of 4 for the philtrum should be given when the pillars are smooth and/or excessively separated in their junction with the vermillion border of the upper lip, and often do not fully reach the base of the nose in their upper third, making this upper area flat without an obvious visible groove. Differentiation between 3 and 4 is certainly subjective and can be difficult, yet the importance for the diagnosis is key, given 3 is considered normal. The same applies to the upper lip. A score of 5 should be given when the groove is hardly seen between the pillars in most of its vertical dimensions, and the overall volume of these pillars is minimal or

non-existent, leading to a completely smooth or flat philtrum area. Specific ethnic lip/philtrum guides are being developed for multiple populations and should be used when possible. It can be a good idea to develop your own lip philtrum guide for the local population. Ideally, after taking photographs of multiple patients in clinic or in a research study, one should select example pictures of individuals that best match the validated Guide by Astley and Clarren. This customized lip-philtrum guide will take into account the observed normal variation in the local sample as well as the concordance of the feature in very clearly affected individuals with FAS who we believe have a smooth philtrum.

10.9 Thin Vermillion of the Upper Lip (Figs. 10.3 and 10.4)

A thin or narrow vermillion border of the upper lip has been considered another hallmark of the FAS since its initial definition. However, not only rating the thickness of the upper lip, but rather scoring the attenuation or lack of the Cupid's bow shape of the upper lip is a more precise way to evaluate this feature as shown for scores 4 and 5 of the lip/philtrum guide. Familial and ethnic variation in the thickness of the lips is great and even age tends to make the upper lip thinner. The thickness of the upper lip itself is greater in populations of African descent than in most other ethnic groups, making the use of ethnic-specific lip-philtrum guides most important for the evaluation of this feature [30].

10.10 Physical Features Not Included in the Diagnostic Criteria

- Long philtrum and short anteverted nose
- Ptosis of the eyelids
- Epicanthal folds
- Strabismus
- Low/flat nasal bridge
- Midface hypoplasia
- Abnormal palmar creases
- Camptodactyly

10.10.1 Long Philtrum and Short Anteverted Nose

Although it has been reported that a long philtrum is a characteristic feature of FAS, Moore et al. have suggested that the ratio between the length of the philtrum and size of the nose is often so disproportionate that it makes individuals with FAS seem to have long philtrums, when they in fact have short noses [31]. Particularly at birth, a short nose with anteverted nares is characteristic of infants with PAE, but is also a common feature in many non-exposed new-borns in varying degrees. Again,

subjectivity of its assessment and familial and ethnic variations have made these features less specific to the condition.

10.10.2 Ptosis of the Eyelids

Ptosis represents drooping of the upper eyelids, usually due to hypotonia or dysfunction of the muscles that elevate the upper eyelid. Very short palpebral fissures can be extreme and lead to blepharophimosis, a term that describes that the eye opening is very small, and this sometimes interferes with vision and can be confused with ptosis.

True drooping of the upper eyelids is seen in a subset of cases of FAS, whether unilateral or bilateral, and is associated with the diagnosis of FAS, and with impaired cognition in several PAE studies. Its frequency in different studies is very variable, perhaps because severe ptosis is seldom present and minor degrees are evaluated in a very subjective way by different examiners. Its evaluation is easier when it is asymmetric by comparing the position of the upper eyelids in both eyes.

10.10.3 Epicanthal Folds

Epicanthal folds represent skin redundancy around the inner-canthi of the eyes. They may cover the inner canthi and should always be pulled outwards in order to accurately access the inner canthus with the ruler when measuring the palpebral fissures. Their presence is often associated with a low underdeveloped nasal bridge in new-borns and infants; at this age, they can be considered normal variations. They often disappear at older ages as the nasal bridge grows. They are frequent in the normal population, particularly of Asian descent. They are very common in other syndromes, and are certainly not specific features of the FAS, but they do occur with increased frequency in children with PAE. Epicanthal folds appear to be also very common in the Finnish population with FASD [15].

10.10.4 Strabismus

Strabismus represents misalignment of the eyes. It can occur in all positions or only in some extreme positions of the eyes. It can be assessed verifying with a light that the red reflex is centered in both pupils at the same time. Strabismus will be called esotropia when one eye deviates inward or exotropia when it deviates outward. Esotropia can often be found in normal infants and often corrects itself spontaneously with time, whereas exotropia almost always represents an abnormal configuration or tone of the extraocular muscles due to an underlying neurologic or muscular dysfunction. Strabismus causes double vision and can lead to amblyopia of one of the eyes, the functional loss of visual acuity due to the use of only the other eye to focus, in order to avoid double vision. Strabismus is often present in cases of FASD,

but it is a very non-specific feature. Decreased visual acuity due to alteration of the visual cortex and decreased size of the optic nerves, as well as refraction errors and hypotonia of the extra-ocular muscles can all contribute to strabismus in these patients.

10.10.5 Low/Flat Nasal Bridge

A low nasal bridge is very dependent on age, familial variation and ethnic background. The bridge is low in most new-borns and grows persistently until adulthood, or even after. It is a less specific feature of FAS, and difficult to quantify on physical examination. A low nasal bridge is very often associated with the presence of epicanthal folds. Not all studies have evaluated its presence in FASD, perhaps due to these reasons.

10.10.6 Midface Hypoplasia

The midportion of the face includes the malar bones and the maxillary bones. Sometimes a difference is made among maxillary hypoplasia and malar hypoplasia, but midface hypoplasia reflects a combination of the two. Underdevelopment of the midface results from deficiency of the neuroectoderm-derived facial mesenchyme, specifically the medial and lateral nasal processes [32]. An objective assessment of this feature is extremely difficult, due to several factors. Familial and ethnic variations are significant, but also the fact that the midface grows mostly in the first decade and the mandible later makes its assessment challenging. In a standing adult, the malar bones and the chin are approximately in the same plane, so any retrusion of the midface behind this plane is an objective measure of midface hypoplasia. However, this is only true if the mandible is of normal size, and varying degrees of micrognathia are frequent in the general population, although not a common or specific feature of FAS. Measurements of the tragomaxillary and tragomandibular arches and calculation of the ratio of both have been the best attempts to quantify the degree of midface hypoplasia. However, great variation in the size of the mandible makes this quotient unreliable and subjective as well. An excellent marker of significant midface hypoplasia is a down-slant of the palpebral fissures. However, many researchers and clinicians will call midface hypoplasia in the absence of down-slanting palpebral fissures. Midface hypoplasia is very prevalent in children with FAS, but is also frequent in normal non-exposed individuals. For all these reasons, this is not a reliable and specific feature of FAS.

10.10.7 Railroad Track Ears

The ears have two curved borders of cartilage around the semi-circular shell-shaped "concha". The outer border is the helix. In its frontal margin, the helix curves backward and ends in the center of the concha; this portion is called the crus of the helix.

The inner border or antihelix crosses the concha in its upper margin usually reaching the space under the folded antihelix. The shape of two parallel rails can be formed by a prominent horizontal crus of the helix that reaches the end of the concha, in combination with a prominent crus of the antihelix and is named the Railroad Track Ears. This is a finding in less than 20% of patients and not very specific of FAS, since it can be present in many genetic syndromes. However, it is very rare in the general population. Although the mechanism leading to this minor anomaly is unknown, we suggest that different amounts or patterns of cell migration from the branchial arches to form the auricle could be responsible.

10.10.8 Abnormal Palmar Creases

Normal hand creases are a reflection of early movement in utero in the late embryonic period. Once the hands are formed, movement will carve the palms and the volar aspect of the fingers with flexion creases that represent these early movements. Palmar creases are visible at 12–13 weeks gestation [33, 34]. The three creases over the volar face of the fingers are visible at 9 weeks, indicating flexion of the fingers precedes flexion of the palm. Absent or decreased early movement can be reflected in absent or very faint creases. Abnormal configuration of the creases can represent a qualitatively different movement with a different angle than the usual. Interestingly, in a normal hand, three primary palmar creases are seen. The thenar crease surrounds the muscle mass of the thenar eminence containing the three muscles that adduct, abduct and oppose the thumb. This is characteristic of primates and includes our most important hand function, opposition of the thumb over the palmar surface. Two other creases cross the palm from side to side. The proximal palmar crease often joins the thenar creases in its medial aspect. The distal transverse palmar crease is usually independent, reaches the ulnar extreme of the hand but usually does not reach its radial end.

Several types of abnormal palmar creases can occur in FAS, but the most characteristic anomaly is the Hockey Stick Crease. In that case, the distal transverse palmar crease curves sharply and reaches the distal end of the palm in the space between digits 2 and 3. The angle must be somewhat close to 90° but is often more obtuse. A straight crease without a curve or a very slight curve reaching the interdigital space is considered a variant of normal. Calling this feature is quite subjective, since minor angulations are often seen. The hockey stick crease is not specific to FAS, but appears to indicate a particular abnormal movement early in gestation. It can also be seen frequently in CHARGE syndrome and other genetic conditions [35]. In FAS, the thenar crease is usually normal, reflecting there is no radial/thumb deficiency and its opposition is normal. The proximal transverse palmar crease can be hypoplastic, much shorter than usual; the segment close to the thenar crease is most often present, but can be short. Given the importance and frequency of the hockey stick crease, this feature is included in the Dysmorphology score as such, and the rest are included as abnormal palmar creases. Extra creases can be present in the palms, and are usually normal variation, often shared by other family members.

10.10.9 Joint Contractures and Incomplete Extension of the Fingers (Camptodactyly)

Limitation of pronation-supination at the elbows below 180° can occur in FAS, and is either a consequence of radioulnar synostosis or due to dislocation of the head of the radius. Joint contractures are rarely seen with the severity typical of the multiple arthrogryposis syndromes, but joint limitations can happen in any large or small joint. Contractures at the interphalangeal joints, causing limitations of full extension of the fingers, are the most frequent type of contractures. Often, the contracture is reducible, but the patient himself is unable to actively fully extend one of more fingers. In cases in which the insult to the brain by PAE occurs prior to the ninth week of pregnancy, when fetal hand movement begins and the interphalangeal creases are formed, these contractures are associated with absent or faint metacarpophalangeal or proximal or distal interphalangeal creases. When no movement has ever occurred in a finger, no creases will be present.

10.10.10 Other Associated Dysmorphic Features

Prognathism has often been noted in FASD and represents an excessive prominence of the mandible, but no objective measurement has been proposed. Midface hypoplasia can lead to apparent relative prognathism, but that does not reflect a primary excess in the growth of the mandible. Hypoplastic nails and hypertrichosis have also been reported frequently. However, these are infrequent features, not specific to FASD. The presence of markedly hypoplastic nails can suggest exposure to anticonvulsants and Toluene embryopathy or a different condition, such as the genetic Coffin Siris syndrome. Hypertrichosis is a frequent feature of immobile fetuses and also present in other syndromes, but is certainly not a specific feature of FASD.

10.11 Correct Assessment of the Dysmorphic Features

We have mentioned several characteristics for the correct assessment of the dysmorphic features in FAS, as part of a detailed and rigorous dysmorphological examination. Measuring the maximal occipito-frontal circumference correctly, approaching the ruler at the correct angle for the correct measurement of the palpebral fissures, and comparing the philtrum and upper lip with the lip/philtrum guides are critical. But most features are indeed somewhat subjective, particularly for non-experts. A good method to improve recognition is to have pictures at the bedside with clear positive cases for these features. Inter-examiner reliability among non-expert trained examiners has been proven to be great in several studies, both for measurements and for recognition of subjective features [36].

10.12 Photographic and Morphometric Analysis of the Dysmorphic Features

Routine 2D photographic assessment has been developed and used by various groups and is currently standard in the Canadian guidelines [29, 37]. Because it is impossible to measure at the required angle of the orbit in a flat picture, photographic measurement of the size of the palpebral fissures results in significantly smaller measures than those obtained through direct examination. In addition, it is the volume of the lip and philtrum that we are trying to assess, even though our model for comparison is the 2D lip philtrum guide. Shades and light effects can cause artifacts in picture that will cause distortion to the pillars and the median groove. Direct observation at an angle appears to be the best way. Also, in practice, we and other groups do not rely on that methodology and believe in the need for direct examination. However, some research studies or a need for diagnosis in remote underserved areas can clearly benefit from photography.

In order to address the fact that 2D photography is not the best method of assessment, a significant body of research has developed using 3D photography [38]. However, the camera is still heavy and costly, and its routine use in clinical settings is not viable. However, the contribution to knowledge about the face in FASD has been very valuable. Face signature graphs show potential for identifying heavily exposed children who lack the classic facial phenotype of FAS on external exam, but who have significant differences in shapes and volumes in the direction of those seen in FAS. These techniques have also made evident that the face in FASD is a continuum, and that assigning discrete features is somewhat artificial. In theory, a system that would digitalize the features of the child, the parents and other siblings could, in the future, serve as an objective measurement of the impact of PAE in the development of the face in a specific individual.

10.13 Differential Diagnosis

A very important role of the physical examination is also the recognition of other dysmorphic features and facial appearances that could suggest a different syndromic condition. Frequent genetic syndromes such as the 22q11.2 deletion syndrome, Williams syndrome, or the De Lange syndrome are used as examples of conditions sharing some features with FASD, and therefore merit necessary consideration in the differential diagnosis. Undertaking as a minimum, where available, a CGH microarray is important, but not strictly necessary. However, when additional dysmorphic features or major malformations not frequent in FASD are present, a consultation with a clinical geneticist, probably followed by more detailed genetic testing, will be necessary. Whereas an informed and trained pediatrician can certainly make a diagnosis of FAS, he/she should also have the ability to recognize those children who have other findings and should establish the need for further workup, by the appropriate specialist.

10.14 Conclusion

FAS and partial FAS are recognizable conditions on physical examination that should ideally be diagnosed by expert dysmorphologists but can also be diagnosed by well-trained pediatricians and other specialists. Unfortunately, the different diagnostic criteria use different combinations of dysmorphic features to make a diagnosis of FAS and pFAS. This fact makes diagnosis difficult for the first-line clinician who has to decide which criteria he will use. At present, a comparison between the five methods used for the diagnosis of an FASD indicates a lack of reliability in diagnosis between the various diagnostic Schemes [9]. Therefore, case series are, and will be, difficult to compare but, most importantly, prevalence figures among studies will not be equivalent. There is a need for consensus diagnostic criteria that will address these important issues.

Also, even though only three dysmorphic features are included in all diagnostic criteria, recognition of all the other features described in this chapter is essential. Their presence only confirms the child's problems are due to PAE and should not suggest that the child has a different syndromic diagnosis. Finally, a correct assessment of these features is essential, and training has been proven to be simple and effective.

Key Learning Points

- The fetal alcohol spectrum disorders (FASD) is an umbrella term for the different diagnoses resulting from prenatal alcohol exposure (PAE) in the unborn child.
- The presence of growth deficits, a small head and three specific facial features (short palpebral fissures, smooth philtrum and narrow vermillion of the upper lip) are used for the diagnoses of the fetal alcohol syndrome (FAS) and partial fetal alcohol syndrome (pFAS).
- This pattern of anomalies is specific enough that a diagnosis of FAS can be made even in the absence of confirmation of PAE.
- Other dysmorphic facial features and anomalies of the joints and hand creases should also be recognized, since they are also characteristic of FAS, although not formally included in the commonly used diagnostic criteria.
- A systematic and precise assessment of these features will be necessary for accurate diagnoses.
- Damage to the developing neural tissue that originates the facial mesenchyme can explain these facial features, and the neurologic dysfunction leading to abnormal embryonic movement can explain features of the joints and abnormal creases.
- Many of these features can also be present in the cases of FASD that do not fulfil criteria for FAS or pFAS.
- There is a need for improved recognition of these features and for the correct interpretation of additional dysmorphic findings in order to establish correct differential diagnoses with genetic syndromes and other exposures to teratogens.

References

1. Smith DW. Dysmorphology (teratology). J Pediatr. 1966;59(6):1150–69.
2. Hennekam RC, Biesecker LG, Allanson JE, Hall JG, Opitz JM, Temple IK, et al. Elements of morphology: general terms for congenital anomalies. Am J Med Genet A. 2013;161A(11):2726–33.
3. Lemoine P, Harousseau H, Borteyru JPMJ. Les enfants des parents alcoholiques: anomolies observees a propos de 127 cas [The children of alcoholic parents: anomalies observed in 127 cases]. Ouest Med. 1968;21:476–82.
4. Jones K, Smith D. Recognition of the fetal alcohol syndrome in early infancy. Lancet. 1973;302(7836):999–1001.
5. National Center on Birth Defects and Developmental Disabilities Centers for Disease Control and Prevention Department of Health and Human Services. Fetal alcohol syndrome: Guidelines for referral and diagnosis: National task force on fetal alcohol syndrome and fetal alcohol effects. 2004. http://www.cdc.gov/ncbddd/fas/documents/FAS_guidelines_accessible.pdf.
6. Hoyme HE, Hoyme DB, Elliott AJ, Blankenship J, Kalberg WO, Buckley D, Abdul-Rahman O, Adam MP, Robinson LK, Manning M, Bezuidenhout H, Jones KL, May PA. A South African mixed race lip/philtrum guide for diagnosis of fetal alcohol spectrum disorders. Am J Med Genet A. 2015 Apr;167A(4):752–5. https://doi.org/10.1002/ajmg.a.37023. Epub 2015 Feb 25. PMID: 25711340; PMCID: PMC4507499.
7. Hoyme HE, Kalberg WO, Elliott AJ, Blankenship J. Updated clinical guidelines for diagnosing fetal alcohol spectrum disorders. Pediatrics. 2016;138(2):e20154256.
8. Chudley AE, Conry J, Cook JL, Loock C, Rosales T, Leblanc N. Guidelines for diagnosis. CMAJ. 2005;172.
9. Coles CD, Gailey AR, Mulle JG, Kable JA, Lynch ME, Jones KL. A comparison among 5 methods for the clinical diagnosis of fetal alcohol spectrum disorders. Alcohol Clin Exp Res. 2016;40(5):1000–9.
10. Sulik KK, Johnston MC, Webb M. Fetal alcohol syndrome: embryogenesis in a mouse model. Science. 1981;214(4523):936–8.
11. Sulik KK. Genesis of alcohol-induced craniofacial dysmorphism. Exp Biol Med (Maywood). 2005;230(6):366–75.
12. Lipinski RJ, Hammond P, O'Leary-Moore SK, Ament JJ, Pecevich SJ, Jiang Y, et al. Ethanol-induced face-brain dysmorphology patterns are correlative and exposure-stage dependent. PLoS One. 2012;7(8):e43067.
13. Martin RA, Jones KL, Benirschke K. Absence of the lateral philtral ridges: a clue to the structural basis of the philtrum. Am J Med Genet. 1996;65(2):117–23.
14. May PA, Gossage JP, Kalberg WO, Robinson LK, Buckley D, Manning M, et al. Prevalence and epidemiologic characteristics of FASD from various research methods with an emphasis on recent in-school studies. Dev Disabil Res Rev. 2009;15(3):176–92.
15. Autti-Ramo I, Loimu L, Hoyme HE. Relationship between dysmorphic features and general cognitive function in children with fetal alcohol spectrum disorders. Am J Med Genet A. 2007;2923:2916–23.
16. Hoyme HE, May PA, Kalberg WO, Kodituwakku P, Gossage JP, Trujillo PM, et al. A practical clinical approach to diagnosis of fetal alcohol spectrum disorders: clarification of the 1996 institute of medicine criteria. Pediatrics. 2005;115(1):39–47.
17. Jones KL, Hoyme HE, Robinson LK, Del Campo M, Manning MA, Prewitt LM, et al. Fetal alcohol spectrum disorders: extending the range of structural defects. Am J Med Genet Part A. 2010;152(11):2731–5.
18. May PA, Fiorentino D, Phillip Gossage J, Kalberg WO, Eugene Hoyme H, Robinson LK, et al. Epidemiology of FASD in a province in Italy: prevalence and characteristics of children in a random sample of schools. Alcohol Clin Exp Res. 2006;30(9):1562–75.
19. Chudley AE, Conry J, Cook JL, Loock C, Rosales T, LeBlanc N. Fetal alcohol spectrum disorder: Canadian guidelines for diagnosis. CMAJ. 2005;172(5):S1–S21.

20. Tze WJ, Friesen HG, MacLeod PM. Growth hormone response in fetal alcohol syndrome. Arch Dis Child. 1976;51(9):703–6.
21. Hellström A, Jansson C, Boguszewski M, Olegård R, Laegreid L, Albertsson-Wikland K. Growth hormone status in six children with fetal alcohol syndrome. Acta Paediatr. 1996;85(12):1456–62.
22. Mattson SN, Jernigan TL, Riley EP. MRI and prenatal alcohol exposure: images provide insight into FAS. Alcohol Heal Res World. 1994;18(1):49–52.
23. Astley SJ, Aylward EH, Olson HC, Kerns K, Brooks A, Coggins TE, et al. Functional magnetic resonance imaging outcomes from a comprehensive magnetic resonance study of children with fetal alcohol spectrum disorders. J Neurodev Disord. 2009;1(1):61–80.
24. Roussotte FF, Sulik KK, Mattson SN, Riley EP, Jones KL, Adnams CM, et al. Regional brain volume reductions relate to facial dysmorphology and neurocognitive function in fetal alcohol spectrum disorders. Hum Brain Mapp. 2012;33(4):920–37.
25. Landgren M, Svensson L, Strömland K, Andersson Grönlund M. Prenatal alcohol exposure and neurodevelopmental disorders in children adopted from eastern Europe. Pediatrics. 2010;125(5):e1178–85.
26. Cook CS, Nowotny AZ, Sulik KK. Fetal alcohol syndrome: eye malformations in a mouse model. Arch Ophthalmol. 1987;105:1576–81.
27. Strömland K, Pinazo-Durán M, Stromland K, Pinazo-Duran MD. Ophthalmic involvement in the fetal alcohol syndrome: clinical and animal model studies. Alcohol Alcohol. 2002;37(1):2–8.
28. Jones KL, Hoyme HE, Robinson LK, Campo M, Manning MA, Prewitt LM, et al. NIH public access. Am J Med Genet. 2011;11:2731–5.
29. Astley S, Clarren S. A case definition and photographic screening tool for the facial phenotype of fetal alcohol syndrome. J Pediatr. 1996;129(1):33–41.
30. Hoyme HE, Hoyme DB, Elliott AJ, Blankenship J, Kalberg WO, Buckley D, et al. A South African mixed race lip/philtrum guide for diagnosis of fetal alcohol spectrum disorders. Am J Med Genet Part A. 2015;167(4):752–5.
31. Moore ES, Ward RE, Wetherill LF, Rogers JL, Jacobson SW, Robinson LK, et al. Unique facial features distinguish fetal alcohol syndrome patients and controls in diverse ethnic populations. Alcohol Clin Exp Res. 2007;31(10):1707–13.
32. Sulik KK, Johnston MC. Sequence of developmental alterations following acute ethanol exposure in mice: craniofacial features of the fetal alcohol syndrome. Am J Anat. 1983;166(3):257–69.
33. Kimura S, Kitagawa T. Embryological development of human palmar, plantar, and digital flexion creases. Anat Rec. 1986;216(2):191–7.
34. Stevens CA, Carey JC, Shah M, Bagley GP. Development of human palmar and digital flexion creases. J Pediatr. 1988;113(1 PART 1):128–32.
35. Lalani SR, Hefner MA, Belmont JW, Davenport SL. CHARGE syndrome. GeneReviews(®). 1993.
36. Jones KL, Robinson LK, Bakhireva LN, Marintcheva G, Storojev V, Strahova A, et al. Accuracy of the diagnosis of physical features of fetal alcohol syndrome by pediatricians after specialized training. Pediatrics. 2006;118(6):e1734–8.
37. Astley SJ, Stachowiak J, Clarren SK, Clausen C. Application of the fetal alcohol syndrome facial photographic screening tool in a foster care population. J Pediatr. 2002;141(5):712–7.
38. Suttie M, Foroud T, Wetherill L, Jacobson JL, Molteno CD, Meintjes EM, et al. Facial dysmorphism across the fetal alcohol spectrum. Pediatrics. 2013;131(3):e779–88.

New Approaches to Diagnosis: the Role of Neurobehavioral Disorder associated with Prenatal Alcohol Exposure (ND-PAE)

11

Julie A. Kable

Chapter Highlights

- Current development in diagnostic approaches in fetal alcohol spectrum disorder (FASD)
- Characterizing the core parts of FASD
- Ways forward in the suture for FASD diagnosis

11.1 Introduction

In a field that has already been criticized by outsiders as being plagued with too many acronyms, many have wondered why neurobehavioral disorder associated with prenatal alcohol exposure (ND-PAE) was created. ND-PAE was proposed to capture the range of mental health problems occurring in individuals with a history of prenatal alcohol exposure (PAE) with or without the associated physical characteristics (i.e., growth deficits, hypoplastic philtrum, thinned upper lip, epicanthal folds, small palpebral fissures). The disorder was included in the most recent revision of the American Psychiatric Association Diagnostic and Statistical Manual, fifth edition under the Conditions for Further Study section [1] and is intended to fill an important void in defining the mental health needs of individuals with fetal alcohol exposure. Clinics worldwide have initiated using the diagnosis to increase access to much needed habilitative care and mental health treatment of individuals with fetal alcohol spectrum disorders (FASDs).

The neurobehavioral consequences of PAE encompass an array of neurocognitive and neurobehavioral impairments with varying degrees of symptom expression

J. A. Kable (✉)
Departments of Psychiatry and Behavioral Sciences and Pediatrics, Emory University School of Medicine, Atlanta, GA, USA
e-mail: jkabl01@emory.edu

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_11

among individuals. A consistent diagnostic formulation of neurobehavioral symptoms associated with PAE has been elusive. In part, this is because of the inconsistency in the consumption patterns of women who drink in pregnancy [2–4] coupled with individual differences in metabolism [5] and other nutrition factors [6] that influence the impact of prenatal alcohol exposure on fetal growth and brain development. Both maternal and child genetic and epigenetic factors [7–10] and postnatal environmental factors have an impact on the functional outcomes of PAE [11]. Various diagnostic systems for FASDs [12–15] have attempted to formulate methods of identifying individuals with alcohol-related neurobehavioral impairment for appropriate diagnosis and to facilitate their access to treatment services but there is a lack of consistency across these systems [15, 16]. In addition, these systems do not directly map onto a unique recognized mental health diagnosis making it difficult to identify individuals impacted by PAE within existing mental health care systems, which would facilitate defining the scope of the problem and appropriate resources needed for care [17]. Historically, individuals seen in FASD diagnostic clinics have been given multiple diagnostic codes to document the range of impact of PAE on their everyday functioning but often did not benefit from the same intervention approaches associated with these diagnoses [18], suggesting the need for the development of targeted interventions that focused in repairing alcohol-specific brain damage [19].

As previously the only formal recognized diagnostic codes for FASDs that could be used were medical disorders described by the International Classification of Diseases (ICD) of the World Health Organization [20] and were limited to those who had facial dysmorphia and growth delays, the development of a specific mental health diagnosis for alcohol-affected individuals may improve recognition and access to diagnostic assessments and habilitative care services for those without alcohol-related dysmorphology. Individuals with PAE who did not meet full criteria for FAS have been found to be at higher risk than those who are diagnosed with FAS/pFAS for a number of adverse life outcomes, including delinquency, school failure, and substance abuse problems [21]. These outcomes are most likely the result of the systemic barriers to accessing habilitative care in the absence of a clear diagnosis.

Although the *Diagnostic and Statistical Manual of Mental Disorders,* fifth edition (DSM-5) has had its criticisms [22], the document is used to define psychiatric disorders for both clinical research and practice by various professionals and the criteria used for identification of the neurobehavioral consequences of PAE had to be formulated within a framework that could be used by these various mental health professionals. This is in stark contrast to traditional FASD diagnostic formulations that were predominantly implemented by geneticists or developmental pediatricians either in independent practice or in the context of multidisciplinary team, which are still viewed as the ideal context for FASD assessments [23]. Although these diagnostic teams are also appropriate for making the ND-PAE diagnosis, there is an increasing awareness that such contexts cannot meet the need relative to the estimates of the prevalence of alcohol-affected individuals [24].

Practice Points:
The Issues That Needed to be Addressed

1. The impact of PAE varies as a result of a combination of factors, including genetics, epigenetics, metabolic functioning, nutritional status, and postnatal environmental factors.
2. Poor recognition of the mental health aspects of PAE may contribute to adverse life outcomes.
3. Although multidisciplinary teams are the "gold standard" for diagnosis, they are not practical for meeting the needs of those impacted by PAE.

11.2 Development of the Criteria

Although PAE is associated with a wide-ranging spectrum of neurobehavioral effects [18, 25], three domains of functioning were proposed as being essential to identifying those who were adversely impacted by PAE and in need of mental health services. They are as follows: [1] neurocognitive, [2] self-regulation, and [3] adaptive functioning. The neurocognitive domain includes five possible symptoms including impairments in global intellectual functioning, executive functioning, learning, memory, or visual-spatial reasoning and the self-regulation domain includes three possible symptoms, including impairment in mood or behavioral regulation, attention deficits, or impairment in impulse control. Within each of these domains, only one symptom is needed to make the diagnosis but for the adaptive domain, two of four possible symptoms (impairments in communication, social, daily living skills, or motor functioning) need to be endorsed and one of the symptoms must be either communication or social impairments. To meet criteria for ND-PAE, the neurobehavioral problems have to cause clinically significant levels of impairment in social, academic, occupational, or other areas of functioning and cannot be explained by substance use, environmental neglect, or by other medical conditions, including head trauma or other genetic disorders. Although many of the symptoms of ND-PAE have overlap with symptoms of other neurobehavioral disorders, the uniqueness of the disorder is derived from the convergence of the domains and the unique symptom characteristics within each domain.

Although establishing a threshold of PAE that produces clinically significant levels of impairment is a difficult task as a result of the variability of symptoms seen in offspring of women who drink during pregnancy [26, 27], a history of "more than minimal" levels of PAE is needed to make the diagnosis, which was defined as being greater than 13 US drinks per month during pregnancy and no more than 2 US drinks on any one occasion. The threshold was chosen to establish a minimum level so that the diagnosis was not over-used as the base rate of drinking any alcohol among women of child-bearing years is relatively high [28] and the impact of low levels of drinking has been controversial [27].

Practice Points:
Crucial Considerations in the Development of the Diagnostic Framework

1. Impairment in neurocognitive, self-regulation, and adaptive functioning are seen as critical for making a diagnosis of ND-PAE.
2. A history of *more than minimal* levels of PAE is needed to make the diagnosis.

11.3 Symptoms Within Each of the Neurobehavioral Domains

Impairment in neurocognitive functioning. Although *general cognitive impairments* or impairments in intellectual functioning were among the first neurobehavioral symptoms identified in children with fetal alcohol syndrome (FAS) [29], there is an increased awareness that most individuals with a fetal alcohol spectrum disorder (FASD) do not meet criteria for "intellectual disability." Estimates of the level of intellectual functioning (IQ) have fallen in the 70s across numerous studies, but the range of IQ is often quite broad among the various samples [30]. For those that meet criteria for an intellectual disability, which is the first possible symptom of neurodevelopmental impairment in ND-PAE, access to much needed intervention services to support the individual's attainment of basic life skills is relatively easy. Unfortunately for those who do not meet these criteria, treatment for their neurodevelopmental impairments is not as readily obtainable. After reviewing the range of potential neurocognitive impairments associated with PAE, which is quite large, four additional symptoms (executive functioning, learning, memory, and visual-spatial reasoning) were selected. The symptoms were selected based on their impact to everyday functioning and the extent to which the symptom could be assessed by the various mental health providers who would be using the diagnostic formulation of ND-PAE. A brief review of these symptoms in individuals with a history of PAE is provided [18].

Executive functioning (EF) impairments involve deficits in higher order cognitive processes involving attentional regulation, planning and organizational thinking, and problem-solving [31]. Individuals with FASDs demonstrate difficulties regulating basic attention, planning and organization, mentally manipulating information, changing strategies or thinking about things in more than one way, using abstract reasoning, and problems generalizing or applying knowledge to new situations [32–34]. These deficits appear to become progressively more acute as individuals age and have been linked to structural and functional brain abnormalities [35–37].

Learning impairments have often been found in children with FASDs using specific learning tasks [30] and specific measures of academic achievement [38, 39]. In addition, alterations in brain development have been found to be differentially related to academic skills in individuals with a history of PAE relative to controls [40]. One of the well-documented areas of relative deficit for individuals

with a history of PAE is in the area of math functioning [41] and evidence of microstructural brain damage in areas associated with mathematical thinking has been found in individuals with FASDs [42]. In addition, Riikonen and colleagues [43] found hypoperfusion in the left parieto-occipital region in a clinical sample of children with an FASD, which is a region associated with mathematical and logical thinking skills [44]. Children with FASD have also been found to have increased levels of negative school-related outcomes, including repeating grades, school failure, and dropping out of school [39, 45, 46]. Of course, these outcomes are determined by complex interactions between the child's neurodevelopmental status, environmental supports for academic success, and emotional stability but point to the need of frequent monitoring of their academic progress.

Memory skills impairments have been in children with FASDs but vary as a function of the task demands and modality of the stimuli used to elicit the responses [30, 47–52]. Reductions in hippocampal volume have been found to mediate the relationship between alcohol-related physical symptoms and performance on memory tasks [50]. Symptoms of memory impairment in everyday life have included difficulty recalling previously learned information, needing frequent reminders, losing or misplacing possessions, and difficulties with performing multi-step mental operations. Deficits in working memory skills have been found to be differentially impacted by PAE and difficulties with managing goals in working memory have been proposed as the underlying mechanism responsible for much of the cognitive impairment seen in children with FASDs [53], including the capacity to carry out many multi-step activities or instructions.

Visual reasoning or spatial thinking impairments are another area of specific cognitive functioning that appears to be differential impacted by PAE [54], which involves encoding and manipulating material presented visually or spatially. Children with FASDs have been found to do poorly on tests assessing visual and spatial memory, perceptual skills, and visual integration skills [41]. Symptoms of visual-spatial reasoning impairments seen in everyday life include having disorganized or poorly planned drawings or constructions, having impairments in perceptions of magnitude or quantity, having difficulties with identifying left from right, and having problems with visual alignment.

Impairment in self-regulation. Behavioral regulation and attentional problems are the most commonly reported problem behaviors by parents and professionals of individuals with FASDs. Early signs of dysregulation include increased stress reactivity [55–57], sleep disruption [58, 59], and increased levels of negative affectivity [60]. Later signs of dysregulation include an increased incidence of externalizing disorders [61–67], increased problems with substance abuse [68–70], and increased legal difficulties [21, 71]. Among adults diagnosed with FAS or fetal alcohol affect (FAE) [46], 94% had a history of mental health problems with the most common diagnoses being attention deficit disorder and depression. It is important to consider the fact that disruption to behavioral regulation or control systems among these individuals may result from a combination of alcohol-related brain damage resulting from PAE and adverse environmental circumstance often associated with maternal substance abuse. Caregiving disruptions of various kinds are the norm, rather

than the exception, when dealing with individuals who have been impacted by PAE and often these individuals originated in families with limited resources. It is also not uncommon for these individuals to also have a history of involvement with child protective services. When FASD children with a history of traumatic events have been compared to those with traumatic events but without any known PAE, a substantial overlap in behavioral symptoms was found, but the severity of behavioral symptoms was greater in those with FASD as was the neurocognitive impairment [72], suggesting the combination of symptom domains may be useful in differentiating those with alcohol-related neurodevelopmental impairments from those with only a history of exposure to adverse events.

Impairments in mood or arousal are commonly reported in individuals with FASDs and considerable knowledge has been amassed in recent years regarding the brain structures that are involved in regulating arousal and reward-seeking behavior. The arousal or reward network, also known as the ventral network, is typically involved in the affective or motivational components of reward processing and the inhibitory control network, also known as the dorsal network, is responsible for cognitive or behavioral control during reward processing [73]. Each of the areas in this neural circuitry have been found to be negatively impacted by PAE. Within the frontal cortex, an overall reduction in size has been reported [74–76]. In addition, alterations in the patterns of functional activation of the frontal cortex have been found [77–80]. The striatum [74], as well as the caudate nucleus [81–86] within the striatum, has been found to be smaller. In addition, the striatum has been found to be asymmetrical [87] and to have altered functional activation [78, 88, 89]. The thalamus has also been found to have reduced size [74, 82, 83, 90], to have altered metabolic characteristics (specifically NAA/Cho and NAA/Cr ratios) [91], and to have poorer diffusion characteristics [92]. Finally, the amygdala [82, 83] has also been found to be altered as a function of PAE. Disruption to the reward neural circuitry results in specific behavioral learning impairments. Children with FASDs have been found to have difficulties with affective learning (stimulus-reward associations) [47, 53, 93, 94], coping with negative feedback of failure [95], and difficulties with handling over-stimulation [96].

Difficulties with sustaining mental effort and being impulsive are commonly reported in individuals with FASD and are the reason that children with attention-deficit hyperactive disorder (ADHD) are commonly used in comparison studies [34, 94, 96–100]. Across several studies, researchers are able to differentiate the two clinical groups [101, 102] and pharmacological evidence suggests that individuals with an FASD do not benefit as well from traditional stimulant medications used to treat these symptoms [18, 103], suggesting that the underlying neurodevelopmental damage is different between the groups.

Impairments in adaptive functioning. Individuals with FASD have been found to have impairments in each of these areas of adaptive skills that are commonly assessed (communication, independent living skills, social, and motor functioning). The *communication impairments* often seen in individuals with FASD include the early acquisition of language and the later use of the pragmatic or figurative use of

language [104, 105]. Such impairments limit their ability to effectively communicate their needs to others and to comprehend communications from others.

Attainment of appropriate levels of basic life skills is vital for an individual to be able to function on his or her own. Individuals with FASD often demonstrate *impairments in independent living skills*, such as dressing, toileting, and making change [106]. They also have a poor understanding of the rules of personal safety [107], placing themselves often in harm's way. Finally, they often struggle with learning to appropriately perceive time intervals and with organizing daily schedules [108].

Social skills impairments are commonly reported in children with FASDs [109], but the symptoms are unique from other developmental disorders known to disrupt social development (i.e., ASD). Individuals with FASDs are often overly friendly with strangers, have difficulty reading social cues, and difficulty understanding social consequences or using social problem-solving skills. These impairments have been found to be independent of the facial dysmorphology, overall intelligence, and cannot be predicted by problem behavior alone, suggesting that this is a unique deficit [110].

Impairments in motor functioning have been identified throughout the lifespan of individuals with FASD [111, 112]. Impairments in neonatal and toddler motor functioning [113] are commonly reported, but impairments in later motor skills have also been found [30, 48, 114, 115]. Impairments in visual-motor integration [30, 116], which involves integrating visual and motor information to produce a response, are also frequently found. Impairments in this area impact handwriting, drawing, and performing math computations where column alignment is needed. Other areas of fine motor functioning have been found to be adversely impacted by PAE, including fine motor strength and coordination [116, 117] and increased motor tremors [114, 118]. Impairments in balance [118, 119], motor clumsiness [120], and gait [118, 121] have also been reported.

11.4 ND-PAE versus ARND

ND-PAE is distinctly different from previous conceptualizations of alcohol-related neurodevelopmental disorder (ARND). As originally proposed by the Institute of Medicine's report on FAS [14], ARND was a category for individuals who had some of the prenatal alcohol-related neurobehavioral characteristics but did not have the physical effects of PAE. Despite attempts to operationalize criteria for ARND by the various FASD clinical diagnostic systems [16], a consistent definition of ARND has yet to be achieved and no formulation has yet been subjected to the criteria needed to formalize a psychiatric diagnosis. ND-PAE was created as an alternative to ARND as the available definitions of ARND could not be reformulated into appropriate DSM-5 criteria and to ensure that all alcohol-affected individuals would have access to mental health services. ND-PAE also differs from ARND in that most FASD diagnostic classification systems incorporate head circumference and neuroimaging results into their criteria and ND-PAE does not. The exclusion of these markers of brain damage is the result of the fact that many mental

health professionals do not conduct physical examinations and do not have the capacity to independently obtain neuroimaging studies, resulting in the need for behavioral-based symptoms that could be assessed by all mental health professionals. The selected ND-PAE symptoms were not intended to capture the full array of neurodevelopmental deficits that have been associated with PAE but were selected to appropriately identify individuals who were negatively impacted by PAE but not individuals with other neurobehavioral problems.

Practice Points:
Core Criteria

1. Criteria for ND-PAE include one symptom from neurocognitive and self-regulation domains and two of four criteria for the adaptive function domains.
2. Neurocognitive impairment symptoms include deficits in overall IQ, executive functioning, learning, memory, or visual-spatial reasoning.
3. Self-regulation impairment symptoms include problems with mood or arousal, impairments in attentional regulation, and problems with impulsivity.
4. Adaptive impairment symptoms include problems with social skills, independent living skills, communication skills, and motor skills.

11.5 Preliminary Data on the Psychometric Characteristics of ND-PAE, What Information Is Still Needed?

Additional diagnostic and taxometric research are still needed for the disorder to establish its reliability and validity. Estimates of the agreement among clinicians making the diagnosis in the context of conducting an archival clinical review of charts have been high with one study reporting an inter-rater agreement of 79% [122] and the other 98% [123], suggesting that the symptoms are able to be reliably assessed among mental health providers.

Establishing the diagnostic coverage, or to what extent do those who have the disorder get appropriately identified, and the descriptive validity of the symptoms, also referred to as the homogeneity of symptoms, are part of basic criteria for evaluating psychiatric classification [124]. To establish a latent trait of disease severity, the symptoms should have a high level of internal consistency and the rate of symptom endorsement should vary as a function of diagnostic status. Symptom endorsement should also provide increased power to accurately differentiate those who are affected or not.

The internal validity of the proposed disorder was evaluated using a sample of children diagnosed with either fetal alcohol syndrome (FAS) or partial FAS who were between 3 and 10 years of age and had enrolled in a math intervention study [125]. Symptoms were coded as present or absent using assessments conducted in the study, including standardized measures of neurocognitive and behavioral

functioning, parent interview, and direct observations of the child. The number of endorsed ND-PAE symptoms was not related to environmental factors, including poverty, exposure to trauma, and disruptions in placements, but was moderately related to the child's age with older children having more endorsed symptoms. ND-PAE symptoms were highly consistent, ranging from 0.74 to 0.77 in the models evaluated, and this did not vary by age. In an epidemiological sample identified as being at risk for ND-PAE based on their mother's report of drinking or their own physical characteristics [126] estimates of the homogeneity of symptoms were also high with Cronbach alphas also ranging in the mid to high 70s [125].

In an archival study conducted using a clinical sample diagnosed with the Canadian guidelines for making FASD diagnoses [122], defining significant levels of ND-PAE symptom impairment using a threshold of 2.0 standard deviation units (SD) from the mean on standardized measures of neurobehavioral outcomes resulted in low sensitivity, particularly with patients diagnosed with FAS or pFAS. A moderate correlation (Cramer V = 0.44) was reported between ND-PAE and those diagnosed with ARND. Among those diagnosed with FAS/pFAS, the agreement was statistically significant but low (Cramer V = 0.05). The authors indicated that ND-PAE symptoms operationalized at a 2.0 standard deviation level resulted in patients being less likely to be identified relative to Canadian diagnostic guidelines and concluded that the ND-PAE symptoms as defined in their study were too restrictive to adequately capture the range of FASD patients in their sample.

Using a clinical sample assessed for symptoms of ND-PAE [127], a factor analysis of symptoms indicated four factors were present with a strong first factor that included IQ, executive functioning, memory, visual-spatial reasoning, adaptive social communication and interaction, and adaptive daily living skills. The second factor included measures related to application of knowledge (learning and adaptive communication). The third factor included symptoms of attentional regulation problems and impulsivity. The fourth factor involved mood regulation. Although the approach was helpful in clarifying the pattern of neurobehavioral impairment in individuals with PAE, their sample size was small (n = 58) and would need to be replicated in larger samples.

Four alternative models for making a diagnosis, including using either a 1.5 or 1.0 cut-off of deviation on standardized measures for establishing symptom severity and models using only one symptom from the adaptive functioning domain or the recommended two symptoms with one of those being either social or communication impairment, were also evaluated in the sample of children enrolled in the math intervention study [126]. Using the proposed adaptive criteria, agreement was 60.7% when symptom threshold was defined as 1.5 standard deviation units and 83.9% when symptom threshold was defined as 1.0 standard deviation units. When the adaptive criteria were modified so only one symptom was needed, agreement improved to 82.1% for the 1.5 SD cut-off level and 89.3% for the 1.0 SD cut-off level. Collectively, the available evidence suggests the ND-PAE adaptive symptoms as proposed may be too restrictive [122, 125, 126] and recommendations have been made for modifying the adaptive functioning criteria to include only one symptom

rather than the 2 of 4 criteria recommended in the DSM-5 to expand the breadth of coverage of individuals adversely impacted by PAE [126].

Data regarding the relative contribution of each of the proposed symptoms in differentiating those affected by PAE from typically developing individuals and individuals with other mental health or developmental disorders is also needed. The discriminate validity of the symptoms in differentiating typically developing individuals has been explored in a sample of individuals enrolled in the large multi-site project attempting to distinguish the unique neurobehavioral profile of alcohol-affected individuals from typically developing individuals and from various contrast groups, including those with ADHD, various behavioral problems, and developmental problems [128]. Using area under the curve analysis to assess the symptom's discriminate validity relative to a diverse contrast group comprised of typically developing individuals and those with ADHD, estimates ranged from the poor (mid 60s) to high (low 80s) depending on the threshold used for endorsing symptoms and whether 1 or 2 adaptive functioning symptoms were required. Modifications of the adaptive functioning criteria were also recommended in this study to improve the discriminative validity of the symptoms [129].

Convergent validity with those diagnosed with ARND using a standard checklist in combination with standardized scores was also evaluated. The authors reported a 90.1% area under the curve for predicting ARND based on meeting criteria for ND-PAE [130]. They concluded that the diagnostic constructs were similar and had a great deal of shared variability.

Practice Points

1. Inter-rater agreement and internal validity of symptoms are in the adequate to high range.
2. Discriminant validity has ranged from poor to high depending on the threshold needed for endorsement of symptoms and adaptations of the criteria used for impairments in adaptive functioning, which appears to be too restrictive as currently stated in the DSM-5.

11.6 Summary

The lack of a specific psychiatric disorder characterizing the neurobehavioral impairments associated with PAE has contributed to the difficulties many of these individuals and their families experience in accessing mental health services. Previous studies have reported that individuals with PAE who did not meet full criteria for FAS are at higher risk than those who are diagnosed for a number of adverse life outcomes. The inclusion of a diagnosis of ND-PAE in the DSM-5 is to facilitate recognition of the treatment needs of individuals negatively impacted by PAE, irrespective of whether or not they have the associated dysmorphic facial features and growth impairment. Although there is substantial support for the inclusion

of the proposed symptoms, the unique formulation of these symptoms may need modifications based on the preliminary results of the studies assessing the internal consistency and discriminative validity of the symptoms of the proposed disorder.

Further Reading

- Glass L, Mattson SN. Fetal alcohol spectrum disorders: a case study. J Pediatr Neuropsychol. 2017;3(2):114–35.
- Hagan JF Jr, Balachova T, Bertrand J, Chasnoff I, Dang E, Fernandez-Baca D, et al. Neurobehavioral disorder associated with prenatal alcohol exposure. Pediatrics. 2016;138(4):e20151553.

References

1. American Psychiatric Association. Diagnostic and statisical manual of mental disorders. 5th ed. Washington, DC: American Psychiatric Association; 2013. p. 86, 798–801.
2. Halmesmaki E, Raivio KO, Ylikorkala O. Patterns of alcohol consumption during pregnancy. Obstet Gynecol. 1987;69(4):594–7.
3. Iversen ML, Sorensen NO, Broberg L, Damm P, Hedegaard M, Tabor A, et al. Alcohol consumption and binge drinking in early pregnancy. A cross-sectional study with data from the Copenhagen Pregnancy Cohort. BMC Pregn Childbir. 2015;15:327.
4. Fortin M, Muckle G, Anassour-Laouan-Sidi E, Jacobson SW, Jacobson JL, Belanger RE. Trajectories of alcohol use and binge drinking among pregnant Inuit women. Alcohol Alcohol. 2016;51(3):339–46.
5. Church MW, Abel EL, Dintcheff BA, Matyjasik C. Maternal age and blood alcohol concentration in the pregnant Long-Evans rat. J Pharmacol Exp Ther. 1990;253(1):192–9.
6. Keen CL, Uriu-Adams JY, Skalny A, Grabeklis A, Grabeklis S, Green K, et al. The plausibility of maternal nutritional status being a contributing factor to the risk for fetal alcohol spectrum disorders: the potential influence of zinc status as an example. Biofactors. 2010;36(2):125–35.
7. Mead EA, Sarkar DK. Fetal alcohol spectrum disorders and their transmission through genetic and epigenetic mechanisms. Front Genet. 2014;5:154.
8. Sulik KK. Fetal alcohol spectrum disorder: pathogenesis and mechanisms. Handb Clin Neurol. 2014;125:463–75.
9. Gilliam DM, Irtenkauf KT. Maternal genetic effects on ethanol teratogenesis and dominance of relative embryonic resistance to malformations. Alcohol Clin Exp Res. 1990;14(4):539–45.
10. Israel Y, Quintanilla ME, Sapag A, Tampier L. Combined effects of aldehyde dehydrogenase variants and maternal mitochondrial genes on alcohol consumption. Alcohol Res Health. 2006;29(4):281–5.
11. May PA, Gossage JP. Maternal risk factors for fetal alcohol spectrum disorders: not as simple as it might seem. Alcohol Res Health. 2011;34(1):15–26.
12. Astley SJ. Validation of the fetal alcohol spectrum disorder (FASD) 4-Digit Diagnostic Code. J Popul Therapeut Clin Pharmacol. 2013;20(3):e416-67.
13. Hoyme HE, May PA, Kalberg WO, Kodituwakku P, Gossage JP, Trujillo PM, et al. A practical clinical approach to diagnosis of fetal alcohol spectrum disorders: clarification of the 1996 institute of medicine criteria. Pediatrics. 2005;115(1):39–47.
14. Stratton K, Howe C, Battaglia F. Fetal alcohol syndrome: diagnosis, epidemiology, prevention and treatment. Washington, DC: National Academy Press; 1996.
15. Astley SJ. Comparison of the 4-digit diagnostic code and the Hoyme diagnostic guidelines for fetal alcohol spectrum disorders. Pediatrics. 2006;118(4):1532–45.

16. Coles CD, Gailey A, Mulle J, Kable JA, Lynch ME, Jones KL. A comparison among five methods for the clinical diagnosis of Fetal Alcohol Spectrum Disorders (FASD). Alcohol Clin Exp Res. 2016;40(5):1000–9.
17. Sanders J. A window of opportunity: the proposed inclusion of Fetal Alcohol Spectrum Disorder in the DSM-5. J Develop Disabilit. 2013;19(3):7–14.
18. Kable JA, O'Connor MJ, Olson HC, Paley B, Mattson SN, Anderson SM, et al. Neurobehavioral disorder associated with prenatal alcohol exposure (ND-PAE): proposed DSM-5 diagnosis. Child Psychiatry Hum Dev. 2016;47(2):335–46.
19. Kable JA, Reynolds JN, Valenzuela CF, Medina AE. Proceedings of the 2013 annual meeting of the Fetal Alcohol Spectrum Disorders Study Group. Alcohol. 2014;48(7):623–30.
20. World Health Organization. International statistical classification of diseases and related health problems. 10th revision ed. 2010. Geneva, Switzerland: World Health Organization; 2011.
21. Streissguth AP, Barr HM, Kogan J, Bookstein FL. Primary and secondary disabilities in fetal alcohol syndrome. In: Streissguth AP, Kanter J, editors. The challenge of fetal alcohol syndrome: overcoming secondary disabilities. Seattle, WA: Seattle University of Washington Press; 1997. p. 25–39.
22. Insel TR. Mental disorders in childhood: shifting the focus from behavioral symptoms to neurodevelopmental trajectories. JAMA. 2014;311(17):1727–8.
23. Carmichael OH. Advancing recognition of fetal alcohol spectrum disorders: the proposed DSM-5 diagnosis of neurobehavioral disorder associated with prenatal alcohol exposure (ND-PAE). Curr Dev Disord Rep. 2015;2(3):187–98.
24. May PA, Chambers C, Kalberg W, Zelnner J, Feldman H, Buckley D, et al. Prevalence of fetal alcohol spectrum disorders in 4 US communities. JAMA. 2018;319(5):474–82.
25. Senturias YS. Fetal alcohol spectrum disorders: an overview for pediatric and adolescent care providers. Curr Probl Pediatr Adolesc Health Care. 2014;44(4):74–81.
26. Abel EL. Fetal alcohol syndrome: from mechanism to prevention. Boca Raton, FL: CRC Press; 1996. 336 p.
27. Reynolds JN, Valenzuela CF, Medina AE, Wozniak JR. Proceedings of the 2014 annual meeting of the fetal alcohol spectrum disorders study group. Alcohol. 2015;49(5):453–60.
28. Tan CH, Denny CH, Cheal NE, Sniezek JE, Kanny D. Alcohol use and binge drinking among women of childbearing age - United States, 2011-2013. MMWR Morb Mortal Wkly Rep. 2015;64(37):1042–6.
29. Jones KL, Smith DW, Ulleland CN, Streissguth P. Pattern of malformation in offspring of chronic alcoholic mothers. Lancet. 1973;1(7815):1267–71.
30. Mattson SN, Riley EP. A review of the neurobehavioral deficits in children with fetal alcohol syndrome or prenatal exposure to alcohol. Alcohol Clin Exp Res. 1998;22(2):279–94.
31. Lyon GR. The need for conceptual and theoretical clarity in the study of attention, memory, and executive functioning. In: Lyon GR, Krasnegor NA, editors. Attention, memory, and executive functioning. Baltimore: Paul Brookes Publishing Co; 1996. p. 3–10.
32. Khoury JE, Milligan K, Girard TA. Executive functioning in children and adolescents prenatally exposed to alcohol: a meta-analytic review. Neuropsychol Rev. 2015;25(2):149–70.
33. Rai JK, Abecassis M, Casey JE, Flaro L, Erdodi LA, Roth RM. Parent rating of executive function in fetal alcohol spectrum disorder: a review of the literature and new data on Aboriginal Canadian children. Child Neuropsychol. 2016:1–20.
34. Kingdon D, Cardoso C, McGrath JJ. Research Review: executive function deficits in fetal alcohol spectrum disorders and attention-deficit/hyperactivity disorder - a meta-analysis. J Child Psychol Psychiatry Allied Discip. 2016;57(2):116–31.
35. Wozniak JR, Muetzel RL. What does diffusion tensor imaging reveal about the brain and cognition in fetal alcohol spectrum disorders? Neuropsychol Rev. 2011;21(2):133–47.
36. Kodali VN, Jacobson JL, Lindinger NM, Dodge NC, Molteno CD, Meintjes EM, et al. Differential recruitment of brain regions during response inhibition in children prenatally exposed to alcohol. Alcohol Clin Exp Res. 2017;41(2):334–44.

37. Gautam P, Nunez SC, Narr KL, Kan EC, Sowell ER. Effects of prenatal alcohol exposure on the development of white matter volume and change in executive function. Neuroimage Clin. 2014;5:19–27.
38. Nash K, Stevens S, Rovet J, Fantus E, Nulman I, Sorbara D, et al. Towards identifying a characteristic neuropsychological profile for fetal alcohol spectrum disorders. 1. Analysis of the Motherisk FASD clinic. J Popul Therapeut Clin Pharmacol. 2013;20(1):e44-52.
39. Howell KK, Lynch ME, Platzman KA, Smith GH, Coles CD. Prenatal alcohol exposure and ability, academic achievement, and school functioning in adolescence: a longitudinal follow-up. J Pediatr Psychol. 2006;31(1):116–26.
40. Glass L, Moore EM, Akshoomoff N, Jones KL, Riley EP, Mattson SN. Academic difficulties in children with prenatal alcohol exposure: presence, profile, and neural correlates. Alcohol Clin Exp Res. 2017;41(5):1024–34.
41. Kable JA, Coles CD. The impact of prenatal alcohol exposure on neurophysiological encoding of environmental events at six months. Alcohol Clin Exp Res. 2004;28(3):489–96.
42. Lebel C, Rasmussen C, Wyper K, Andrew G, Beaulieu C. Brain microstructure is related to math ability in children with fetal alcohol spectrum disorder. Alcohol Clin Exp Res. 2010;34(2):354–63.
43. Riikonen R, Salonen I, Partanen K, Verho S. Brain perfusion SPECT and MRI in foetal alcohol syndrome. Dev Med Child Neurol. 1999;41(10):652–9.
44. Dehaene S, Cohen L. Cerebral pathways for calculation: double dissociation between rote verbal and quantitative knowledge of arithmetic. Cortex. 1997;33(2):219–50.
45. Autti-Ramo I. Twelve-year follow-up of children exposed to alcohol in utero. Dev Med Child Neurol. 2000;42(6):406–11.
46. Streissguth AP, Barr HM, Kogan J, Bookstein FL. Understanding the occurrence of secondary disabilities in clients with fetal alcohol syndrome (FAS) and Fetal Alcohol Effects (FAE). Final Report. Seattle, WA: University of Seattle Washington Publication Service; 1996.
47. Kerns KA, Don A, Mateer CA, Streissguth AP. Cognitive deficits in nonretarded adults with fetal alcohol syndrome. J Learn Disabil. 1997;30(6):685–93.
48. Janzen LA, Nanson JL, Block GW. Neuropsychological evaluation of preschoolers with fetal alcohol syndrome. Neurotoxicol Teratol. 1995;17(3):273–9.
49. Schonfeld AM, Mattson SN, Lang AR, Delis DC, Riley EP. Verbal and nonverbal fluency in children with heavy prenatal alcohol exposure. J Stud Alcohol. 2001;62(2):239–46.
50. Coles CD, Goldstein FC, Lynch ME, Chen X, Kable JA, Johnson KC, et al. Memory and brain volume in adults prenatally exposed to alcohol. Brain Cogn. 2011;75(1):67–77.
51. Manji S, Pei J, Loomes C, Rasmussen C. A review of the verbal and visual memory impairments in children with foetal alcohol spectrum disorders. Dev Neurorehabil. 2009;12(4):239–47.
52. Kodituwakku PW. Defining the behavioral phenotype in children with fetal alcohol spectrum disorders: a review. Neurosci Biobehav Rev. 2007;31(2):192–201.
53. Kodituwakku PW, Handmaker NS, Cutler SK, Weathersby EK, Handmaker SD. Specific impairments in self-regulation in children exposed to alcohol prenatally. Alcohol Clin Exp Res. 1995;19(6):1558–64.
54. Mattson SN, Roesch SC, Fagerlund A, Autti-Ramo I, Lyons KL, May PA, et al. Toward a neurobehavioral profile of fetal alcohol spectrum. Alcohol Clin Exp Res. 2010;34(9):1640–50.
55. Haley DW, Handmaker NS, Lowe J. Infant stress reactivity and prenatal alcohol exposure. Alcohol Clin Exp Res. 2006;30(12):2055–64.
56. Hellemans KG, Sliwowska JH, Verma P, Weinberg J. Prenatal alcohol exposure: fetal programming and later life vulnerability to stress, depression and anxiety disorders. Neurosci Biobehav Rev. 2010;34(6):791–807.
57. Oberlander TF, Jacobson SW, Weinberg JA, Grunau RE, Molteno CD, Jacobson JL. Prenatal alcohol exposure alters biobehavioral reactivity to pain in newborns. Alcohol Clin Exp Res. 2010;34(4):681–92.
58. Pesonen AK, Raikkonen K, Matthews K, Heinonen K, Paavonen JE, Lahti J, et al. Prenatal origins of poor sleep in children. Sleep. 2009;32(8):1086–92.

59. Scher MS, Richardson GA, Coble PA, Day NL, Stoffer DS. The effects of prenatal alcohol and marijuana exposure: disturbances in neonatal sleep cycling and arousal. Pediatr Res. 1988;24(1):101–5.
60. Olson HC, Jirikowic T, Kartin D, Astley SJ. Responding to the challenge of early intervention for fetal alcohol spectrum disorders. Infants Young Child. 2007;20(2):172–89.
61. Fryer SL, McGee CL, Matt GE, Riley EP, Mattson SN. Evaluation of psychopathological conditions in children with heavy prenatal alcohol exposure. Pediatrics. 2007;119(3):e733–41.
62. Disney ER, Iacono W, McGue M, Tully E, Legrand L. Strengthening the case: prenatal alcohol exposure is associated with increased risk for conduct disorder. Pediatrics. 2008;122(6):e1225–30.
63. Walthall JC, O'Connor MJ, Paley B. A comparison of psychopathology in children with and without prenatal alcohol exposure. Mental Health Aspects of Developmental Disabilities. 2008;11(3):69–78.
64. Peadon E, Elliott EJ. Distinguishing between attention-deficit hyperactivity and fetal alcohol spectrum disorders in children: clinical guidelines. Neuropsychiatr Dis Treat. 2010;6: 509–15.
65. Nanson JL, Hiscock M. Attention deficits in children exposed to alcohol prenatally. Alcohol Clin Exp Res. 1990;14(5):656–61.
66. Streissguth AP, Bookstein FL, Barr HM, Sampson PD, O'Malley K, Young JK. Risk factors for adverse life outcomes in fetal alcohol syndrome and fetal alcohol effects. J Dev Behav Pediatr. 2004;25(4):228–38.
67. O'Connor MJ, Paley B. Psychiatric conditions associated with prenatal alcohol exposure. Dev Disabil Res Rev. 2009;15(3):225–34.
68. Baer JS, Ginzler JA, Peterson PL. DSM-IV alcohol and substance abuse and dependence in homeless youth. J Stud Alcohol. 2003;64(1):5–14.
69. Barr HM, Bookstein FL, O'Malley KD, Connor PD, Huggins JE, Streissguth AP. Binge drinking during pregnancy as a predictor of psychiatric disorders on the structured clinical interview for DSM-IV in young adult offspring. Am J Psychiatry. 2006;163(6):1061–5.
70. Yates WR, Cadoret RJ, Troughton EP, Stewart M, Giunta TS. Effect of fetal alcohol exposure on adult symptoms of nicotine, alcohol, and drug dependence. Alcohol Clin Exp Res. 1998;22(4):914–20.
71. Fast DK, Conry J. Fetal alcohol spectrum disorders and the criminal justice system. Dev Disabil Res Rev. 2009;15(3):250–7.
72. Henry J, Sloane M, Black-Pond C. Neurobiology and neurodevelopmental impact of childhood traumatic stress and prenatal alcohol exposure. Lang Speech Hear Serv Sch. 2007;38(2):99–108.
73. Passarotti AM, Sweeney JA, Pavuluri MN. Differential engagement of cognitive and affective neural systems in pediatric bipolar disorder and attention deficit hyperactivity disorder. J Int Neuropsychol Soc. 2010;16(1):106–17.
74. Sowell ER, Leow AD, Bookheimer SY, Smith LM, O'Connor MJ, Kan E, et al. Differentiating prenatal exposure to methamphetamine and alcohol versus alcohol and not methamphetamine using tensor-based brain morphometry and discriminant analysis. J Neurosci. 2010;30(11):3876–85.
75. Burke MW, Palmour RM, Ervin FR, Ptito M. Neuronal reduction in frontal cortex of primates after prenatal alcohol exposure. Neuroreport. 2009;20(1):13–7.
76. Coles CD, Lynch ME, Kable JA, Johnson KC, Goldstein FC. Verbal and nonverbal memory in adults prenatally exposed to alcohol. Alcohol Clin Exp Res. 2010;34(5):897–906.
77. O'Hare ED, Lu LH, Houston SM, Bookheimer SY, Mattson SN, O'Connor MJ, et al. Altered frontal-parietal functioning during verbal working memory in children and adolescents with heavy prenatal alcohol exposure. Hum Brain Mapp. 2009;30(10):3200–8.
78. Fryer SL, Tapert SF, Mattson SN, Paulus MP, Spadoni AD, Riley EP. Prenatal alcohol exposure affects frontal-striatal BOLD response during inhibitory control. Alcohol Clin Exp Res. 2007;31(8):1415–24.

79. Sowell ER, Lu LH, O'Hare ED, McCourt ST, Mattson SN, O'Connor MJ, et al. Functional magnetic resonance imaging of verbal learning in children with heavy prenatal alcohol exposure. Neuroreport. 2007;18(7):635–9.
80. Spadoni AD, Bazinet AD, Fryer SL, Tapert SF, Mattson SN, Riley EP. BOLD response during spatial working memory in youth with heavy prenatal alcohol exposure. Alcohol Clin Exp Res. 2009;33(12):2067–76.
81. Astley SJ, Aylward EH, Olson HC, Kerns K, Brooks A, Coggins TE, et al. Magnetic resonance imaging outcomes from a comprehensive magnetic resonance study of children with fetal alcohol spectrum disorders. Alcohol Clin Exp Res. 2009;33(10):1671–89.
82. Chen X, Coles CD, Lynch ME, Hu X. Understanding specific effects of prenatal alcohol exposure on brain structure in young adults. Hum Brain Mapp. 2011;33(7):1663–76.
83. Nardelli A, Lebel C, Rasmussen C, Andrew G, Beaulieu C. Extensive deep gray matter volume reductions in children and adolescents with fetal alcohol spectrum disorders. Alcohol Clin Exp Res. 2011;35(8):1404–17.
84. Mattson SN, Riley EP. Brain anomalies in fetal alcohol syndrome. In: Abel EL, editor. Fetal alcohol syndrome: from mechanism to prevention. Boca Raton, FL: CRC Press; 1996. p. 51–68.
85. Archibald SL, Fennema-Notestine C, Gamst A, Riley EP, Mattson SN, Jernigan TL. Brain dysmorphology in individuals with severe prenatal alcohol exposure. Dev Med Child Neurol. 2001;43(3):148–54.
86. Roussotte FF, Bramen JE, Nunez SC, Quandt LC, Smith L, O'Connor MJ, et al. Abnormal brain activation during working memory in children with prenatal exposure to drugs of abuse: the effects of methamphetamine, alcohol, and polydrug exposure. NeuroImage. 2011;54(4):3067–75.
87. Willford J, Day R, Aizenstein H, Day N. Caudate asymmetry: a neurobiological marker of moderate prenatal alcohol exposure in young adults. Neurotoxicol Teratol. 2010;32(6):589–94.
88. Clark CM, Li D, Conry J, Conry R, Loock C. Structural and functional brain integrity of fetal alcohol syndrome in nonretarded cases. Pediatrics. 2000;105(5):1096–9.
89. Cortese BM, Moore GJ, Bailey BA, Jacobson SW, Delaney-Black V, Hannigan JH. Magnetic resonance and spectroscopic imaging in prenatal alcohol-exposed children: preliminary findings in the caudate nucleus. Neurotoxicol Teratol. 2006;28(5):597–606.
90. Mattson SN, Riley EP, Jernigan TL, Ehlers CL, Delis DC, Jones KL, et al. Fetal alcohol syndrome: a case report of neuropsychological, MRI and EEG assessment of two children. Alcohol Clin Exp Res. 1992;16(5):1001–3.
91. Fagerlund A, Heikkinen S, Autti-Ramo I, Korkman M, Timonen M, Kuusi T, et al. Brain metabolic alterations in adolescents and young adults with fetal alcohol spectrum disorders. Alcohol Clin Exp Res. 2006;30(12):2097–104.
92. Lebel C, Rasmussen C, Wyper K, Walker L, Andrew G, Yager J, et al. Brain diffusion abnormalities in children with fetal alcohol spectrum disorder. Alcohol Clin Exp Res. 2008;32(10):1732–40.
93. Chasnoff IJ, Wells AM, Telford E, Schmidt C, Messer G. Neurodevelopmental functioning in children with FAS, pFAS, and ARND. J Dev Behav Pediatr. 2010;31(3):192–201.
94. Coles CD, Platzman KA, Raskind-Hood CL, Brown RT, Falek A, Smith IE. A comparison of children affected by prenatal alcohol exposure and attention deficit, hyperactivity disorder. Alcohol Clin Exp Res. 1997;21(1):150–61.
95. Kodituwakku PW, Kalberg W, May PA. The effects of prenatal alcohol exposure on executive functioning. Alcohol Res Health. 2001;25(3):192–8.
96. Kooistra L, Crawford S, Gibbard B, Kaplan BJ, Fan J. Comparing Attentional Networks in fetal alcohol spectrum disorder and the inattentive and combined subtypes of attention deficit hyperactivity disorder. Dev Neuropsychol. 2011;36(5):566–77.
97. Jacobson JL, Dodge NC, Burden MJ, Klorman R, Jacobson SW. Number processing in adolescents with prenatal alcohol exposure and ADHD: differences in the neurobehavioral phenotype. Alcohol Clin Exp Res. 2011;35(3):431–42.

98. Khoury JE, Milligan K. Comparing executive functioning in children and adolescents with fetal alcohol spectrum disorders and ADHD: a meta-analysis. J Atten Disord. 2019;23(14):1801–15.
99. Crocker N, Vaurio L, Riley EP, Mattson SN. Comparison of verbal learning and memory in children with heavy prenatal alcohol exposure or attention-deficit/hyperactivity disorder. Alcohol Clin Exp Res. 2011;35(6):1114–21.
100. Crocker N, Vaurio L, Riley EP, Mattson SN. Comparison of adaptive behavior in children with heavy prenatal alcohol exposure or attention-deficit/hyperactivity disorder. Alcohol Clin Exp Res. 2009;33(11):2015–23.
101. Coles CD. Fetal alcohol exposure and attention: moving beyond ADHD. Alcohol Res Health. 2001;25(3):199–203.
102. Mattson SN, Crocker N, Nguyen TT. Fetal alcohol spectrum disorders: neuropsychological and behavioral features. Neuropsychol Rev. 2011;21(2):81–101.
103. Doig J, McLennan JD, Gibbard WB. Medication effects on symptoms of attention-deficit/hyperactivity disorder in children with fetal alcohol spectrum disorder. J Child Adolesc Psychopharmacol. 2008;18(4):365–71.
104. Coggins TE, Timler GR, Olswang LB. A state of double jeopardy: impact of prenatal alcohol exposure and adverse environments on the social communicative abilities of school-age children with fetal alcohol spectrum disorder. Lang Speech Hear Serv Sch. 2007;38(2):117–27.
105. Thorne JC, Coggins TE, Carmichael Olson H, Astley SJ. Exploring the utility of narrative analysis in diagnostic decision making: picture-bound reference, elaboration, and fetal alcohol spectrum disorders. J Speech Lang Hear Res. 2007;50(2):459–74.
106. Jirikowic T, Kartin D, Olson HC. Children with fetal alcohol spectrum disorders: a descriptive profile of adaptive function. Can J Occup Ther. 2008;75(4):238–48.
107. Coles CD, Strickland DC, Padgett L, Bellmoff L. Games that "work": using computer games to teach alcohol-affected children about fire and street safety. Res Dev Disabil. 2007;28(5):518–30.
108. Astley SJ. Profile of the first 1,400 patients receiving diagnostic evaluations for fetal alcohol spectrum disorder at the Washington State Fetal Alcohol Syndrome Diagnostic & Prevention Network. Can J Clin Pharmacol. 2010;17(1):e132–64.
109. Kully-Martens K, Denys K, Treit S, Tamana S, Rasmussen C. A review of social skills deficits in individuals with fetal alcohol spectrum disorders and prenatal alcohol exposure: profiles, mechanisms, and interventions. Alcohol Clin Exp Res. 2012;36(4):568–76.
110. Kully-Martens K, Denys K, Treit S, Tamana S, Rasmussen C. A review of social skills deficits in individuals with fetal alcohol spectrum disorders and prenatal alcohol exposure: profiles, mechanisms, and interventions. Alcohol Clin Exp Res. 2012;36(4):568–76.
111. Osborn JA, Harris SR, Weinberg J. Fetal alcohol syndrome: review of the literature with implications for physical therapists. Phys Ther. 1993;73(9):599–607.
112. Bay B, Kesmodel US. Prenatal alcohol exposure - a systematic review of the effects on child motor function. Acta Obstet Gynecol Scand. 2011;90(3):210–26.
113. Coles CD, Smith I, Fernhoff PM, Falek A. Neonatal neurobehavioral characteristics as correlates of maternal alcohol use during gestation. Alcohol Clin Exp Res. 1985;9(5):454–60.
114. Aronson M, Lyllerman M, Sabel KG, Sandin B, Olegard R. Children of alcohol mothers. Developmental, perceptual, and behavioural characteristics as compared to matched controls. Acta Paediatr Scand. 1985;74(1):27–35.
115. Kyllerman M, Aronson M, Sabel KG, Karlberg E, Sandin B, Olegard R. Childern of alcohol mothers. Acta Paediatr Scand. 1985;74:20–6.
116. Conry J. Neuropsychological deficits in fetal alcohol syndrome and fetal alcohol effects. Alcohol Clin Exp Res. 1990;14(5):650–5.
117. Barr HM, Streissguth AP, Darby BL, Sampson PD. Prenatal exposure to alcohol, caffeine, tobacco, and aspirin: effects on fine and gross motor performance n 4-year-old children. Dev Psychol. 1990;26:339–48.
118. Marcus JC. Neurological findings in the fetal alcohol syndrome. Neuropediatrics. 1987;18(3):158–60.

119. Roebuck TM, Simmons RW, Richardson C, Mattson SN, Riley EP. Neuromuscular responses to disturbance of balance in children with prenatal exposure to alcohol. Alcohol Clin Exp Res. 1998;22(9):1992–7.
120. Steinhausen HC, Nestler V, Spohr HL. Development of psychopathology of children with the fetal alcohol syndrome. J Dev Behav Pediatr. 1982;3(2):49–54.
121. Conn-Blowers EA. Nurturing and educating children prenatally exposed to alcohol: the role of the counsellor. Int J Adv Couns. 1991;14(2):91–103.
122. Sanders JL, Breen RE, Netelenbos N. Comparing diagnostic classification of neurobehavioral disorder associated with prenatal alcohol exposure with the Canadian fetal alcohol spectrum disorder guidelines: a cohort study. CMAJ Open. 2017;5(1):E178–E83.
123. Bell CC, Chimata R. Prevalence of neurodevelopmental disorders among low-income African Americans at a clinic on Chicago' south side. Psychiatr Serv. 2015;66(5):539–42.
124. Blashfield RK, Draguns JG. Evaluative criteria for psychiatric classification. J Abnorm Psychol. 1976;85(2):140–50.
125. Kable JA, Coles CD. Empirical evidence supporting the internal validity of the ND-PAE diagnosis. Alcohol Clin Exp Res. 2015;39(SI):226A.
126. Kable JA. Update on the empirical evidence supporting the reliability and validity of the Neurobehavioral Disorder associated with Prenatal Alcohol Exposure (ND-PAE) diagnosis. American Psychiatric Association 168th Annual Meeting, Toronto, Canada; 2015.
127. Sanders JL, Netelenbos N, Dei SO. Construct and factorial validity of Neurobehavioral Disorder associated with Prenatal Alcohol Exposure (ND-PAE). BMC Psychol. 2020;8(1):53.
128. Mattson SN, Roesch SC, Glass L, Deweese BN, Coles CD, Kable JA, et al. Further development of a neurobehavioral profile of fetal alcohol spectrum disorders. Alcohol Clin Exper Res. 2013;37(3):517–28.
129. Mattson SN, Goh P, Sadler M, Riley EP. Validation of nd-pae diagnostic criteria and suggestions for improvement. American Psychiatric Association 168th Annual Meeting; Toronto, Canada; 2015.
130. Johnson S, Moyer CL, Klug MG, Burd L. Comparison of alcohol-related neurodevelopmental disorders and neurodevelopmental disorders associated with prenatal alcohol exposure diagnostic criteria. J Dev Behav Pediatr. 2018;39(2):163–7.

Diagnosing FASD in the Context of Other Overlapping Neurodevelopmental Presentations

12

Raja A. S. Mukherjee

Chapter Highlights

- How the behavioural symptoms present
- Overlapping presentation with other comorbid outcomes such as autism and attention deficit hyperactivity disorder (ADHD)
- How other risk factors in pregnancy, such as wider drugs, impact the developing fetus

Fetal alcohol spectrum disorders (FASD) often present as one of their common comorbid conditions to clinical services. It has been shown in a systematic review of the literature published in 2016, that there are over 428 comorbid conditions linked to FASD [1]. This highlights that clinical services will be more likely to have referrals for one of these conditions, rather than for an FASD diagnosis itself [2]. It is important, however, to consider these issues as comorbidities. As the understanding of the relationship between FASD and these comorbid conditions improves, better and more accurate modifications to treatment pathways occur [3].

Complicating this, is how to disentangle the effects of prenatal alcohol exposure from other aetiological factors. This is not always straightforward. This chapter will begin to address some of these issues. The chapter will provide an overview and focus more on the approaches to differentiate presentation and allow the clinician to understand the principles and basic methods by which this may take place.

R. A. S. Mukherjee (✉)
National FASD Specialist Behaviour Clinic, Surrey, UK
e-mail: Raja.Mukherjee@sabp.nhs.uk

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_12

12.1 Principles of Assessment

Fetal alcohol syndrome has been identified for over 40 years. Recently, it has also been recognised to be the condition that requires exclusion of other diagnoses, as well as identifying classic diagnostic factors [4]. In order to be confident of a diagnosis, it is important to consider a range of influences on the development of body and brain. The effects of these need to be excluded through careful analysis and assessment. This can be achieved either through a careful history, or as evidence grows, through differentiating psychometric properties found in the tests carried out. Only once this has been completed, can an FASD diagnosis be made with confidence. Due to the complexity of human behaviour and the interaction of some of the confounding factors, this is not always possible. In these situations, it is important to list all those factors involved in leading to the identified presentation. Diagnosis and assessment are discussed in other chapters.

12.2 Social Versus Biological Considerations

Diagnostically, when trying to differentiate the effects of different confounding factors, it is important to look at the biological basis of these presentations and not necessarily just the social effects. This is especially important when related to prenatal teratogenic compounds. For example, compounds such as heroin and cocaine both have societal impacts. Both are banned 'Class A' substances in the United Kingdom, yet each has very different biological effects on the brain and body. It is therefore important, when considering a biological process of teratogenic damage, to focus on the biological mechanisms that underpin the presentation. This is not to diminish these social impacts, more merely to highlight that diagnostic differentiation can only occur by understanding the biological processes involved. Figure 12.1 highlights the different areas that need consideration.

12.3 Information Gathering

A lack of accurate information is often the most common reason for not obtaining a diagnosis. Because so many of the areas highlighted in Fig. 12.1 are reliant on history, observation and gathering of information, it is an area that may well often be missing. For example, whilst records may suggest that prenatal drugs or alcohol were used, rarely is the amount, timing and dosage recorded. In fact, often the specific drugs are unknown. The same issue is seen for later trauma. Whilst evidence has shown that severe trauma and neglect can cause neurodevelopmental outcomes, some of this is based on the Romanian orphanage study [5]. The level of neglect described in those populations is not often seen in UK populations. Other studies have found lesser impact on neurocognitive outcomes where the levels of neglect are less [6]. Therefore, whilst that impact of neglect in extreme deprivation is clear,

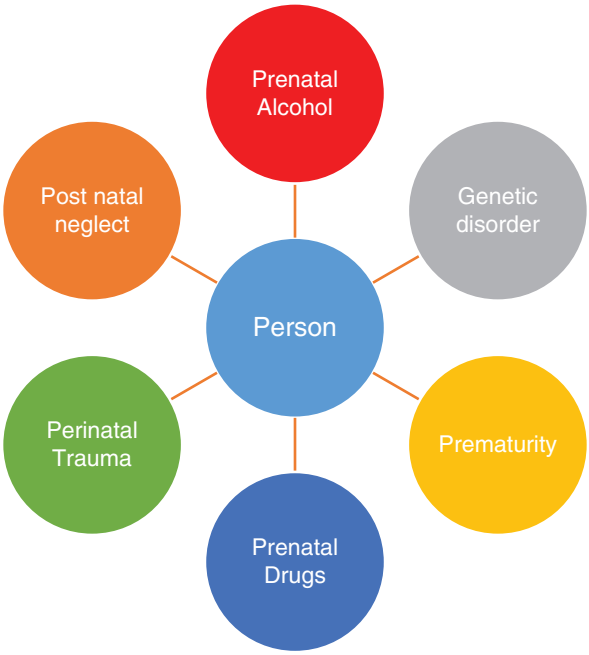


Fig. 12.1 Summary of main factors to rule out in diagnostic process when considering factors causing impact on early neurological development

whether or not this same level of neglect occurs or can be attributed to UK populations is unclear.

When the individual is adopted or fostered, often the required histories can be missing. Recording the information prior to placement is vital to later diagnosis [7]. The birth mother may well also not wish her medical records passed on. This means, once again, information can be missing. Even when the birth mother is present, because people do not always realise, they are pregnant or keep accurate records of their lifestyles, information is often based on subjective memories rather than accurate descriptions. Accurate information therefore is often lacking.

Whilst there is still a great deal not known and specific research to partition out effects is still needed, some understanding is available now. These will be considered in the next sections.

12.4 Causes and Outcomes

Part of the confusion around comorbidity relates to the relationships between different conditions. In the scientific literature, conditions which lead to underlying damage and those which are outcomes of that damage are often compared. This can sometimes be inappropriate.

In many ways, both are based on phenomenology. That is, taking descriptive symptoms and then applying strict criterion to identify that condition. Even when comparing outcomes this can remain a challenge to define strict criteria. In many

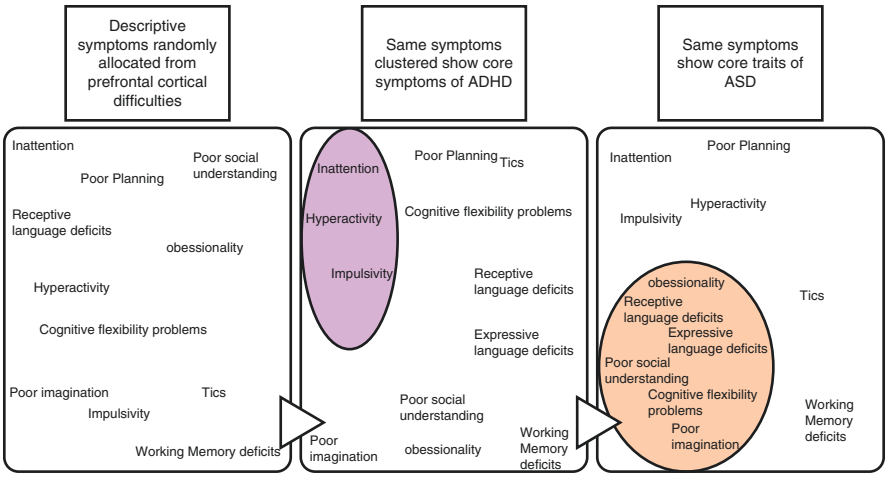


Fig. 12.2 Symptoms and diagnosis: Relationship between symptoms and phenomenology

cases, the symptoms overlap. Figure 12.2 highlights how attention deficit hyperactivity disorder (ADHD) and autism have similar overlapping characteristics. When taken from the same set of purely symptom descriptors, listed here from executive functioning symptoms, both disorders can be attributed from the same original list. For a condition such as FASD, nearly all these symptoms may well be seen. FASD, however, is a syndrome that has caused the damage to the developing brain and body rather than ADHD and autism, which are the outcomes of that underlying damage. Outcome diagnoses may well be comorbid conditions also seen, and as suggested the route by which FASD presents to clinical services. For example, ADHD is probably the most common behavioural diagnosis and pathway these children are presented to. When picking apart what has caused the damage, other factors that are considered aetiological need to be looked at.

12.5 Biological Considerations in Ruling Out Other Aetiological Factors

12.5.1 Genetic Disorders

By far the most significant group of aetiological conditions to rule out is genetic disorders. The majority of disorders seen in individuals can be attributed to an underlying genetic aetiology. Ever increasing sophistication in identifying these disorders has meant more conditions have had an attributable cause attached to them. The idea of single gene disorders leading to significant deficits has waned in that the understanding of underlying mechanisms and how multifactorial genetics, and more recently epigenetics, that is the way genes are modified by environmental influence, have changed this understanding [8].

Whilst it is not possible to rule out in its entirety the genetic effect, evidence has suggested that in the absence of the dysmorphology, a micro-array as a minimum would be sufficient to define that no significant genetic influence is occurring for the majority of cases [9]. In this situation, a normal result is preferred, as it indicates that genetic conditions are unlikely. Where dysmorphology exists or an abnormal finding is identified, it cannot be stated with absolute certainty, even of a benign variant, that no influence has been made on the presentation. In this situation, it is best to label this alongside the prenatal alcohol is an aetiological component to the overall presentation. Where a normal result is obtained and no dysmorphology is seen, it is possible to state with good confidence that genetic factors are unlikely to be contributing to the presentation.

12.5.2 Prematurity

Prematurity is another area where the level of clinical evidence of poor developmental outcomes is relatively clear [10]. The earlier the birth and the smaller the size of the fetus when born, the greater the likelihood of disability. Despite this, with modern care significant improvements have been made in longer-term outcomes. Generally, where the child is of a larger size or if the prematurity is after 34 weeks, defined as late preterm infants, the level of neurodevelopmental outcome falls away progressively. This is especially true if neonatal intensive care is not needed for any prolonged period [11]. By 37 weeks, the data would suggest almost normality in terms of the neurodevelopmental consequences compared to those born at full term. This area of late preterm infants is still a developing area. Research and clinical practice that improves outcomes is a constantly developing area.

Prematurity is an area which is often easy to track as the birth records follow the child and it is rare that this information is not known. Where prematurity, especially at a very early age, is seen, the neurodevelopmental consequences cannot be solely attributed to the alcohol. Case studies of individuals seen in clinical settings where there is significant prematurity and alcohol as well would suggest similar presentations to those where there is alcohol alone and no prematurity. The research in this area has not necessarily always considered the confounding effect of alcohol and therefore more research is needed before the effects of early prematurity and alcohol can be separated. As such, it is best in these situations with extreme prematurity before 34 weeks, to label both aetiological factors together. Between 34 and 37 weeks, the wider consideration of size and initial status would guide the outcome more than the term of gestation in itself.

12.5.3 Smoking

Smoking is a common associated behaviour seen in women who also consume alcohol. Whilst the rates of smoking have fallen, for example in the UK, it is still a common overlap with alcohol consumption by some. Tobacco smoke contains a mixture

of chemicals and substances with nicotine crossing the placenta [12]. Early data was suggestive that smoking caused a wide range of deficits including ADHD, conduct disorder, growth and behavioural issues as well as wider physical difficulties such as cleft lips and respiratory distress [13–15]. It is clear, however, that developmental delays and impacts on the individual can be seen, where genetic vulnerability has been taken into account, the studies have suggested that the genetic component may explain the presentation more than the smoking. For example, more recent studies have suggested that ADHD may not be attributed to smoking [16].

Whilst the scope to further delineate effects is needed, the suggestion that the symptoms seen in ADHD are not necessarily attributable to smoking allows, even now, some delineation from the effects of other teratogens.

12.5.4 Cannabis

Cannabis is a compound that is known to cross the placenta. The active compound of Delta-9-tetrahydrocannabinol, the main psychoactive component, has been shown in mothers who consumed high levels to cross and have impacts on development [12]. It has been shown to have effects on opiate receptors and can act as an endorphin. Binding sites for cannabis are distributed throughout the brain with highest density in frontal lobes, hippocampus, cingulate gyrus and cerebellum. Therefore, it could be speculated that these areas may well be impacted by heavy usage [17].

Some of the wider lifestyle issues related to cannabis use may lead to difficulties in identifying its outcomes in isolation when compared to other compounds. Evidence from human and animal literature point to some effects, but the other lifestyle factors may be interacting to have a greater impact. For example, a healthy diet can be protective of its effects [18].

Long-term effects on cognitive functioning have suggested broader reductions in general attainment even when controlling for other factors. There has been some suggestion that executive function may be affected, but the long-term ongoing effects remain uncertain [18].

Interestingly, in one study of children aged 9–12 highlighting the six psychometric tests to assess executive function, subtests looking at impulse control, visual analysis and hypothesis testing were the most affected [19]. On the contrary, WISC-III Block Design and Picture Completion seem to be those most impacted by cannabis usage, allowing some differentiation from damage caused by other compounds [19].

12.5.5 Cocaine

Cocaine acts on mesolimbic dopaminergic pathways to increase dopamine and serotonin within the synapses. Activation of noradrenergic and serotonergic sites in the basal forebrain and cerebral cortex have also been noted to be affected. These

can have frontal lobe connections leading to some overlap in pathology with alcohol impacted areas [12, 17].

Where studies have been conducted on cocaine exposed neonates, concerns have been raised with the methods used and the overlap of other substances, especially alcohol [18]. High-level cocaine use has been noted to have effects on peoples' growth and behaviour, but the long-term effects are still uncertain and consensus is yet to be established [18].

Due to the lack of ability to separate the overlap based on current research, should cocaine be reported at high-level usage alongside alcohol it may not be possible at this time to differentiate them from an aetiological point of view. Therefore, both aspects should be considered as having an impact on the underlying presentation.

12.5.6 Opiates

Opiates are chemical substances used widely in medicine, but also are common drugs of abuse. Opiates are known to rapidly cross the placenta. Primarily, they bind to opioid receptors which are found in the central and peripheral nervous system as well as in the gastrointestinal tract. Many different types of opioid receptors have been found, up to 17 in total, although three major ones. These are distributed across parts of the cortex both pre- and post-synaptically. Research would suggest that it is the number of receptors that determines the overall effect [12, 17].

Many opiates have quick onset of action, but also abrupt withdrawal symptoms. These withdrawal symptoms are seen commonly in the neonates with significant withdrawal symptoms being an important management consideration [20].

Opiate use can often be seen alongside other poly substance abuse, with one recent study suggesting that where opiates have been consumed, but denied alcohol, 44% of cases were found to have actually also consumed alcohol therefore incorrectly reporting exposure [21]. The overlap of risk is therefore significant.

Longer-term studies of the impact of opiates are limited. No clear pattern of neurological findings has been identified. The limitations with these studies to date, would suggest they cannot be seen as conclusive [22]. The consensus is not yet clear. Where longer-term studies have been conducted, poly substance use, rather than opiates on their own, have suggested negative outcomes [23]. It is therefore not possible to attribute the outcomes to opiates alone. A consensus statement completed by the American Academy of Paediatrics suggested that there was broadly limited consensus and whilst some effects had been seen, these were not of a strong relationship [18].

Opiates have been used therapeutically for opiate addiction where usage is monitored and poly substance use therefore avoided, even during pregnancy, fewer pregnancy complications, greater birthweight and better developmental behaviour outcomes were seen compared to where uncontrolled heroine exposure was used [17]. These studies have limitations and do not, once again, offer definitive information.

Separation of these effects clearly needs greater study, but the impact of opiates in isolation from other compounds at present, seems to be of lesser severity, especially on long-term outcomes.

12.5.7 Others

Many other drugs of abuse were consumed potentially during pregnancy which have been shown to have mixed effects. Studies, for example, with methylphenidate and stimulants are limited. Their effects on brain and cognition have not been studied in great depth. It is therefore difficult to comment on longer-term impacts. Amphetamines are known to increase catecholamine concentrations in the brain via multiple mechanisms [12]. Consistent with other findings in human studies, overlapping usage with other compounds, such as alcohol and tobacco, is common. While some studies have shown neuropsychological deficits, other studies have shown no difference. Long-term conclusions, therefore, cannot be easily drawn [18, 24].

Antipsychotics, antidepressants and other medication in pregnancy is often more controlled and monitored. Whilst they are known to have some effects, especially physical, overlaps can be seen. For example, sodium valproate has similar impacts on the fetus as alcohol due to mechanisms of action overlapping. Other medications such as antidepressants or antipsychotics all have more controlled usage and monitoring which offer better insight into the impact on development than the uncontrolled usage in generic population-based samples and the cognitive impact is less [12, 25, 26].

It is only by taking accurate histories and identifying risks early that understanding will improve and interventions be developed. A good clear alcohol and drug history, based around both assessment of pre-pregnancy behaviour, as an indicator of later patterns, before exploring pregnancy behaviour is a necessity. It is important that this is conducted in an enquiring manner and not just as a 'tick box' exercise. Developing rapport with the individual, often involving midwifery, is essential in creating a therapeutic relationship. Bearing in mind, the majority of pregnancies are unplanned and that pregnancy-related behaviours do not often establish themselves until the pregnancy is confirmed, pre-pregnancy patterns of behaviour will be important to assess as well as pregnancy behaviour.

12.6 Comorbid Diagnoses Seen as an Outcome of the Underlying Neurological Damage Caused in FASD Cases

Whilst there is a fairly classic phenotype in individuals who have fetal alcohol spectrum disorders, for the majority of cases who are simply exposed to prenatal alcohol compared to those who develop fully diagnosed disorders, the level of difficulty in

social communication, inattention and hyperactivity do not reach a threshold by which they are considered disordered. When considering that exposure rates in some countries have been estimated to be 60% of the population [27], it is clear that not all of these cases that are exposed to alcohol are damaged. Therefore, there appears to be a graded response to the level of exposure and the amount of damage seen. There may well be some effect on the individual, without being sufficient in nature and degree, that would warrant a disorder diagnosis. This can lead to diagnostic confusion as some symptoms may be present, but not enough to meet any specific diagnostic threshold. As highlighted above, as well as in boxes one and two, outcome diagnoses such as ADHD and ASD are identified by meeting the characteristic symptoms over defined periods. For many, this may not be the case, leaving them with lower level difficulties not meeting the threshold for any particular diagnostic outcome other than the FASD.

Box 12.1 DSMV Diagnostic Criteria ADHD [28]

- Inattention criteria (9 symptom descriptors, must reach 6/9)
- Hyperactivity/Impulsivity (9 symptom descriptors, must reach 6/9 Children or 5/9) adults)
- Onset before the age of 12
- Must cause a functional deficit
- Must be seen across different situations

Box 12.2 DSM V Criteria ASD [28]

- Qualitative deficits in social communication in the three domains (3/3 domains must be met)
  - Social and emotional reciprocity
  - Non-verbal communication deficits
  - Making and maintaining friendship difficulties
- Restrictive and repetitive patterns of behaviour (2/4 domains must be met)
  - Stereotyped or repetitive movements
  - Insistence of sameness and inflexible adherence to routines
  - Highly restricted and repetitive interests
  - Hyper or hypo arousal to sensory inputs
- Symptoms present in early developmental period
- Symptoms cause significant impairment
- Symptoms not better explained by intellectual levels of communication

12.7 Developing Understanding and Relationship Between Comorbid Disorders

Increasingly, relationships and overlaps between conditions have been identified and delineated. As suggested above, cause and outcome are now better understood. ADHD and Autism are now known to not be caused by a single condition and are in fact most likely to be a range of conditions with different causes that have, for ADHD, impulsivity, inattention and hyperactivity at their core. For Autism, the same is also true. A range of conditions caused by different factors with social communication deficits, non-verbal communication issues and restrictive and repetitive patterns of behaviour being central. The rationale for a spectrum relates to the understanding that different causes can have different presentations within the spectrum for example, different types of social engagement, with variability within the same broad domain. This will, therefore, have different implications on diagnosis [29]. Figure 12.3 shows how the diagnostic rates vary between different research projects identified in the scientific literature showing even where structured approaches are used, inconsistency is seen.

Sociability within these different syndromes also varies. This adds to the diagnostic conundrum. Figure 12.4 summarises a piece of research conducted in Birmingham, UK, which identified differing sociability between different genetic syndromes. Many also met diagnostic criteria for wider ASD. In conducting the same test with each genetic disorder, different levels of sociability were identified. Those with less sociability appeared to be more in keeping with classical descriptions of Autism, whilst those with higher sociability did not. Even then, some met stricter ASD criteria [31].

The same can be said of an Autistic presentation in people with FASD. Whilst research studies have identified both ASD and FASD as conditions, both having

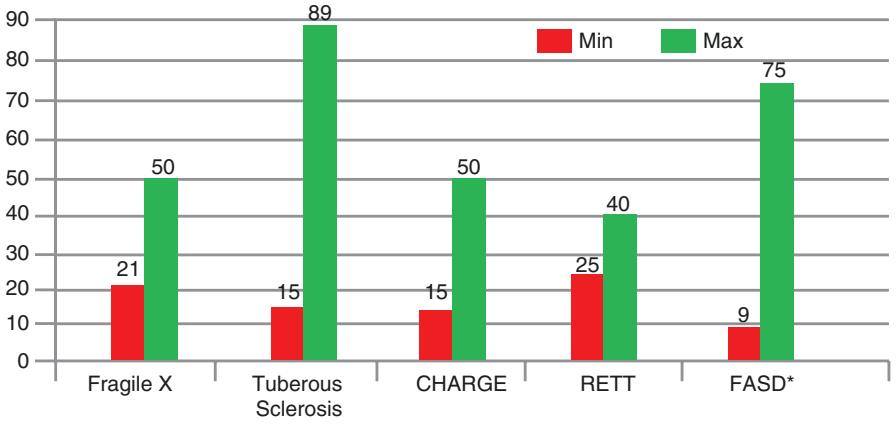


Fig. 12.3 Percentage rates of ASD diagnosed in different genetic syndrome [29]: *FASD data taken from wider research [30]

Fig. 12.4 Diagrammatic representation of different types of social functioning in people with different aetiologies [31]. Note: The FASD position is based on wider research [30]

social communication as a core deficit, due to the differences in sociability between the two syndromes, they have sometimes therefore been attributed in the literature as separate conditions. Instead however, increasingly, the relationship has been demonstrated to be less straightforward. Prenatal alcohol exposure and FASD are potential causes of the underlying neurological deficit, whilst the autism and ADHD outcomes of that underlying damage. Instead of being a classic Autism-type presentation, where an aloof sociability is seen, in keeping with the study conducted in Birmingham, because they are a much more pro-social group, yet still retain the social communication deficit at its core (alongside the required restrictive behaviours), they may not appear to be classically autistic. Yet when this group is systematically studied, many of its constituents meet ASD criteria. In the most extreme cases, the presentations may be far more complex. For example, in our specialist diagnostic clinic in the UK, where ASD and ADHD are routinely and thoroughly assessed in every patient alongside clarification of the FASD, it is not uncommon to see a wide overlap of individuals who have both of these common neurodevelopmental conditions [30, 32].

Rather than trying to delineate the presentation of ASD or ADHD from FASD, it becomes possible to delineate the type of ADHD or Autism seen in someone who is also diagnosed with FASD. This highlights the comorbid nature of the presentations with FASD.

For ADHD, those inattentive symptoms of distractibility, forgetfulness, inability to concentrate, poor organisation and distractibility are far more common than some of the hyperactivity symptoms. Impulsive symptoms, seen frequently with the classic FASD phenotype, are associated with problems in starting and stopping behaviours. Sustained attention and executive function deficits are commonly described; therefore, making it unsurprising that ADHD is a common presenting feature [3, 32].

The same can be said for Autism. Whilst the classic phenotype of autism is unusual, social communication difficulties are described as a core feature in many people who have prenatal alcohol exposure and FASD. Rigidity and the need for routine is another common description in this group. When considering the expanded criterion in Box 12.2, it becomes evident that some individuals with FASD may well reach a threshold for this presentation. The type of Autism seen in this group is not of the social communication style of classic Autism rather more pro-social approach,

with a lack of ability to modify or mitigate their behaviour to other people's needs, thus remaining part of the wider autistic spectrum. They may well show some evidence of sympathy, but lack true empathy for others individuals. It also means it can lead to diagnostic confusion [33]. The understanding of this relationship continues to be established.

The rationale here, consistent with the process undertaken in the UK National Clinic, would suggest that it is appropriate to evaluate everybody who is diagnosed with FASD to see whether or not they also meet the diagnostic outcome of ADHD and Autism as well.

The conditions, therefore, should not necessarily be seen as separate, but related. Understanding the pathways and relationships becomes important in terms of diagnosis, management and subsequent support for individuals.

Where the two conditions present themselves together, they should be analysed and assessed thoroughly as it modifies the treatment regime. Examples of modifying treatment are particularly seen in ADHD. Recent consensus guidance has highlighted that different approaches may well be warranted [3]. Where it is understood that the aetiological factors can influence the outcome and response to treatment, the importance of identifying what has caused the problem is vital. The same is true for Autism.

These outcomes represent two commonly seen comorbid conditions. Many more exist and influence the management. Delineating the differences between what are causes and outcomes is important in order to identify the importance of identification and recognition as this leads to different treatment approaches.

Practice Points

- Comorbidities are common in FASD.
- FASD is related to the factors causing damage to the brain and body and needs to be delineated from outcomes of that damage.
- When considering other aetiologies, a biological perspective is important to understand to look at the effects of each as a teratogen.
- Outcomes are common, such as ADHD and ASD, in this group but they may not be classic presentations.

References

1. Popova S, Lange S, Shield K, Mihic A, Chudley AE, Mukherjee RAS, Bekmuradov D, Rehm J. Comorbidity of fetal alcohol spectrum disorders: a systematic review and meta-analysis. Lancet. 2016;387:978–87.
2. Mukherjee RAS, Carlisle CS, Livesay AC. Neuropsychological aspects of prevention and intervention for FASD in Great Britain. J Paediatr Neuropsychol. 2017;3(1):61–7.
3. Young S, Absoud M, Blackburn C, Branney P, Colley B, Farrag E, Fleisher S, Gregoru G, Gudjonsson GH, O'Malley KD, Plant M, Rodriguez A, Ozer S, Takon I, Woodhouse E, Mukherjee R. Guidelines for the identification of individuals with ADHD and associated FASD based on expert consensus. BMC Psychiatry. 2016;16(324):1–14.

4. Science BBo. Fetal alcohol Spectrum disorders, a guide for healthcare practitioners update. London; 2016.
5. Sonuga-Barke EJS, Kennedy M, Kumsta R, Knights N, Golm D, Rutter M, Maughan B, Scholtz W, Kreppner J. Child to adult neurodevelopmental mental health trajectories after early life deprivation: the young adult follow up of the longitudinal English and Romanian adoptees study. Lancet. 2017;389(10078):1539–48.
6. DeBellis MD. The psychobiology of neglect. Child Maltreat. 2005;10:150–72.
7. Gregory G, Reddy V, Young C. Identifying children who are at risk of FASD in Peterborough: working in a community clinic without access to gold standard diagnosis. J Adopt Foster. 2015;39(3):225–34.
8. Nafee TM, Farrell WE, Carroll WD, Fryer AA, Ismail KMK. Epigenetic control of fetal gene expression. BJOG. 2008;115:158–68.
9. Douzgou S, Breen C, Crow YJ, Chandler K, Metcalfe K, Jones E, et al. Diagnosing fetal alcohol syndrome: new insights from newer genetic technologies. Arch Dis Child. 2012:1–6.
10. Allen MC. Neurodevelopmental outcomes of preterm infants. Curr Opin Neurol. 2008;21:123–8.
11. Boylan J, Alderdice FA, McGowan JE, Craig S, Perra O, Jenkins J. Behavioural outcomes at 3 years of age among late preterm infants admitted to neonatal intensive care: a cohort study. Arch Dis Child Fetal Neonatal. 2014;99:359–65.
12. Schafer C, Peters P, Miller RK. Drugs during pregnancy and lactation. 3rd ed. London: Elsevier; 2015.
13. Gilles P, Wakefield M. Smoking in pregnancy. Curr Obstet Gynaecol. 1993;3:157–61.
14. Jauniaux E. Short and long term outcomes of smoking in pregnancy. Early Hum Dev. 2007;83:697–8.
15. Wacschlang LS, Pickett KE, Cook E, Benowitz NL, Leventhal BL. Maternal smoking during pregnancy and severe antisocial behaviour in offspring: a review. Am J Public Health. 2002;92(6):966–74.
16. Thapar A, Rice F, Hay D, Boivin J, Langley K, van den Bree M, Rutter M, Harold G. Prenatal smoking may not cause ADHD: evidence from a novel design. Biol Psychiatry. 2009;66:722–7.
17. Preece PM, Riley EP. Alcohol, drugs and medication in pregnancy, vol. 2011. London: MacKeith; 2011.
18. Behnke M, Smith VC. Prenatal substance abuse; short and long term effects on the developing fetus. Paediatrics. 2013;131:e1009–e24.
19. Fried PA. Behavioural evaluation of the older infant and child. In: Slikker W, Chang LW, editors. Handbook of developmental Neurotoxicology. San Diego: Academic Press; 1998. p. 469–86.
20. MacMullen NJ, Dulski LA, Blobaum P. Evidence based interventions for neonatal abstinence syndrome. Paediatr Nurs. 2014;40(4):165–203.
21. Abernethy C, McCall K, Cooper G, Favretto D, Vaiano F, Bertol E, Mactier H. Determining the pattern and prevalence of alcohol consumption in pregnancy by measuring biomarkers in meconium. Arch Dis Child. 2017;103(3):F216–20.
22. Onroy A, Segal J, Bar-Hamburger R, Greenbaum C. Developmental outcome of school age children born to mothers with heroin dependency: importance of environmental factors. Dev Med Child Neurol. 2001;43:668–75.
23. Bada HS, Bauer CR, Shankaran S, Lester B, Wright LL, Das A, Poole K, Smeriglio V, Finnegan LP, Maza PL. Central and autonomic system signs with utero drug exposure. Arch Dis Child Fetal Neonatal Ed. 2002;87:f106–f12.
24. Humphreys C, Garcia-Bournissen F, Ido S, Koren G. Exposure to ADHD medication during pregnancy. Can Fam Physician. 2007;53:1153–5.
25. Christensen J, Gronborg TK, Sorensen MJ, Schendel D, Purner ET, Pedersen LH, Vestergaard M. Prenatal valproate exposure risk of ASD and childhood autism. JAMA. 2013;309(16):1696–703.
26. Rai D, Lee BK, Dalman C, Golding J, Lewis G, Magnusson C. Parental depression, prenatal antidepressant use during pregnancy and risk of ASD: population based case control study. BMJ. 2013;346:1–15.

27. Popova S, Lange S, Probst C, Gmel G, Rehm J. Estimation of national, regional, and global prevalence of alcohol use during pregnancy and FAS: a systematic review and meta analysis. Lancet Global Health. 2017;17:1–10.
28. APA. DSM V. Washington: American Psychiatric Association; 2013.
29. Moss J, Howlin P. Autism spectrum disorders in genetic syndromes: implications for diagnosis, intervention and understanding the wider autism spectrum disorder population. J Intellect Disabil Res. 2009;53:582–873.
30. Mukherjee RAS, Layton M, Yacoub E, Turk JT. Autism and autistic traits in people exposed to heavy prenatal alcohol: data from a clinical series of 21 individuals and a nested case control study. Adv Mental Health Intellect Disab. 2011;5:43–9.
31. Moss J, Nelson L, Powis L, Waite J, Richards C, Oliver C. A comparative study of sociability in Angelman, Cornelia de Lange, fragile X, down and Rubinstein Taybi syndromes and autism spectrum disorder. Am J Intellect Dev Disabil. 2016;121(6):465–86.
32. Mukherjee RAS. The relationship between ADHD and FASD. ADHD Pract. 2016;8:4–7.
33. Mukherjee RAS, Wray E, Curfs L, Hollins S. Knowledge and opinions of professional groups concerning FASD in the UK. J Adopt Foster. 2015;39:212–24.

Paediatric Medical Assessment of the Child with FASD

13

Inyang Takon

Chapter Highlights

- The clinical process linked to fetal alcohol spectrum disorders (FASD) referrals.
- Medical examination and practical approaches to assessment.
- Investigations and approaches to evaluating children referred for assessment of FASD.

> 'We are concerned about T's behaviour, she was adopted at 1 year of age and we felt she would have settled a few years after; however, T has been displaying behaviours which we struggle to understand and manage as a family'. T has been seen by various professionals over the years. She was felt to have attachment difficulties and we have been through several sessions with the psychotherapist in CAMHS. T is now 7 years of age and she has struggled to settle in the school. She has been assessed for attention deficit hyperactivity disorder (ADHD) and autism spectrum disorder (ASD) but we were told she did not have enough features to be given the diagnosis of either ADHD or ASD. We want to understand the cause of her challenging behaviours as this is impacting on the whole family. We are struggling so much..... (Information received from T's adoptive parents).
>
> Please kindly see J who is a 5-year-old boy with very difficult behaviour at home and at school. J is extremely lively and struggles to concentrate. He has been displaying very risky behaviours. Parents have attended several parenting courses to try and support J; however, his behaviour remains very challenging. J now attends the school part time as the teachers are unable to manage his behavioural challenges towards the staff and his peers. J lives with his adoptive parents and he is one of four children in the family. Referral from GP for J assessment.

Fetal alcohol spectrum disorder (FASD) remains one of the commonest causes of intellectual disability; however, the awareness amongst clinicians who see children with a possible FASD diagnosis remains limited. Children with FASD can present

I. Takon (✉)
East and North Hertfordshire NHS Trust, Welwyn Garden City, UK
e-mail: Inyang.takon@nhs.net

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_13

with physical as well as neurodevelopmental disorders. Professionals seeing children should be aware of the various ways in which the children can present at each developmental stage. Early detection of children with FASD and accompanying early intervention has been shown to result in a more favourable outcome for the children. There remains significant underrecognition and underdiagnosis of FASD in children seen by various clinicians including Paediatricians. Surveys have shown that many paediatricians are aware of FASD; however a significant number feel less confident in assessing children with suspected FASD [1].

The assessment of a child for FASD requires a multidisciplinary approach to help identify the neurocognitive and behavioural complication of this disorder. This chapter describes the step-by-step process carried out in the medical assessment of the children for fetal alcohol spectrum disorder.

There is no single test for diagnosing FASD; hence, the process of assessing children for FASD is similar to some extent to the assessment of children for other neurodevelopmental conditions such as ASD and ADHD. However, ADHD and ASD which have standardised international diagnostic criteria such as International Classification of Diseases, tenth revision (ICD-10) and Diagnostic and Statistical Manual of Mental Disorders, fifth edition (DSM-5) against which diagnosis is usually made. On the contrary, there are several diagnostic criteria used in different countries to make a diagnosis of FASD.

13.1 Referrals

Children with behavioural and developmental difficulties are usually referred to the community/neurodevelopmental paediatrician to assess and consider if their pattern of behaviour is in keeping with neurodevelopmental disorders such as ADHD and ASD. In some cases, children are referred for the assessment of FASD when the referring professional is aware of a history of prenatal alcohol exposure. Children may also be referred based on parental request for an assessment. Adoptive parents who have children with behavioural challenges and neurocognitive difficulties may seek a referral for assessment of FASD.

It is important that professionals regularly obtain a history of prenatal alcohol exposure in all children being referred for neurodevelopmental assessment.

13.1.1 When Should FASD Be Suspected?

- Children suspected to have facial features of FAS and accompanying developmental delay, behavioural difficulties or cognitive difficulties.
- Children with prenatal alcohol exposure who are displaying challenging behaviour or learning difficulties.
- Children with failure to thrive and growth delay with poor weight gain despite increased calorie intake.
- Children with a sibling diagnosed with FASD who are also displaying neurocognitive impairment.

In determining if the criteria for having the diagnosis of FASD have been met, I will go through the stage-by-stage process of assessing the children referred. There is no gold standard process or pathway that has been agreed for carrying out assessments of children for FASD. The guidelines and pathways differ in several countries with some countries having clearer and well-defined pathways and services whist services are non-existent in other countries or regions within the same country. Chapter 19 describes some of these pathways. The core assessments remain the same irrespective of the pathways. Assessments should explore evidence of neurocognitive and behavioural impairment, positive history of alcohol consumption by the mother during her pregnancy and assessment for facial features of FAS (small palpebral fissure, smooth philtrum and thin upper lip) required to make the diagnosis.

13.2 Information Gathering

Preparation is key when assessing a child for FASD. It is important to explore history of prenatal alcohol exposure from several sources as this information is essential in determining if the child meets criteria for an FASD diagnosis. Lack of information on maternal alcohol consumption in pregnancy is frequently seen in children being assessed for FASD. This lack of alcohol history does have significant consequences when a diagnosis cannot be confirmed particularly in children who do not have the full facial features of FAS.

Professionals should remember to seek consent from parents before reviewing their medical records.

Sources of information on maternal alcohol exposure include the following

1. Care proceeding reports.
2. Child looked after reports.
3. Statements/reports from relevant professionals—Some professionals may have observed a child's mother being drunk or drinking excessively during pregnancy.
4. Social care records—reports on the state of the home may reveal empty bottles of alcohols being found in the home on a regular basis. This may suggest excessive alcohol consumption in the home.
5. Antenatal records.
6. Parenting assessment reports including reports from additction clinics if available.
7. Psychiatric/Psychological assessments on parents.

Permission for obtaining information to determine prenatal alcohol exposure from antenatal records of the mother should be obtained if this information is not available in the birth records or obtainable from the social worker.

It is important to gather information on the child's biological family history prior to the assessment. Many children seen for FASD assessment are living with their adoptive parents or foster carers. In many cases, biological parents do not attend the

assessment of the children. Social workers may be aware of a family member that is able to provide some information on the family history and history of excessive alcohol consumption.

Obtaining photographs of the child from an early age is also helpful as part of the assessment. Children may have had more obvious facial features at a younger age which may not be as obvious at the time of the assessment. Family photographs also gives the clinician the opportunity to see the whole family and compare the child's facial appearance to his/her parent's appearance. The clinician can also have an idea of parental height from the photographs.

13.3 Current Concerns

Alcohol exposure does result in a wide range of medical and developmental issues in the child. Review of the literature has suggested there may be 428 comorbid conditions, both physical and mental linked to those diagnosed with FASD [2]. The paediatrician seeing a child for FASD assessment will need to be aware of the wide spectrum of presentation of difficulties in children with FASD.

The clinician seeing the child for assessment should actively ask about the child's strengths. This allows the opportunity for the carers to share some of the things the child is good at and also helps in the intervention stage.

Concerns which carers/parents may have include the following.

1. Delayed development—Parents may report that the child has been slow in achieving their milestones. The child may have delay in their motor or language milestones.
2. The child may present with difficulties with their learning and make poor progress at school.
3. Attention and concentration difficulties. Parents of children with FASD frequently describe the child having difficulty staying on task and focusing. The child may be reported as having these concerns at home and at school.
4. Children may struggle to settle and be constantly on the go. Carers may find it difficult managing the children at home and outdoors. Parents may report the child is extremely lively and disruptive.

History from Carers/Parents It is important to find out when and why carers are concerned about the child's behaviour. Detailed exploration of the concerns expressed by parents/ carers should be done in the diagnostic interview. Some assessment teams have developed proformas/ questionnaires to help obtain a structured history [3].

Box 13.1 shows the types of information obtained during a structured interview.

Box 13.1: Questions to Be Asked in Differing Domains

Concerns	Detailed history
Developmental history	• Clinician's knowledge of normal developmental milestones in children is an essential part of this assessment • Information on the child's gross motor, fine motor, social skills and speech and language skills is important • Play skills and behaviour • Interaction with other children at school and outside the home setting • Toileting history- children with FASD may have delay in attaining continence. • Independence skills • Non-verbal interaction as a baby such as pointing and sharing and eye contact
Birth history	• Was the child full term or preterm? (Carers may not have this information so it is important to check the child's records, hospital discharge summaries of any clinic letters that have this information) • History of maternal alcohol use in pregnancy. Document if the history is present and describe the amount of alcohol consumed • Was the baby born by normal delivery? Or through the use of instruments such as forceps or Ventouse. Was the child delivered by caesarean section? • Did the child come out in good condition at birth? Did they need any resuscitation? If yes, for how long was • Was the child admitted to special care baby unit? If yes, how many days was the child admitted for? • Did the child appear jittery after birth? If so what happened after the child's birth. Mothers with prenatal alcohol consumption may also have consumed other substances which may then result in neonatal abstinence syndrome in the child • Was the child given any medication after birth? If yes, how long was this medication taken for?
Attention and concentration	• Is the child able to focus on their play, school work, if so, what is the duration of time they are able to focus • Is the child easily distractible? • Is this worse when the family are outdoors? • Does the child focus better with 1: 1 support?
Impulsivity	• Some children with FASD have impulsive behaviours • Does the child do things without thinking about it? • Do they learn from consequences? • Is the child aware of danger?
Motor coordination	• Some children with FASD have marked difficulties with their coordination. They may be clumsy, fall over easily and have bruises from falls • How are they doing in school? Do they have special equipments for use in school such as special pens? Or special chairs? • Is the child able to dress themselves? • Can they feed themselves? If yes, has this been done properly?

Concerns	Detailed history
Sensory difficulties	• Is the child sensitive to loud noises? Does the child show preference for certain textures? • Does the child have constant urge to move about?
Sleep problems	• Children with FASD do suffer with sleep difficulties very often. Obtaining a detailed sleep history would help clarify the nature of the sleep difficulties
Social skills	• Does the child have friends, if so, what do they do together? • Is the child able to initiate conversations with his/her peers? • Does the child play better with peers or children much younger? • Can the child share toys with other children in play?

School History It is important to get detailed information on the child's schooling history. The clinician should obtain history about the child's behaviour in playgroups, nursery, primary school and secondary school.

- Description of the difficult behaviours.
- Strategies used by school to address the behaviours.
- Information on behaviour and interaction with the child's peers at school.
- History on participation in the classroom activities, understanding of the work and requirement for additional support.
- Information on the child's memory including short- and long-term memory.
- Information on the child's abilities with reading, writing and maths.

13.4 Feeding History

Children with FASD have been reported to have feeding difficulties which in turn may impact on their nutrition and growth. Thorough physical examination of the children is important to check for congenital abnormalities such as cleft lip and palate which may also occur as a result of the teratogenic effect of alcohol. Feeding difficulties may be behavioural in nature or due to a physical abnormality. Children with FASD may have nutritional difficulties as a result of their poor oral intake. Iron deficiency has been reported in children with FASD. It is important that physical assessments also include examination of the child's skin, nails and hair which can affected secondary to nutritional deficiencies. Very few studies have been conducted specifically to look at the feeding difficulties in children with FASD. It is important that a detailed feeding history is obtained as part of the medical assessment of children with FASD.

Physical—This may result from cleft palate, digestive system abnormalities which have been noted as associated difficulties that may occur in children with FASD. Children with cardiac defects resulting from prenatal alcohol exposure may also present with feeding difficulties and poor growth.

Behavioural—Children with FASD commonly present with sensory sensitivities which can also impact on their ability to tolerate certain food textures and swallow certain foods. The behavioural feeding difficulties seen in some of the children may impact on the quantity and quality of food they consume.

Growth—Prenatal alcohol exposure can impact significantly on the child's growth. Alcohol exposure impacts on the child's physical and cognitive development. Review of the child's growth should start from before birth. It is important to ask if there were any concerns with the fetal growth during antenatal monitoring. Prenatal alcohol exposure needs to be considered in children with primary growth problems being seen in failure to thrive clinics or general paediatrics outpatient clinics. Some useful screening questions include the following

- Were there any concerns with fetal growth in the antenatal period? If so, how often was the mother followed up and what investigations were carried out?
- Information on maternal nutrition and any maternal health concerns that may also have impacted on the fetal growth?
- Information on maternal use of other substances in addition to the alcohol.
- What was the child's birth weight? Prenatal alcohol exposure can cause low birth weight and small for gestational age babies. Some studies have shown that extensive drinking is associated with more extensive growth restriction.
- What was the child's head circumference and length?—Prenatal alcohol exposure can impact on the child's overall head growth and lead to small head size (microcephaly).
- Information on the child's feeding pattern in the early years.
- Review of the child's red book to check the child's weight and height velocity. Children with significant growth restriction from alcohol exposure do have difficulty putting weight on despite taking enough calories.
- Information on the child's dietary intake- encourage parents to keep documented food diaries which can be reviewed.
- Information on parental weight and height. This information is useful in considering if growth problems may be partly genetic in nature.
- Information on the child's bowel habits to ensure there are no concerns suggestive of malabsorption.
- Useful to obtain history on any other illnesses the child may have as this could also impact on the child's growth.
- History of any other congenital abnormalities in the child.
- History of medication use in the child. Some children being assessed for FASD may already have been diagnosed for ADHD and treated with stimulant medications. These medications can impact on growth velocity.

Children with growth restriction from prenatal alcohol exposure do tend to remain smaller than their peers [4]. There are few studies looking at the long-term outcome of growth in these children.

13.5 Behaviour

Description of the child's behaviour difficulties should be obtained. Trigger factors for the behavioural challenges, duration and frequency should be obtained. Attachment difficulties are fairly common in children with FASD. Children with significant anxiety and mood problems may present with behavioural challenges.

In older children and young people, history of drug and alcohol use, sexual history and offending behaviours should be obtained. Children and young people with FASD can be impulsive and easily led.

13.6 Services Involved with the Child

Children referred for FASD assessment have been seen by various professionals prior to being referred for a formal assessment. Some children have received multiple diagnoses such as ADHD, dyspraxia and ASD over the years with the child still presenting with behavioural challenges. Some children with a diagnosis of ADHD may have been treated with medication with some of them showing poor response to the medication. It is important to document the services where the child is being seen and all the professionals seeing the child. This helps with sharing the outcome of the assessment and also planning interventions alongside other professionals.

The following professionals are typically involved in the assessment of a child with suspected ADHD.

1. ASD support team
2. ADHD clinic review
3. Othoptist
4. Ophthalmologist
5. Audiologist
6. Geneticist
7. Therapists
8. Psychologists
9. Speech and Language Therapists
10. Occupational Therapists

13.7 Medical History

- Obtain detailed history of other medical conditions the child may have
- Ask about reccurent infections- immune deficiencies have been described in some children with FASD
- Screen for cardiac problems through the history and refer for further cardiac assessment if there are any concerns
- Ask about medication which the child is having

- Screen for seizures- Epilepsy has been reported in a few children with FASD [5]
- Screen for continence problems-Children with FASD have been reported as having continence problems such as nocturnal enuresis and daytime urinary incontinence. Congenital kidney and genitourinary problems are prevalent in children with FASD

Sensory Difficulties Children with FASD have increased prevalence of sensory processing difficulties. Prenatal alcohol exposure has been noted to affect the brain in a widespread and diffuse way. Affected children are noted to display sensory seeking behaviours and have less awareness of processing what goes on around them. The sensory seeking behaviours in the children can result in behavioural responses that may result in the child being given the wrong diagnosis of ADHD. It is important to find out if the child has been seen by the Occupational Therapist and the interventions that have taken place till date. Paediatricians should screen all children seen for sensory difficulties.

Sleep Sleep difficulties are common in children with FASD. These range from delayed sleep onset, fragmented sleep pattern with disrupted sleep and overall poor quality of sleep. Wengel et al., found sensory processing difficulties was a significant cause of sleep related difficulties in children with FASD. The brain dysfunction and cortical damage seen in children with FASD is felt to contribute to the sleep difficulties also seen in the children [6]. Sensory processing difficulties are also felt to contribute to the sleep problems seen in children with FASD. Sleep difficulties can also impact on the quality of life of the family. Obtaining a proper history helps classify the type of sleep problems the child has and hence helps with the management. Children with sleep difficulties are commonly prescribed melatonin which does not have a significant effect in improving their sleep. Children may benefit better from use of behavioural programmes to address the sensory processing difficulties which results in the sleep problems seen in the children.

Detailed sleep history can be obtained by direct interview history taking from the care giver or use of sleep questionnaires which the care giver can complete.

Some useful sleep screening questionnaires and sleep diary are available through the American Sleep Association website.

13.7.1 Screening Questions for Sleep Problems

- Bedtime routines
- When does the child go up to bed? What time does the child fall asleep?
- Disruptions/fragmented sleep patterns including number of times these occur at night
- Electronics in the bedroom- TV, computer and use before bedtime
- History of snoring

13.7.2 Behavioural Presentations of Children with Sensory Seeking Behaviours

1. Constantly moving, restlessness
2. Sleep difficulties with restlessness in sleep [6]
3. Disruptive behaviours
4. Anxiety
5. Feeding difficulties- children may present with restricted eating pattern
6. Aversion to certain clothing textures
7. Aversion to touch and loud noises

Caregivers could complete a questionnaire assessing the child's sensory profile and the severity of symptoms. The short sensory profile questionnaire is a useful screening tool to assess these difficulties. Information on strategies used by caregivers to help manage these sensory difficulties is important.

Physical History Important to ask about any gross and fine motor difficulties in the child. Children with FASD do commonly have motor coordination difficulties which may impact on their academic ability and participation in physical activities. Information on the activities the child or young person takes part in outside school is important.

Personal Care Skills It is important to ask age related questions on the child's personal care skills. Children with FASD may have normal IQ's but struggle in the area of achieving Independence. The adaptive behaviour questionnaire (ABAS-III) or Vinelands Adaptive Behaviour Questionnaire does help with screening their personal skills and level of independence.

Home Situation Information on the carer's/parents home environment is important. Children with FASD and behavioural challenges may display significant challenges in noisy, busy home environments. It is important to ask how many people do live at home, the child's behaviour towards other children and family members. Activities which the child participates in outside the school and home environment as well as activities which the child enjoys doing.

- How do the carers respond to the child's behavioural challenges?
- Understanding of the impact of the child's behaviour on carers and other family members

Family History Children being assessed for FASD may have other affected siblings who may or may not have been assessed. It is important to ask the carer if there are concerns in the other children.

- Ask about family history of developmental difficulties and learning difficulties. Although FASD is caused by prenatal alcohol exposure, children may also have genetic abnormalities which needs further investigations.
- Information on the alcohol consumption pattern in Parents is important. The mother of the child with suspected or confirmed FASD may still continue to drink and present further risk to future children she has.
- Information on Parental height and weight is important. This information also helps when assessing the child's growth.
- History of health problems and mental health concerns in the child's biological family is also important. Children with FASD do have a high risk of developing mental health problems; however, the added impact of a family history of mental health concerns may increase the child's risk of developing these problems.

Physical Examination Examination should start with observation of the child. The child's face should be assessed for distinctive facial features, inspection for include facial assessment for dysmorphic features, assessment of the child's skin for unusual birth marks, observation of the hands and feet, ear lobes and general appearance for any unusual features. There are some genetic syndromes that have some features that can be seen in children with FAS.

Thorough examination of the child's facial features, mouth, including looking into the inside of the mouth, checking the palate, and teeth are important. It is important to measure, record and plot the child's weight, height and head circumference on a growth chart. The child should then have a general physical examination cardiovascular, respiratory, abdominal and neurological examination done. Examination should include assessment of the child's gait and how the child performs activities in the clinic.

The child should have an eye examination done. An initial eye assessment in the clinic to look for squints or major eye abnormalities should be done. Children should also be referred to the Ophthalmologist for further assessment. Eye abnormalities seen in children with FASD include optic nerve hypoplasia, microphthalmia and colobomas [7].

13.8 Facial Assessment

Assessment for facial features of FAS is a key part of this assessment. Lip Philtrum Guides which offers a pictorial guide to rating the child's lip and philtrum is the commonest tool used for the facial assessment. The Lip Philtrum Guide can be obtained from the FASDPN Network in Washington and is fairly straightforward to use.

Palpebral fissure measurements are done using a very thin small transparent ruler to measure the palpebral opening. The measurements are read off on a palpebral fissure chart and helps determine if the child has small palpebral fissure.

Computerised software for facial assessment is also available.

New developments such as the use of 3D scanning of the face are available in some centres and allow better resolution of the images. May allow other features and more accurate classification, but for now remain a tool primary for research.

13.9 Clinic Observation

Children seen for FASD assessment may vary from being compliant to being extremely non-compliant. It is important that the clinician observes the child's behaviour from the waiting room until the end of the consultation. Information on the child's neurological functioning, behaviour, speech, social skills and facial appearance can be obtained by observation and also engaging the child in a conversation. Children who are non- compliant for the initial assessment should be seen again for a subsequent assessment.

13.10 Observation by Other Professionals

Reports from assessments by other professionals should be reviewed. This is helpful when addressing some of the concerns described by the carers. Children with significant hyperactivity may appear very quiet in the clinic but may display hyperactive behaviours that have been observed by other professionals in a different setting.

Children with social communication difficulties or autism spectrum disorders may communicate well on a 1–1 in the clinic but display difficult behaviours when seen in a different setting.

Children with significant behavioural challenges may not display these behaviours in the clinic setting.

13.11 Investigations

There are no specific investigations for FASD; however, any child being assessed for FASD should have genetic investigations done (microarrays) [8]. Children being assessed for FASD may have coexisting genetic abnormalities. There are also some chromosomal abnormalities that may have similar facial features and presentation to that seen in children with FAS.

Children with microcephaly as a result of FAS do not need to have an MRI done; however, children being seen in outpatient clinics for concerns of microcephaly should also be screened for FAS.

Children with growth delay do need to have investigations to check the underlying cause of their growth delay. Growth hormone deficiency may be detected in these children; however, their response to growth hormone treatment is usually suboptimal.

Children with FASD should have their hearing and vision assessed by the relevant specialists. Hearing and vision abnormalities are prevalent in children with FASD. There is report of visual and hearing abnormalities in children with FASD.

13.12 School Assessments

Medical assessment of children for FASD also involves obtaining information from the schools. There is still poor awareness amongst educational staff regarding how children with FASD present in the school environment.

The following information should be requested

- Report from the Class teacher or School SENCo regarding the child's behaviour and learning
- Copies of Reports by Educational psychologists if available. If not, then request for assessment by the Educational/Clinical Psychologists should be made
- Strength and Difficulties Questionnaires
- ADHD screening questionnaires- Vanderbilt (Teacher and Parent), Conner's (Teacher and Parent)
- Social Communication Questionnaires to screen for ASD
- Sensory Profile questionnaires to screen for sensory processing difficulties
- Adaptive Behaviour Questionnaires—Vineland, ABAS to help assess the child's adaptive behaviour functioning

Developmental Assessment In younger children, depending on the age of the child, the child's development can be assessed using one of the standardised developmental assessment tools. These assessments are done as part of assessment for central nervous system (CNS) abnormalities which would include developmental delay and neurocognitive impairment.

Examples of Developmental Assessment Scales.

- Bayley's scale
- Griffith's scale

Developmental screening in younger children can be carried out using the Schedule of Growing skills. Children with prenatal alcohol exposure may show delay in their development during the assessment. Some affected children may still pass the early developmental screening and develop cognitive difficulties later. It is important that children with significant exposure to alcohol are monitored closely and clinicians should avoid giving early reassurances to families where the children have passed the early developmental assessment.

13.13 Summary

- Children with FASD may present in different clinical settings within the hospitals as a result of the physical abnormalities that result from the teratogenic effect of prenatal alcohol exposure. Clinicians without the knowledge of the varying presentations of FASD may not refer the children for further assessment.
- Effective medical assessment of the child with FASD is dependent on good information gathering to reflect prenatal alcohol exposure.
- Structured approach to history taking will help detect physical and neurocognitive impact of prenatal alcohol exposure.
- Clinicians seeing children with growth delay and microcephaly may avoid the need to carry out extensive investigations in children whose physical difficulties is due to FAS.

References

1. Gahagan S, Sharpe TT, Brimacombe M, Fry-Johnson Y, Levine R, Mengel M, et al. Pediatricians' knowledge, training, and experience in the care of children with fetal alcohol syndrome. Pediatrics. 2006;118(3):e657–68.
2. Popova S, Lange S, Shield K, Mihic A, Chudley AE, Mukherjee RAS, Bekmuradov D, Rehm J. Comorbidity of fetal alcohol spectrum disorders: a systematic review and meta-analysis. Lancet. 2016;387:978–87.
3. Fitzpatrick JP, Latimer J, Ferreira M, Martiniuk AL, Peadon E, Carter M, et al. Development of a reliable questionnaire to assist in the diagnosis of Fetal Alcohol Spectrum Disorders (FASD). BMC Pediatr. 2013;13:33.
4. Carter RC, Jacobson JL, Molteno CD, Dodge NC, Meintjes EM, Jacobson SW. Fetal alcohol growth restriction and cognitive impairment. Pediatrics. 2016;138(2):e20160775.
5. Boronat S, Vicente M, Lainez E, Sanchez-Montanez A, Vazquez E, Mangado L, et al. Seizures and electroencephalography findings in 61 patients with fetal alcohol spectrum disorders. Eur J Med Genet. 2017;60(1):72–8.
6. Wengel T, Hanlon-Dearman AC, Fjeldsted B. Sleep and sensory characteristics in young children with fetal alcohol spectrum disorder. J Dev Behav Pediatr. 2011;32(5):384–92.
7. Brennan D, Giles S. Ocular involvement in fetal alcohol spectrum disorder: a review. Curr Pharm Des. 2014;20(34):5377–87.
8. Douzgou S, Breen C, Crow YJ, Chandler K, Metcalfe K, Jones E, et al. Diagnosing fetal alcohol syndrome: new insights from newer genetic technologies. Arch Dis Child. 2012;97(9):812–7.

The Role of Formal Psychometric Assessment in Fetal Alcohol Spectrum Disorder (FASD)

14

Alexandra Carlisle and Alexandra Livesey

Chapter Highlights

- Practical approaches to psychometric testing.
- Tests to use in different situations.
- Subtleties that influence the presentation and interpretation of psychometric testing.

14.1 Rationale for Psychometric Assessment

The main benefit of carrying out a psychometric assessment in any clinical or educational psychology setting is that it allows more objective assessment of an individual's underlying cognitive strengths and difficulties, rather than relying on clinical judgement alone. Although each individual is unique in their strengths and difficulties, psychometric testing allows assessment scores to be compared to a set of normative data; that is, individuals' abilities can be compared to those of a sample which provide data about what is usual in a defined population at a specific point in time (see Fig. 14.1 for normal distribution curve). Another benefit of psychometric assessments is that they are standardised, whereby they are designed in such a way that the questions, conditions for administering, scoring procedures, and interpretations, are consistent and are administered and scored in a predetermined manner, as far as possible. Subtests within psychometric assessments are statistically chosen for their validity and reliability. In other words, subtests are included in a testing battery if they measure what they are supposed to measure (validity) and results are consistent (reliability). There is not one approach to this form of testing.

A. Carlisle (✉) · A. Livesey
FASD Clinic, Redhill, UK
e-mail: Alexandra.carlisle@sabp.nhs.uk; Alexandra.livesey@sabp.nhs.uk

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_14

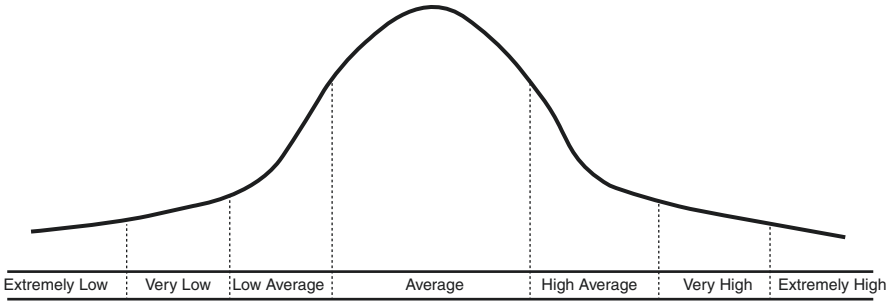


Fig. 14.1 Normal distribution curve

This chapter sets out the approach used in the UK National Fetal Alcohol Spectrum Disorder (FASD) behavioural specialist clinic.

The benefit of carrying out psychometric assessment as part of the assessment of FASD is that Cognition, Memory and Executive Functioning are consistently included as three of the nine neurobehavioural domains to be considered during assessment for FASD. However, there are variations as to what constitutes 'impairment' in these domains, according to the various FASD guidelines. For example, Astley et al.'s [1] FASD guidelines purport that 'significant' impairment is generally defined as performance that is two or more standard deviations below the mean on a standardised test. However, according to the 2005 Canadian guidelines for FASD diagnosis [2] a domain is considered 'impaired' when, taking into account the reliability of the specific measure and normal variability in the population, on a standardised measure:

1. Scores are two standard deviations or more below the mean.
   - For example, we would expect that two-thirds of the population would have intelligent quotient (IQ) scores between 85 and 115, with the mean IQ score being 100. An IQ/Index score of 70 or below is two standard deviations or more below the mean.
2. There is a discrepancy of at least one standard deviation between Subdomains.
   - For example, with an Index score of 85 on the Verbal Comprehension Index and an Index score of 100 on the Fluid Reasoning Index there is a discrepancy of one standard deviation.
3. There is a discrepancy of at least one and a half to two standard deviations among Subtests on a measure.
   - A Subtest Scaled score of 1 on Digit Span and a Subtest Scaled score of 7 on Picture Span would indicate a 2 standard deviation difference between Working Memory Subtests.

The 2016 Canadian guidelines [3] vary slightly from the above. Specifically, within the Cognition and Memory domains, a cut-off of two standard deviations below the mean on a Composite score (such as IQ) or major Subdomain score (such as Verbal Comprehension Index) is considered to be the primary evidence of impairment, with differences between Subdomains only being considered if the discrepancy size occurs with a very low base rate in the population (≤3% of the population), *and* where the

lower of the two discrepant scores is at least one standard deviation below the mean. Within the Executive Function domain, impairment as measured by direct assessment is present when multiple Subetst scores below the clinical cut-off are obtained. It is also recommended that a domain should not be considered impaired on the basis of a single Subtest score from one assessment measure.

Whichever criteria are used, unless psychometric assessment is carried out, it would be difficult to demonstrate whether these criteria have been met. Where it is not possible for a standardised assessment to be carried out, clinical assessment with converging evidence from multiple sources and DSM-5 diagnostic criteria for certain disorders may also be considered in specific domains which are not easily assessed by standardised tests. In the UK Fetal Alcohol Spectrum Disorder Behavioural Specialist Clinic, standardised cognitive and executive function assessments are routinely carried out by the psychologist as part of a wider assessment. Please note that the 2016 FASD Canadian guidelines [3] highlight the importance of using clinical judgment to determine whether a true deficit is present—for example, when test data is inconsistent within a domain. In these situations, the decision should be supported by clinical observation and history.

14.1.1 Cognitive Assessment

In the UK Fetal Alcohol Spectrum Disorder Behavioural Specialist Clinic, we begin with an assessment of the Cognitive Domain using a Wechsler assessment battery, such as the Wechsler Intelligence Scales for Children-Fifth Edition (WISC-V) [4], as this allows an overall estimate of cognitive ability to be calculated in the form of a Full Scale IQ score. In the WISC-V there are 10 core Subtests, seven of which contribute to the Full Scale IQ score. The 10 Subtests further divide into five Index scores—Verbal Comprehension, Visual Spatial, Fluid Reasoning, Working Memory and Processing Speed. The Full Scale IQ (FSIQ) score is not considered clinically meaningful if there is too much variability between the seven Subtest scores of which it is comprised. For example, if the difference between the highest and lowest Subtest equals or exceeds five points on the WISC-V, the FSIQ is still statistically reliable, but may not be clinically meaningful [5]. In reality, this means that although the FSIQ is still seen as a reliable indicator of overall intelligence, because it is a Composite score, there is a risk that simple interpretation of *only* this score may mask the relative strengths and weaknesses in the cognitive profile. Where there is less than a five-point difference between Subtest scores in an Index, the risk of this is minimised.

In the instances when the FSIQ is not considered clinically meaningful, there are several processes a clinician can follow. Further Index scores can be computed, which may not be affected by the Subtest score difference. The WISC-V allows for an alternative estimate of overall cognitive ability (called the General Ability Index—GAI) to be calculated, as well as the Non-Verbal IQ. The GAI can be a useful score to report if the individual's performance on the Verbal Comprehension, Visual Spatial and Fluid Reasoning, Subtests is similar—however, the GAI does not take account of the Working Memory or Processing Speed Subtests. The Non-Verbal IQ can be helpful if performance on Verbal

Comprhension is significantly more impaired or elevated than ability on the four other Indexes. In the UK FASD clinic, we often find there is a lot of variability in the cognitive profile such that neither the Full Scale Index nor the General Ability Index provide a meaningful overall intelligence score. In these instances, it is possible to refer to the Canadian guidelines as well as to compare scores using the WISC-V scoring criteria, to determine if there is significant difference between Subdomains or between Subtest scores.

14.1.2 Executive Function

When it comes to testing executive function (EF) it is not advisable to conceptualise EF as a unitary construct in the same way that IQ or intelligence ('g') can be understood. For example, McCloskey and Perkins [6] identify four principles to consider in relation to EF assessment (Rapid reference 5.1, pp. 132–133):

1. tasks that measure EF also measure other cognitive constructs
2. tasks that measure cognitive constructs also measure EF
3. all assessment tasks are measures of multiple aspects of EF
4. the amount and nature of EF in any assessment task varies greatly depending on the format, content and complexity of the task

Therefore, since EF assessments usually include multiple tasks and/or multiple scores based on variations of administration format, item content and task complexity, their interpretation must occur at the subtest/task level and below. In the UK FASD clinic, we currently use four subtests of the Delis-Kaplan Executive Function System (D-KEFS) [7] specifically Trail Making, Verbal Fluency, Colour-Word Interference and Twenty Questions. The D-KEFS has been chosen as it has been widely used in FASD studies (as cited in 7) and these particular subtests were chosen in order to assess elements of executive functioning that have been observed within the FASD population as being common areas of cognitive difficulty, namely inhibition, cognitive switching and problem solving. As stated above, unlike the WISC test battery, it is not possible to derive an overall EF score from the D-KEFS; however, it is possible to determine how much difference there is from the norm, or between Subdomains or Subtest scores, as per the Canadian FASD guidelines [2] and [3].

14.2 Further Considerations When Undertaking Psychometric Assessment

The research literature has identified that EF can be seen as a continuum of 'Cool' and 'Hot' [8]. Cool EF involves tasks where handling of complex information, inhibition and problem solving is key—but where there is little emotional response elicited during the task [9]. In comparison, Hot EF occurs in situations that require control of emotion as part of the decision-making process, and an individual's

actions are affected by the consequence of the action. Research has suggested that within the FASD population, EF deficits can be found across the continuum, but they are more likely to be in Hot EF area (e.g [9, 10].). We hypothesise that Hot EF difficulties would be more highly linked to observed functional difficulty. However, it is important to note that the D-KEFS tasks (and many other standardised measures of EF) are not necessarily measuring 'Hot' EF as there is little emotional response elicited during many of the tasks. Therefore, the interpretation of the D-KEFS or other EF assessment scores should be considered with caution (Fig. 14.2).

For example, in the UK FASD clinic we examined relationships between outcomes from caregiver reports using the Behaviour Rating Inventory of Executive Function (BRIEF) and the clinic-based assessment using the D-KEFS, as part of an ongoing service evaluation [11]. Pearson's Correlations between all 11 BRIEF scales and the 18 selected D-KEFS subscales showed little relationship. The BRIEF showed a profile of clinically significant elevations in all three Index scores and seven out of the eight Scale scores, whilst several D-KEFS tests showed Below Average EF. These results indicate that both executive function measures have separate clinical utility in demonstrating executive function difficulties in FASD however caregiver reports display little relationship to neuropsychological tests and we considered it likely the caregivers are reporting more 'Hot' EF type functioning.

Therefore, clinicians should be aware that different aspects of EF are seen in different settings and future research should focus on identifying tests that better relate findings from clinical settings to behaviour in daily life. When the element of emotional regulation is added to elicit a 'Hot' EF response, it is likely that increased maladaptive behaviour occurs. For example, a child with FASD may be able learn to apply inhibition with repeated opportunities for learning such as learning to stop, look and listen when they cross a road in everyday circumstances. However, if an element of arousal is involved (e.g. a puppy is seen across the road) it is often

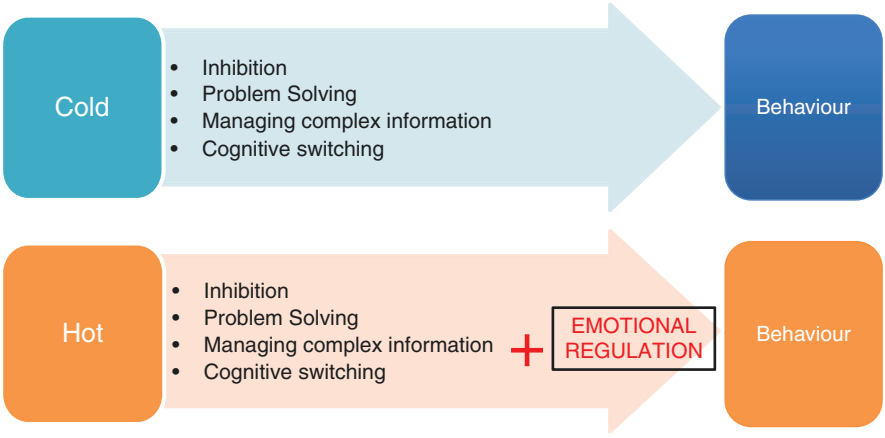


Fig. 14.2 'Hot' versus 'Cold' executive function

reported that the impulse to cross the road without looking overwhelms the inhibition that is otherwise in place. Therefore, it is important to highlight and explain that 'Hot' EF may not have been measured in a clinic setting especially where behavioural report from caregivers' contrast with the 'Cool' EF test results.

When interpreting any psychometric test results, consideration should also be given to whether an individual's scores have been influenced by motivation, attention, interest, emotion and opportunities for learning. For example, if an individual is very anxious at the beginning of the assessment this may result in them doing poorly on the first subtest administered (e.g. Block Design on the WISC-V). It is important therefore to take anxiety or other factors such as fatigue into account where they may have affected the outcome. As far as possible, the person administering the test should allow time before beginning the assessment to put the individual at ease as far as possible—whilst balancing this with encouragement to do ones best. Testing conditions are often very different to a normal classroom environment. During our assessment individuals are given breaks as necessary and may be prompted to re-focus or be given motivation (only where appropriate according to what is permissible in the standardised instructions). Therefore, it should be made clear in the interpretation section that the results of the assessment are a reflection of individuals' ability in a quiet, one-to-one environment. This is particularly important as many of the people we assess have significant attention difficulties and may show less difficulty in the more structured formal assessment environment than in the more distracting classroom, home or work environment.

We are also aware that it would be preferable to carry out more psychometric assessment than we are at present able to carry out, due to time limitations. For example, we would add a more in-depth assessment of memory functioning. However, even if many more tests were completed, it is important to convey that psychometric test scores do not equate to skills and strengths that an individual might be capable of using/developing to assist him/her in achieving success and happiness in future life with adequate levels of support. Rather, test scores can be seen indication of the individual's level of current cognitive and executive functioning in comparison to a normative sample, and they can provide evidence regarding the type of support that is required in learning and vocational environments.

14.3 Behavioural Observations

Aside from the quantitative results from a neuropsychological assessment, behavioural observations throughout the clinic assessment are key to the final FASD diagnosis and recommendations. Within the clinic report behavioural observations are included which may include observations around the individual's:—attention, impulsivity and hyperactivity; use of nonverbal communication; social and emotional reciprocity; sensory seeking behaviours; repetitive or stereotyped behaviour; perseveration; and talking out loud. These areas are known to be commonly problematic for individuals with FASD and can provide essential clinical information. However, as stated before, a major caveat with presenting information from a

clinic-based assessment surrounds the environment this information is gained from, namely a one-to-one assessment outside of the individual's home, vocational or school environment. We routinely see individuals who are reported as having considerable attentional difficulties both at home and in vocational settings, yet when in the clinic environment appear to cope well. This apparent discrepancy may be due to the behavioural modifications implemented and adapting the assessment procedure to meet the needs of the individual.

Observation of engagement is also important to report on. The individual's overall engagement can be affected by a number of factors, including motivation, understanding of the reason for the assessment, mental health and emotional difficulties, social communication and interest in the assessment. Engagement in the specific subtests is also important as it may subsequently impact the assessment scores and in some cases, skew the cognitive profile they receive. In some cases, their engagement might be so poor that the quantitative assessment results should not be presented as it is not a true reflection of their abilities. This can be especially apparent in individuals who have a social communication disorder where they have a special interest in some topics and apparent lack of understanding of the need to complete the assessment. Neuropsychological strengths and weaknesses may be influenced by how much the task captures their attention and therefore how motivated they are to complete the task.

We also take time to observe the individual's confidence and ability to separate from their carers or parents. This can provide evidence of confidence and self-esteem as well as information on how the individual copes without carer support. For some individuals it is not possible or optimal for the individual to be assessed without their carer there, in these cases acknowledgment of why this was not possible is important. For some people with FASD, especially those who are seen as more able and whom achieve average range IQ indexes, observing their awareness of their difficulties is essential when considering self-esteem and suggesting interventions that would allow them to reach their potential. We have witnessed that as children progress into teenage years, this self-awareness of psychological differences can have detrimental effects on mental health and educational progress and often leads to social withdrawal. Thus, identifying and implementing strategies for understanding and overcoming these differences is essential as part of the FASD assessment.

A comorbid diagnosis of an Autistic Spectrum Disorder (ASD) is often seen in individuals with FASD [12] and the neuropsychological assessment can contribute to this diagnosis. However, a formal social communication assessment is not currently part of the remit of the cognitive assessment, but instead clinical observations are made and reported on. Specific elements of social communication that are especially observable within the assessment include eye contact, tone of voice, social disinhibition, ability to follow up cliff-hangers, the ability to engage in two-way reciprocal conversation, and ability to modify communication to the needs of the listener. Working in a multidisciplinary manner for example with a speech and language therapist allows a more comprehensive assessment of ASD to be considered as part of the FASD assessment.

14.4 Specific Tools Used

14.4.1 Wechsler Intelligence Scales for Children—Fifth Edition (WISC-V)

Verbal Comprehension Index

The Subtests in Verbal Comprehension Index (VCI) evaluate skills in understanding verbal information, thinking and reasoning with words, and expressing thoughts as words. Tests require individuals to give conceptual similarities between words (Similarities), to provide definitions of words (Vocabulary) and to draw on general knowledge and awareness of social situations (Comprehension). With regard to the Comprehension subtest in particular we often find people with FASD show some understanding of social convention; however, they are unable to give sufficient information to earn full credit. This is especially apparent where the level of reasoning suggests elements of socially appropriate behaviour have been rote learned, but the individual does not understand why certain behaviours are appropriate and does not demonstrate an understanding of another person's perspective. It would therefore be likely the individual would have difficulty generalising their learned skills to new and different situations and may find it difficult to modify and utilise social behaviours effectively. Furthermore, individuals often have islands of ability depending on their educational experience as well as their special interests.

Non-Verbal Reasoning Indexes

In the previous version of the WISC, the WISC-IV [13], there were four Indexes; one of which was Perceptual Reasoning Index (PRI). Perceptual Reasoning examined a broad spectrum of abilities including both visual-spatial and more traditional non-verbal reasoning. In the most up-to-date WISC-V, aspects related to non-verbal reasoning are assessed in two distinct Indexes; Visual Spatial and Fluid Reasoning. A brief description of each Index is given below; however, the experience of the UK FASD clinicians is historically based on the WISC-IV, thus we are in the infancy of understanding the profile of people with FASD in these two new Indexes. Observations from use of the PRI and the two new Indexes are presented below.

Visual Spatial Index

This Index indicates how well an individual does on tasks that required them to examine and think about things such as patterns, designs and spatial relationships and pictures in order to replicate 2D and 3D designs from a picture. Visual spatial skills requires a specific aspect of non-verbal reasoning—namely visual spatial reasoning, as well as the integration of visual and motor abilities, hand-eye-hand coordination and working quickly and efficiently with visual information.

Fluid Reasoning Index

This Index (FRI) assesses the ability to consider and identify the conceptual rules linking visual objects and to apply rules to solve visual problems. The FRI requires the combination of visual processing, abstract thinking, multitasking and inductive

reasoning. Fluid reasoning is usually believed to be one of the most reliable indicators of intelligence and does not rely on educational learning.

It has been our impression in the UK FASD clinic that the PRI of the WISC-IV and the Wechsler Adult Intelligence Scales—Fourth Edition (WAIS-IV) [14] is often a relative strength for people with FASD, frequently being more than one standard deviation above the other indexes, although further research is needed to verify this clinical impression. This can be the case despite the Index itself often falling within the low average range or lower. Similar findings are beginning to emerge for the VSI and FRI. Clinical reasons for this apparent preservation is not definitively known; however, there are a number of working hypotheses. In terms of the nature of the tasks, they all involve visual stimuli and on the face of it appeal to the individuals more quickly than their verbal comprehension counterparts. Observationally, we often note that people with FASD gravitate towards practical, less verbally loaded tasks. Engagement, motivation to do well and attention all tended to be higher on the non-verbal reasoning tasks, which in turn could impact on the scores achieved.

We also often notice more of a tendency with perseverance on the non-verbal reasoning tasks, this might be especially apparent in individuals who have ASD traits and/or rigidity to their thinking. Whilst with Block Design perseverance does not inflate an individual's scores as the timing element actually penalises those who may ultimately be able to get to the right answer but need longer to do so, Matrix Reasoning is not timed and therefore does allow the individual time to work out the answer. It is possible that this is why higher scores are often seen on this task in comparison to timed tasks. With the addition of Figure Weights and Visual Puzzles which both are timed, it will be interesting to see how the FASD population fare. Although some non-verbal tasks are often relative strengths for people with FASD, we notice that given additional time, rather than being placed under a time pressure, allows them to achieve even more success. In fact, one of the most common recommendations is that children with FASD may benefit from more time to complete problem-solving activities and failure to do this may result in poorer performance and subsequently impact on their self-belief.

We frequently suggest that if the non-verbal reasoning abilities are a relative strength, using similar tasks in school or vocational settings will help to increase a child's self-esteem, motivation and confidence. Interspersing cognitive strengths with areas of more challenge usually also helps people to meet their potential. However, conversely when considering actual functioning verses neuropsychological assessment results, when an individual with FASD has a relative strength in non-verbal reasoning, this strength can be masking other areas of difficulty. Thus, although it can be a considerable positive that an individual has this area of cognitive strength, it is important to be aware that others might overestimate their level of ability across many tasks based on their apparent high level of perceptual reasoning ability. Without acknowledgment of both strengths and areas of weaknesses, expectations of task and functional performance may be misaligned and lead to decreased self-esteem and confidence.

Working Memory Index

The subtests in the Working Memory Index (WMI) score indicate how well an individual can perform tasks requiring them to learn and retain information in memory while using the learned information to complete a task. These tasks also measure skills in attention, concentration and mental reasoning and can be affected by anxiety. On one Subtest (Digit Span), individuals are required to repeat a sequence of numbers in either the same or reverse order as presented by the assessor. The second Subtest (Picture Span) is new to the WISC-V and assesses visual working memory. Working Memory is known to be a particular area of difficulty for individuals with FASD and is well documented elsewhere in the FAS literature [15]. Also important to consider, is the relatively simple nature of the information assessed in the WMI and some individuals may have less difficulty with remembering sequences of numbers and pictures than they do 'real life' settings where an individual is required to remember a mixture of information presented in a variety of formats. This is where the wider assessment carried out by Speech and Language Therapy colleagues can be helpful in determining if there is a specific problem such as poor memory for language.

Processing Speed Index

Processing Speed is an interesting area of neuropsychology in people with FASD. The Processing Speed Index (PSI) indicates how well an individual does on timed tasks requiring them to quickly scan symbols and make judgments about them. Clinically, we often see that it falls within the below average to impaired ranges on the WISC-V or WAIS-IV. Individuals often present with difficulty completing the tasks, which involve multiple skills such as visual scanning, working memory and fine motor skills, quickly and accurately. As with Block Design the timing element of the PSI tasks is often to the detriment of people with FASD. But since the nature of the PSI is to assess the speed at which cognitive information is processed, the scores obtained on the PSI are usually a true reflection of their functional ability. For some individuals this cognitive difficulty is highly linked to their attentional difficulties as it appears the cognitive load needed for the tasks is too taxing. If the speeded element of the task was removed, many people would perform accurately; however, the tasks would then not be assessing processing speed.

In some cases, people with FASD appear to receive inflated scores on the PSI, in comparison to their other indexes. These situations often appear to occur in individuals who have acute ADHD symptoms at the time of the assessment. Individuals who have struggled to attend to many of the other neuropsychological tasks, appear to find the nature of the PSI tasks helps them attend and concentrate better. It is likely that the combination of using motor skills, being timed, being a short two-minute task and the use of visual stimuli aides the individual in this case. However, when this occurs, reporting the PSI score and making recommendations need to be considered carefully. The scores the individual receives may well be a good reflection of their optimal processing speed ability in a very controlled environment. However, in other environments where there are more distractions, task length is

longer and individuals are not being timed, an individual's performance is likely to be much more impaired. Therefore, their functional ability is likely to be significantly lower than their optimal ability.

14.4.2 Delis-Kaplan Executive Functioning (D-KEFS)

As previously noted, when assessing EF (Executive Functioning), it is important to consider the Hot and Cold differentiation [8]. To date, there is little way of assessing the Hot element of EF using standardised procedures. Thus, clinicians are reliant on the more typical Cold EF measures, such as the D-KEFS [7]. Please see Table 14.1 for a suggested list of tests of executive function that can be used [16, 17] although there are other EF assessments available. Each clinician has their own preference for using one of these, or other EF assessments, although clinic-based assessments are usually carried out alongside caregiver ratings such as the BRIEF (Behavioural Rating Inventory for Executive Functioning) [18]. In the National UK FASD clinic, we have historically used the D-KEFS as it is standardised for children and adults

Table 14.1 Tests of executive function

Name of EF battery	Age range	Executive function-related tests
Delis Kaplan Executive Function System [7]	8–89	• Trail making test • Verbal fluency test • Design fluency • Sorting test • Colour word interference test • Twenty questions test • Word context test • Tower test • Proverb test *(NB include error scores where possible)*
Behavioural Assessment of the Dysexecutive Syndrome in Children [16]	8–16	• Playing card test • Water test • Key search test • Zoo map test 1 • Zoo map test 2 • Six-part test
NEPSY-II [17] (as well as EF tasks the NEPSY-II also measures social perception, language, memory and learning, sensorimotor, and visuospatial processing)	3–16	• Animal sorting • Auditory attention • Response set • Clocks • Design fluency • Inhibition • Statue

and has reliably been used in FASD populations globally. The following sections detail considerations and observations from using four tasks of the D-KEFS. Please note, in addition to considerations detailed below, when using the D-KEFS it is vitally important to consider the Error analyses (Uncorrected and Self-Corrected) scores as individuals often appear to do adequately on the speed tasks (detailed below)—however, the error analyses often reveal that the accuracy is greatly reduced. Therefore, it is important to always check the Error scores as the increased complexity of the tasks can often compromise the ability to carry out the task accurately, even though the task may still be carried out at speed.

Trail Making

The Trail Making task has five conditions. In the Visual Scanning condition, the child is required to find and mark a certain number on a page of letters and numbers as quickly as possible. In the Number Sequencing and Letter Sequencing tasks the child is required to connect numbers or letters in the correct order, as quickly as possible. In the Motor Speed task, the child is asked to follow a marked trail as quickly as possible. In the Number-Letter Switching task, which is the most complex of the Trail Making tasks, the child is required to switch between joining numbers and letters whilst still keeping them in sequence. Research has indicated that individuals with FASD do not usually demonstrate significant difficulty in the four baseline conditions (Visual Scanning, Number Sequencing, Letter Sequencing and Motor Speed); however, they do often exhibit difficulties on the Number-Letter Switching condition, revealing higher-level executive function deficits that are not related to deficits in basic skills [19]. Our own experience in the clinic would also be that whilst overall cognitive ability can be uncompromised, and the basic sequencing tasks are completed adequately, individuals begin to show difficulty with the introduction of the higher-level switching task—that is, they lack cognitive flexibility. In every day scenarios people may be able to function when there is one clear repetitive task but when they are required to abandon a previous response in order to generate a novel response, they have difficulty engaging in the task to the same level.

Verbal Fluency

The Verbal Fluency task provides four scores: letter fluency, category fluency, category switching and category switching accuracy. Whilst the letter fluency and category fluency subtasks provide useful information on baseline categorical knowledge and ability to retrieve semantic information quickly, the category switching and the category switching accuracy scores are the measures of the executive functioning ability of verbal switching, as they require the individual to alternate between saying words from two different semantic categories. Common observations with the FASD group appear to indicate that categorical information is often more intact that semantic letter retrieval; however, performance on verbal switching ability is more variable. Why retrieving information based on a common starting letter is more challenging than retrieving sematic categorical information (animals, boys' names) is not known. It is possible that verbal comprehension difficulties or limited

vocabulary skills have an effect, but it is also possible that the individual's interest and engagement with the task impacts on performance. Memory (another area of common difficulty in FASD; [20]) is clearly also crucial for all three subtasks, but each requires a different element, that is, working, long-term and short-term memory. Establishing FASD specific areas of memory difficulty is work in progress, therefore it is difficult to hypothesise how memory difficulties impacts on the FASD population's performance on verbal fluency. In addition, not all people with FASD exhibit the same pattern of differences on the Verbal Fluency task, with many appearing to have intact abilities in retrieval but challenges in switching. Often difficulties in score interpretation occur when the individual's ability to verbally switch (which is what is assessed in this task) varies considerably to cognitive switching in other D-KEFS tasks such as Trail Making and Colour-Word Inferencing and WISC-IV and WAIS-IV Letter-Number Sequencing. The key consideration is that although all these tasks assess the ability to switch thinking patterns, some involve visual stimuli and visual switching, others involve sequencing as well as switching and verbal fluency relies on categorical knowledge and switching. Thus, it is entirely possible that an individual has variable ability across all these areas of executive functioning. This needs to be explicitly laid out in the assessment report as these differences may not be implicitly understood by all readers of the report.

Colour-Word Interference

On the Colour-Word Interference subtests, the child is presented with four different tasks. The first involves naming colour blocks (Colour Naming) and the second involves reading words printed in black ink that denote colours (Word Reading). The third is an interference task where the child must inhibit reading the actual word (a colour name) in order to name the dissonant ink colour the words are printed in (Inhibition). The fourth and most complex task requires the examinee to switch back and forth between naming the dissonant ink colours and reading actual text of the words (Inhibition-Switching). Research has indicated that individuals with FASD do not demonstrate significant difficulty in the two baseline conditions; however, they do significantly differ from control subjects on both the Inhibition and Inhibition-Switching [19]. Our own experience would support this. Occasionally, a clinician is faced with the challenge of interpreting a paradoxical situation where a child has scored in the impaired range for the simple Inhibition task, but is far more successful in the supposedly more complex Inhibition/Switching task. It is difficult to explain this variance as logic would suggest that if the child finds inhibiting their natural responses hard, then adding an additional load such as switching would impair them further. However, we have observed that occasionally, the added element of switching allows the child to perform better perhaps as they do not have to sustain their level of concentration on inhibition for as long as in the Inhibition alone task. In essence, the switching allows a reprieve from the effort of inhibition and they can reset before carrying on. This is only a working hypothesis and needs to be researched further, but may explain why some children present with apparently confounding results.

Twenty Questions

The Twenty Questions task is designed to assess an individual's ability to problem solve using verbal reasoning. The individual is given a stimulus picture containing 30 items and instructed to ask questions to find out which item the examiner is thinking of in as few questions as possible and following specific rules. The individual receives three scores, an Initial Abstraction Score, a Total Number of Questions Score and Total Weighted Achievement score. The Initial Abstraction Score identifies how well they plan their first approach to solving the problem based on how many items are eliminated from the possible solutions after their first question. This is very useful for the FASD assessment as it gives a strong indication of whether they can plan and initiate an effective problem-solving strategy with very few prompts. The Total Questions Asked and Weighted Achievement score are more indicative of an individual's ability to alter their strategy if needed and think effectively about categorical information whilst using this to problem-solve. A low score in any of these areas is suggestive of difficulty in verbal problem solving; either planning or initiating strategy, perseverance, ability to alter strategies and also knowledge and use of categorical information. In our experience, the Twenty Questions task can identify people with a specific difficulty in these areas and rarely gives a 'false negative' result; people who struggle with this task truly have an executive functioning weakness in this area. However, it is not uncommon for people to receive an average level score and thus not be identified as having a difficulty in this area despite reportedly finding this type of task challenging outside of the assessment environment. We have noted that it is quite possible for an individual to receive a 'false positive' result when they have in fact simply been guessing specific items and been lucky in identifying the correct item quickly. We consider that there is a conceptual link between abilities assessed in the WISC-V and WAIS-IV subtests of Similarities and Picture Concepts as all three tasks require categorical information. For example, it would be unusual for an individual to achieve a low score on the Similarities and Picture Concepts task yet score highly on the Twenty Questions task. Therefore, it is imperative that the assessor interprets the received scores alongside other Subtest scores and behavioural observations in order to produce conclusions about their verbal problem-solving ability that are valid. Finally, this task usually appears at the end of the assessment process which can be a long and tiring period for the individual. It is important to consider this when interpreting the results, as fatigue and the desire to finish the assessment can mean that the individual is less engaged with it in comparison to early subtests. However, the nature of the task is such that most people enjoy it from younger children through to adults and it can be a positive and rewarding end to the neuropsychological assessment.

14.5 Frontal Lobe Paradox

George and Gilbert [21] observe that patients with prefrontal cortex damage may appear proficient within clinical interview and perform normally on traditional assessments and yet exhibit marked limitations within adaptive functioning. This is referred to as Frontal Lobe Paradox (FLP) where individuals may be able to describe

what they should be doing but in practice fail to use this knowledge to guide their actions. In FLP, individuals perform better on externally prompted tasks such as clinic assessment but have difficulties in:

- Non-routine situations.
- Long-term rule maintenance.
- Multi-step tasks or tasks involving greater mental effort.
- Social cognition difficulties.

In FASD, as in patients with prefrontal cortex damage, we also find that superficial presentation in structured settings/self-report/clinic-based assessments may or may not reflect the extent of the level of functional difficulty and it is important to take caregiver reports into account. Therefore, as well as clinic-based assessment, caregiver questionnaires/interviews and observations of unstructured activities are vital to inform the neurobehavioural aspect of diagnosis.

Table 14.2 highlights measures routinely used by the clinic (Table 14.3).

Table 14.2 Measures used in the UK FASD clinic

Assessment	Description	Related neurodevelopmental domain (2016 Canadian Guidelines—3)
Diagnostic interview for social communication disorders	The DISCO [22] is a 600 question developmental history that was developed by Lorna Wing and Julie Gould of the National Autistic Society the results of which can lead to diagnosis on the Autistic Spectrum giving ICD-10 and DSM-V diagnosis	• Adaptive behaviour/ social skills/ social communication • Affect regulation • Motor skills • Cognition • Executive function including impulse control and hyperactivity • Attention
Conners 3 parent and teacher forms	Developed by Keith Conners [23] for assisting in the assessment of ADHD	• Executive function including impulse control and hyperactivity • Attention
ADHD screening questionnaire	Clinical tool developed by Dr. Raja Mukherjee which is based on the diagnostic criteria from the DSM-V diagnostic manual	• Impulse control and hyperactivity • Attention
Vineland adaptive behaviour scales-II	The Vineland Adaptive Behaviour Schedule [24]. Divided into four broad areas of social communication domain, daily living skills, socialisation, and motor skills. Whilst there is a bias and slight standardisation towards an American population it remains useful worldwide in terms of its ability to classify functional ages	• Adaptive behaviour/ social skills/social communication • Language

(continued)

Table 14.2 (continued)

Assessment	Description	Related neurodevelopmental domain (2016 Canadian Guidelines—3)
Developmental behavioural checklist (primary carer and teacher version)	The Developmental Behaviour Checklist [25] which was designed specifically for a developmental delayed population. The use of this tool is as much qualitative in that it is used to understand behaviour in different settings	• Adaptive behaviour/ social skills/social communication • Academic achievement • Affect regulation • Impulse control and hyperactivity • Attention
The sensory processing measure (SPM)	Developed by Parham and Ecker [26] the tool is used for a broader sensory assessment for school age children, and enables assessment of sensory processing issues, praxis, and social participation	• Motor skills (sensory motor) • Adaptive behaviour/ social skills/ social communication
Semi-structured FASD screening questionnaire	The semi structured FASD Screening Questionnaire was developed by Dr. Raja Mukherjee. In essence it is a semi structured history based around the main parameters that are known to be in deficit with some with fetal alcohol spectrum disorders	• Cognition • Language • Academic achievement • Memory • Executive function including impulse control and hyperactivity, attention • Affect regulation • Adaptive behaviour/ social skills/social communication
Social communication questionnaire	The Social Communication Questionnaire [27] is a tool developed by Professor Michael Rutter and colleagues to screen people for an Autistic Spectrum Disorder. It has been demonstrated to have good reliability as a screening tool, rather than a diagnostic tool	• Adaptive behaviour/ social skills/social communication
Short sensory profile	The short sensory profile [28] is a tool developed by Winnie Dunn, an expert in sensory integration, as a screen for sensory disorders. It is not a comprehensive assessment in itself, rather a tool to identify areas where sensory deficits may lie	• Motor skills (sensory motor)
The behaviour rating inventory of executive function (BRIEF)	Developed by Gioia et al. [18] is used qualitatively to determine broader executive function in home and school environments	• Executive function including impulse control and hyperactivity • Attention • Affect regulation

Table 14.2 (continued)

Assessment	Description	Related neurodevelopmental domain (2016 Canadian Guidelines—3)
Wechsler intelligence scales—Child and adult	Depending on age, the tool used for the cognitive assessment is either the Wechsler Adult Intelligence Scale—Fourth Edition [14] or the Wechsler intelligence scale for children—Fifth Edition [4]	• Cognition. • Memory (working memory)
The Delis Kaplan [7] executive functioning system (D-KEFS)	The Delis Kaplan [7] executive function test is currently used for children and adults aged eight and over	• Executive function including impulse control and hyperactivity
Clinical evaluation of language fundamentals—fourth edition UK (CELF-4)	The standardised tool used for assessing communication is the Clinical Evaluation of Language Fundamentals—Fourth Edition UK [29]	• Language
Children's communication checklist second edition (CCC-2)	The Children's communication checklist second edition (CCC-2) [30] is a 70 item questionnaire developed by Dorothy Bishop which screens for communication problems in children aged 4–16 years	• Language • Adaptive behaviour/ social skills/social communication
Informal language assessment	A less structured assessment provides opportunities for the SALT to observe broader language ability such as reciprocity, self-directed language, and higher level social inferencing abilities	• Language • Adaptive behaviour/ social skills/social communication

Table 14.3 Rapid reference summary of psychometric testing considerations

1. Psychometric testing can be a statistically valid and reliable way to determine if there is impairment in the domains of Cognition, Memory, and Executive Functioning
2. In terms of FASD diagnostic criteria, 'impairment' is frequently defined as Composite or Subdomain scores that are two standard deviations or more below the mean, although there are other additional considerations included in some diagnostic criteria
3. Where it is not possible for a standardised assessment to be carried out, clinical assessment with converging evidence from multiple sources and DSM-5 diagnostic criteria for certain disorders may also be considered in specific domains which cannot be easily assessed
4. As Subtest scores within domains can often be variable in people with FASD, it is helpful to describe the whole profile of scores. Variability between Index and/or Subtest scores in itself can be evidence of impairment in FASD according to the Canadian guidelines [2, 3]
5. Wider assessment, including information from carers and teachers and behavioural observations during the assessment, is also of vital importance when considering interpretation of psychometric assessment data as many different factors can influence scores
6. The structured/externally prompted nature of the assessment environment may result in scores that do not correlate with daily functioning in unstructured settings

References

1. Astley SJ. Diagnostic guide for fetal alcohol spectrum disorders: the 4-digit diagnostic code. 3rd ed. Seattle: University of Washington Publication Services; 2004.
2. Chudley AE, Conry J, Cook JL, Loock C, Rosales T, LeBlanc N. Fetal alcohol spectrum disorder: Canadian guidelines for diagnosis. CMAJ. 2005;172(5 Suppl):S1–S21.
3. Cook JL, Green CR, Lilley CM, Anderson SM, Baldwin ME, Chudley AE, Rosales T. Fetal alcohol spectrum disorder: a guideline for diagnosis across the lifespan. Can Med Assoc J. 2016;188(3):191–7.
4. Wechsler D. Wechsler intelligence scale for children. 5th ed. Bloomington, MN: Pearson; 2014.
5. Flanagan DP, Alfonso VC. Essentials of WISC-V assessment. Hoboken: Wiley; 2017.
6. McCloskey G, Perkins LA. Essentials of executive functions assessment. Hoboken: Wiley; 2013.
7. Delis DC, Kaplan E, Kramer JH. Delis-Kaplan executive function system (D-KEFS). San Antonio: Psychological Corporation; 2001.
8. Zelazo PD, Muller U. Executive function in typical and atypical development. In: Goswami U, editor. Blackwell handbook of childhood cognitive development. Oxford: Blackwell Handbooks of Developmental Psychology; 2002. p. 445–69.
9. Kodituwakku PW, May PA, Clericuzio CL, Weers D. Emotion-related learning in individuals prenatally exposed to alcohol: an investigation of the relation between set-shifting, extinction of responses, and behavior. Neuropsychologia. 2001;39(7):699–708.
10. Kully-Marten K, Treit S, Pei J, Rasmussen C. Affective decision-making on the Iowa gambling task in children and adolescents with fetal alcohol spectrum disorders. J Inter Neuropsychol Soc. 2013;19(2):134–44.
11. Mohamed Z, ACS C, Livesey AC, RAS M. Comparisons of the BRIEF parental report and neuropsychological clinical tests of executive function in Fetal Alcohol Spectrum Disorders: data from the UK national specialist clinic. Child Neuropsychol. 2018; https://doi.org/10.1080/09297049.2018.1516202.
12. Mukherjee RAS, Layton M, Yacoub E, Turk JT. Autism and autistic traits in people exposed to heavy prenatal alcohol: data from a clinical series of 21 individuals and a nested case control study. Adv Ment Health Intellect Disabil. 2011;5:43–9.
13. Wechsler D. Wechsler intelligence scale for children. 4th ed. San Antonio: Psychological Corporation; 2003.
14. Wechsler D, Coalson DL, Raiford SE. WAIS-IV technical and interpretive manual. San Antonio, TX: Pearson; 2008.
15. Rasmussen C. Executive functioning and working memory in fetal alcohol spectrum disorder. Alcohol Clin Exp Res. 2005;29(8):1359–67.
16. Emslie H, Wilson FC, Burden V, Nimmo-Smith I, Wilson BA. Behavioural assessment of the dysexecutive syndrome for children (BADS-C). London, UK: Harcourt Assessment/The Psychological Corporation; 2003.
17. Korkman M, Kirk U, Kemp S. NEPSY. 2nd ed. San Antonio, TX: Pearson; 2007.
18. Gioia GA, Isquith PK, Guy SC, Kenworthy L. Behavior rating inventory of executive function. Odessa, FL: Psychological Assessment Resources; 2000.
19. Mattson SN, Goodman AM, Caine C, Delis DC, Riley EP. Executive functioning in children with heavy prenatal alcohol exposure. Alcohol Clin Exp Res. 1999;23(11):1808–15.
20. Stratton K, Howe C, Battaglia FC. Fetal alcohol syndrome: diagnosis, epidemiology, prevention, and treatment. National Academies Press; 1996.
21. George M, Gilbert S. Mental Capacity Act (2005) assessments: why everyone needs to know about the frontal lobe paradox. The Neuropsychologist. 2018, 5:59–66.
22. Wing L, Leekam S, Libby S, Gould J, Larcombe M. Diagnostic interview for social and communication disorders: background, inter-rater reliability and clinical use. J Child Psychol Psychiatry. 2002;43:307–25.
23. Conners CK. Conners 3rd ed manual. Toronto, ON: Multi-Health Systems; 2008.

24. Sparrow SS, Cicchetti DV, Balla DA. Vineland Adaptive Behavior Scales: Second edition (Vineland II), survey interview form/caregiver rating form. Livonia: Pearson; 2005.
25. Einfeld SL, Tonge BJ. Manual for the developmental behaviour checklist. Clayton, Melbourne and Sydney: Monash University for Developmental Psychiatry and School of Psychiatry, University of New South Wales; 1992.
26. Parham LD, Ecker C, Miller H, Henry DA, Glennon TJ. Sensory processing measure. Los Angeles: WPS; 2007.
27. Rutter M, Bailey A, Lord C. The social communication questionnaire. Los Angeles: Western Psychological Services; 2003.
28. Dunn W. The sensory profile: user's manual. San Antonio: Psychological Corporation; 1999.
29. Semel E, Wiig EH, Secord WA. Clinical evaluation of language fundamentals. 4th ed. Toronto, Canada: The Psychological Corporation/ A Harcourt Assessment Company; 2003.
30. Bishop D. Children's communication checklist. 2nd ed. London: Harcourt Assessment; 2003.

The Role of Speech and Language Therapy for Individuals with FASD

15

Rachelle Quaid and Lesley Wilson

Chapter Highlights

- Feeding a child with fetal alcohol spectrum disorder (FASD).
- Communication and language in FASD.
- Example of a case and complex presentation related to language.

The role of a Speech and Language Therapist (SALT) for people with fetal alcohol spectrum disorder (FASD) is both complex and extensive. The role not only looks at supporting the feeding and wider communication needs of the person therapeutically but also increasingly plays an important role in the diagnosis and assessment of people with FASD. This chapter will focus on both aspects.

15.1 Feeding Infants with FASD

Many infants with FASD will not be breastfed, in an effort to help eliminate any alcohol being transferred through the breast milk. Therefore, the following advice is written specifically for bottle feeding, although may also be useful for those babies who will be breastfed.

R. Quaid
Auckland District Health Board, Level 3, Starship Hospital, Auckland, New Zealand
e-mail: rachelleq@adhb.govt.nz

L. Wilson (✉)
FASD Specialist Behaviour Clinic, Surrey and Borders Partnership NHS Foundation Trust, Redhill, UK
e-mail: Lesley.wilson@sabp.nhs.uk

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_15

Infants with FASD may have varying degrees of dysmorphia/craniofacial abnormalities that can impact on feeding. Typically, the physical features impacting on feeding include:

- A large mouth and tongue: may create difficulties with the infant being able to fit a teat into the mouth and difficulties with lip seal.
- A thin tented upper lip: may create great difficulties with lip seal around the teat, thus making the suck weak and inefficient.
- Retro or micrognathia: again may create difficulties with lip seal around the teat, as well as affecting the efficiency of tongue stripping, and therefore milk expression during the suck.

15.2 Infants 0–6 Months

15.2.1 Common Characteristics/Difficulties

Newborns with FASD tend to have difficulty establishing routines. They tend to sleep poorly, are irritable, hypersensitive to touch light and sound, and are often poor feeders [1].

These infants tend to be difficult to settle despite being keen suckers on a pacifier. The rooting reflex is often described as poor [2]. The suck is characterised by poor lip seal (often due to their physical characteristics), and a frantic fast suck rate leading to an inefficient suck with poor milk expression. These are the babies that seem to suck ferociously for little volume at the end of the feed. As a result, they are often fed for prolonged feed times (in excess of 30 min), or in very short frequent bursts. This in turn further contributes to difficulties establishing a feeding/sleeping routine.

These characteristics will be further exacerbated if the infant is experiencing alcohol withdrawal.

15.3 Feeding Tips

Infants with FASD often handle transitions poorly, so it is important to establish a strict and predictable routine, particularly around feeding [1].

Routine feed times, rather than on demand, will be more beneficial as it can be difficult to differentiate hunger cues from withdrawal cues/tiredness/general grumpiness, especially as these babies are often voracious suckers.

To help with the baby's feeding routine, there should be a consistent feeder, particularly if in a ward environment. The feeding area should ideally be darkened, quiet, free of distractions and familiar. Babies should be fed in the same place, position and with the same bottle and teats. It can be tempting to constantly change the teat and bottle of a poor feeder, in an attempt to 'find the best one'; however, these constant changes will often only serve to confuse and further unsettle the infant.

All feed times should start the same way, in a manner that is achievable for the family. For example:

- Starting with a nappy change
- Putting on a bib
- Being swaddled
- A familiar song etc.

The end of feed time should also be indicated again in a set way, for example:

- Wiping the mouth
- Burping
- Removing the swaddle
- Saying 'all done' etc.

Swaddling infants during feeds may help them feel secure, therefore enabling them to settle. By having limbs contained they are less likely to be distracted by flailing limbs, especially if they are withdrawing. This will also benefit those infants with altered motor tone.

When introducing a bottle, the rooting reflex should always be elicited first. This is what integrates all the cranial nerves and muscles needed for effective feeding [3]. If the bottle is just put in the mouth without the rooting reflex, the infant will have great difficulty coordinating an efficient and safe suck and swallow. The rooting reflex can be elicited by touching the teat to the sides of the mouth or top lip. The baby needs to be given time to turn towards the teat with a wide open mouth. This will help achieve good latching to the teat and gives the best foundation for an effective suck.

If babies are not able to demonstrate the rooting reflex, or appear sluggish with this, then it is possible to help to stimulate this by firm yet gentle stroking from the ear lobe to the corner of the mouth, 3–4 times on each side. This will encourage the infant to turn towards the stimuli, thus increasing the likelihood of the rooting reflex occurring.

Feed times should be limited to 30 min. This will give the infant the best opportunity to rest and sleep in preparation for the next feed. The infants that are fussy, coming on and off the bottle are unlikely to consume many more calories after 30 min and are better off receiving the balance of their feed via a tube rather than trying to the bottle feed. Prolonging feed times can lead to a vicious cycle of the baby being too sleepy and grumpy to feed well next time, leading to longer feed times and increased carer stress and so on.

If the infant has found their rhythm when feeding, then there is no need to stop for regular burping breaks unless the infant is showing discomfort. This will be shown by the infant wriggling, drawing their legs up, grimacing and tuning away from the bottle. At this point the bottle should be removed, the baby burped, and then the rooting reflex elicited again before re-offering the bottle.

If the infant is prone to gagging during feeding, then it may be that the rooting reflex has not been properly elicited, and this should be carefully checked. If, despite an appropriate rooting reflex, gagging persists, then it is possible the infant has a hypersensitive oral cavity. Shorter teats may be useful in the short term for these infants. In addition, encouraging the baby to suck on fingers, toys, swaddling etc. will all help to desensitise the oral cavity and normalise the gag. Should gagging be persistent during oral feeds, then it is often better to limit oral feeding and rely on tube feeding for nutrition until the gagging is reduced, as otherwise the infant is at risk of developing both on oral and food aversion.

Setting up these early routines will likely help the infant progress more easily onto solid foods.

15.4 Feeding Equipment (Bottles and Teats)

There are a great variety of teats and bottles on the market, each proclaiming to be superior to their counterparts for various reasons. There is little evidence to support one bottle and teat over another with this population. It is not unusual for carers to try many teats and bottles in the hope of finding the one that will work best for their baby. This may only serve to confuse and unsettle the baby, who is likely orally hypersensitive to even small changes in teat size and shape. It is often better to choose one bottle and teat and then stick with this. With this in mind, the following advice provides guidelines only, rather than being prescriptive.

If lip seal/latching to the bottle is problematic then using a different teat may be useful. The literature is contradictory about whether wide-based teats are better vs. standard neck teats. It is argued that the wide base teats allow the infant to create a seal with less effort, whereas others will argue that this encourages jaw slackness, and that alternatively, a standard neck teat will be more beneficial.

When considering flow rate of the teat, it is important to think of the safety and efficiency of the infant's feeding as well as volume taken. It can be tempting to use a faster flow teat, especially when only small volumes appear to be taken; however, this can have significant impact on the infants' ability to suck and swallow safely. Using the faster flow teats tends to lead to a negative feeding experience for both the infant and the feeder, setting the baby up for future potential feeding difficulties. Because of the often voracious nature of the FASD infant suck and the tendency towards a faster suck rate, a slower flow teat may be considered. This can have the side effect of further reducing the volume of milk taken however. It is probable that the infant will need supplemental tube feeding initially. This takes the pressure off the infant to achieve a certain volume, and allows the baby to feed within its ability, as well as helping the baby achieve the best practice for oral feeding.

The material the teat is made from should also be considered. Latex teats may be prone to flattening out with the inner edges sticking together, particularly with a fast and frantic suck. This further inhibits the baby's ability to express milk from the bottle. With this in mind, a silicone teat may be useful as it is more robust and resistant to being flattened.

15.5 Infants 6 months +

15.5.1 Common Characteristics/Difficulties

Introduction of solids is often delayed in this population with the median age for introduction of solids not until 9 months [2]. These infants are also slow to acquire self-feeding behaviours (1.08 years vs. 1 year). Possibly these two behaviours are related. The reasons for this delay is unclear in the literature, however, it may be due to delayed oromotor skills, altered tone, hypersensitivities, social situations or a combination of all of these.

Some of these children may experience oral or food aversion, likely associated with the hypersensitivity experienced in the newborn period. It is important to differentiate between these two terms. An oral aversion is characterised by the child becoming upset at any touch around the mouth or face. Teeth brushing is likely to be traumatic as is face washing, kissing games etc. In contrast, the child with a food aversion may well enjoy touch around their mouth, but will only experience difficulty when asked to eat specific disliked foods. This dislike may be based on colour, taste, smell, texture or a combination of any of these. Older children may have a tendency to be picky eaters and have a poor appetite.

15.6 Feeding Tips

Meal time routines and predictability are as important in this phase of eating as with bottle feeding, if not more so. Ideally families will eat together for most meals of the day. There should be a predictable routine that food time is coming up—this could be putting on a bib, sitting at the same chair, same plate and spoon etc. There should also be a predictable routine for the end of the meal—e.g. saying 'last spoon', wiping the face, removing the bib and then getting down from the table. Meal times should be no more than 20–30 min, especially if they are fraught.

Where possible, baby food should be introduced at 6 months of age. This will maximise the child's opportunity for accepting new tastes and developing those oromotor skills required for efficient and safe feeding. In a typically developing child, solids would be introduced once the child is showing an interest in adult food/eating behaviours, is becoming hungrier, and has sufficient head and trunk control to be able to sit for a short period of time. If these cues/milestones are not being shown, then small tastes only of smooth puree (e.g. 1–2 baby spoons) once a day should still be introduced. Ensure the child is supported in an upright position, and the feed time should be relaxed and enjoyable. The aim is for a pleasurable experience, rather than to consume calories. Amounts offered should be gradually increased as food is accepted and enjoyed, with a new taste introduced every 3–4 days. Some babies have difficulty accepting puree off a spoon. When this occurs, offering tastes on the lips via a caregiver's finger is a good way of working around this, saving the spoon for later on.

It is quite normal for babies to pull funny faces/poke out their tongues when trying new flavours. This is a normal reaction to a new taste, with many sources stating a new taste needs to be offered as many as 14 separate occasions before being recognised as an accepted familiar food by the baby. If, however, there is gagging or overtly aversive behaviour, then offers of puree should be stopped for a week or so, and then started again.

Spitting out of food may occur due to delayed oromotor skills, picky eating, overstuffing the mouth or a combination of all 3. It is important to distinguish which behaviours are causing the problem. Overstuffing can be managed by reducing the size of pieces given to the child, and ensuring their mouth is clear before having the next piece of food. For the child that is old enough, it can be a useful exercise to have them check their mouth in a mirror after each mouthful. This will help them marry up the sensory feedback in their mouth in a visual way. For the child with delayed oromotor skills, then the techniques mentioned in texture progression will help. The picky eater is more difficult to resolve and may need the use of reward charts/motivation to manage.

15.7 Oral Aversion

The child who is genuinely orally aversive is unlikely to accept any food in their mouth. Therefore, trust has to be built up between the child, the carer and food. Food should never be forced or coerced. Using messy play is a useful way of engaging the child with food in a nonthreatening way. For those children where weight gain is of concern, then the pressure of consuming calories can be alleviated by temporarily inserting a nasogastric tube, although this also has some negative experiences associated with the oral cavity.

15.8 Food Aversion

Some children seem to get stuck on smooth purees only, and will gag or vomit when offered more textured foods. This may be due to an oral hypersensitivity, delayed oromotor skills or a combination of both.

When attempting to progress the child through new textures, it needs to be done in a slow and controlled way. By beginning with accepted/preferred textures, slow changes/increases to texture can be created by adding tiny amounts of baby cereal or thickener to purees. The amounts must be barely visible, and may not even be discernible to the parent's palate. Adding ½ teaspoon of baby rice into a pureed meal should be the maximum amount of change at a time.

Children tend to accept smooth runny purees first. Before they can accept mixed textures (e.g. crunchy cereal in milk, carrots in a casserole) they must first be able to tolerate a thicker texture and manipulate this around their mouth. This requires a degree of maturation of oromotor skills. Textures should be gradually thickened to a mash potato consistency. Within this consistency, small soft lumps can then be

'hidden' within the texture. This gives the child the opportunity to practise those sideways tongue movements with minimal risk of stray bits of food getting 'lost in the mouth' and causing distress. These small lumps can gradually be increased in size, with increasing differences in texture. Again, it should be stressed that these changes need to be slow and subtle, particularly with the child displaying hypersensitivity to foods in the mouth.

Young infants are only able to achieve a backward and forward tongue movement, and this is all that is required for liquid consistencies and runnier purees. With maturation and eating practice, they are then able to achieve the sideways tongue movements and independent jaw movements needed for chewing.

15.8.1 Messy Play

Messy play is always recommended for those children with an oral or food aversion. This can be offered easily in a high chair with a tray (thus helping to limit mess). The more often messy play is offered, the more quickly you are likely to see results. Messy play will ideally be all food based, with any attempt by the child (accidentally or otherwise) to put the food to their mouth or lips given lots of exaggerated praise. There should be no coercion for the child to taste the food.

Messy play comes in a hierarchy of difficult textures, with easier textures being those that will leave no residue on the hands (e.g. uncooked pasta) up to those which are very slimy and sticky (e.g. whipped cream) and everything in between. Start to offer those textures your child is comfortable with, then slowly challenge the child with increasingly wet/messy textures. By doing this you are allowing the child to explore smell, texture, viscosity and temperature in a safe and contained way.

To give the child with FASD the best opportunities for successful and positive feeding experiences, mealtimes need to be routine, predictable and pleasant. Infants should be offered a taste/texture they are comfortable with, and if changes are required these should be made slowly and subtly. How quickly a child will be able to develop and mature their oromotor skills may be limited by their CNS dysfunction. Currently, it is unclear whether the presence of early feeding difficulties in babies with fetal alcohol exposure is predictive of later more generalised neurological problems, and would be an interesting area for future research.

15.9 The Role of Communication Assessment in Diagnosis

A communication assessment forms an integral part of a clinical diagnosis of Fetal Alcohol Syndrome (FASD). For example, in the assessment clinic at Surrey and Borders NHS Foundation Trust we utilise a combination of assessments techniques to evaluate the communication skills of those who attend. The use of historical information from the person themselves/family members etc. and formal and informal testing is used to reach overall conclusions about a person's communication strengths and areas of development.

Formal testing, using the Clinical Evaluation of Language Fundamentals, fourth UK Edition (CELF 4), is supplemented with informal assessments including sequencing, nonverbal recognition and inferencing tasks. In combination these provide information about the person's receptive and expressive language skills together with their use of pragmatics. Whilst it should be recognised that the information gathered is a snap shot and possibly may not fully illustrate people's communication styles and abilities in day-to-day situations, the combination of formal and informal testing plus information from the individual being assessed and their families gave us a good understanding of their strengths and a general baseline of information.

There are some reservations from our clinical experience about the use of the CELF 4 as it is not always standardised for the age group it is used for. However, there is not another appropriate standardised test on the market that does the work so efficiently.

The people that attend the FASD clinic are a particularly disparate group. Age ranges widely as do their backgrounds, educational and career development. There is a huge variation in their skill sets but there are also many similarities. Often people attending the clinic will describe difficulty at school with concentration and attention and difficulties with working memory. Either the person themselves or their family are looking for answers, trying to understand why the individual is experiencing this particular group of problems and wanting to know how best to assist them. Either the children that attend, or their parents, are seeking help to understand the root of the difficulties that they are experiencing, and are looking for tangible strategies that they may adopt to improve performance. Many adults are in the same position but are beginning to develop work arounds in their lives.

The remaining part of the chapter will consider some of the clinical trends seen in the communication skills of the people attending for assessment.

15.10 Concentration/Attention

Many people tested had a level of attention lower than might be expected for their age and maturation. During assessment it was often necessary to break tasks into short components and agree joint goals. Agreements such as 'when we reach number 15 we will break and do…..' helped people to manage their attention levels more efficiently. Without this strategy people more quickly reached a point where they simply 'switched off' and did not concentrate at all.

For younger people, a breakdown in concentration would result, as might be expected, in them utilising a number of distraction techniques. Wanting to go to the toilet, have a drink, fiddling, or in their general behaviour becoming harder to manage. For people past their school years, there could often be a much more passive response. They did not get up and move around or employ the distraction techniques of their younger counterparts but would rather sit passively, staring at the test

material hoping that a prompt may materialise or that the examiner would end the session. On questioning, it became clear that this was a technique that many people had utilised during their school years. In a group of 30 children, it is easy to sit quietly and wait for the lesson to be over. Giving a break either literally or by substitution of another task often means that they are able to return to the task and effectively complete the next part of the subtest.

If there were marked attention/concentration problems, interventions were suggested. Such as recognise and acknowledge the problem to yourself and with others around you. Let people know what would help. Adapt tasks to match abilities. For example, for many, understanding that situations where they are given large chunks of information all at once, such as lectures, is going to be difficult. Therefore, either avoid such situations or employ coping mechanisms such as recording the information and listening to it back in smaller chunks. For individuals experiencing these attention/concentration problems, it is important that factual information is given in small chunks, and backed up by being written down.

15.11 Understanding Complex Visual Information

Clinical experience has identified that some people who have attended the clinic find complex visual information confusing. So, for instance if given a set of cards to place sequentially in a line to tell a story, they might fail to notice sufficient detail in the pictures to enable them to do this successfully. For example, one element of the informal testing is to ask people to arrange 3 sets of sequential cards to form a story. It was quite usual for people to experience difficulty, for example, in sequentially arranging a set of 6 pictures depicting a shopping expedition to the market, even though they had previously correctly arranged other sequences. This was because the market sequence was visually very busy and it was more difficult to isolate the key elements that made it possible to work out the sequence of events. In this case the number of items that had been placed in the shopping basket. Once this had been observed, it allowed a judgement to be made as to which stalls had been visited and in which order. Whilst we often give advice to people who have attended the clinic, to make the best use of visual information, whether it is written or pictorial, to enhance understanding and memory. We also note that it is important that this information is clear and uncluttered or it itself becomes a distraction. These informal tests add to an understanding of how the person may function in a broader setting and at least offers important qualitative information to an evaluation.

It is also worth noting that unusually a small number of people who attended the clinic did not automatically defer to the accepted left/right line scanning technique that most of us would automatically use. So, for example, when looking at a line of small line drawings of 7/8 items in a line and asked which picture is to the right of the car they would ask whether they should start from the left or the right.

15.12 Working Memory Problems

Some working memory problems can be seen in many of the test results, particularly obvious where people have been asked to recall a sentence given moments before. Often this presented a problem, people were able to internalise short sentences containing simple language structures to provide accurate recall. But as the sentence increased in length and complexity, they would begin to substitute words or whole phrases, sometimes this resulted in a change of meaning to the sentence or a loss of the richness of the information contained with it. This clearly has implications in terms of how normal information is given to them in a non-clinical context and can be used to ascertain how to best support the individual and the level of scaffolding needed by that individual.

This recall is required in many aspects of life, e.g. following directions, instructions, note taking and learning new vocabulary.

This difficulty with recall was also often noted in the recall of short paragraphs. This task involves listening to one of three short paragraphs and then after each answering a set of five questions some of which require the recollection of facts, others that require the person to infer information from the facts that they had heard.

To perform well in this task, information must be processed and stored in the intermediate memory.

A difficulty with these skills suggests that someone will have difficulty in everyday life processing conversations and those difficulties will become more marked as:

- Sentences become longer
- More complex
- More time elapses between hearing the information and acting upon it

Where the information must not only be retained and processed but also used to infer and predict future information or actions. It is therefore important to be aware that it will be difficult to process complex language especially if there is a considerable volume of it. If given verbal instructions, it is important to ask whoever is delivering the instructions to give them in short bursts and to check out that it has been fully understood before offering the next layer of complexity.

It is also worth noting at this stage that it is not unusual for people to show a paucity of vocabulary, experiencing difficulty in explaining the meaning of some words. An ability to define words clearly illustrates a skill in broadening word meaning to form concepts, acquire new word meanings and develop an understanding of word use. It has been found that, trying to make sense of the words that are not understood, using the context of the situation to tease out what the person thinks the meaning of a particular word might be, is not always helpful. As it leads to individuals filling gaps and presenting with confabulation.

Essential too for listening and reading competence is the ability to recognise similarities and make semantic links in the meaning of words and from word associations. In this group, once again this can be a variable presentation. For example,

some people falling well below expected level whilst others seemingly function better.

The following case example illustrates some of the issues and also how there may be variability in the presentation. It also demonstrates why the qualitative information is important in supplementing the quantitative findings in these groups.

15.13 Case of B (Fully Anonymised)

B is 12 years old young man who was referred to the FASD diagnostic clinic. His birth mother was known to have drunk heavily during and after pregnancy and B suffered a period of neglect before he was finally taken into the care of the local authority. He was adopted as a 16-month-old baby.

B was experiencing problems at school. He was not concentrating during lessons. He was not disruptive, but often simply opted out of lessons. He was described by his teachers and parents as being easily led, often not thinking through the consequences of what his actions might be. Like all people referred to the FASD diagnostic clinic, a speech and language assessment is completed as part of the assessment clinic. B had already received an initial assessment by the Consultant and a psychological assessment. Seven of the CELF4 subtests were administered together with some informal pragmatic language tasks.

Table 15.1 below has the standard scores and percentile ranking for those seven subtests. This shows the wide variability in scores.

B responded appropriately to social greetings, understood about turn taking in conversations and how to make his needs known verbally. In general, he played the passive partner in conversations. He would respond to questions and comments but not introduce new topics of conversation and on the whole, not extend a conversation beyond supplying an answer to a given question. He did not make eye contact during the assessment and when he was having difficulty maintaining his attention or experiencing difficulty with a task he would drum his fingers on the table or fiddle with his hands. This drumming and fiddling behaviour disappeared when he was fully engaged in a task and performing well.

Table 15.1 Scores from CELF on 7 domains assessed in B

	Percentile
Recalling sentences	25
Formulating sentences	63
Word classes: Receptive	50
Expressive	25
Total	37
Word definition	25
Understanding spoken paragraphs	5
Sentence assembly	50

Most subsections of the test had to be broken down into smaller units and interspersed with each other to ensure he maintained concentration but B did participate well during the 2-h assessment.

Recalling sentences	Percentile: 25

B was able to internalise short sentences to understand and recall the meaning, structure and intent of spoken words. He consistently made minor substitutions of words where he had internalised the general meaning of sentence but imposed his own, slightly immature grammatical structures on the sentence so 'the class that sells' became 'the class who sells'.

As sentences become longer in length, he made far more errors. He showed a definite pattern of remembering the first part of the sentence and then being unable to recall the second half. He would say the words that he did recall and then stop speaking. He showed some discomfort, by drumming his fingers on the table, aware that he had not repeated everything he had heard, but was unable to even guess at the missing content. He made no comment about his performance but silently, passively waiting for the therapist to comment or move on.

Formulated sentences	Percentile: 63

B was given a picture and a word and asked to describe the picture with a sentence that incorporated that word he had been given. He was much more comfortable with this task and was able to formulate complex sentences using either given words and/or illustrations. There was no concern about the grammatical content of the sentences.

Word classes: receptive	Percentile: 50
Expressive	25
Total	37

B was given four words, two of which were semantically similar and asked if he could identify the words that went together and explain what he thought the connection was.

B could intuitively recognise that some words were semantically connected, for example, window and glass although as the vocabulary became more complex he struggled more, simply because he did not recognise the words and therefore know their meaning or relationship to other words. Sometimes, he was able to understand that there was relationship between words but was not able to explain that relationship. It was again very noticeable that B did not make any attempt to guess if he did not know the answer but exhibited behaviours similar to the previous subtest.

Word definitions	Percentile: 25

B struggled with this task because it required some concentration and, marshalling of thought. His responses were often near to a definition of the target word but lacked a sharpness that would give a full definition. So, for example, when asked to define the word 'priority', he responded 'goal' rather than the first, or top of the list.

Understanding spoken paragraphs	Percentile: 5

B experienced considerable difficulty with this element of the test. It involved listening to short paragraphs that included facts as well as inferring meaning and then remembering the information to use in order to answer verbal questions.

B did not retain some of the factual evidence and experienced difficulty inferring information from the information he had heard.

Sentence assembly	Percentile: 50

Given words in a written form, B was asked to arrange these into first one and then another semantically different and grammatically correct sentence. B was able to do this and score was average for his age but he struggled with the task. He tended to see the first sentence immediately and then could not discard this from his mind to make another new sentence using the same words. He would repeat the same sentence aloud to himself time and again not realising it was identical to the last.

B's performance across the subtest was fairly consistent except for his performance on listening to spoken paragraphs where a number of language functions are required at one time. The qualitative aspects of his presentation; however, when taken alongside the overall scores suggest someone with subtle but important difficulties. If not identified and addressed through reasonable adjustments, it would impact on his functioning and learning. This highlights the importance of conducting detailed assessments of someone's communication ability. It also demonstrates how a language and communication assessment can complement the evaluation of other groups such as psychologists but presenting information that adds to the overall profile of an individual.

References

1. Godel J. Fetal alcohol syndrome. 2002. https://www.ncbi.nlm.nih.gov/pmc/articles/PMC2794810/#b42-pch07161.
2. Amos-Kroohs RM, et al. Abnormal eating behaviours are common in children with FASD. J Pediatr. 2016;169:194–200.
3. Wolf and Glass. Feeding and swallowing disorders in infancy. Therapy Skill Builders; 1992.

The Complexity of Attachment Issues in FASD

16

Helen Oakwater

Chapter Highlights

- The relationship between attachment parenting and prenatal alcohol
- How different factors influence the relationship between parent and child
- Parenting and fetal alcohol spectrum disorder (FASD)

If you met my children briefly, you wouldn't know they had fetal alcohol spectrum disorder (FASD). If you spent the day with them, you might not know. If you spent a week with them, you'd get an inkling something wasn't right. Spend a year with them and you would *know* something was wrong, but be unable to identify what. You'd also find yourself frustrated by their inexplicable, inconsistent behaviour. When these three children, aged 2, 4 and 5, walked through my front door 1 day in the early 1990s, it took me 2 years to realise that their functioning was 'odd'. It took two decades for me to fully appreciate the nature and magnitude of their dysfunction. Early on, I thought maybe it was poor attachment, or perhaps the impact of trauma. Much later, I discovered that during each pregnancy, their birth mother had consumed alcohol. Only much, much later did I realise that the intersection of the attachment process with fetal alcohol exposure almost certainly compounded the complexity of the difficulties experienced by my children and many others.

In this chapter, I have constructed four stereotypical examples (Sandy, Tim, Aamy and Bob) to illustrate how theory, research, neuroscience, biology and functioning are intertwined.

H. Oakwater (✉)
FAB Parents, London, UK
e-mail: helen@helenoakwater.com

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_16

16.1 What Is Attachment in a Typically Developing Child?

The connection between an infant and its mother is generally referred to as 'attachment' and is built over time, through regular interactions with a (ideally) nurturing mother, or caregiver. Its essentially a relationship, vital for survival and protection from which the child can explore the surrounding world and move towards adult independence.

Box 16.1 Further Reading on Attachment
The work of Bowlby and Ainsworth, the pioneers who first developed theory around attachment—see also reference [1]:

- Attachment theory into practice. Briefing paper No.26, British Psychological Society: www.bps.org.uk.
- Attachment Across The Lifecourse. David Howe 2011. Red Globe Press®, Springer Nature Ltd., London. ISBN 978-0-230-29,359-5.

The process of attachment also functions as a 'brain programmer' for the developing brain, which affects current and future behaviour. Segments of the attachment circuitry within the brain have been explored through animal models. These circuits are massively receptive in the first year of life, providing a unique window of opportunity for developing a strong attachment between infant and mother, designed to keep the child safe. Importantly, this attachment occurs, *regardless* of the quality of care received, even if it is abusive or neglectful (although these factors might alter the "character" of the attachment).

Neuro-behavioural processes involving complicated hormonal, neurological and behavioural interactions occur in mothers after birth, which drive her to connect to the infant. Of these, the hormone oxytocin has been best researched [2].

For the infant, the attachment experience is sensory. Loving eyes gazing down, familiar smell, known soothing voice, soft stroking, warm milk, whilst any discomfort is alleviated quickly. There is an abundance of norepinephrine (noradrenaline) released from the locus coeruleus in the brain stem, which supports the attachment process [3].

Although there is currently little discussion about the Autonomic Nervous System (ANS) in relation to attachment, it is known that the ANS is involved in the regulation of arousal levels in the nervous system as a whole. The ability to self-regulate and retain some control over our level of arousal is crucial: by remaining physiologically calm, we are able to think and act in our best interest. Indeed, this is an essential life skill [4]. It is far harder to think clearly when angry, or dissociated.

As an example, a nurturing, yet sleep-deprived, stressed caregiver has to control their own arousal level and focus on calming a screeching infant. Competent

self-regulation is sensed by the infant who intrinsically acquires the calming skills of deep breathing, slower heart rate and being grounded. This 'dance of attunement' happens frequently. You will spot it every time an adult rhythmically bounces a baby, quietly sings a lullaby, or a myriad of other nurturing behaviours.

Infants do need to learn to know *how* to regulate, or calm themselves. As development and behaviour become more complicated with increasing age, this is learned and reinforced slowly over many years through thousands of sensitive interactions with attuned caregivers.

Attachment is a dynamic process. It's at the heart of our relationship with ourselves, and between 'self' and others. The quality and nature of our early infant attachment experience determines our subsequent 'attachment style' as first described and predicted by Bowlby in the late 1950s [1].

16.2 Secure Attachment (Creating SANDY)

Consider the experience of an infant who is nurtured, loved, soothed and has her needs met regularly and repeatedly: hour by hour. Day by day. Month after month. As shown by the diagram below (Fig. 16.1).

From these repeated experiences, SANDY (Securely Attached Normal Developing Youngster) starts generalising and creates a representation of herself and the world: her inner working model. If she is upset or hurt, a familiar face responds to alleviate her distress. Life is consistent, predictable. She learns to trust. The next diagram suggests her possible belief system and sense of self. She can tolerate occasional and minor inconsistencies. Her parents do not need to be perfect (Fig. 16.2).

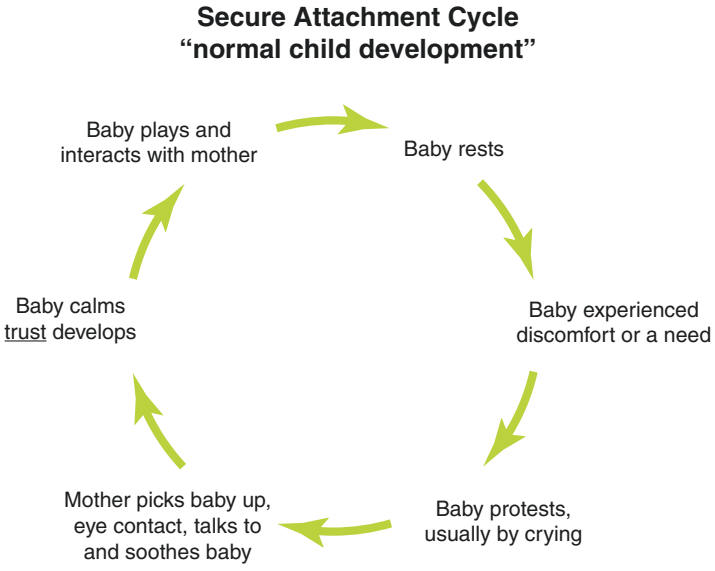


Fig. 16.1 Secure attachment cycle

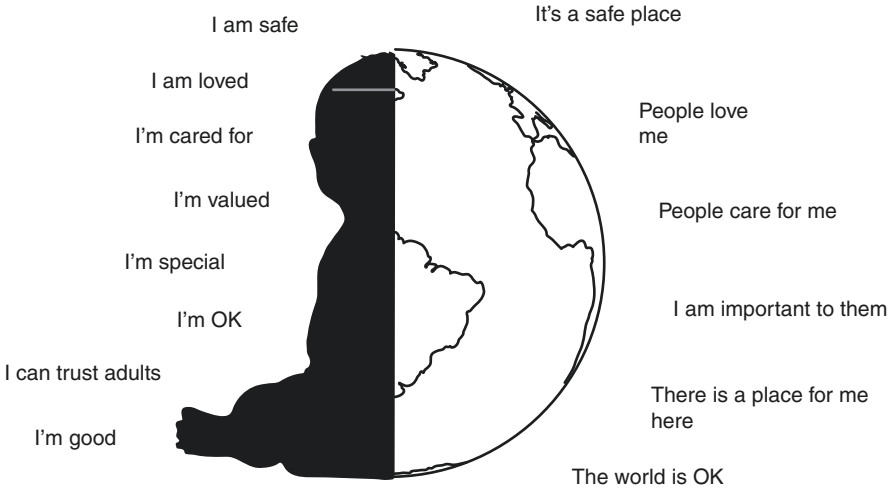


Fig. 16.2 Secure attachment: Child's view of self and world

16.3 A Continuum of Parenting

Fortunately, most children have attuned, empathic caregivers who are capable of putting their child's needs ahead of their own. They are, to use Winnicott's phrase, "good enough parents" [5].

However, for a wide range of reasons, some caregivers do not have that capacity. Parenting requires a mixed bag of skills, which change over time. The negotiating skills and boundary setting needed to manage a teenager are wasted on a howling toddler, who does not have the executive functioning and thinking skills to participate in such dialogue. But, both require the adult to maintain a calm state.

There are a variety of different dysfunctional parenting styles which can have profound long-term impacts on child development and behaviour [6, 7].

Abusive behaviours such as physical and emotional maltreatment directed towards children, as well as neglect, can also significantly adversely affect children's social and emotional development path to adulthood. It is likely that many such parents have themselves experienced poor parenting role models, in combination with poor attachment. They may not have developed emotionally mature ways of resolving inter-parental conflict, or their own internal conflicts.

Forward [8] describes "Toxic Parents having negative patterns of behaviour which are consistent and dominant in a child's life … The adult children of toxic parents suffer similar symptoms—damaged self-esteem, leading to self-destructive behaviour… They almost all feel unlovable and inadequate." (Several decades later, the description 'toxic parent' is probably a harsh description for styles of parenting that are highly likely to result from the adults' own disordered attachment patterns. This explains, rather than blames individuals or couples with dysfunctional relationships possibly including partner conflict and the use of drugs and/or alcohol.)

The behaviours expressed as part of this type of parenting will, over time, disrupt the 'normal' attachment process. What was at that time unclear—and still to large extent remains so—is whether these types of parenting styles have a greater impact on children who are already affected by underlying brain damage (such as that associated with prenatal alcohol exposure). In other words, in the context of a child with neurodisability, does the adverse parenting style make the situation worse than it might otherwise have been, or is the presentation due to the pre-existing damage alone and independent of parenting style? (Fig. 16.3).

Typically, these are developing infants who might be regularly ignored, neglected, screamed at, handled roughly, left alone, who are often hungry, thirsty, too hot, too cold, or might have sores from extensive nappy rash or pain from an undiagnosed medical condition, and living in a hostile world full of fear, with significant adverse sensory experiences.

Trauma can be defined as either, a belief you might die or an overwhelm of affect (feeling/emotion) which you are unable to integrate. Hence living with a toxic parent is truly "traumatic" which creates 'TIM' (Traumatised Infant, Maltreated). (Fig. 16.4).

There are several different types of insecure attachments, and there is evidence to support the causal link between how a dysfunctional parent behaves and the subsequent likely attachment style of the child [9]. This is summarised in Fig. 16.5:

Pause for a moment and think of behaviours in parents who struggle to manage their emotions, which may be regulated through the use of alcohol and drugs. Sometimes they seem happy, then euphoric, then maybe teary or shouting, then maybe comatose. Tim witnesses each phase. On a scale of zero to ten, how safe and secure do you imagine he feels at each stage?

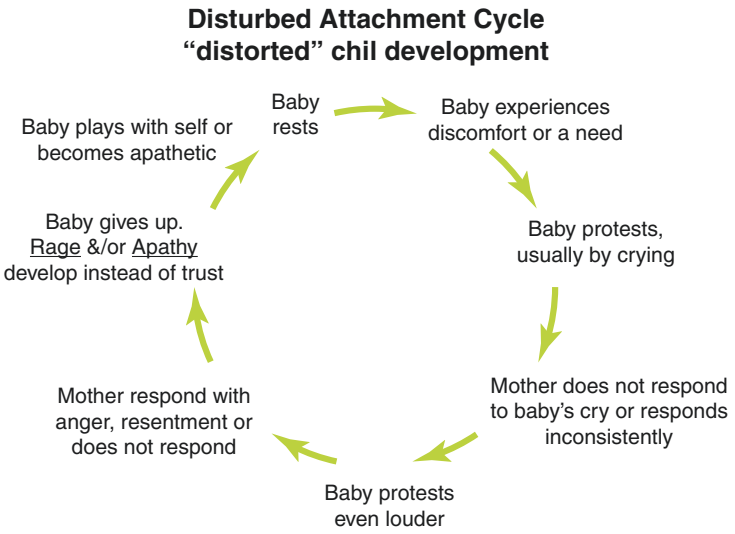


Fig. 16.3 Disturbed attachment cycle (creating TIM)

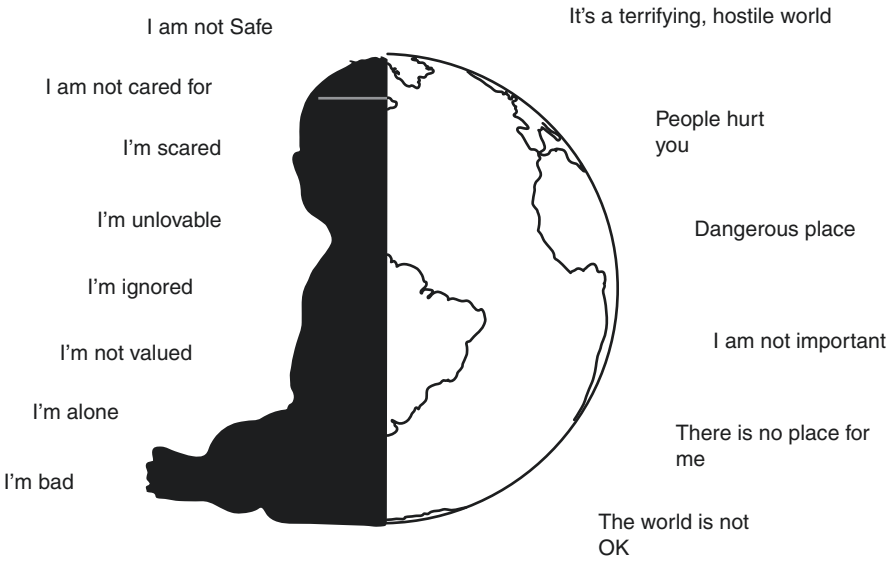


Fig. 16.4 Disturbed attachment: Child's view of self and world

Attachment type	***Ambivalent***	***Avoidant***	***Disorganised***
Parent/care-giver characteristics	Insensitive Under-involved Inconsistent Anxious Uncertain	Rejecting Hostile Cold Conditional Intrusive Controlling	Frightening Frightened Helpless Abdication of responsibility

Attachment type	***Ambivalent***	***Avoidant***	***Disorganised***
Typical associated behaviour in children	Hyperactivation of attachment Coercive Passive Dependent Need & Anger	Defended Emotions inhibited Compliant Self-sufficient Independent Anger & Fear	Fear/compliance Compulsive caregiving Rage + Fear + Sadness = controlling aggressive

Fig. 16.5 Relationship between parent and child attachment styles

16.4 Feeling Safe? (the Amygdala and Beyond…)

The amygdala plays a role as part of the fear-centre of the brain [3], operating akin to a smoke detector. Incoming sensory information is compared and contrasted with old records to decide; is this a threat: yes or no? Hence the circumstances/associations when 'records' (memories) are created will determine the level of activation of the amygdala in the future when similar associations are experienced. Children living with significant parental conflict develop excellent and effective early warning systems. Their smoke alarm is hypersensitive and remains so.

There are recent findings from animal research which indicate the complexity of these issues and how they change during early development. There seems to be a change from a maternal presence-influenced attachment system towards an amygdala-dependent threat system as they transition to independence [10]. During the infants' first year, their attachment circuitry is programmed for survival through connection to the caregiver. It seems that the ability of the amygdala to learn fear is suppressed during that first year's intensive attachment process.

Significantly, this combined activation of the attachment circuitry and suppression of the fear circuitry (amygdala plasticity) seems to produce a particular vulnerability to later mental health difficulties as well as increased mortality in later life, as demonstrated by various studies into the impact of Adverse Childhood Experiences (ACE) [11, 12]. This vulnerability in the FASD group is compounded by the fact that they have pre-existing brain pathology which somehow seems to reduce the resilience and increase the sensitivity of the brain to the impacts of these adverse events.

Neuroception, the ability to detect signs of safety or threat, is thought by some to be a functional component of the Autonomic Nervous System [13–15]. It has been suggested that this too can be 'mis-wired', becoming rather like a constantly pinging radar system, forever warning of some unspecified danger. If you don't feel safe in your body, you can't feel safe anywhere.

16.5 Attachment, Empathy and Future Relationships

Those with secure attachments are able to establish healthier relationships. Their capacity to connect to others has not been compromised by confusing signals from early caregivers. Functional relationships require empathy. Empathy is a complex, active process.

In his book, Zero Degrees of Empathy [16], Baron Cohen describes empathy as "… our ability to identify what someone else is thinking or feeling, and to respond to their thoughts and feelings with an appropriate emotion". So, there are two stages to empathy: recognition and response.

He argues that "developing empathy proceeds well only if it is safe to imagine another person's thoughts and feelings". Please note the language here: "only if it is safe". For a child growing up in the context of persistent abuse from a caregiver/parent, connecting to that mind could be terrifying, so avoidance or dissociation might be a better strategy.

He goes on to discuss at least ten specific interconnecting brain regions which constitute the '*empathy circuit'* and develops his theory to suggest that humans

express a spectrum of varying degrees of empathy, with seven empathy bands. At level zero, "an individual has no empathy at all", (psychopathic). At level six, an individual is constantly focused on others' feelings. Whilst empathy can be situational and fluctuate, he considers that the band that we mostly inhabit is broadly fixed. Even though he considers there may be a genetic component to empathy, he concludes empathy is also fundamentally the result of 'experiencing parental love'. Clearly, therefore, there is an overlap between attachment and empathy.

For people with social communication disorders such as Autism, it has been shown that empathy is innately affected as a core feature [17] and this may be related to poor development of their 'theory of mind'. [18] There is some suggestion that, irrespective of parenting, this is an innate difficulty in children with ASD, meaning that they are more likely to form insecure attachments [19]. Theory of mind difficulties have also been shown to be seen commonly in FASD [20], making it very likely that they too would be likely to establish insecure attachment irrespective of parenting style [21].

16.6 It's a System: Attachment, ANS and Maternal Alcohol Consumption

So how might the brain be different with respect to these aspects in the child who was exposed to alcohol as a developing fetus? It is known that there is damage to brain structures as well as neural connections in FASD. This will inevitably have an impact on how information is processed. Metaphorically, this can be seen like a road network, transporting messages through the brain and carrying motor and sensory information to the rest of the body. What if some of these roads were poorly constructed? What should have been a four-lane highway is a dirt track. Bridges stop in mid-air. Some messages would get lost, arrive at the wrong destination, or simply disappear. Others would be delivered appropriately. One particular road is open…unless it's raining. When there is an 'r' in the month, another pathway narrows from three lanes to one. It's a dysfunctional communication network, but one that is 'reliably inconsistent'. The exact nature of this dysfunctional communication system might be uniquely determined by the timing, quantity and frequency of alcohol exposure experienced by the fetus related to the particular stage of fetal nervous system development when the exposure occurs. The result, by reducing the child's ability to easily adapt to different situations, probably reduces what has been called 'resilience' – making disordered attachment more likely.

The relationship between developmental abnormalities and postnatal adverse experiences and attachment is interesting and merits further research. A recent small study looking at neurodevelopmental outcomes in those children with FASD with and without any postnatal neglect, found that the prenatal alcohol exposure appeared to act independently of postnatal environment with respect to neglect, which in itself did not change the neurodevelopmental or brain-related outcomes [22]. This suggests that attachment in this group is not just about parenting, but also about the underlying disorder. Both have to be considered to understand the outcomes seen in the child. Furthermore, it also explains why interventions around attachment may not actually be helpful.

This offers us some clarity. Especially for parents/caregivers. It allows us to understand and acknowledge a child's inconsistent behaviour. Many parents and

teachers are frustrated by a child whom they believe is deliberately oppositional, and realising the child actually "can't", rather than "won't", dissipates much emotional charge, reduces frustration and results in improved relationships.

16.7 Muddying the Waters: Pre- and Post-Birth Factors

Keeping it quite simple, we can look at two factors: was the child's nervous system affected before birth by fetal alcohol exposure, and/or affected in early infancy due to maltreatment? Figure 16.6 below gives us a template to help conceptualise these factors. We can then try to estimate whereabouts in the quadrant a specific child might reside. This could help guide us to an improved understanding of this individual child's problems and thus, how those issues might be best addressed.

In reality, we try to make sense of the behaviour of the child who shows up in our clinic, school or home. We wonder: "is their 'not normal' functioning due to neglect, FASD, poor parenting, underlying developmental disorder, attachment disorder?" Detective work is required to join up the dots and pinpoint the root cause(s). The overlap of symptoms between attachment disorders, FASD and neurodevelopmental disorders with need to identify or exclude other medical disorders means that reaching the correct diagnosis(-es) is difficult and requires specialist training and experience. Accurate diagnosis is essential to manage, support and parent the child,

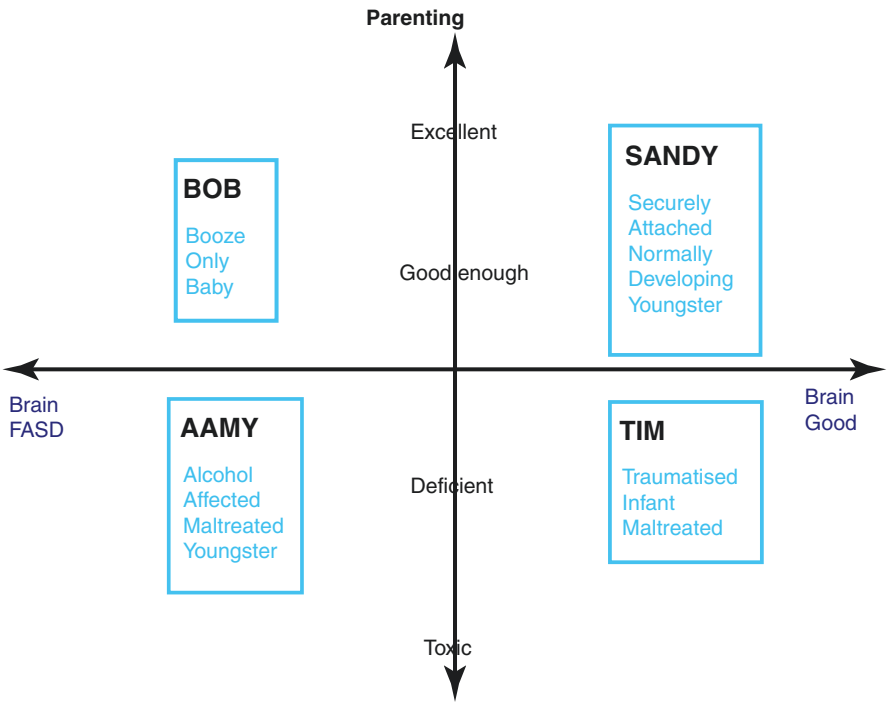


Fig. 16.6 Model of pre- and post-birth factors influence on the child

because from the correct diagnosis springs the correct understanding of the nature of the problems and potential for the correct help and therapeutic approach.

From a non-specialist perspective—if you know their internal communication system is structurally flawed, then to continue the road system analogy, traffic flow needs to be slow and gentle. Reduce crashes by keeping stress levels low. A neurodevelopmentally abnormal brain is likely to need much more external support to function. A child with compromised memory, benefits from instructions in bite size chunks, and likewise, defects in planning and executive function require stepwise instructions and often structured external support.

16.8 Parenting Expectations

If parents think they are parenting Sandy, they will expect her to meet normal developmental milestones and display age-appropriate behaviours. If they are actually parenting some variation of Bob, Aamy or Tim they often become disappointed or exacerbated.

Let's for a moment focus on Bob. His birth mother, a high-flying professional had, during pregnancy, 'just one daily glass of chardonnay' to unwind after a hard day. Her post-birth parenting potential was better than 'good enough', but Bob struggles with breast feeding, he is hard to comfort, his development is slow, milestones are not being met. Her friends are enjoying rapport with their children, but she is not. Is Bob's neurodevelopmentally abnormal brain hindering secure attachment for him and preventing fulfilling parenting for her? If Bob does not have FASD facial features, will anyone question whether his poor functioning is due to prenatal alcohol exposure? That's why diagnosis and honesty is so important. After the shock of diagnosis, she can modify her parental dreams and implement appropriate parenting strategies for Bob.

Sometimes, due to continued toxic parenting, Aamy and Tim are unable to stay with their birth families, so the courts decide permanence via fostering or adoption is the child's best option. Many children adopted from the care system have FASD, often undiagnosed, but strongly suspected. Many foster and adoptive parents knew our children had suffered significant maltreatment and believed their challenging behaviour was the result of trauma and attachment disorder. Unaware of the profound impact of maternal alcohol misuse; we parented Tim, not Aamy.

16.9 Schooling

Fast forward a few years into a classroom containing Sandy, Bob, Aamy and Tim. What will their behaviour be like? Do their parents, teachers or the education system realise the level of their brain/body functioning? What is each child capable of doing, and what can't they do? Is this seen as 'can't' or 'won't' by the supervising adult? Might they just be considered "oppositional" rather than disabled.

For many years, I've talked about 'bubble wrapped children' describing how during abuse or neglect, children swaddle themselves, in metaphorical bubblewrap, as a protective strategy [23]. Effective at the time, but later, its continued presence, distorts their perspective of the world, and the world's view of them. What if, below the bubblewrap, there is a neurodevelopmental abnormality due to prenatal alcohol exposure? Then the therapeutic reparenting strategies (undo the damage and fill in the gaps) for Tim, may not work with Aamy. Many adoptive parents (myself included) were unaware that our children also had underlying neurological damage. With this retrospective knowledge, I can now make sense of what seemed to be their 'nonsensical' behaviour. I wore "trauma glasses" to comprehend my kids, not knowing "FASD goggles" existed.

16.10 Parenting with FASD

Fast forward again. Sandy, Tim, Aamy and Bob are now legally adults, with the biological ability to produce offspring. In this context, I want to focus on one specific disability associated with FASD: the often-deep discrepancy between their physical age and the various different domains of development (Table 16.1):

What if this is Aamy's profile; or Roberta's, (known as Bob)? She struggles to manage money, time, relationships and stress levels. What if she is unsupported and living alone? She can't budget, plan appointments, make healthy friendships or protect herself. She is vulnerable to sexual exploitation, abuse or coercive control.

What is her understanding of sex, contraception, love, pregnancy, childbirth and parenting? Can she perceive a baby as a 24/7 commitment, or does she just want a living doll? (Please don't think I'm being harsh; this perspective comes from first-hand experience).

How does a pregnant woman like Aamy or Roberta (Bob) present to the outside world? How does she appear to a doctor, midwife, social worker, housing or police officer? She can talk the talk, but stumbles with the walk: repeatedly. Does any

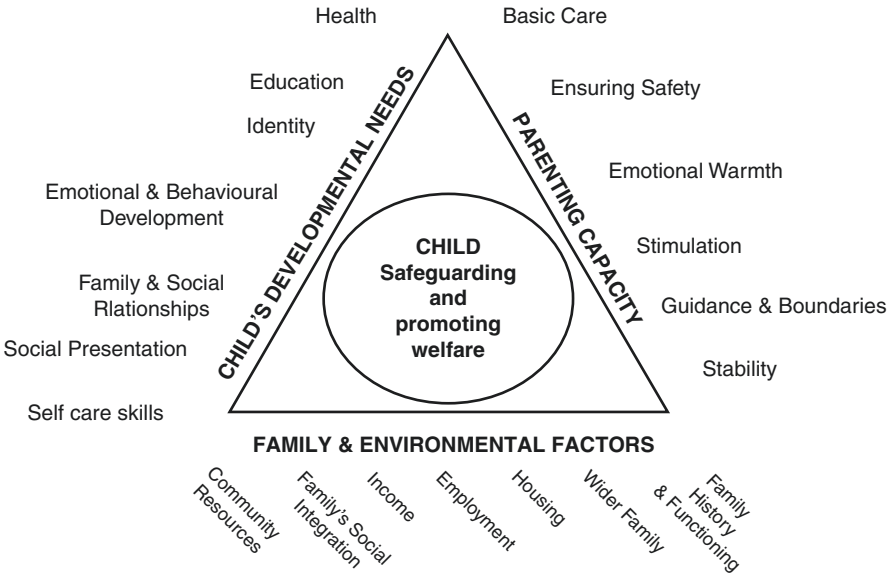
Table 16.1 Developmental profile example

Developmental Profile (Simplified example, not including all domains)	
Actual age: Level of sexual maturity:	16 years 16 years
Domain of development	Developmental age
Visual comprehension	15
Expressive language	14
Receptive language and linguistic processing	8
Executive function	10
Ability to handle money	9
Social skills	11
Emotional maturity	8 (variable)

professional spend sufficient time with her to detect that pattern? What is their knowledge of adult FASD?

What criteria might they use to assess her? Subjectively, from her good expressive language or objectively, through observed behaviour and appropriate testing?

Would the Common Assessment Framework (diagram below) fully reveal the underlying issues? Would the appropriate support be identified? Could it be provided?



Do Aamy or Bob have the capacity to be good enough parents? Could they support? If so, how much and in what form? Do professionals see them as feckless women or vulnerable adults with undiagnosed FASD and/or attachment disorder? What're their empathy levels? Can they create an environment for an infant to thrive? Do they use alcohol to manage their anxiety: before, during or after pregnancy?

Do they create a mini-me? A better or worse version of themselves? Are we looking at inter-generational repeated patterns?

So many questions. So many scenarios. No easy answers.

References

1. Bowlby J. Attachment and loss: retrospect and prospect. Am J Orthopsychiatry. 1982;52(4):664–78.
2. Galbally M, Lewis AJ, van IJzendoorn M, Permezel M. The role of oxytocin in mother-infant relations: a systematic review of human studies. Harv Rev Psychiatry. 2011;19(1):1–14. https://doi.org/10.3109/10673229.2011.549771.
3. Sullivan RM. Unique characteristics of neonatal classical conditioning: the role of the amygdala and locus coeruleus. Integr Psychol Behav Sci. 2001;36:293–307. https://doi.org/10.1007/BF02688797.

4. Porges SW, Furman SA. The early development of the autonomic nervous system provides a neural platform for social behaviour: a polyvagal perspective. Inf Child Develop. 2011;20:106–18. https://doi.org/10.1002/icd.688.
5. Winnicott DW. The theory of the parent-infant relationship. Int J Psychoanal. 1960;41:585–95.
6. Kumnig M, Höfer S, Huber A, et al. Patterns of dysfunctional parenting styles and psychological disturbances in offspring. Z Psychosom Med Psychother. 2013;59(4):356–68.
7. Bornstein L, Bornstein MH. Parenting styles and child social development. In: Tremblay RE, Barr RG, Peters RDV, editors. Encyclopedia on early childhood development [online]. Montreal, QC: Centre of Excellence for Early Childhood Development; 2007. p. 1–4. Available at: http://www.childencyclopedia.com/documents/BornsteinANGxp.pdf. Accessed 4 Dec 2020.
8. Forward S. Toxic parents: overcoming their hurtful legacy and reclaiming your life. New York: Bantam Books, Random House; 1989. ISBN 0-553-38140-7.
9. Howe D. Adoption and attachment. Adopt Foster. 1995;19(4):7–15. https://doi.org/10.1177/030857599501900403.
10. Sullivan RM, Opendak M. Neurobiology of infant fear and anxiety: impacts of delayed amygdala development and attachment figure quality. Biol Psychiatry. 2021;89(7):641–50. https://doi.org/10.1016/j.biopsych.2020.08.020.
11. Chapman D, Dube S, Anda R. Adverse childhood events as risk factors for negative mental health outcomes. Psychiatr Ann. 2007;37:359–64. https://doi.org/10.3928/00485713-20070501-07.
12. Felitti VJ, Anda RF, Nordenberg D, et al. Reprint of: relationship of childhood abuse and household dysfunction to many of the leading causes of death in adults: the adverse childhood experiences (ACE) study. Am J Prev Med. 2019;56(6):774–86. https://doi.org/10.1016/j.amepre.2019.04.001.
13. Porges S. Neuroception: a subconscious system for detecting threats and safety. Zero Three. 2004;24(5):19–24. ISSN: ISSN-0736-8038. https://www.zerotothree.org/resources/series/journal-archive. Accessed 4 Dec 2020.
14. Damasio A, Carvalho G. The nature of feelings: evolutionary and neurobiological origins. Nat Rev Neurosci. 2013;14:143–52. https://doi.org/10.1038/nrn3403.
15. Oakwater H. Want to adopt: how to prepare yourself to parent a child from the care system. London: FAB Parents; 2019. ISBN-10 1913040003. ISBN-13 978-1913040000.
16. Baron-Cohen S. Zero degrees of empathy: a new theory of human cruelty. London: Penguin, Random House; 2011. ISBN: 9780718193348.
17. Yirmiya N, Sigman MD, Kasari C, Mundy P. Empathy and cognition in high-functioning children with autism. Child Dev. 1992;63:150–60. https://doi.org/10.1111/j.1467-8624.1992.tb03603.x.
18. Baron-Cohen S. Theory of mind and autism: a review. Int Rev Res Ment Retard. 2000;23:169–84. Academic Press. ISSN 0074-7750. ISBN 9780123662231. (http://www.sciencedirect.com/science/article/pii/S0074775000800105). https://doi.org/10.1016/S0074-7750(00)80010-5.
19. Sivaratnam C, Newman L, Rinehart N. Emotion-recognition and theory of mind in high-functioning children with ASD: relationships with attachment security and executive functioning. Res Autism Spectr Disord. 2018;53:31–40, ISSN 1750-9467 (http://www.sciencedirect.com/science/article/pii/S1750946718300771). https://doi.org/10.1016/j.rasd.2018.05.005.
20. Rasmussen C, Wyper K, Talwar V. The relationship between theory of mind and executive functions in children with fetal alcohol spectrum disorders. Can J Clin Pharmacol. 2009;16(2):e370–80. https://jptcp.com/index.php/jptcp/article/view/561/489.
21. O'Connor MJ, Kogan N, Findlay R. Prenatal alcohol exposure and attachment behavior in children. Alcohol Clin Exp Res. 2002;26:1592–602. https://doi.org/10.1111/j.1530-0277.2002.tb02460.x.
22. Mukherjee PC, Norgate S, Price A. Neurodevelopmental outcomes in individuals with fetal alcohol spectrum disorder (FASD) with and without exposure to neglect: clinical cohort data from a national FASD diagnostic clinic. Alcohol. 2019;76:23–8. ISSN 0741-8329. https://doi.org/10.1016/j.alcohol.2018.06.002.
23. Oakwater H. Bubble wrapped children: how social networking is transforming the face of 21st century adoption. London: MX Publishing; 2012. ISBN-10 1780920970. ISBN-13 978-1780920979.

Children Who Are Looked After: The Impact of FASD

17

G. A. Gregory

Chapter Highlights

- Identification and prevalence of fetal alcohol spectrum disorder (FASD) in the care system.
- How to overcome barriers to diagnosis for children in care.
- Developing resilience.
- Planning for adulthood after leaving care.

Children in Care (CIC) are a particularly vulnerable group of children and young people who have an increased risk of prenatal drug and alcohol exposure, and therefore, of fetal alcohol spectrum disorders, FASD [1]. Health assessments offer the ideal opportunity for identification and assessment of neurodevelopmental disorders, but unless the referring social worker and the health professionals, carrying out the assessments, have sufficient knowledge and recognition of the risk factors and presenting difficulties, this is a missed opportunity for the child. Because placements in care are not all long term, and disruption can occur, it is vital that appropriate assessment is offered as early as possible, and that interventions for the child can be implemented as early as possible. This may be complicated as not all children exposed to alcohol prenatally will develop FASD, and children may not present with symptoms suggestive of a disorder before they are of school age. Neurodevelopmental difficulties may not even become apparent until the child is around 8 or 9 years old. It is important that carers for children who have, or might have FASD, are informed as how best to support the child in the long term. Services for children and health professionals need to develop pathways for assessment and

G. A. Gregory (✉)
Integrated Paediatric and Neuro-development Service, Cambridgeshire and Peterborough NHS Foundation Trust, Peterborough, UK
e-mail: ges.gregory1@nhs.net

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_17

management of children in care with FASD, and to be able to complete these in a timely way. Once identified, there needs to be advanced planning in preparation for affected young people Leaving Care.

17.1 Children in Care

The definition of looked-after children (LAC) or Children in Care (CIC) is found in the Children Act 1989. A child is looked after by a local authority if a court has granted a Care Order to place a child in care, or a council's children's services department has cared for the child for more than 24 h. (A local authority in the United Kingdom, UK, is an organisation that is officially responsible for all the public services and facilities in a particular area). Any child under the age of 18 years can potentially be placed in care, and the circumstances which result in a child or young person being first placed in care are likely to be somewhat different at different ages.

In the United Kingdom, the local authority can be awarded joint parental responsibility for the child or young person by the Family Court. A common outcome is a placement with foster carers. In a small number of cases, the child may stay with parent(s), but on a Care Order. Older children and young people over the age of 16 years may be placed in some form of supported lodging. In 2012, the number of children living in homes, hostels or secure accommodations was about 9% of the total number of children looked after. In 1978, that figure was 32%. The emphasis on using foster placements wherever possible has meant that children's residential homes have increasingly come to be used principally for older children with more serious difficulties [2, 3]. By definition, these young people are more likely to have multiple adverse risk factors including neglect, child abuse, parental substance misuse and mental health difficulties, and so neurodevelopmental disorders and FASD are more likely [4, 5].

17.2 Prevalence of FASD for Children in Care

Knowledge and prevalence of FASD in communities is outlined in the section on Public Health and Policy in this book.

It is no surprise that Children in Care have a higher rate of FASD than in a general population, but to what extent has been less clear. A meta-analysis of published and unpublished studies, calculated the pooled prevalence estimates of diagnoses of FAS and FASD for children and young people, was reported on in 2013. This gave an overall estimate for FAS at 6% and for FASD at 17% for children in a number of different care settings [6]. This analysis drew on information from a number of studies from a total of eight countries (Brazil, Chile, Canada, Israel, Russia, Spain, Sweden and the United States). The prevalence of FASD diagnosis ranged from 0% to 52%. This data did not include any information on children from the UK.

There are no prevalence studies in the UK; however, an audit of children in care in Peterborough, published in 2015, identified that this was a vulnerable population with a high risk for FASD in the Looked After Children of 27%, and that for those children being placed for adoption, the risk was much higher at 75% [1]. Children placed for adoption are usually younger than the population of children in care, and it is possible that maternal drug and alcohol misuse may be one reason for babies, in particular, being removed from their mothers' care.

Ethnicity and country of birth are also factors linked to a higher risk of FASD. In one study, 31% of children from Poland adopted by Dutch families had a diagnosis of FASD, while a further 21% were suspected as having the condition, but not yet diagnosed at the time of the study [7]. In Sweden, 52% of children adopted from Eastern European orphanages had diagnoses of FAS, partial FAS and alcohol-related neurodevelopmental disorders [8].

Local authorities in England have only been required to collect data on ethnicity of children since 2000/2001. A social care study in 2011 looked at reasons for children of minority ethnic backgrounds coming into care and then placed for adoption. Parental drug and alcohol misuse was one of three main factors, and this was almost entirely a problem for white and mixed ethnicity mothers. 30% of mixed ethnicity children had been born with neonatal abstinence syndrome (NAS) or had signs of FASD [9].

17.3 Health Assessments

There is a statutory requirement that all children in care have an initial health assessment within 20 working days from the date of initially being in care and, following this review, health assessments every 6 months if under 5 years of age and ever year if over 5 years of age [10]. This provides the ideal opportunity for recognition and assessment of FASD in this high-risk group. The social worker referring for the initial assessment needs to provide some information on why the child is in care and at this early stage it is likely that any information on prenatal drug and alcohol exposure is going to be easier to ascertain and report on [11]. If this is not provided, then the assessor should make enquiries in regards to the possibility of alcohol drunk in pregnancy, and if there is a history of exposure, then quantity and timing in relation to gestation age of the fetus are important pieces of information in order to predict the pattern of risk to the child [12, 13]. Any health professional carrying out assessments should have adequate training and knowledge of FASD sufficient to recognise when a further detailed assessment is required.

As noted previously, not all children who have had exposure will develop FASD, and in many cases where there are no facial features or growth deficiencies, a child may appear to be unaffected until into primary school after which neurodevelopmental difficulties may become more apparent [14, 15]. Professionals and carers need to be aware that lack of any concerns for the child at an early age may not necessarily mean there will not be a longer-term problem. Chasnoff et al. collected data on 547 children in care or adopted who underwent detailed multidisciplinary

assessment. Over 80% had a missed diagnosis of FASD, while of those who had a diagnosis made previously, it was incorrect in 6.4%. He noted that 13.5% had received a correct diagnosis [16].

Children are placed in care for a variety of adverse health risk factors. Many will have had recent significant neglect, abuse, both emotionally and physically and/or sexual abuse. Attachment difficulties are common and children may take some time to adjust to a new care environment. This may be initially complicated by various different contact arrangements with birth families stipulated by the courts. As such, even if there is a clear history of prenatal alcohol exposure, it may not be appropriate, or in the interest of the child, to do a detailed assessment for FASD until some stability is in place for the child. Documentation of the risk factors in the child's records, therefore, is vital, and records should also record the source of the information, in case of a dispute at a later date.

Siblings of children in care may themselves be taken into care as a protective measure against risk of abuse and neglect, and in these cases, the information known for older siblings may be very useful in identifying risks. Additionally, a younger child in care who has been identified as having FAS or FASD should trigger an enquiry into the risk factors for older siblings. Patterns of alcohol misuse in women typically show increasing alcohol misuse over time; however, it is also known that some women who may have binged during one pregnancy may not do so in future pregnancies once they are aware of the risks.

Health assessments for CIC are carried out by a number of different professionals, but the statutory guidance dictates that the Initial Health Assessment should be done by a medical practitioner. It is an ideal opportunity to gain as much history about the prenatal period as possible, as often mothers will attend this appointment especially for newborn babies. It is important that this is done sensitively with emphasis that it is for the purpose of ensuring the child's well-being, and any future health and developmental needs can be met. It is also an opportunity to ensure that the mother is aware of risks of prenatal alcohol exposure for any future pregnancies. At each health assessment, a Health Action Plan (HAP) is produced for each child seen, which lists actions that need to be completed and who is responsible for these. It is prudent that an action in this list would be for consideration of any difficulties suggestive of FASD at the subsequent health assessment in order that this is considered each time the child is seen.

17.4 The Barriers to Making a Diagnosis for Children in Care (CIC)

A meeting of health professionals in the UK in 2011, facilitated by a national FASD charity, the FASD Trust, agreed that—in the absence of any alternative UK guidance—the Canadian guidelines on diagnosis, as set out by Chudley et al. in 2005, should be adopted as the most appropriate for use in the UK [17].

For children in care where FASD is suspected and further assessment is warranted, the pathway for assessment will need to be considered at a local level. It is

likely to necessitate a separate appointment to the health assessment in order to complete a comprehensive assessment.

The clinical assessment should be done by a paediatrician who has knowledge of fetal alcohol spectrum disorders and should include

- Detailed history of timing and quantity of prenatal exposure
- Highlight if other siblings could be at risk or affected
- Detailed physical examination including growth
- Developmental assessment
- Screening assessment for ADHD, ASD and other mental health difficulties
- Assess need to refer for Speech and Language assessment
- Assess need to refer for occupational therapy assessment
- Investigation to exclude other causes of difficulties or coexisting conditions

The child may then require further assessment of neurodevelopmental comorbid conditions as appropriate [18, 19].

If indicated, access to an occupational therapy assessment for coordination difficulties and sensory processing disorders should be available, although the exact arrangements for this may vary between different NHS trusts. A Speech and Language Therapy assessment may be required for the child, but services for assessment for a child with no apparent difficulty may not be available in all areas due to limited resources. Assessment of verbal comprehension may best be tested as part of a cognitive assessment. Consideration may be given to the use of screening tools/checklists which can be used by non-specialist professionals (see Chap. 14). However, further research is needed in this area.

Children in Care have the advantage in that they have access to Specialist Psychology Services because of the high level of emotional and behavioural difficulties in this group of children and young people. In some cases, the psychology services may also be able to carry out detailed cognitive assessments and assessments of adaptive behaviours, and daily living skills. This can be a problem in areas where resources are stretched, which can be a barrier for these assessments for children who may not meet the referral criteria for local child and adolescent mental health or neurodevelopmental services. Detailed Executive Functioning testing also may not be available, although this is crucial to the assessment of children with the complex neurodevelopmental difficulties associated with FASD, as these tests are not routine in assessment for other conditions, such as ADHD or Autistic Spectrum Disorder. Consideration of where and how children can have FASD assessments completed needs to be addressed as more paediatricians, child psychiatrists, social workers and carers of children recognise the possibility of FASD and will request appropriate comprehensive assessment. The age at which these detailed executive functioning assessments can be carried out with validity must also be considered, as a child needs to be able to be functioning at an appropriate cognitive level in order for some testing, and in most cases, this would not be considered until after 6 years of age. This can itself be an issue when social care wish to have assessments completed as early as possible if considering placing a child for permanency, such as

adoption, and delaying assessment can be seen as a barrier to being able to provide full information to the prospective adopters.

A further obstacle to starting or completing assessments in a timely manner is that children quite often change placements and have a change of carer, move schools or fail to engage in assessment. Another barrier is that the catchment area for CIC is often wider than that of local community paediatric, neurodevelopment and mental health services. That means, a child may have health assessments done in one area, but will not have access to the other local services there, which can cause confusion in who is responsible for what part of an assessment, which can result in a delay in bringing everything together in a comprehensive report with appropriate feedback and advice on completion.

Whilst international structures differ, there is limited access in the UK for specialist assessment, if this cannot be provided at a local level. Consideration of development of specialist assessment pathways for all children, but in particular, this vulnerable population of children and young people, must be a consideration for all NHS trusts in the future.

There are now a few specialist assessment centres in the UK for FASD. 'The Fetal Alcohol Spectrum Disorders Clinic' in Surrey was the first one set up in England, offering assessment for children over the age of 6 years into adulthood, and more recently 'The Centre for FASD' in East Anglia does offer comprehensive assessment for children. More are likely to develop. Although commissioning assessments through this service may be an issue, it offers a solution for those cases where assessment is not otherwise possible, or the complexity of the case merits this approach. Local authorities and doctors for Children in Care in the UK should be aware of these services, alongside consideration of continuing local development. The Scottish Intercollegiate Guidelines Network (SIGN) has produced a National Clinical Guideline in January 2019, and the National Institute of Clinical Excellence (NICE) released its draft quality standard on fetal alcohol spectrum disorder (FASD) in August 2020 for further public consultation. This will require all local commissioning groups in the UK to pay attention to the development needs of this group.

17.5 Adverse Childhood Experiences

Adverse Childhood Experiences (ACEs) are an increasing international concern. ACEs are stressful experiences occurring during childhood that directly harm a child (neglect, emotional, sexual or physical abuse) or affect the environment in which they live (growing up in a house with domestic violence, parental mental illness, parental separation, parental substance misuse and a relative who has been incarcerated). There is a growing body of evidence that our experiences during childhood can affect health throughout life. Children who experience stressful and poor quality childhoods are more likely to adopt health-harming behaviours during adolescence, which can themselves lead to mental health illnesses and diseases such as cancer, heart disease and diabetes later in life. This is not just a concern for health as experiencing ACEs means individuals are more likely to perform poorly in school

and to more likely to be involved in crime. The risk is higher for those who have experienced a higher number of ACEs and significantly higher with four or more ACEs [20].

Studies have now been done that show a relationship for developmental delay for children at 2 years of age [21], and also child outcomes at 3 years of age for internalising and externalising behaviours [22], particularly when the mother has experienced three or more ACEs.

The ACEs identify living in a household with a problem drinker or alcoholic, and living with anyone using illegal drugs or abusing prescription medications. They do not identify exposure to alcohol in the prenatal period, and it is therefore unclear to what extent the difficulties of any children or young people affected by ACEs may also have FASD. Most studies do not specify how many children affected by ACEs are or have been in the care of the local authority, although the majority of children in foster care have a history of maltreatments which collectively includes ACEs [23]. There is emerging evidence that the two may well act in separate manners and that much more evidence is needed in this area to allow us to delineate these effects. We need to be mindful that when considering assessments for FASD that the impact of ACE's and the long-term health difficulties associated with these, in addition to any prenatal alcohol exposure, need to be taken into consideration as part of any assessment.

17.6 Attachment

There has been a tendency for Children in Care presenting with longstanding behavioural difficulties to be labelled as having attachment difficulties. While this may the case for many children, it has perhaps been historically overused instead of considering if neurodevelopmental disorders may be the main cause or a confounding factor. While it may seem obvious that attachment disorders are not unexpected for a child placed in care, following longstanding neglect and abuse and then having multiple placements when in care, it may not seem to be as clear-cut for a baby placed with fosters carers from birth and then with adopters at an early age. Adopters will rightly challenge the attachment theory as to an adequate explanation for challenging behaviours when the child is older.

The differential diagnosis of a neurodevelopmental disorder must be considered. Children with FASD may not understand the consequences to their actions and the impact this may have on others. They also have difficulties with social interaction, although seemingly appearing very engaging and chatty when young, and some may also fulfil the criteria for an ASD when older.

The Coventry Grid [24] is an attempt to summarise the differences between the behaviour of children with Autistic Spectrum Disorder and those with significant attachment problems. It is based upon clinical work with children rather than research. There is an emerging body of research which is clarifying the range of social and communication difficulties seen in children and young people who have experienced early adversity [25, 26]. In practice, this is a useful tool for

consideration when starting to unpick reasons for why children with suspected FASD behave as they do, however, does not replace the need for a comprehensive neurodevelopment assessment.

17.6.1 Cognitive Ability and Adaptive Functioning

A diagnosis is especially important for children in care particularly in regards to understanding the child's needs, but is also often a key to getting appropriate long-term support in education and other services. The diagnostic process should include detailed assessment of cognitive ability, and a measure of executive functioning and or adaptive functioning. Children with FAS and FASD are described as having both primary and secondary disabilities.

The primary disability refers to the brain damage from prenatal exposure that has a direct effect on development and cognition. Streissguth [27] reported on 473 individuals with FAS and Fetal Alcohol Effects, FAE. The range of IQ in those with FAS was from 29 to120 with a mean of 79. For FAE the range was from 42 to142 with a mean IQ of 90. The functional abilities, however, were lower than expected for the IQ measurement. In reality, this means that although a large number of children with FASD have lower IQ levels than average, most will be above the cut-off level of IQ 70 that would enable support from learning Disability Services. Those with borderline IQ levels may get some additional support if it is clear that their ability to function on a daily basis is limited, however, many will not. Those with FASD (FAE) have, on average, an even higher cognitive ability on testing and will be further disadvantaged in this respect [28]. Further discussion around the cognitive presentation in people with FASD can be found in Chap. 14 (Table 17.1).

Secondary Disabilities are those not present at birth, but occur as a result of the primary disabilities. Secondary disabilities can presumably be prevented or lessened by better understanding and appropriate interventions. Streissguth reported on secondary disabilities ascertained from life history interviews of 415 individuals with FASD (aged 6–51 years) using 450 questions [27]. Mental Health Problems were experienced by 94% of the full sample. During childhood, 60% of children with FASD had ADHD. During adulthood, most adults with FASD had clinical depression. Disrupted School Experience (suspension or expulsion or drop out), was experienced by 43% of children of school age. By the time students with FAE reached adulthood, the rate of disrupted school experience peaked at 70%. Trouble with the Law (involvement with police, charged or convicted of crime) was

Table 17.1 Understanding the occurrence of secondary disabilities in clients with Fetal Alcohol Syndrome (FAS) and Fetal Alcohol Effects (FAE)

Diagnosis and number of cases	IQ Range	Mean IQ
FAS 178	29–120	79
FAE 295	42–142	90

Final Report, Centers for Disease and Control Prevention. August 1996

Table 17.2 Understanding the occurrence of secondary disabilities in clients with Fetal Alcohol Syndrome (FAS) and Fetal Alcohol Effects (FAE)

Secondary disabilities	≤12 years of age	≥12 years of age
Mental health problems (reported problems or treatment with counsellor or psychotherapist: Depression, suicidal intention, psychosis, ADHD, panic attacks)	60% ADHD	94% overall Depression 50% Suicide threats 43% and attempts 23%
Disrupted school experience (suspension or expulsion or drop out from education)	43%	70%
Trouble with the law (involvement with police, charged or in trouble for criminal activity)	42%	60%
Confinement (inpatient treatment for mental health, alcohol/drug problems, or incarceration for crime)	23%	Over 60%
Inappropriate sexual behaviour (repeated problems with sexuality or sentenced to sex offender program)		45% FAE: Men 65%
Alcohol and or drug problems (alcohol abuse, drug abuse, participation in alcohol / drug treatment programs)	–	30% FAE: Women 70% FAE men 53%

Final Report Centers for Disease and Control Prevention. August 1996

experienced by 42% of those in the study, and by about 60% of those age 12 and over. Confinement (inpatient treatment for mental health, alcohol/drug problems or incarceration for crime) was experienced by 60% of those aged 12 and over. 20% of adults had been confined for substance abuse treatment. Inappropriate sexual behaviour including promiscuity was reported in 45% of those aged 12 and over, and 65% of adult males. And alcohol and/or drug problems were experienced by 30% of individuals aged 12 and over. Of the adults with FAE, 53% of males and 70% of females experienced substance abuse problems. This was more than five times that of the general population (Table 17.2).

17.7 Risk of Exploitation, Sexual Offending and Criminal Behaviours

Young people with FASD need guidance and support as they will lack the ability to make good choices, self-regulate behaviours and understand consequences to their actions as a direct result of the primary injury to their brain. Secondary disabilities are common and failure to support young people in the long term makes it inevitable that many will end up in serious trouble. Even when work is done after they have offended or been abused, they may revert to the same behaviours once intervention for support is stopped, as the primary disability remains unaltered and they remain particularly vulnerable to exploitation. It would make sense that understanding of what FASD means to a child and young person, and subsequently appropriate support and supervision to prevent secondary disabilities and entrenched patterns of behaviours must give a better outcome for the young person [28].

17.8 Leaving Care

It is not just enough to meet the needs of children while they are in care, but important to begin planning for life for those leaving care. Every year, increasing numbers of children who have been identified with FASD will be transitioning out of care and into the community. The shift to independence is difficult for a young person with any disability. This is due in part to the significant differences of services available for children and adults. The move to independence for young people with FASD is further complicated by the nature of their disability. They are often not eligible for services related to cognitive impairments because their level of intellectual functioning is above the eligibility criteria. There are few, if any, adult services directly related to FASD. As adults, their disability tends to be invisible, but their behaviour can still present many challenges. Long-term planning for children with FASD needs to include special attention to their transition into adulthood, and conceptualisation and consideration of lifespan planning should begin in childhood [29, 30].

Success for a young person leaving care directly relates to experiences in stability of placement, stability in education and development of good attachment to a significant mentoring adult. A significant number of young people who are in care over the age of 16 years are placed in semi-independent living arrangements with a lower level of supervision than in a foster placement. Managing finances, organising meals and making good choices in daily activities as well as friendships are all challenging when you have FASD, and the vulnerability of this particular age group cannot be underestimated.

In practice, this means that the stresses they will face when attempting to manage their lives without the support structures around them are likely to have a huge impact on long-term mental health. Transition plans need to address the particularly problematic outcome areas of employment, income, housing, social relationships and mental health.

17.9 Resilience

The literature identifies that for children with FASD, stability in placements and education, and a lasting relationship with at least one adult are important for a successful transition when leaving care. It is known that challenging behaviours as seen in FASD are linked to disruption of placements, especially for adolescents in teenage years. This, along with multiple changes of social workers, carried the greatest potential for disruption of schooling and planning for independence. The type of placement appeared to be an important contributor to stability. Recognising that stability is critical to children in care, it is important to enhance the ability of foster parents to deal with the particular issues of adolescence in this group so that long-term foster placements do not break down [31, 32]. It is also important that some support is available for those adoptive families to enable them to cope with the challenges they may face early on rather than when at a breaking point.

Teenagers must have stable placement from which to launch themselves towards adulthood. Many children leaving care reconnect with their biological families. The conditions under which these children enter care is most often related to the conduct of their parents, which will include alcohol and/or drug use, and as such, these young people are very vulnerable and will risk themselves repeating the cycle.

The message for Social Care, adoption agencies and all professionals working with young people with FASD is that stability of care when growing up aids resilience in coping as an adult, and also that a structured environment and support with an advocate or mentor is needed in the long term.

Practice Points

- Children in Care and those placed for adoption are at a high risk for FASD
- There is a risk of missed diagnosis or misdiagnosis of FASD for Children in Care
- ACEs need to be taken into consideration when formulating assessments for FASD
- Local pathways for assessment of Children in Care need to be established
- Specialist services to support children, carers and families in the UK still need to be developed
- There must be recognition of the particular risks for vulnerable young people with FASD Transition planning and leaving care plans for those with FASD need careful consideration
- In FASD, stability of care when growing up aids resilience in coping as an adult

References

1. Gregory G, Reddy V, Young C. Identifying children who are at risk of FASD in Peterborough: working in a community clinic without access to gold standard diagnosis. Adopt Foster. 2015;39(3):225–34.
2. Popova S, Lange S, Burd L, Rehm J. Canadian children and youth in care: the cost of fetal alcohol spectrum disorder. Child Youth Care Forum. 2014;43:83–96.
3. Berridge D, Biehal N, Henry L. Living in children's residential homes. Department for Education: Research Report DFE-RR201. 2012. http:/www.eduction.gov.uk/publications.
4. Herrick K, Hudson L, Burd L. The elephant in the cradle; fetal alcohol spectrum disorders. Zero to Three. January 2011.
5. Streissguth AP, Brookstein FL, Barr HM, Sampson PD, O'Malley K, Young JK. Risk factors for adverse life outcomes in fetal alcohol syndrome and fetal alcohol effects. J Develop Behav Pediatr. 2004;25(4):228–38.
6. Lange S, Shield K, Rehm J, Popova S. Prevalence of fetal alcohol spectrum disorders in child care settings: a meta-analysis. Pediatrics. 2013;132(4):e980-95.
7. Knuiman S, Rijk CHAM, Hoksbergen RAC, Van Barr AL. Children adopted form Poland display a high risk of fetal alcohol spectrum disorders and some may go undiagnosed. Acta Paediatr. 2015;104(2):206–11.

8. Landgren M, Svensson L, Strömland K, Grönlund MA. Prenatal alcohol exposure and neurodevelopmental disorders in children adopted from Eastern Europe. Pediatrics. 2010;125(5):e1178-85.
9. Selwyn J, Wijedesa D. Pathways to adoption for minority ethnic children in England – reasons for entry to care: child and family. Soc Work. 2011;16:276–86.
10. Department for Education and Department of Health. Promoting the health and wellbeing of looked-after children, Statutory Guidance. March 2015.
11. Clarke M. What social workers need to know about foetal alcohol spectrum disorders. TACT Fostering & Adoption November 30, 2015 in Child safeguarding, Children http://www.communitycare.co.uk.
12. Mukherjee R, Cook PA, Flemming KM, Norgate SH. What can be done to lesson morbidity associated with fetal alcohol spectrum disorders? Arch Dis Child. 2016;0:1–5.
13. BMA publication. Alcohol in Pregnancy. Preventing and managing fetal alcohol spectrum disorders. June 2007 (updated February 2016).
14. O'Leary CM, Taylor C, Zubrick SR, Kurinczuk JJ, Bower C. Prenatal alcohol exposure and educational achievement in children aged 8-9 years. Pediatrics. 2013;132(2):e468-75.
15. Burd L, Cohen C, Shah R, Norris J. A court team model for young children in foster care: the role of prenatal alcohol exposure and fetal alcohol spectrum disorders. J Psychiatry Law. 2011;39(1):179–91.
16. Chasnoff I, Wells AM, King L. Misdiagnosis and missed diagnosis in foster and adopted children with prenatal alcohol exposure. Pediatrics. 2015;135(2):264–70.
17. Chudley AE, Conry J, Cook JL, et al. Fetal alcohol spectrum disorder: Canadian guidelines for diagnosis. Can Med Assoc J. 2005;172:s1–s21.
18. Young S, Absoud M, Blackburn C, Branney P, Colley B, Farrag E, Fleisher S, Gregory G, Gudjonsson GH, O'Malley KD, Plant M, Rodriguez A, Ozer S, Takon I, Woodhouse E, Mukherjee R. Guidelines for the identification of individuals with ADHD and associated FASD based on expert consensus. BMC Psychiatry. 2016;16:1–14.
19. Popova S, Lange S, Shield K, Mihic A, Chudley AE, Mukherjee RAS, Bekmuradov D, Rehm J. Comorbidity of fetal alcohol spectrum disorders: a systematic review and meta-analysis. Lancet. 2016;387:978–87.
20. Public Health Wales NHS Trust. Welsh adverse childhood experiences (ACE's) study. www.nationalarchives.gov.uk/doc/open-government-licence/version/3/. ISBN 978-1-910768-23-5.
21. Folger AT, Eismann EA, Stephenson NB, Shapiro RA, Macaluso M, Brownrigg ME, Gillespie RJ. Parental adverse childhood experiences and offspring development at 2 years of age. Pediatrics. 141(4):e20172826. (ISSN Numbers: Print, 0031-4005; Online, 1098-4275). https://doi.org/10.1542/peds.2017-2826.
22. McDonalda SW, Madiganc S, Racinec N, Benziesd K, Tomfohrc L, Tougha S. Maternal adverse childhood experiences, mental health, and child behaviour at age 3: the all our families community cohort study. Prev Med. 2019;118:286–94.
23. Jamora MS, Brylske PD, Martens P, Braxton D, Colantuoni E, Belcher HME. Children in foster care: adverse childhood experiences and psychiatric diagnoses. J Child Adolesc Trauma. 2009;2:198–208.
24. The Coventry Grid Version 2 (Modified Jan 2015) Differences between Autistic Spectrum Disorder (ASD) and attachment problems based upon clinical experience and observations.
25. Shonkoff JP, Garner AS, Siegel BS, Dobbins MI, Earls MF, Garner AS, McGuinn L, Pascoe J, Wood DL. The committee on psychosocial aspects of child and family health, committee on early childhood adoption and dependent care, and section on developmental and behavioural paediatrics. Pediatrics. 2012;129:e232.
26. Fisher PA, Beauchamp KG, Roos LE, Noll LK, Flannery J, Delker BC. The neurobiology of intervention and prevention in early adversity. Annu Rev Clin Psychol. 2016;12:331–57.
27. Streissguth AP, Barr HM, Kogan J, Bookstein FL. Understanding the occurrence of secondary disabilities in clients with fetal alcohol syndrome (FAS) and fetal alcohol effects (FAE). Final report centers for disease and control prevention grant no R04/CCR008515 August 1996.

28. Peadon E, Rhys-Jones B, Bower C, Elliott EJ. Systematic review of interventions for children with Fetal alcohol Spectrum disorders. BMC Pediatr. 2009;9:35.
29. Temple V, Shewfelt L, Tao L, Casati J, Klevnick L. Comparing daily living skills in adults with fetal alcohol spectrum disorder (FASD) to an IQ matched clinical sample. J Popul Ther Clin Pharmacol. 2011;18(2):e397–402.
30. Zarnegar Z, Hambrick EP, Perry BD, Azen SP, Peterson C. Clinical improvements in adopted children with fetal alcohol spectrum disorders through a neurodevelopmentally informed clinical intervention: a pilot study. Clin Child Psychol Psychiatry. 2016;21(4):551–67.
31. Fuchs D, Burnside L, Marchenski S, Mudry A. Transition out-of-care: issues for youth with FASD. Ottawa: Centre of Excellence for Child Welfare; 2008.
32. Joy S. Holloway. Outcome in placements for adoption or long term fostering. Arch Dis Child. 1997;76:227–30.

International Adoption and Undiagnosed FASD

18

Mary Mather

Chapter Highlights

- An overview of intercountry adoption.
- The differences between domestic and international adoption in the UK.
- Summary profiles of people with FASD in the adoption setting.

Any child adopted from any country in the world could have been exposed to alcohol prenatally. Intercountry adopters are advised to obtain comprehensive, local information about alcohol misuse in pregnancy in their chosen country. This information is frequently difficult to obtain and may not be reliable. Diagnosis in intercountry adopted children is difficult because the history of alcohol exposure is usually unknown. Growth delays, learning, emotional and behavioural problems in institutionalised children may have other causes. Even if the information is incomplete, an intercountry adopted child should not be denied a diagnosis because early diagnosis improves the long-term prognosis. The preparation of intercountry adopters for the possibility that their child may have been exposed to alcohol is essential. Although not currently recommended in most of the UK, all intercountry adopted children, irrespective of their country of origin, deserve to have a comprehensive health assessment and screening after adoption.

18.1 Introduction

Alcohol is the most globally available teratogen on our planet. In most of the world, drinking alcohol is a perfectly legal activity, which humans have enjoyed for thousands of years. Whilst alcohol has long been associated with social disorder and

M. Mather (✉)
IAC - The Centre For Adoption, London, UK
e-mail: marymather@doctors.org.uk

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_18

violence, it is only in the last 50 years that the consequences of drinking in pregnancy have been recognised. Prenatal exposure to alcohol is now accepted as the commonest and most preventable cause of mental retardation in the world. In every country where women drink in pregnancy, children are born with fetal alcohol spectrum disorders (FASD). Despite strenuous public health campaigns in most countries, women continue to drink in pregnancy.

To date, most countries do not have population prevalence data for either alcohol use in pregnancy or fetal alcohol syndrome (FAS). A comprehensive epidemiological survey in 2017 estimated that globally about 10% of women drink in pregnancy and 1 in 67 women delivers a child with FAS. This means that on average, about 15 of every 10,000 live births worldwide will have FAS, translating into 119,000 affected children born globally every year [1]. The effects of exposure range from devastating physical and learning disabilities to more subtle brain damage causing impulsive behaviour, violence and criminality.

18.2 Intercountry Adoption and The Hague Convention (1993)

Apart from a small number of expert practitioners, intercountry adoption (ICA) is poorly understood in the United Kingdom. There are no accurate UK statistics, little research and widespread professional prejudice against the practice. In England and Wales, the Department of Education (DfE) keeps records of applications to adopt overseas but not the numbers of children adopted. Not all applications result in adoption and adopters may adopt more than one child. In the 6 years between January 2002 and December 2008, there were thought to be 2232 applications [2]. These figures are only a general guide, they cannot be validated and exclude applications to Hague Convention countries from residents in Wales, Scotland, Northern Ireland and the Isle of Man. The number of intercountry adopted children living in the UK, the outcomes for the children and the number affected by exposure to alcohol are unknown. In comparison to the growing body of findings from the USA, Scandinavia and the Netherlands, intercountry adoption in the UK is a very neglected area.

There is a widely held perception that ICA is primarily a humanitarian response to poverty, war or social crises because the practice started in North America as a philanthropic response to the devastation following World War II and initially involved children moving from orphanages in Europe to North America [3]. As a more global phenomenon, it has grown rapidly since 1990 when the world first became aware of the desperate plight of children living in Romanian orphanages.

In affluent societies, increasing demands for children, particularly babies, coupled with a marked decrease in domestic adoption have fuelled this growth. The internet has also increased public awareness about the availability and unmet needs of children in developing nations from where the vast majority of intercountry adoptions now originate. Altruism is however only part of the story and does not explain the worldwide trends and clusters of children from various countries.

Over the last 20 years in the UK, adoption numbers from one country have peaked and then slowly declined to be replaced by adoption from another country. The process is driven by the availability of children and the procedures and regulations in both the sending and receiving countries. In the UK over the past 20 years, Romanian orphans were followed by girls from China, then Russian orphans and currently the numbers of children from Ethiopia, India and Pakistan are increasing [4]. In line with global trends, the overall number of internationally adopted children in the UK is declining, the unofficial DfE figure for 2016 was 60 applications.

Alongside the growth in intercountry adoption, the international community has made very significant attempts to control the process. The underpinning ethical principles were first introduced in Article 21 of the 1989 UN Convention on the Rights of the Child [5]. The detailed implementation was left to The Hague Convention on the Protection of Children, which was agreed in the aftermath of the Romanian crisis in 1993 [6].

The Hague Principles seek to put the best interests of children first. Contracting states must ensure that the abduction, sale and trafficking of children is prevented. Children must be protected against the risks of illegal, irregular and ill-prepared adoptions abroad. The child must have been freely given up for adoption. No financial inducements of any kind can be made to the birth parents. Efforts must have been made to place the child in a family in their home country. The receiving state must confirm that the adopted child will be given permanent residence and that potential parents have been comprehensively assessed as suitable adopters. The UK ratified The Hague Convention in 2013, and at the time of writing it is one of the 97 countries that have now ratified or acceded to the Convention [7].

Few adopters appreciate that the status of their chosen country under The Hague Convention will also affect the health assessment which their child receives in the UK after adoption. Children from convention countries come into the UK already legally adopted, whereas children from non-convention countries must be legally adopted in the UK. Only this latter group has local authority surveillance of the placement which includes medical reports. Apart from Northern Ireland, there is no statutory requirement for intercountry adopted children to have a health assessment after placement. Although the child is entitled to both primary and secondary National Health Service (NHS) care, there are few GPs or paediatricians with expert knowledge of the unique problems of intercountry adopted children. As one mother reported ‘I have had to become the expert in my child’s health, because out of 10,000 patients in our GP practice my child is the only one adopted from an orphanage abroad’.

18.3 How Intercountry Adoption Differs from Domestic Adoption

The prospective intercountry adopter must firstly choose a country from which to adopt and the availability of children changes rapidly. A country accepting applicants could stop at any time and a country that did not have an adoption programme

may introduce one. Britain has recently suspended adoption from certain countries. Other countries only allow their own citizens to adopt. Each country has unique eligibility criteria, placing restrictions on adopters which do not exist in the UK, such as age limits or marital status or religion. Adopters therefore must become experts in the adoption practices of their chosen country and also make all initial enquiries themselves. Intercountry adopters must be both resilient and committed. When medical reports, travel, accommodation, time off work and legal expenses are included, the minimum cost is at least £20,000, with some paying as much as £50,000. The entire process can take up to 3 years or even longer.

Many intercountry adopters have lived or worked in their chosen country, have family members living there or are aware of other parents who have successfully adopted babies from that country. Most children adopted from abroad are infants or very young children and there is an incorrect public perception that these children are less 'risky' than domestically adopted or older children. Adopters can be overwhelmed by the complexity of ICA and miss the public health risks to their child. They have usually heard about the long-term implications of exposure to alcohol but are frequently more concerned about the impact of institutionalisation on development. Few realise the lifelong implications of alcohol exposure, the range of neurodevelopmental problems their children might experience and the difficulties in establishing the diagnosis in the absence of a reliable maternal history. It is often only during the assessment that they realise the extent of alcohol abuse in their chosen country. For example, South African adopters are often shocked to discover the incidence of FAS/D in the Western Cape and that there are no medical tests which might exclude the diagnosis.

Having chosen their country, intercountry adopters then undergo the same assessment and approval process as domestic adopters [8] (Adoption Agencies Regulations: England and Wales 2013, Scotland 2005). This includes statutory checks, a home study and discussion by an adoption panel. After approval, the prospective adopters' papers are sent to the Department of Education and then placed on a waiting list in the child's State of Origin.

In the UK, there is a specific set of government policies favouring domestic over inter-country adoption. Before taking a decision to proceed with a match, domestic adopters have access to a wealth of information including the medical and social history of the birth family and all the relevant records. They meet the foster carer, the child's social worker, paediatrician and possibly the birth parents. Whilst waiting for an adoptive placement, the child will have regular developmental and medical assessments. Using this information, the adopter's skills as carers can be matched with the child's likely future needs. Adopters are given time to read all the written reports, consider verbal information, see videos and have their questions answered. All the costs are met by the agency.

For the intercountry adopter, the only information they receive comes from the institution abroad. Information is very limited and outside the control of either the adopters or the agency. Increasingly countries, including Hague signatory countries, are utilising online processes to match children to applicants. There is no mechanism for matching the adopter's skills to the child's future needs and they have a

very limited time frame in which to make any decision. It is only after they have made the decision to proceed that they will travel abroad and meet their child.

18.4 The Global Picture of Alcohol Misuse

The incidence of FAS/D in a country mirrors the incidence of alcohol abuse. In the absence of reliable information about an individual child, intercountry adopters can only turn to more generalised statistics. One of the most useful is The World Health Organisation Global Status Report on Alcohol and Health (2014) [9]. This report presents a comprehensive perspective broken down into global, regional and individual country statistics on the consumption of alcohol, patterns of drinking, health consequences and policy responses. The statistics are helpfully broken down by age and gender, the percentage of illegal or unrecorded alcohol use which are not reflected in official statistics and the alcohol content of local beverages. The report however has no data about alcohol consumption in pregnancy or the numbers of alcohol affected children.

A recent systematic review and meta-analysis suggested that the estimated prevalence of alcohol use in pregnancy was the highest in Russia, the United Kingdom, Denmark, Belarus and Ireland. The lowest prevalence was in Oman, Kuwait, The United Arab Emirates, Saudi Arabia and Qatar; all predominantly Muslim countries following a religion which is associated with high rates of abstinence from alcohol [1]. However, the risk exists for every child and although some parts of the world have a lower risk, intercountry adopters need to realise that *any* child adopted from *any* country could have been exposed to alcohol prenatally.

18.5 Diagnostic Challenges in Intercountry Adoption

For the clinician, making a diagnosis of alcohol exposure is not easy when a child has been adopted abroad because so much relevant information is missing.

18.5.1 A History of Alcohol Exposure Before Birth

This information is usually unknown to either the adoptive family or the assessing clinician. The health information currently given to intercountry adopters is totally inadequate, as shown by a recent analysis [10].

IAC-The Centre for Adoption in North West London assesses and approves intercountry adopters from 29 local authorities in London and the Southeast of England and undertakes approximately 85% of UK intercountry adoptions. A retrospective review analysed 120 medical reports from 23 countries written about children matched with adopters from IAC between April 2010 and November 2014. The quality and quantity of medical information varied widely but was generally of poor quality and largely based on a single isolated physical examination done by a

doctor, without access to previous medical information. There was incomplete screening for important medical conditions, inadequate medical histories and virtually no assessment of development [10].

Out of 120 reports, 93 had no information at all about the birth mother, father or extended family. Ninety-three reports had no information about the antenatal period or the mother's lifestyle in pregnancy. Seventy-four reports had no information about the delivery. Seventy-one reports had no recorded birth weight. Ninety-one reports had no neonatal information. Twenty-nine children were being adopted from Russia, a recognised high-risk area. Out of these, only one medical report made the comment 'mild FAS features: philtrum not well expressed' but without further explanation or information. There were no comments about alcohol in the other 119 reports.

The preparation of adoptive families for the possibility that their child may have FAS/D varies, as do their ability and willingness to grapple with the realities of parenting the affected child. ICA is a service that needs experienced medical and social work support. If at the time of matching, the adopters have concerns about the child's medical status, they should where possible request for more information from the agency abroad and should be able to discuss their concerns with an experienced paediatrician. American adoption agencies have recognised this fact for many years and provide more support and more services, both before and after adoption, to the children and their families [11].

18.5.2 Physical and Facial Features

Children exposed to alcohol can have a variety of abnormal physical features. The characteristic facial appearance is usually under-recognised in the neonatal period, even amongst the children of known alcoholics [12] but becomes more apparent in late infancy and early childhood. In a large prospective study, only 50% of those identified at the age of 4 with FAS had been identified at birth. Many of the facial features become less prominent as the children enter adolescence [13]. Recognition is more difficult in Asian children with epicanthic folds and the very young child who may not cooperate with an examiner. In the absence of the birth parents, familial facial features cannot be compared.

Overseas agencies usually send photographs to the potential adopters. FAS cannot be readily diagnosed in this way but a good video and/or photograph can raise suspicions. Unfortunately, these photographs do not usually show the face in repose but are usually of a smiling child. Smiling distorts the features and makes even a normal upper lip and philtrum disappear. However, if the lips and philtrum can be clearly seen and both are abnormal and these appearances are combined with microcephaly (small head) plus living in a high-risk area of the world, parents should be concerned. American intercounty adoption agencies now routinely teach parents, meeting their child abroad, how to take accurate photographs themselves, with a measuring sticker on the child's face. They can then be emailed back to the USA for computer analysis.

Computer-assisted analysis of facial photographs can identify affected children with a much greater degree of accuracy but this technology is not usually available in developing countries, even in those counties where alcohol misuse is extremely common. This technology is invaluable in the later diagnosis of the adopted child who is showing difficulties suggestive of exposure.

18.5.3 Growth

Growth depends on a complex interplay of ethnicity, genes, parental heights, gestation, intercurrent illness, nutrition and nurture. Growth delays are very common in intercountry adopted children. Many children have low birth weights, which can be difficult to interpret in the absence of accurate gestational information. Malnutrition may worsen during institutional life. Familial factors are usually unknown. In three distinct populations of adoptees (Romanian, Russian and Chinese), the length of institutionalisation was directly related to delays in linear growth [14–16]. For every 3–5 months spent in an orphanage, the children lost 1 month of linear growth.

Although malnutrition is common in most new adoptees, nearly all children exhibit rapid catch-up growth after adoption [17]. Children who fail to show this catch-up need careful evaluation and investigation. Genetic, cardiac, gastrointestinal, renal and endocrine disease should be excluded, plus screening for infectious diseases and parasites, before alcohol is suspected.

Children affected by prenatal alcohol exposure can also be unusually small in weight and/or height at birth and later. Growth deficiency caused by alcohol exposure becomes more likely when poor growth is not explained by other factors and there is no catch-up growth pattern in the first year. Children who are unusually short compared to others raised in a similar environment should also raise concerns about exposure to alcohol. In most orphanages, children are cared for in units where all the children are roughly the same age. If a potentially adopted child is the smallest child in such a unit, the parents should request for more information about the background of the child and question possible exposure to alcohol.

Accurate monitoring of the adopted child's growth should start as soon as they arrive in the UK and be plotted on the centile chart in the parent-held record. Accurate growth measurements are unlikely to have been done before placement. In the analysis of the 120 reports received by the Centre for Adoption, although 94 children had their height, weight and head circumference recorded, these were single recordings done in a situation where the accurate age of the child was not known. Trends were impossible to establish, the ethnicity of the child was unknown, local centile charts were unavailable and some measurements, when replotted on standard UK charts, were clearly wrong [10].

US adopters are now taught how to accurately measure their child's head circumference themselves, practicing with a non-stretchable measuring tape calibrated in centimetres [18]. They need to wrap the tape snugly around the widest possible circumference – from the most prominent part of the forehead (often 1–2 fingers above the eyebrow) to the widest part of the back of the head. They are asked to

remeasure three times and to record the largest number. Microcephaly (head circumference less than the second centile or below) in the growth chart can be evidence of brain damage from alcohol. It is a more useful prognostic factor in infants and toddlers, because meeting early, largely motor milestones, does not rule out later difficulties with learning and behaviour.

18.5.4 Neurobehavioural and Neurodevelopmental Difficulties

Poor maternal health, no antenatal care, the complications of pregnancy labour and delivery, malnutrition in infancy and exposure to infections or toxins will all affect child development. Life in an orphanage often involves a lack of stimulation, play and nurture, emotional or even physical abuse. This pre-adoption information is largely unavailable to the new adoptive parents and the assessing clinician. After adoption, the child must make a complicated transition to a new family, culture and language. For the older child, this additionally means new educational experiences, attachment and identity issues.

The diagnosis of alcohol exposure is not easy in these circumstances. The functional disabilities from alcohol damage are usually not apparent before school age. Most affected intercountry adopted children are referred for diagnosis in the early years of primary school with the increasing sense that 'something is not quite right'. Alcohol affects multiple domains of brain functioning. The children can present a complex mix of problems including inattention and impulsivity, low IQ scores, specific learning impairments especially with mathematics, understanding cause and effect, organisation and planning, social and communication impairments, coordination difficulties, sleeping and eating difficulties. Teasing this out can require wide-ranging testing by professionals familiar with alcohol effects. This type of assessment is not easy to access in the UK where there are very few specialist diagnostic clinics. Many children are not identified early enough and their difficulties incorrectly attributed to attachment or adjustment problems, attention-deficit disorder or an autistic spectrum disorder.

Intercountry adopted children should not be denied a diagnosis if strict diagnostic criteria cannot be readily applied. Despite the absence of a maternal history, the combination of facial features, microcephaly, failure to show catch-up growth and neurobehavioral difficulties strongly suggests the diagnosis in intercountry adoptees, especially those from high-risk areas.

18.6 The Outcomes for Intercountry Adopted Children in the UK

The outcomes for this small but disadvantage group of British children are largely unknown. There are only three published studies which were confined to groups of children with unique characteristics who were all adopted from the

same country at about the same time. The only longitudinal study, the English and Romanian Adoptee Study, looked at the outcomes for 111 Romanian children adopted into the UK in the early 1990s. All had suffered severe institutional deprivation and neglect [19]. Two further retrospective studies looked only at the adjustment of British intercountry adopted children. The first study looked at children adopted from Vietnam [20] and the second, the British Chinese Adoption Study looked at 100 adult women aged between 40 and 50 adopted from orphanages in Hong Kong in the 1960s [21]. One small study of 35 children adopted into Hampshire between 1990 and 1995 emphasised the importance of screening the health and development of the children, given the high number of unsuspected medical problems found, many of which were significant [22]. It also identified several key issues for adoption agencies to consider when preparing and assessing applicants for intercountry adoption, and the importance of a multi-disciplinary approach in helping to support the adopters in their task of caring for the child. Two further small-scale reviews of UK adopters from a range of countries did not specifically focus on medical issues but noted that adopters found their child to be in poor or very poor health at the first meeting [23, 24]. Previous exposure to alcohol was not considered in any of these studies.

18.7 The Importance of a Health Assessment

Regardless of their country of origin, numerous studies across the world have shown that intercountry adopted children have a myriad of special health care needs and an increased incidence of physical, developmental and mental health concerns [25–29]. Although some of these concerns may be addressed before adoption, many problems persist, continue to be significant or do not become apparent until after the time of placement in an adoptive home. Dr. Dana Johnson from the University of Minnesota International Adoption Clinic has said that the chances of an institutionalised orphanage child having normal growth, health and development are essentially zero. He also says the second most common reason children are institutionalised is termination of parental rights because of neglect and/or abuse, often alcohol related.

Regardless of the country concerned, numerous studies across the world have shown that the prevalence of the fetal alcohol spectrum disorders is higher in children who are in the care of the state or adopted. In one US study, 71 children adopted from Eastern Europe were assessed 5 years after adoption [30]. Fetal alcohol spectrum disorders (fetal alcohol syndrome (FAS), partial FAS and alcohol-related neurodevelopmental disorders) were identified in 52% of children. FAS was found in 30%, partial FAS in 14% and alcohol-related neurodevelopmental disorders in 9%. Alcohol-related birth defects were found for 11% of children, all of whom were also diagnosed as having FAS. Mental retardation or significant cognitive impairment was found for 23% of children, autism for 9%, attention-deficit/hyperactivity

disorder for 51% and developmental coordination disorder for 34%. Although there are no prevalence studies in British intercountry adopted children, it would not be surprising to find a considerable number of undiagnosed affected children in this population.

Other receiving states acknowledge these issues more readily than the UK. All receiving countries have to tackle inadequate reports, absent medical histories, incomplete records, the impact of institutional care and a high prevalence of undiagnosed medical problems including prenatal exposure to alcohol. Most see further assessment of the child on arrival as essential. This issue has detailed coverage on the US State Department website [31]. All children new to the USA must have an immigration medical, ideally carried out by a specialist, within 2 weeks of arrival. This includes not only a physical examination but also a developmental assessment, vision and hearing testing and screening tests dependent upon age and country of origin. US adopters are advised that medicals done in the country of origin are not reliable. Similar advice is given in Canada. In Northern Ireland, the adoption regulations (2010) state that every intercountry child coming into the province should be seen by a social worker, a general practitioner and a health visitor within 7 days and by a paediatrician within 21 days of arrival [32]. In other areas of the UK, there is no statutory requirement for the child to have any health assessment at all.

18.8 Conclusion

The numbers of children concerned are small but their needs are important and they are disadvantaged compared to domestic adoptees. Many families struggle unfairly with prejudice and even professional hostility. General practitioners, general paediatricians and child psychiatrists see few intercountry adopted children. There is often a general reluctance to comprehensively assess children or to refer for further specialist advice. There are no specialist intercountry adoption clinics in the UK and for many UK families, other adopters, advice lines and American or Canadian websites are their main sources of advice and information.

The difficulties which alcohol affected intercountry adopted children experience are lifelong and untreatable. FAS/D is not however a hopeless diagnosis. Consistent, patient, loving, ‘industrial-strength’ parenting with structure and appropriate expectations plus support in school can help alcohol affected children to reach their full potential. That potential will be limited by alcohol-related brain damage but setting the bar at the right height, and identifying what they can do versus what they cannot do, will help them achieve success in their life. Early diagnosis will hopefully prevent some of the ‘secondary disabilities’ of depression, acting out and aggression, victimisation, trouble with the law and substance abuse. On the plus side, children who are adopted internationally will have a better prognosis than children who stay in a poor-quality institutional environment abroad.

Practice Points

- Compared to domestic adopters, intercountry adopters have been through a very different process which an assessing clinician must consider.
- Unlike domestic adoption, intercountry adopters are advised to obtain comprehensive, local information about alcohol misuse in pregnancy in their chosen country. This information is frequently difficult to obtain and may not be reliable.
- Any child adopted from any country in the world could have been exposed to alcohol prenatally. Exposure is more likely in children adopted from Eastern Europe, especially Russia and the former Russian states. It is much less likely in a child adopted from a predominantly Muslim country.
- The preparation of intercountry for the possibility that their child may have FASD is essential. Parents need to honestly assess their capacity for parenting the affected child and should not be afraid to say no to a placement.
- The prenatal history of alcohol exposure is usually unknown in intercountry adopted children. Adopters should be advised to obtain as much information as possible from the agency or institution aboard and if possible, have this information reviewed by a knowledgeable paediatrician before accepting a referral.
- Although not currently recommended in most of the UK, all intercountry adopted children, irrespective of their country of origin, deserve to have a comprehensive health assessment and screening after adoption.
- Even when the information required for a diagnosis is incomplete, an intercountry adopted child should not be denied a diagnosis. Early diagnosis improves the child's long-term prognosis and allows the parents and school to use appropriate strategies.
- Growth, learning, emotional and behavioural problems in intercountry adopted children may have other causes than prenatal alcohol.
- Adoption may ameliorate some of the long-term difficulties of intercountry adopted children exposed to alcohol.

References

1. Popova S, et al. Estimation of national, regional and global prevalence of alcohol use during pregnancy and fetal alcohol syndrome: a systematic review and meta-analysis. Lancet Glob Health. 2017;5(3):E290–9. https://doi.org/10.1016/S2214-109X(17)30021-9.
2. CDickinson K, Feuchtwang N. Children adopted from abroad. In: Merredew F, Sampeys C, editors. Promoting the health of children in public care. London: British Agencies for Adoption and Fostering; 2015. p. 223–5.
3. Selman P. The demographic history of intercountry adoption. In: Selman P, editor. Intercountry adoption: developments, trends and perspectives. London: British Agencies for Adoption and Fostering; 2000. p. 15–35.
4. Selman P. Intercountry adoption in the new millennium; the quiet migration revisited. Popul Res Policy Rev. 2002;21:205–25.

5. Office of the United Nations, High Commission for Human Rights, Convention on the Rights of the Child. Geneva; 1990.
6. Hague Convention on the Protection of Children and Co-operation in respect of Intercountry Adoption. Permanent Bureau, Hague conference on private international law. In: Proceedings of the seventeenth session, Tome 11. 1993.
7. Hague Convention on the Protection of Children and Co-operation in respect of Intercountry Adoption. A full and up to date list of ratifying and acceding states. 1993 Available on www.hcch.net/en/instruments/conventions/status-table/?cid=69. Accessed 20 Feb 2017.
8. Department for Education and Skills. Statutory guidance on adoption, annex C, adoptions with a foreign element. 2005. https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/270102/annex_c_adoptions_with_a_foreign_element.pdf.
9. World Health Organisation, Global information system on alcohol and health (GISAH). Geneva: World Health Organisation; 2014. http://www.who.int/substance_abuse/publications/global_alcohol_report/en/. Accessed Mar 2017.
10. Mather M, Bell N, Way J, Haworth G. The quality of medical information given to prospective intercountry adopters in England. Adopt Foster. 2017;4(1):52–66. http://journals.sagepub.com/doi/pdf/10.1177/0308575916684996
11. Adoptive families of British Columbia, Fetal alcohol syndrome/effects and intercountry adoption: know the risks. https://www.bcadoption.com/resources/articles/fetal-alcohol-syndromeeffect-and-intercountry-adoption-know-risks.
12. Stoler JM, Holmes LB. Under recognition of pre-natal alcohol effects in infants of known alcohol abusing women. J Paediatr. 1999;135:430–6.
13. Streissguth AP, Barr HM, Sampson PD, Bookstein FL. Prenatal alcohol and offspring development: the first 14 years. Drug Alcohol Depend. 1994;36:89–99.
14. Albers L, Hostetter M, Iverson S, Johnson D, Miller L. Health of children adopted from the former Soviet Union and Eastern Europe: comparison of pre-adoptive medical records. JAMA. 1997;278(11):922–4.
15. Miller LC, Hendrie NW. Health of children adopted from China. Paediatrics. 2000;105:e76.
16. Johnson DE, Miller LC, Iverson S, et al. Health of children adopted from Romania. JAMA. 1992;268:3446–51.
17. Van Ijzendoorn MH, Bakermans-Kranenburg MJ, Juffer F. Plasticity of growth in height, weight, and head circumference: meta-analytic evidence of massive catch-up after international adoption. J Dev Behav Pediatr. 2007;28(4):334–43. https://doi.org/10.1097/DBP.0b013e31811320aa.
18. University of Minnesota International adoption clinic how to measure head growth https://adoption.umn.edu/services-and-resources/how-measure-head-growth.
19. Rutter M, Beckett C, Castle J, Kreppner J, Stevens S, Sonuga-Burke E. Policy and practice implications from the English and Romanian adoptees(era) study. Forty five key questions. BAAF London; 2009. All other publications available at the ERA website http://www.kcl.ac.uk/ioppn/depts/mrc/research/Previous-Research/theenglishandromaniandoptee(era)project.aspx.
20. Bagley C, Young L, Scully A. International and transracial adoptions: a mental health perspective. Brookfield, MI: Avebury Press; 1993.
21. Feast J, Grant M, Rushton A, Simmonds J, Sampeys C. Adversity, adoption and afterwards, a mid- life follow up study of women adopted from Hong Kong British Association for Adoption and Fostering, London; 2012.
22. Harnott C, Robertson R. Intercountry adoption: implications for adoption agencies and medical advisers. Adoption & Fostering. 23(4):26–34.
23. Mason K. UK families experience of the adoption process. In: Ryvgold A, Dalen M, Saeterdals B, editors. Mine - yours - ours and theirs: adoption, changing kinship and family patterns. Oslo: University of Oslo; 1999.
24. International Bar Association. The intercountry adoption process from the UK adoptive parents' perspective. A report from the working party on intercountry adoption of committee 4 (family law), section on general practice, International Bar Association, London; 1991(ibanet.org).

25. Juffer F, Van Ijzendoorn M. Behavior problems and mental health referrals of international adoptees. JAMA. 2005;293(20):2501.
26. Bolton M, Day D. A systemic evidenced based literature review of medical and developmental issues of international adoptees with emphasis on the need for immediate, Annual Symposium: Graduate Research and Scholarly Projects. Wichita, KS: Wichita State University; 2007. p. 135–6.
27. American Academy of Pediatrics. Medical evaluation of internationally adopted children for infectious diseases. In: Pickering LK, editor. Red book: report of the committee on infectious diseases. 25th ed. Elk Grove Village, IL: American Academy of Pediatrics; 2000. p. 148–52.
28. Miller L. The handbook of international adoption medicine: a guide for physicians, parents, and providers. Oxford: Oxford University Press; 2004.
29. Mather M, Kerac M. Caring for the health of children brought into the UK from abroad. Adopt Foster. 2002;26(4):44–54.
30. Landgren M, Svensson L, Strömland K, Grönlund MA. Prenatal alcohol exposure and neurodevelopmental disorders in children adopted from eastern Europe. Pediatrics. 2010;125(5):e1178–85. https://doi.org/10.1542/peds.2009-0712.
31. US Department of State. Intercountry Adoption. undated. https://travel.state.gov/content/adoptionsabroad/en.html. Accessed Mar 2017.
32. Northern Ireland. Adoption, Regional policy and procedures, Chapter 13. 2010. http://coram-baaf.org.uk/webfm_send/2565. Accessed Mar 2017.

Service Delivery: Organisation and Models of Care

19

Raja A. S. Mukherjee

Chapter Highlights

- Areas to focus care
- Locations of care
- Models of care and management at different ages

19.1 Introduction

Fetal Alcohol Spectrum Disorders (FASD) are a set of conditions that often do not present to clinical settings as a referral for an assessment of FASD itself. As highlighted in a systematic review of comorbid disorders in those diagnosed with FASD, 438 different ICD 10 attributable conditions could be identified [1]. These range from psychiatric presentations to physical health issues. It is most often for these wider comorbid presentations and conditions that are the reasons behind why referrals to clinical services are made. This can also mean that generic services are in most likelihood, already seeing some cases. They are just not being recognised. For this reason, developing specialist diagnostic processes as well as therapeutic interventions is challenging. This does not mean they are not needed. Often, the biggest issue raised by families is that generic approaches do not work. As such, the underlying condition is missed when standard treatments fail. Further, different commissioning structures in different countries add an extra layer of complexity to this already complex scenario.

This chapter will present some basic principles and considerations that should be applied to different settings, in order to facilitate service delivery across a range of

R. A. S. Mukherjee (✉)
National FASD Specialist Behaviour Clinic, Surrey, UK
e-mail: Raja.Mukherjee@sabp.nhs.uk

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_19

Fig. 19.1 Areas of care delivery

areas. As other chapters will be addressing prevention and primary support as well as the identification and prevention of drinking by midwives and other practitioners, this chapter will focus more on the needs of the child/adult affected and resources required to support them through the diagnostic pathways.

Broadly commissioning for those affected can be separated into three areas. These are pre-diagnostic, diagnostic and post diagnostic, as highlighted in Fig.19.1.

The separation of these three areas allows not only the individual components to be broken down and considered but also the responsibilities to be divided. For example, health may not in many places provide respite for families where difficulties are seen, as it is the responsibility of social care; yet in some places, health is the primary commissioner of these services if there is an associated disability. Another scenario where there are differences includes, for example, therapeutic interventions. In many places, supportive interventions may well be part of a health-funded resource; yet increasingly in many places, including the UK, this is provided post diagnostically by third sector agencies. These factors may need local consideration, and a chapter such as this will not be able to address all local variances; however, the principles may be applicable across settings.

19.2 Pre-diagnostic Issues

Families of children potentially affected by prenatal alcohol exposure face challenges in knowing how to navigate what can be a complex system [2, 3]. Whilst looked after children and those adopted have been identified as being high risk for this disorder [4], it is highly probable that in the broader population, many children will be in families who may or may not be willing to consider or even accept this as a possible diagnosis. The sense of guilt and anger as well as unwillingness to believe the diagnosis has been shown to play a part in both seeking and preventing access to diagnosis [5]. It is ultimately when the child's difficulties become evident that investigations are required to confirm the diagnosis. It is then that consideration is given to accessing a pathway towards referral. It is important to recognise however, the diagnosis is not the end point but only really the start of the journey for improving the outcomes for these children. A diagnosis in itself is often not enough.

Due to the complexity of the diagnostic process, several issues need to be addressed. Firstly, it is vital that information is available with regard to alcohol

exposure in pregnancy. When this is missing, it means the majority of cases cannot be appropriately labelled. The proportion of those who have a presentation where a diagnosis can be made without the alcohol history is currently small. Even when dysmorphic features are present, it is wise to always seek out information on alcohol exposure risk. Without this, whilst in the future it may be possible to identify alcohol exposure without the recorded history, at this point only a probable diagnosis can be attributed to those without this confirmed history.

As such, ensuring information follows the child about prenatal risk factors from birth can have significant benefits. Having a recording of prenatal alcohol exposure in the child development record, which is transferred to the primary care record via specific Read codes (used by general practitioners (GPs) especially in the UK to record a medical history), will be one simple way of identifying exposure risk. Whilst this will not provide the full information regarding the extent of exposure, it will offer some insight, which is often currently missing. Midwifery and paediatrics have a clear role in this process.

This broader information gathering and identification, of what is probably the most vital piece of information, can be supported by various agencies. At the early stages, if going through an adoption and fostering process, social workers have the ability to note down and seek information regarding a birth mother's alcohol consumption in pregnancy. Knowing that alcohol exposure in itself did not occur is as important as recording that it did take place. It should be noted however, with caution that many people who state they did not drink are later identified as having alcohol exposure [6]. For example, 80% of women in the UK consume alcohol. Not all of this group will give up alcohol consumption at conception. More reduce when pregnancy is identified but some will continue throughout pregnancy [7]. Various studies have identified that both the level of exposure and frequency of exposure are greater than would have been suspected and that people underestimate what they consume [8]. The UK has been identified as the fourth highest consumer of alcohol in pregnancy [9].

For birth mothers, offering diagnostic counselling and support to seek help with the potential feelings of guilt, then supporting and helping them through a process of grief. This is necessary in order to move them to a place where they are able to best support the child [5]. Rather than expecting the grief response not to be present, supporting the process may be a more beneficial therapeutic intervention for both the family and the individual. Similar issues may be needed for adoptive families where the information was initially missing but comes to light later.

Teachers may well be supportive of this process. Many of the behaviours that children present with can be seen in school, as well as at home. A teacher who is willing to support the family to seek appropriate help, gives an accurate description of the individual child and listens to families will also facilitate the process. Research has shown often the opposite to be true and families are blamed for behaviour rather than supported [10].

A similar role can be taken by general practitioners. Rather than dismissing the family's presentation, as it has been described by those seeking help [10], where the general practitioner or the primary health care physician is willing to listen and

support access to the diagnostic process, it has been shown this can lead to significant help and support for the individual family. Families have reported on numerous occasions where they have had help from a supportive primary care physician that the journey has been far easier for them [2, 10]. Whilst it would not necessarily be the place for the primary care physician to have a detailed diagnostic knowledge of FASD, as the complexity of the presentation is more suitable towards secondary or tertiary care assessments, having sufficient insight to refer to a more appropriate clinician is vital. Having the pathways in place to facilitate this is therefore also necessary.

This is another area where third sector organisations and family support groups can offer input. Before entering a diagnostic process, most services do not offer specific work to engage families. Third sector organisations however may well be able to guide a family through the types of information that is needed for a referral, and also where these can be obtained. Strategies learned by others can be shared with mutual engagement. Working collaboratively and closely with health sector can minimise the impact and cost. By using resources as far as possible that already exist in various areas, rather than trying to develop completely new structures and practices in all cases, will reduce costs. Sharing responsibility will offer potential benefits for all. This does not mean both groups have to be part of the same organisation; however, partnership relationships can often have mutual benefits.

Various pieces of information are useful and should be gathered. These include the following:-

1. Alcohol exposure information as accurately as possible.
2. Other drug and medication exposure in pregnancy as accurately as possible.
3. History of early upbringing and developmental milestones, including considering neglectful and traumatic experiences.
4. Medical records of other investigations and observations made, including genetic, are available.

19.3 Diagnostic Issues

19.3.1 Process of Getting Diagnosis and Levels of Care

Whilst the prevalence of FASD may vary across different regions, in some areas such as the UK, the exact figure is yet to be established. It is likely to be far more common than had been previously expected. Estimated prevalence rates for Europe based on pooled prevalence stand at around 2% of the population [11]. A separate screening prevalence study offering an estimate suggested rates could be as high as 17% in the general population [12]. As a result, the number of individuals in the general population who may be affected is likely to be far higher than is currently presenting. As highlighted above, it may well be hidden and presenting to other services. FASD remains a condition that is poorly understood by a majority of professionals and public both in the UK and wider afield [13].

It is also clear that there are different diagnostic methods that exist. When added to the complexity and range of factors that have to be addressed, can mean for some, this diagnosis remains highly specialist service. It means it is a pathway that is not necessarily always accessible to families, even potentially at the secondary care level. An overview of the diagnostic measure has been summarised in the British Medical Association (BMA) board of science review. It highlighted that at the time the UK consensus was published, it would lean towards the older Canadian 2005 guidance [14]. Whilst this has been updated recently [15], this update was supported by the Scottish Intercollegiate Guidelines Network (SIGN) 156 review [16] and has more recently been adopted by the English National Institute for Clinical Excellence (NICE). Increasingly diagnostic services are turning towards more established diagnostic manuals such as Diagnostic and Statistical Manual of Mental Disorders (DSM) and International Classification of Diseases (ICD) to describe the parameters of features that are necessary. To this extent, the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM V) has suggested diagnostic criteria of Neurobehavioural Disorder Associated with Prenatal Alcohol Exposure (ND-PAE). This is currently in the evaluation phase prior to final acceptance [17].

What is clear, however, is the parameters that need to be sought in order to make the diagnosis consistent across each layer of service provision. The difficulty relates to defining the thresholds between each layer of service provision and what these different layers need to do in order to complete an assessment. Cases which are more straightforward may well be suitable for more local assessment with fewer resources but due to the ease of identification they are not always needed. Whereas, a complex individual may well have to be seen by more specialist service, where more tools and assessment methods can be used to identify more subtle presentations. Figure 19.2 highlights from an overall population of how each tier of service will link to provide for the complexity of care.

19.4 Roles of Different Providers and Levels of Expertise

As highlighted above in Fig. 19.2 (although not to scale), the proportion of the population seen by increasingly specialist clinics decreases, as the complexity of cases being seen increases. The complexity and range of assessments completed will also increase through the different levels. For example, it will be envisaged that the secondary care practitioner, who is seeing the majority of more straightforward diagnostic and management cases, would often be working as an individual practitioner in an already commissioned generic service with limited resource. Even though all diagnostic criteria require a multidisciplinary review, this can be gathered via tools that assess the different areas and therefore manage in a more cost restricted environment.

Whereas a specialist service is more likely to be a multidisciplinary team with access to materials and tools, it is likely that the majority of cases will be seen at the secondary care level. When moving to a more regional level, the cases will become more complex. This is where the multidisciplinary specialist assessment team is

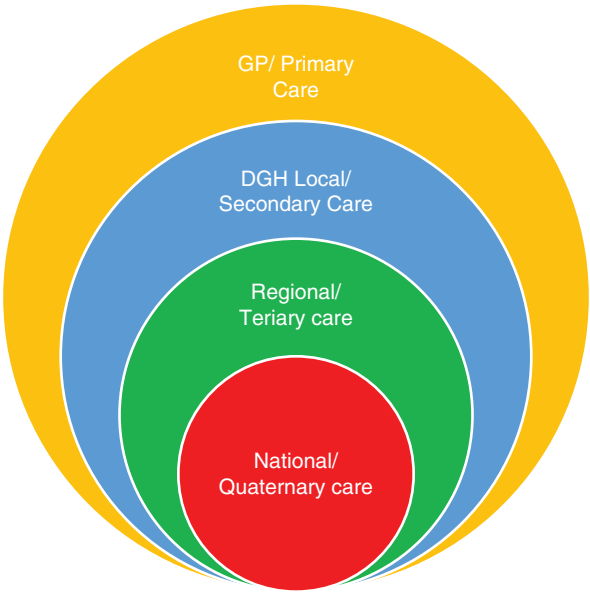


Fig. 19.2 Levels of care based on population need and complexity

required and more direct face-to-face assessment used, rather than replying just on questionnaires. They will be needed to offer a more detailed assessment of the wider functioning deficits and have the capacity to rule out other disorders, as described in other chapters.

The threshold to move between layers would be best expedited through expert supervision of the lower layers of service provision. Moving to the National Centre will again be reserved for the most complex cases where the definitions and expertise to pull apart the subtleties of the presentation would be necessary. These should only be directed through to the national service where regional services lack the equipment or expertise to do this. For example, the use of three-dimensional (3D) technology in facial recognition is currently limited and not available everywhere. At a national level, it would be expected that they would use these more detailed evaluations which are not necessary in other tiers of service delivery. Eventually as services mature and regional services can offer the same level of expertise, national services may not be required at all.

It is important to recognise that many practitioners have expressed the view that they lack the expertise to diagnose FASD in the UK [13]. As highlighted in Fig. 19.3, the hub and spoke model of service delivery, as described in the BMA board of science document [3], is a model used in other specialties, balancing local service provision which is already in place, supported by expertise and supervision. Each tier would support the lower level. For example, local supported by regional and regional subsequently by national services. This would support decision-making to be taken about suitability of cases for each layer but also facilitating the majority of

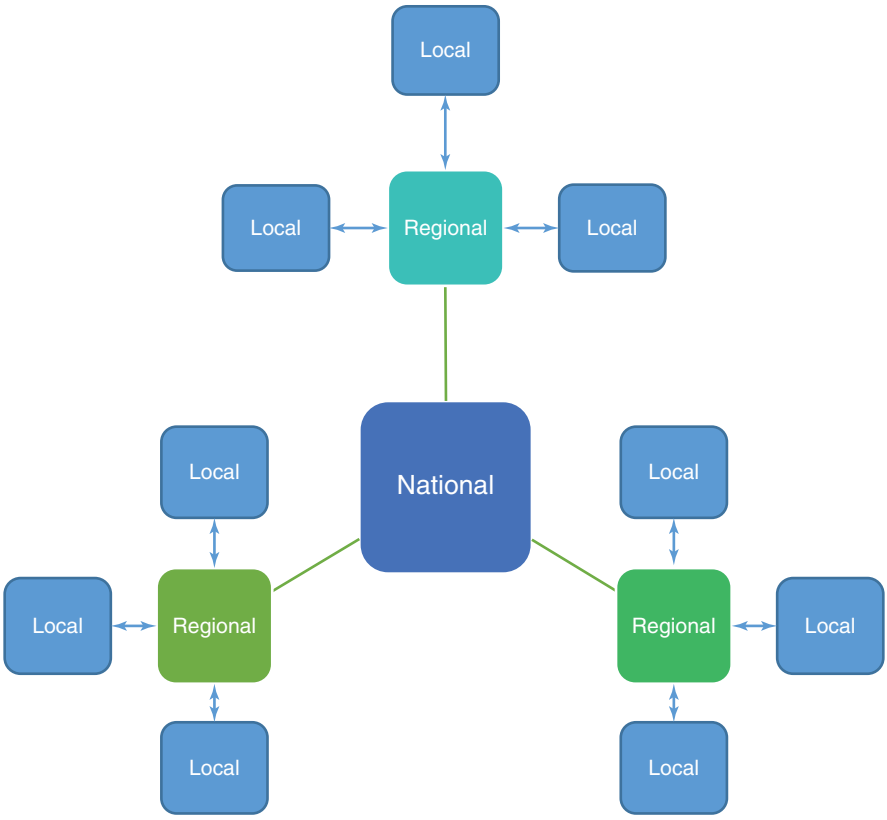


Fig. 19.3 Hub and spoke model of service delivery [3]

cases to be managed close to home in the local setting with expert support. This will allow cost to be kept to a minimum, expertise to be shared and knowledge increased across the board. It would also facilitate service and therapeutic care delivery to individuals when they need it.

Whilst there are financial implications to the implementation of this approach, with a condition as potentially prevalent and complex as FASD, the overall impact is likely to minimise these costs. This will also help reduce expensive secondary disabilities. It would be in keeping with an investment to prevent model financial saving. When the evaluation process is tailored for the individual to meet their needs and guide future management, prevention of secondary mental health issues, criminal justice system involvement including incarceration as well as preventing further generations of problem are more likely.

Underlying these aspects is the commissioning responsibility to both recognise and provide support for these service developments. Whilst government has a responsibility to direct and support development, different countries have different approaches. In a country such as Scotland where central government is able to

allocate funds more directly, support can be tailored to the needs of the country. In an area such as England where commissioning is decided at a local level by clinical commissioning groups (a group of GPs who come together to decide on local needs and commission local services), this can be more difficult as the area is initially too small for the investment needed. When the focus is only at the local level and not even regional, trying to establish a consensus about needs can be challenging. In these situations, where the number of cases per local area may be small comparatively, the foresight to develop the above, more strategic approach, can be limited. It is in trying to have a more cohesive and considered approach that these improvements will eventually be seen.

A similar situation will arise where the funding of health care is via insurance. When this is primarily geared towards physical interventions rather than psychiatric and neurological, it is important that the establishment of clear approaches, with diagnostic formulations which meet criterion, preferably through consensus, is achieved. To not do so means that many of these children will not meet threshold in further obtaining services and will therefore develop the secondary disabilities that are well described with this condition [18].

19.5 Child vs. Adult

The challenge, not only in terms of diagnosis but also wider service delivery, becomes more challenging as an adult. Whilst clinical services for children have a developmental perspective and are tailored to meet the holistic needs of the child, this is not necessarily the case in an adult. In adult services, presentations and care tend to be delivered in different specialty areas, akin to silos. Whilst there is sometimes cross collaboration, the holistic approach to care is often missing. The developmental perspective is also rare.

Specific, specialised neurodevelopmental services, which can understand and manage the whole person, do not exist in many parts of the UK or wider in the world. This means that for many, especially those who do not have an intellectual disability (IQ below 70), fall through service gaps and struggle to gain management interventions. Even those who are diagnosed struggle to access care and support. This can be due to the nature of expectation placed upon them. As a child, education services offer ongoing development and support, but as an adult the expectation is on the individual to find employment and support themselves. This is not always possible. Vulnerabilities and the developmental perspective are rarely taken, leading to many finding life overwhelming and suffering with the secondary disabilities well described, including mental health problems and involvement of the criminal justice system [18].

Further challenges occur diagnostically. As an individual becomes older, whilst the history of struggles becomes more evident, the complexity of disentangling the longer history of the presentation can cause problems. Not having access to early records, including sometimes to the alcohol history during pregnancy, becomes an issue. These challenges to the diagnostic process often require more specialist

assessment. This means that local services often lack expertise to undertake this, leading to the need for more regional and national services. It is entirely possible however that those regional and national services could serve both children and adults due to the established availability of a multidisciplinary team. Commissioning intention, as discussed above, would need to have the foresight to recognise this and the teams would have to have the skill set to be able to manage both groups. The UK national clinic is an example where this already occurs.

19.6 Post-Diagnostic Support and Ongoing Involvement

As highlighted, diagnosis is only one part of the process. When an individual has received the diagnosis and formulated an understanding of the underlying issues, management of these needs to be delivered via variety of approaches. Figure 19.4 illustrates some of the areas around the individual that need to be considered and supported in order for the person themselves to receive the best support and care. Only a part of this is health related. Joint engagement with social care, education, criminal justice system, housing as well as third sector charitable sector support for individuals and their families is often vital. Commissioning for this, with most countries' resources and budgets allocated separately rather than jointly, leads to a lack of joined-up decision-making. Unfortunately, individuals do their best when there is a coordinated approach. For children in England, approaches such as the Team Around the Child have been developed to pull together such resources. Circle of friends approaches can also be used in order to facilitate this. In later years

Fig. 19.4 Examples of the relationships and network around and individual

however, the complexity increases with the need for clear evaluation of the support needs in order to provide and maintain an individual at the centre of their own care.

More work to consider therapeutic intervention, medical management and also psychoeducational approaches need development. Strategies to support local resource and facilitate access to education, housing, employment either via the health, social care or third sector support route in many areas need better focus and development. These areas remain in their infancy in many settings. The lack of evidence base for many areas means developing commissioning intentions for these is often challenging but remains the necessary focus for future.

19.7 Conclusions

FASD is a complex condition that often requires differing levels of care to diagnose and support individuals. This often needs clear intentions for a coordinated approach to care.

Practice Points

- Support can be separated into three parts. Pre-diagnostic, diagnostic and post diagnostic.
- Different levels of complexity require different approaches to diagnosis and management.
- Only when the right information is collected can the diagnosis be made but different levels of complexity offer different requirements for assessment.
- Investing in service can offer cost savings overall.

References

1. Popova S, Lange S, Shield K, Mihic A, Chudley AE, Mukherjee RAS, Bekmuradov D, Rehm J. Comorbidity of fetal alcohol spectrum disorders: a systematic review and meta-analysis. Lancet. 2016;387:978–87.
2. All-Party Parliamentary Group (APPG) for FASD. Initial report of the inquiry into the current picture of FASD in the UK today. London; 2015.
3. BMA. Fetal alcohol Spectrum disorders, a guide for healthcare practitioners update. London; 2016.
4. Gregory G, Reddy V, Young C. Identifying children who are at risk of FASD in Peterborough: working in a community clinic without access to gold standard diagnosis. J Adopt Foster. 2015;39(3):225–34.
5. Thomas R, Mukherjee RAS. Experience of birth mothers of children diagnosed with FASD. EUFASD 4, London; 2016.
6. Abernethy C, Mcall KE, Cooper G, Favretto D, Vaiano F, Bertol E, Mactier H. Determining the pattern and prevalence of alcohol consumption in pregnancy by measuring biomarkers in meconium. Arch Dis Child. 2018;103:f216–f20.

7. Nykjaer C, Alwan NA, Greenwood DC, Simpson NAB, Hay AWM, White KLM, et al. Maternal alcohol intake prior to and during pregnancy and risk of adverse birth outcomes: evidence from a British cohort. J Epidemiol Commun Health. 2014;68(6):542–9.
8. BMA. Alcohol misuse: tackling the UK experience. London; 2008.
9. Popova S, Lange S, Probst C, Gmel G, Rehm J. Estimation of national, regional, and global prevalence of alcohol use during pregnancy and FAS: a systematic review and meta analysis. Lancet Global Health. 2017;17:1–10.
10. Mukherjee RAS, Wray E, Commers M, Hollins S, Curfs L. The impact of raising a child with FASD upon carers: findings from a mixed methodology study in the UK. J Adopt Foster. 2013;37(1):43–56.
11. Roozen S, Peters GJ, Kok G, Townend D, Nijhuis J, Curfs L. Worldwide prevalence of fetal alcohol spectrum disorders: a systematic literature review including meta-analysis. Alcohol Clin Exper Res. 2016;40(1):18–32.
12. McQuire C, Mukherjee R, Hurt L, Higgins A, Greene G, Farewell D, et al. Screening prevalence of fetal alcohol spectrum disorders in a region of the United Kingdom: a population-based birth-cohort study. Prev Med. 2019;118:344–51.
13. Mukherjee RAS, Wray E, Curfs L, Hollins S. Knowledge and opinions of professional groups concerning FASD in the UK. J Adopt Foster. 2015;39:212–24.
14. Chudley AE, Conry J, Cook JL, Loock C, Rosales T, LeBlanc N. Fetal alcohol spectrum disorder: Canadian guidelines for diagnosis. Can Med Assoc J. 2005;172(5 Suppl):S1–S21.
15. Cook JL, Green CR, Lilley CM, Anderson SM, Baldwin ME, Chudley A, Conroy JL, LeBlanc N, Loock CA, Lutke J, Mallon BE, McFarlane AA, Temple VK, Rosales R. FASD, a guideline for diagnosis across a lifespan. Can Med Assoc J. 2016;188(3):191–7.
16. SIGN. SIGN 156. Edinburgh: Health Improvement Scotland; 2018.
17. APA. DSM V. Washington: American Psychiatric Association; 2013.
18. Streissguth AP, O'Malley KD. Neuropsychiatric implications and long term consequences of fetal alcohol spectrum disorders. Semin Clin Neuropsychiatry. 2000;5(3):177–90.

Part III

Management

Section 3: Overview of Management

20

Raja A. S. Mukherjee and Neil Aiton

20.1 The 'So What?' Question

For many, a significant reason for not considering diagnosis of fetal alcohol spectrum disorder (FASD) relates to the understanding that there is nothing that can be done once a diagnosis is made. As has been highlighted in previous chapters, diagnosis on its own without necessarily considering individual needs is not always enough. It does, however, signpost the individual and clinician to strategies that may ultimately benefit long-term outcomes.

By understanding the needs of individuals and their families, support can be put around both groups in order to provide the best function and outcome. For example, by educating families around how to communicate, modify environments including regarding the sensory world, knowing how to get access to appropriate and adequate support modified to the specific needs of these individuals, allows the development of bespoke intervention. These factors reduce future demand on services and improve long-term outcomes for the individuals and their families.

Much of the focus has been on the negative secondary disabilities linked to not intervening. The opposite therefore also becomes true. By directing and identifying appropriate interventions rather than just for those where resource is available hopefully improves the longer term outcomes. This also has an economic benefit, in that it reduces the cost impact on the wider health, social and educational economy.

R. A. S. Mukherjee (✉)
National FASD Specialist Behaviour Clinic, Surrey, UK
e-mail: Raja.Mukherjee@sabp.nhs.uk

N. Aiton
The One Stop Clinic, Royal Sussex County Hospital, Brighton, UK
e-mail: neil.aiton@nhs.net

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_20

Ultimately however, it is through directed intervention that individuals will improve. It is also only through understanding how the evidence base has grown in this area can there be a shift from the assumption that there is no point to intervene. This then needs to move to answering the 'so what' question in a positive manner, which ultimately supports those who need it.

20.2 Need to Change the Trajectories for People

As highlighted, secondary disabilities are common for those who are not recognised and not appropriately treated. The best-known study took place in the clinic population following up nearly 500 people over 30 years. Ninety per cent of that cohort went on to have mental health problems. Fifty per cent at some point had been incarcerated and involved with the criminal justice system and a third had gone on to develop their own addictions. All of these have long-term cost implications to wider society.

Whilst the narrative here appears to be poor, this is partly because the subsequent management, post diagnosis, was inadequate. Support structures were not put in place and therapeutic interventions to support and guide were not properly addressed. Whilst specific medication is not available to treat FASD, the management of associated comorbidities and how they can be modified are now increasingly understood. Psychological and educational approaches have also been developed. Increasingly, where this has been used, better outcomes are seen. It is therefore important not to presume that there is a negative outcome. Rather, by appropriate early recognition, then intervention, better outcomes can be seen. This is particularly pertinent when considering the fact that the majority of those exposed, often with lesser degrees of difficulty, do not present to clinical services. It may well be that in actual fact those with lesser levels of difficulty are either to be found in different situations, mislabelled and with secondary disabilities or have found ways to manage and cope. This is not truly known. What is seen however, in those who are most affected that intervening does make a difference.

20.3 Consensus and Evidence

Unfortunately, the level of evidence that has been collected is more of a cohort-level evidence effect. Randomised controlled trials in this group have been rare. The full benefits of therapeutic intervention compared to other outcomes are therefore based more on consensus and cohort effects. While several interventions have been identified and developed, they are yet to be fully evaluated. This does not mean that they have no benefit, only that more work is yet to be done and this remains the focus of future work.

Increasingly, consensus statements are being developed around evidence through expertise, and whilst this is a lower level of evidence in the hierarchy of quality, it is a starting point to try and identify approaches that will be useful. This allows an

understanding of how treatments need to be modified from traditional approaches. Some of these are discussed in the subsequent chapters.

20.4 Overview of Chapters

The following chapters present what is known already and how to manage individual approaches. These include supporting the individual, the family as well as issues with transition from childhood to adulthood. Managing the individual in the classroom and in other settings will be presented. Also, understanding what it is from an individual and families' perspective will be presented. The final section presents a broader overview of some of the knowledge around prevalence and strategies to help intervene and develop service in order to facilitate care for these groups.

The Value of Support Groups and Advocacy in Helping Families

21

Diane Black

Chapter Highlights

- Overview of the needs of families of those with FASD.
- The needs of individuals in the families of people with FASD.
- Importance of the family voice in caring for a person with FASD.

Raising a child with a FASD places a great burden of care and stress on families, whether the child lives with the birth parents or is placed with a foster or adoptive family. Due to the burden of care, a high percentage of children with FASD are raised in foster or adoptive families. Caregivers need information on FASD, emotional support, respite care, advice on dealing with difficult behaviors, and guidance with supporting their children into adulthood. In addition, birth mothers often suffer from social stigma as they are blamed for the child's disability. Both research and parent reports show that local parent-led support groups are very good at providing emotional support and help with advocacy. The role of the clinician is central, with respect to providing evidence-based information and referrals to services. Clinicians are encouraged to be aware of local parent-led support groups. In care for the child or adult with FASD, the physician is encouraged to be proactive in offering care such as contraception.

21.1 Introduction

Raising a child with FASD imposes a heavy burden of care on families [1]. Parents, whether birthparents, foster parents, or adoptive parents, are often tired, frustrated, and overwhelmed. Parents worry about developmental delays, behavior, medical

D. Black (✉)
European FASD Alliance, Landskrona, Sweden
e-mail: diane.black@eufasd.org

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_21

problems, poor social adaptation, and low performance in school. Primary caretakers, often the mother, are exhausted from the permanent round of visits to the pediatrician, psychologist, play therapist, speech therapist, and physical therapist. The child's problems are complex; not only do they have FASD, but in many cases they carry a burden of early abuse, failed placements in foster homes, as well as multiple medical and psychiatric comorbidities [2]. Parents are anxious about the future—what will become of my child when he/she is an adult? Who will take care of my child when I am gone? Finances can be a source of worry, due to cost of therapies not paid by the health insurance and loss of income if one parent cuts down on or quits paid work to devote more time to the child. The parents lose their social contacts, as many friends drift away. Parents feel helpless when the usual child-raising techniques do not work with this child, but they often get no advice from the professionals beyond "just love him/her, and everything will turn out alright."

The goal, therefore, of this short contribution is to give some insight into the needs of families raising a child with FASD and to suggest how clinicians can support the parents by judicious referrals to other professionals and to parent-led support services.

21.2 Types of Families

Children with FASD may be living with foster parents, adoptive parents, or birthparents. Due to their complex needs, many children with FASD cannot be raised in their birth family, so they may be living in foster or adoptive homes. The high prevalence of FASD in foster and adoptive families illustrates this problem. For example, an international review by Lange et al. [3] reported that active case ascertainment studies give the prevalence of FASD among children in the social care systems as 158 per thousand in Chile and 40 per thousand in Israel; and among adoptees from eastern Europe in Sweden, 521 per thousand. Indeed, these authors state that the severity of the disability seems to directly predict the chance of a child not being raised in the birth family.

21.3 Needs of Parents

Within each family, each person is affected by FASD in a different way: fathers, mothers, siblings, and the child with FASD himself or herself. Much of the research has been focused on caregivers, usually the mother, with much less attention given to the needs of other family members such as fathers who are likely not the primary caregiver.

The burden of raising a child with a disability has been multiply documented: research shows that such parents suffer high rates of emotional distress and poor health-related quality of life [4]. While raising a child with autism spectrum disorder is often considered to be highly stressful, one study concluded that FASD causes even higher stress [4]. In comparing types of families, birth and adoptive families

experience higher stress than do foster care families [5], and caregivers for teens and adults experience more stress than those caring for younger children [6]. Foster parents are likely to be better prepared for a child with FASD than other parents: they may know that the child has FASD at the time of placement and they may already be experienced in caring for difficult children. And if the burden on the family becomes too high, foster parents have the option of ending the placement. In any case, the job of the foster parents is likely to end when the child reaches adulthood. Adoptive parents, on the other hand, may not realize that their long-hoped-for child has FASD, or they may not realize what that means. They may be inexperienced and lacking confidence as parents, likely have no support from social services after the adoption, and have no idea what is wrong or where to get help. In these cases, the general practitioner or pediatrician can play a key role in referring the parents to diagnostic and support services. Another key aspect for both adoptive and birth parents is that they are financially responsible for the child, and this responsibility continues into adulthood. Finally, birthmothers experience all the above stresses, and in addition, they may be stigmatized as they are seen as responsible for their child's disability [7].

The evidence base for the needs of families raising children with FASD is small, but compelling. One study specifically identified the need for individualized information, practical support, respite care, and the opportunity to share with other caregivers [6]. Another study found that 69% of parents want to talk to other parents about their feelings about the child; 51% want to learn how to deal with unusual behavior; and 48% want to understand why the child acts difficult or oddly [8]. Similar needs were identified by focus groups of professionals: need for timely diagnosis, emotional support, social support, understanding behavior, and advocacy [9].

21.3.1 The Right Information at the Right Time

The parents want the information they need, when they need it. The first need, often even before the official diagnosis, is for information about FASD. Does my child have FASD? What does that mean? Many parents suspect FASD and have searched for information on the internet. Even so, the moment of receiving the actual diagnosis is a shock. Despite the shock, many parents feel relieved as their suspicions and fears are confirmed, as they can finally give a name to the problems. As some parents say, "the puzzle pieces all fell into place." At the time of diagnosis, the diagnostician gives information, but parents may feel too overwhelmed to absorb and remember all they hear at that time. Thus, it is worthwhile to provide parents with a factsheet or brochure with some key points, and also with a list of resources for further support and information.

Many parents have sought a diagnosis due to behavioral problems. Research confirms the complaints of many parents: the child does odd things, shows no fear, and does not respond to normal child-rearing methods [10]. Thus, parents are already searching for information on how to manage behavior, and indeed, the

search for these answers may have been the driving force in seeking a diagnosis. Perhaps first and foremost, it is often a relief for parents to realize that many of the odd behaviors are due to the FASD, rather than poor parenting. Indeed, parents who understand that their child has neurological damage more easily adapt their parenting styles [11].

There are many sources of information on raising children with FASD, including books, websites, support groups, and more. Some of these resources are partly or wholly based on parental experience rather than research, yet they have stood the test of time. Examples are the books *Trying Differently Rather Than Harder* [12] or *Strategies Not Solutions* [13]. In the last few years, more research has been devoted to developing evidence-based interventions. A 2009 review of five interventions identified several success factors: parent education, teaching of specific skills, and embedding the programs into the existing care plan [14]. Petrenko and Alto [15] reviewed the increasing number of evidence-based programs, finding that some programs reduced caregiver stress and improved child behavior. Unfortunately, these programs are home or community based, and thus of limited availability. A newer approach, training by Internet, could overcome the geographical limitations. While such programs are in their infancy, a training program for parents of children with autism has been tested in a randomized controlled trial; parents were assigned to a self-learning group or therapist-assisted (by video-conference) group. Both groups improved in several measures such as stress and perception of the child, though the therapist-assisted group showed higher improvement [16]. Such programs are appearing for parents of children with FASD; one is the Canadian Strongest Families FASD program, which is under development and testing [17]. If such programs are shown to be effective, they could be used more widely, but would require translation into local languages and adaption to the local culture and healthcare system.

21.3.2 Emotional Support

Parents need social support in the difficult task of raising their child, and guiding them through the transition into adulthood. Many adoptive and foster parents find that their former friends gradually disappear during the year after the adoption as they simply do not understand their former friends who have become stressed, tired, anxious, and emotional. Even with the best of relationships, it is difficult for parents of normally developing children to relate to the problems experienced by their friends.

To some extent, parents depend on their child's pediatrician, but he/she does not have the time to provide daily support. Furthermore, parents may be hesitant to discuss embarrassing topics such as public masturbation and hallucinations with even the most open-minded doctor—they feel as parents they may be blamed for "causing" the behaviors. Finally, living with these children can be such a strain that parents do not feel "love" for them, and they do not dare to admit this to anyone. For such parents, it can be a real relief to meet other parents who have experienced these feelings and occurrences and can put them into perspective. It is not surprising that parents dealing with an unpredictable child 24/7 feel anxious, fearful, and exhausted

rather than relaxed and loving. Such parents need support, education/information on managing the child, and above all, respite care so they can get some rest and time for themselves. Getting funding for respite care and then finding appropriate care givers is difficult, and just the search for such care adds another stress to the level of care [18].

21.3.3 Advocacy

Persons with FASD need lifelong support services. Anecdotal reports from parents show that children may have 10 or more professional support persons: case manager, guardian, speech therapist, play therapist, physical therapist, specialized orthodontist, special education teachers, pediatrician, psychologist, psychiatrist, respite care workers, etc. Older teens and adults with FASD need support with finances, housing, finding work, and arranging appropriate free-time activities. They may need the support of social workers, housing officers, work supervisors, psychologists, financial guardians, and perhaps judges, police, and probation officers as well. Each child/adult with FASD has individual needs, and the required support systems vary. Usually, these children and adults need a caretaker to arrange the necessary support: often, the parents continue this role into advanced age. In the best of cases, the role can be later taken over by members of the extended family, including spouses, or by social work or healthcare professionals.

Clinicians can support advocacy by relevant referrals, for example, referring parents to an organization which can help obtain a budget to buy in care, and providing research articles for the parents to give the orthodontist on orofacial anomalies with a goal of getting insurance coverage for care. The clinician can also refer parents to local parent-led support groups. Parents share information on local schools, a FASD diagnostic clinic in the area, or tips on financial planning for their adult children with FASD.

21.3.4 Sources of Support

Parents look first to their doctor for advice. However, the research shows that most professionals feel they have inadequate knowledge to provide support for management [19]. Thus, referring parents to other sources, such as social media groups or local organizations, can be helpful.

There are countless e-mail groups and Facebook groups, mainly run by parents. Research suggests that these groups can effectively provide information, emotional support, and share advice on raising children with special needs [20]. In the case of support groups for families living with FASD, it is clear that the flexibility and level of personal engagement is high. A big advantage of these groups is the availability 24 h a day, 7 days a week. Parents can often find a listening ear or get some good tips even on weekends or the middle of the night. Parents report that benefits include "blowing off steam," "getting advice on problems," and "gaining courage from success stories." Another big plus is the language and cultural specificity of

country-based groups: it is much easier to express emotions and difficult problems in your own language, and then to get advice which is relevant to the local health and social care systems. A respected international English language group is faslink (http://www.acbr.com/FAS/faslink.htm). There are also such e-mail groups, for example, in the Netherlands, in Germany, and in Spain. A clear disadvantage of this type of group is that the quality of information is variable, depending on who answers. Another problem is that while some of these groups serve as an adjunct to face-to-face support groups, in many cases there is no personal contact. This can lead to anonymity which can result in emotional outbursts and accusations. However, a strong moderator can keep these groups on track.

Formal associations and foundations exist in many countries, states, and regions around the world. These organizations are able to provide information, support, training, and materials in the local language and in a manner relevant to the local culture and healthcare systems. Research supports the value of these organizations in providing information, social support, and fostering advocacy [21]. It is common for the parent-led FASD organizations to provide websites, folders, flyers, and books geared to professionals as well as materials targeted to parents. Generally, formal groups maintain good contacts with professionals, organizing and attending conferences for researchers and parents, and these organizations usually have a scientific advisor or board. Thus, the information provided is likely to be of high quality. These groups are usually run by volunteer parents, which confers advantages and disadvantages. Parents may be very knowledgeable, based on own experience, following the scientific literature and attending conferences. They are very concerned about helping others. There is no cost for what is sometimes very extensive support, and support is available pretty much 24/7 via the social media and e-mail. The downside is that these parents may be asked to provide advice which should be the realm of professionals, although they may not have the training or the broad understanding that a professional should bring to the problem. Finally, these parents find the tasks fulfilling and worthwhile, but are at high risk of fatigue and burnout.

Box 21.1: Where to Find Support and Information

In many countries, there are strong local organizations that can provide information and support in various local languages. These can be found by searching on the Internet or checking with umbrella organizations, including the following.

- NOFAS (USA) www.nofas.org
- EUFASD Alliance (Europe) www.eufasd.org
- FASD Global (international) www.fasdglobal.org
- National FASD (UK) www.nationalfasd.org.uk

21.3.5 Planning for Adulthood: Work, Housing, Money, and Free Time

The adult with FASD is likely to experience difficulty in finding and holding a job [22], as well as with managing finances and locating and paying for housing. In many cases, a child has had a good support system until the age of 18, but then services stop abruptly. Thus, planning for the transition to adulthood is imperative, yet parents again face the hurdles of arranging the necessary supports. Experienced parents can support other parents in preparing their children with a disability for adulthood, with information, emotional support, and affirmation [23]. Planning should be made early, so supports are in place on the 18th birthday, to avoid a gap when childhood supports fall away. If the young adult is finished with school, then plans must be made for work or some sort of daily activity. The young adult with FASD is not ready socio-emotionally to live alone, so in many cases, the young adult with FASD lives with the parents or moves to a sheltered living environment. In some cases, parents may buy an apartment nearby for the adult child, so there is still supervision and contact, while the young adult has their own space. However, owning property may jeopardize the right to public financial support. As adults with FASD generally are very poor at managing money, arrangements have to be made, depending on the legal possibilities in the country, for such arrangements as a financial guardian to manage the finances and give weekly pocket money. Parents should find a knowledgeable financial advisor and prepare their testament with care. Depending on the legal possibilities in each country, it may be possible to leave an inheritance under control of a judge or financial guardian, so as to ensure wise usage of the property.

21.4 Needs of Siblings

Normally developing siblings in a family with a child with FASD may feel neglected and may suffer stigma from their peers. Anecdotal reports show that the healthy children in the family may feel that all the attention goes to the child with FASD and that they do not receive enough support themselves. A systematic review of interventions to support siblings of children with a chronic illness or disability found that some interventions reduced anxiety as well as benefiting mood and behavior. However, due to widely varying study approaches, it was not possible to identify the intervention characteristics that were beneficial [24]. Furthermore, the healthy child may be stigmatized for having an unmanageable and unpredictable brother or sister. Various organizations, either parent led or professionally led, provide some degree of support for healthy siblings, for example, picture books to explain FASD to a healthy young sibling.

21.5 Needs of the Child/Adult with FASD

Young persons and adults with FASD are individuals, all with their own hopes and aspirations for the future. Though the transition to adulthood is fraught with high risks of unemployment, broken relationships, and mental illness, these are considered secondary handicaps, that is, they are largely avoidable with good support [22]. It is currently rather rare to ask youth and adults with a disability what *they* want. However, the United Nations Convention on the Rights of Persons with Disabilities lays down some highly relevant principles: "Respect for inherent dignity, individual autonomy including the freedom to make one's own choices" and "Respect for the evolving capacities of children with disabilities" [25]. There is extensive discussion on how these principles can be applied to adults with mental disabilities, for example, by supported decision-making [26]. Due to slow maturation and poor planning capabilities, many teens and adults need extensive support in the transition to adulthood [27]. This support may include financial planning, support with finding and keeping work, and also support with sexuality and relationships. Research publications tend to focus on deviant or criminal behavior, rather than on supporting healthy sexuality. There is some research on fostering healthy sexuality in adolescents with a disability; recommendations for the clinician include anticipatory guidance, provision of contraception, and taking a broad view of sex education [28].

Self-advocacy groups can be supportive for adults with FASD: one study showed that participation in such a group promoted self-esteem and empowerment among adults with an intellectual disability [29]. Numerous organizations provide specific support groups for adults with FASD, both in person at family camps and weekends, as well as by Facebook (examples are FASD Deutschland, the Swedish FAS Förening).

Another issue is whether and how to inform employers, supervisors, neighbors, and others about the FASD diagnosis. Even just giving information about FASD can contribute to stigma. There has been little research on how to give information without promoting stigma [30]; thus, it is clear that some care and judgment should be exercised in giving information.

Box 21.2: Seven Tips for Parents and Families

- Get a clear diagnosis—ask your general practitioner for a referral to a diagnostic clinic.
- Obtain reliable information about FASD from one of the organizations listed above.
- Make contact with other parents, either via a local face-to-face group or an e-mail group.
- Recognize that the difficult behavior is due to brain damage, not ill will.
- Take care of yourself: get enough sleep, eat well, and exercise.
- Take care of your relationship and the other children: the difficult task of raising these children can be hard on a family.
- Involve the older child and adult in their own care and future plans.

21.6 Going a Further Step: Prevention and Political Action

Since there is no cure for FASD, many parents feel deeply that it is important to help prevent further cases of FAS and to become engaged in political action both to gain attention of the policy makers to prevention of FASD and to provide sufficient funding and services. Regarding prevention, many parents engage in activities on International FASDay, the ninth of September, each year. There are a plethora of available programs, posters, and actions in English but also in other languages. Surprisingly, there has been very little research on which programs are actually effective in prevention, and many well-meaning activities may be ineffective or contribute to stigmatizing persons with FASD and their birthmothers. Thus, there is a need to apply evidence-based principles of health promotion campaigns [31]. These principles include providing evidence-based information and also promoting self-efficacy with positive messaging [32]. Developing effective campaigns, therefore, implies that professionals and parent-led organizations should work together, harnessing the expertise and enthusiasm of both.

21.7 Why Should We Listen to Families?

Medical care for children is increasingly a matter of family-centered care [33]. From earliest days of FASD awareness, parents have been involved in dissemination of information about FASD (e.g., the book *The Broken Cord* [34] and the early website FAS Community Resource Center [35], which first went online in 1995). Research conferences on FASD are unique in the world of scientific conferences, involving parents, researchers, and adults with FASD as equals. Parents and parent-led organizations such as the NOFAS and the EUFASD Alliance are actively involved in research [7, 31] and dissemination of research. While medical research has traditionally been top-down, involvement of families and adults with FASD can be empowering [36]. Participatory research involves citizens directly in research: a recent example is the Australian involvement of members of the public in developing an instrument for diagnosis [37]. An example of research planned and carried out by adults with FASD and supported by researchers is an extensive international survey on health conditions among adults with FASD, presented at the seventh International Conference on FASD Research in Vancouver, Canada, in March 2017 [38].

21.8 Conclusion and Recommendations for Clinicians

Families living with FASD look to their doctors as the primary source of information on FASD. Thus, clinicians fill a key role in supporting families of children and adults living with FASD. The needs for support vary according to the family and the individual needs of the person. The following specific recommendations can be made.

- Provide evidence-based information to parents on what FASD is and on what it means in terms of behavior.
- Provide referrals to appropriate therapies as relevant: speech therapy, play therapy, psychologist, and special education.
- Become familiar with local parent-led support groups and parent-supported websites. Do they give good-quality information? Then refer parents to these groups as well.
- Be sensitive to needs of healthy siblings.
- Support teens and adults with FASD with information and referrals as necessary. Provide contraception.
- Offer to support local parent-led groups on their scientific advisory council.

Acknowledgments I thank the many parents of children and adults with FASD who shared their insights for this contribution, especially parents in the Dutch, German, and Spanish mailing groups.

References

1. Mukherjee R, Wray E, Commers M, Hollins S, Curfs L. The impact of raising a child with FASD upon carers: findings from a mixed methodology study in the UK. Adopt Foster. 2013;37:43–56.
2. Popova S, Lange S, Shield K, Mihic A, Chudley AE, Mukherjee RAS, Bekmuradov D, Rehm J. Comorbidity of fetal alcohol spectrum disorder: a systematic review and meta-analysis. Lancet. 2016;387:978–87.
3. Lange S, Shield K, Rehm J, Popova S. Prevalence of fetal alcohol spectrum disorders in child care settings: a meta-analysis. Pediatrics. 2013;132:e980–95.
4. Vonneilich N, Lüdecke D, Kofahl C. The impact of care on family and health-related quality of life of parents with chronically ill and disabled children. Disabil Rehabil. 2015;8288:1–7.
5. Paley B, O'Connor MJ, Frankel F, Marquardt R. Predictors of stress in parents of children with fetal alcohol spectrum disorders. J Dev Behav Pediatr. 2006;27:396–404.
6. Bobbitt SA, Baugh LA, Andrew GH, Cook JL, Green CR, Pei JR, Rasmussen CR. Caregiver needs and stress in caring for individuals with fetal alcohol spectrum disorder. Res Dev Disabil. 2016;55:100–13.
7. Corrigan PW, Lara JL, Shah BB, Mitchell KT, Simmes D, Jones KL. The public stigma of birth mothers of children with fetal alcohol spectrum disorders. Alcohol Clin Exp Res. 2017;41(6):1166–73. https://doi.org/10.1111/acer.13381.
8. Olson HC, Oti R, Gelo J, Beck S. "Family matters": fetal alcohol spectrum disorders and the family. Dev Disabil Res Rev. 2009;15:235–49.
9. Baskin J, Delja JR, Mogil C, Gorospe CM, Paley B. Fetal alcohol spectrum disorders and challenges faced by caregivers: clinicians' perspectives. J Popul Ther Clin Pharmacol. 2016;23:e114–30.
10. Tsang TW, Lucas BR, Carmichael Olson H, Pinto RZ, Elliott EJ. Prenatal alcohol exposure, FASD, and child behavior: a meta-analysis. Pediatrics. 2016;137:e20152542.
11. Petrenko CLM, Pandolfino ME, Roddenbery R. The association between parental attributions of misbehavior and parenting practices in caregivers raising children with prenatal alcohol exposure: a mixed-methods study. Res Dev Disabil. 2016;59:255–67.
12. Malbin D. Trying differently rather than harder. Diane Malbin; 2002.
13. Edmonton and Area Fetal Alcohol Network. Strategies not solutions. Edmonton, AB: Edmonton and Area Fetal Alcohol Network; 2007.

14. Bertrand J. Interventions for children with fetal alcohol spectrum disorders (FASDs): overview of findings for five innovative research projects. Res Dev Disabil. 2009;30:986–1006.
15. Petrenko CLM, Alto ME. Interventions in fetal alcohol spectrum disorders: an international perspective. Eur J Med Genet. 2016;60:79–91.
16. Ingersoll B, Wainer AL, Berger NI, Pickard KE, Bonter N. Comparison of a self-directed and therapist-assisted telehealth parent-mediated intervention for children with ASD: a pilot RCT. J Autism Dev Disord. 2016;46:2275–84.
17. Hundert AS, Huguet A, Green CR, et al. Usability testing of guided internet-based parent training for challenging behavior in children with fetal alcohol spectrum disorder (strongest families FASD). J Popul Ther Clin Pharmacol. 2016;23:e60–76.
18. Doig JL, McLennan JD, Urichuk L. "Jumping through hoops": parents' experiences with seeking respite care for children with special needs. Child Care Health Dev. 2009;35:234–42.
19. Gahagan S, Sharpe TT, Brimacombe M, Fry-Johnson Y, Levine R, Mengel M, O'Connor M, Paley B, Adubato S, Brenneman G. Pediatricians' knowledge, training, and experience in the care of children with fetal alcohol syndrome. Pediatrics. 2006;118:e657–68.
20. DeHoff BA, Staten LK, Rodgers RC, Denne SC. The role of online social support in supporting and educating parents of Young children with special health care needs in the United States: a scoping review. J Med Internet Res. 2016;18:e333.
21. Law M, King S, Stewart D, King G. The perceived effects of parent-led support groups for parents of children with disabilities. Phys Occup Ther Pediatr. 2001;21:29–48.
22. Streissguth ANNP, Young JK, Ed M. Risk factors for adverse life outcomes in fetal alcohol syndrome and fetal alcohol effects. Pediatrics. 2004;25:228–38.
23. Kingsnorth S, Gall C, Beayni S, Rigby P. Parents as transition experts? Qualitative findings from a pilot parent-led peer support group. Child Care Health Dev. 2011;37:833–40.
24. Hartling L, Milne A, Tjosvold L, Wrightson D, Gallivan J, Newton AS. A systematic review of interventions to support siblings of children with chronic illness or disability. J Paediatr Child Health. 2014;50:E26–38.
25. United Nations. Convention on the rights of persons with disabilities. 2006. http://www.un.org/disabilities/documents/convention/convoptprot-e.pdf.
26. Craigie J. A fine balance: reconsidering patient autonomy in light of the UN convention on the rights of persons with disabilities. Bioethics. 2015;29:398–405.
27. Lynch ME, Kable JA, Coles CD. Prenatal alcohol exposure, adaptive function, and entry into adult roles in a prospective study of young adults. Neurotoxicol Teratol. 2015;51:52–60.
28. Holland-Hall C, Quint EH. Sexuality and disability in adolescents. Pediatr Clin North Am. 2017;64:435–49.
29. Clarke R, Camilleri K, Goding L. What's in it for me? The meaning of involvement in a self-advocacy group for six people with intellectual disabilities. J Intellect Disabil. 2015;19:230–50.
30. De Pietro N, De Vries J, Paolozza A, Reid D, Reynolds JN, Salmon A, Wilson M, Stein DJ, Illes J. Ethical challenges in contemporary FASD research and practice. Camb Q Healthc Ethics. 2016;25:726–32.
31. Roozen S, Black D, Peters G-JY, Kok G, Townend D, Nijhuis JG, Koek GH, Curfs LMG. Fetal alcohol spectrum disorders (FASD): an approach to effective prevention. Curr Dev Disord Rep. 2016:10–5.
32. France KE, Donovan RJ, Bower C, Elliott EJ, Payne JM, D'Antoine H, Bartu AE. Messages that increase women's intentions to abstain from alcohol during pregnancy: results from quantitative testing of advertising concepts. BMC Public Health. 2014;14:30.
33. Schor EL, American Academy of Pediatrics Task Force on the Family. Family pediatrics: report of the task force on the family. Pediatrics. 2003;111:1541–71.
34. Dorris M. The broken cord. New York: Harper Perennial; 1990.
35. Kellerman T. FAS Community Resource Center. http://www.come-over.to/FASCRC/.
36. Powers LE, Garner T, Valnes B, Squire P, Turner A, Couture T, Dertinger R. Building a successful adult life: findings from youth-directed research. Exceptionality. 2007;15:45–56.

37. Jones HM, McKenzie A, Miers S, et al. Involving consumers and the community in the development of a diagnostic instrument for fetal alcohol spectrum disorders in Australia. Health Res Policy Syst. 2013;11:26.
38. Himmelreich M, Lutke CJ, Travis E. The lay of the land: final results of a health survey of 500+ adults with diagnosed FASD. In: 7th International Conference on fetal alcohol spectrum disorders research, results and relevance; 2017.

The Importance of the Multidisciplinary Approach

22

Jon Skranes and Gro Christine Christensen Løhaugen

Chapter Highlights

- The role and responsibility of different professionals in an MDT for FASD.
- An overview of assessment and treatment approaches to FASD.
- Support approaches for FASD.

22.1 Background

It is well established that a multidisciplinary team is crucial in the early and comprehensive assessment and diagnosing children with FASD [1, 2]. Several classification systems for the diagnosis of FASD have been developed and implemented in clinical care. However, when it comes to clinical management, research is lacking with regard to both standardized tailored management programs and recommendation plans/guidelines for the comprehensive provision of healthcare, educational, and social services.

The South-Eastern Health Region of Norway established a regional resource center offering services to children and adolescents aged 2–18 years with prenatal alcohol and/or illegal drug exposure. The resource center is the first of its kind in Scandinavia and has two main objectives: (1) Provide specialized health services for children referred from hospitals in the South-Eastern Health Region of Norway for diagnostic services and intervention planning. (2) Provide information, educational courses, and seminars focusing on the identification, diagnostic process, and treatment planning for children with a history of prenatal alcohol/drug exposures. Much of the content in this chapter is based on experience-based knowledge from working with patients and families in this resource center.

J. Skranes · G. C. C. Løhaugen (✉)
Regional Competence Center for Children with Prenatal Alcohol and/or Drug Exposure, South-Eastern Health Region, Department of Pediatrics, Sørlandet Hospital, Arendal, Norway
e-mail: jon.skranes@sshf.no; gro.lohaugen@sshf.no

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_22

In most FASD cases, a multidisciplinary approach to management is necessary since the child may present with deficits and impairments in several organ systems. The clinical consequences may include medical, physical, cognitive, neuropsychological, adaptive, and behavioral/mental health problems. It is therefore important to establish a ***core team of professionals*** that work together in coordinating the child's support needs, with a common understanding of the child's different and sometimes hidden disabilities. A multidisciplinary management will be able to detect and treat impairments in different domains at an early time point as they appear through childhood and adolescence. In addition, different professionals working together and thereby adding their specific competence are essential for making effective treatment recommendations, implementing needs, and ensuring access to relevant resources. A successful approach to management therefore includes the involvement of different professionals in the treatment of the child with FASD and support for the family.

22.2 Organizing a Multidisciplinary Approach to Management: Teams at Different Levels

It would be most beneficial for the child and the family to organize the management by establishing multidisciplinary teams at different levels of proximity to the daily functioning of the child and family. A core team of professionals should be the basis for the management, while a supplementary team of specialists could assist in more specific problems that are present or may appear with time.

22.2.1 The Core Team

The core team is the group of professionals working most closely with the family for treatment recommendations and implementation. The core team may vary according to available health resources and the age and clinical picture of the specific child and family, but would ideally include the following persons:

The Core Team

- The family physician/primary care physician/pediatrician.
- The child development specialist, for example, a physiotherapist, occupational therapist, speech-language pathologist, or a clinical psychologist.
- The main educator/special education teacher in kindergarten or school.
- The core team leader: A key support person who knows the family well and who understands the child's comprehensive needs. This person should secure good communication between medical, educational, and community services and other professionals. This could be the social worker in the community or the public health nurse.

The main responsibilities and relevant tasks for the different professionals in the core team are presented in Table 22.1.

Table 22.1 Members of ***the core team***: their responsibilities and tasks in management

Core team member	Responsibilities and tasks in management
Primary care practitioner/family physician	• Execute treatment and/or referral to specialist services for medical treatment • Consider start treatment of mental health comorbidities or refer to child psychiatrist • Assess and treat any feeding problems or refer to a nutritionist • Assess and treat any growth deficiency or refer to a pediatrician • Assess and treat any sleeping disorders • Treat comorbidities like epilepsy or refer to a child neurologist • Follow-up of sensory defects (hearing and vision) or refer to ophthalmologist and ENT doctor • Create a management plan for the child with regular follow-up visits • Involve other medical professionals when necessary
Child development specialist (could be a clinical psychologist/ neuropsychologist, physiotherapist, occupational therapist, or speech)	• Assess level of psychomotor development and cognition – Denver developmental screening test – Age-appropriate Wechsler IQ test (WPPSI/WISC/ WAIS/WASI) • Assess neuropsychological deficits and impairments in the following cognitive domains: Attention/ executive, language, visual motor, sensory motor, learning, and memory. Performing the assessments requires that professionals be experienced so that they can adapt the methods and sequence of examinations to the child's focus and as their motivation changes – Neuropsychological test battery: NEPSY-II battery, Delis-Kaplan, Beery-Buktenica developmental test of visual-motor integration, clinical evaluation of language fundamentals (CELF), sensory profile • Assess executive functions in daily living – The behavior rating inventory of executive function – BRIEF questionnaire • Assess adaptive functioning – Vineland adaptive behavioral scale • Based on the assessments above: Develop an individualized pedagogic and social program based on the child's strength and weaknesses for use in kindergarten and school

(continued)

Table 22.1 (continued)

Core team member	Responsibilities and tasks in management
Main educator at school or in kindergarten	• Initiate early intervention program in kindergarten • Execute implementation of educational support/ pedagogic program/special services in kindergarten or school, in collaboration with speech therapist to help with learning and behavior issues • Facilitate learning by offering the child small-group teaching, shorter work sessions, increased amount of repetitions, teaching at age-appropriate level, and avoid too much homework • Initiate PC use in school work, especially if problems with writing due to fine motor and eye-hand coordination problems and/or slow processing/motor speed
Social worker or public nurse	Could be the coordinator in the core team: • Secure good communication between medical, educational, and community services • Help families access appropriate resources, for instance, adequate social care/funding and arrange respite care for families • Help working out an individual habilitation plan for the child Public nurse: • Perform well-baby care, secure good nutrition and hygiene, monitor growth, and give advices for any feeding problems and sleep disorders • Be in charge of the child's vaccination program • Recommend appropriate vitamin and nutritional supplements • If the child is still living with his/her birth mother with a dependency problem, encourage treatment of the mother's alcoholism to enable better parenting and to prevent future pregnancies from being affected

22.2.2 The Supplementary Team

Depending on the child's age, symptoms, and problem areas, a supplementary team of professionals may be engaged in the patient and his/her family in addition to the core team. This may include (depending on what is "missing" in the core team) the following:

The Supplementary Team
- A clinical psychologist or a neuropsychologist.
- A physiotherapist or an occupational therapist.
- A speech-language therapist.

See Table 22.2 for responsibilities and relevant tasks for the members of the supplemental team.

Table 22.2 Members of ***the supplemental team***: their responsibilities and tasks in management

Supplemental team member	Responsibilities and tasks in management
Psychologist/ neuropsychologist (if not in core team)	• Assess level of psychomotor development and cognition • Assess neuropsychological deficits and impairments in the following cognitive domains: Attention/executive, language, visual motor, sensory motor, learning, and memory (NEPSY-II, Delis-Kaplan, Beery-Buktenica developmental test of visual-motor integration, clinical evaluation of language fundamentals (CELF), sensory profile) • Assess executive functioning in daily functions (the BRIEF questionnaire) • Assess adaptive functioning (the Vineland adaptive behavior scales second edition) • Assess the foundations of social perception (the NEPSY-II for children between 2 and 16 years of age) • Based on the assessments above: Develop an individualized pedagogic and social program based on the child's strength, skills, and weaknesses for use in kindergarten and school • Perform cognitive and neuropsychological assessments at regular intervals • Offer behavioral intervention programs • Offer family and network education
Physiotherapist (if not in core team)	• Evaluate motor function (movement ABC) • Assess visuo-motor function (Beery VMI) • Evaluate adaptive functioning (VABS) • Start early intervention program if sensory-motor or motor problems • Initiate motor program to improve gross and fine motor functioning in kindergarten • Monitor and perform follow-up of any foot deformities, leg length discrepancy (anisomelia), and scoliosis that are common in children with FASD
Occupational therapist (if not in core team)	• Assess level of psychomotor development (Denver DST) • Evaluate fine motor function (movement ABC) • Assess visuo-motor function (Beery VMI) • Assessment of sensory issues ***(see later chapter)*** • Evaluate adaptive functioning (VABS) • Help the child to independently carry out everyday tasks with more confidence and independence by suggesting changes to the environment, at home, kindergarten, or school • Participate in individualized treatment programs based on present functioning and training to achieve the "next step" in development • Introduce the use of special equipment which will help with daily activities

(continued)

Table 22.2 (continued)

Supplemental team member	Responsibilities and tasks in management
Speech and language pathologist (if not in core team)	• Assessment of language function should be performed at level of basic language functions like phonological processing but also focus on higher order aspects of language as understanding metaphors, the ability to understand abstract concepts, and the ability to extract the main points from a conversation or lecture • In pre-school age children: Assessment focusing on vocabulary and comprehension to evaluate language development • In school-aged children: Assessment of reading speed and reading comprehension in addition to tests of grammar and spelling to ensure realistic level of expectations • For older children: Complete wider communication assessments including communication passports and identify communication disorders • Initiate early intervention program if speech delay or language disorders • Monitor special education programs geared toward the child's specific needs and learning style
Child psychiatrist	• Assessment of behavioral problem • Diagnosing of ADHD, which is a common comorbidity in children with FASD • Responsible for the psychiatric evaluation that includes assessing the FASD child's behavioral and mental health (CBCL, ADHD rating scale, PSI) • If the diagnostic criteria for attention deficit hyperactivity disorder (ADHD) or other mental health conditions are met, the appropriate recommendations regarding treatment should be outlined • Recommend and start appropriate medication of children with FASD with psychiatric comorbidities ***(see later chapter about medication)*** • Treat any mental health comorbidities in collaboration with primary care practitioner/family physician ***(see later chapter about ADHD)***

However, some children may also be in need of more specialized medical treatment and care. A ***secondary team of medical specialists*** with competence in pediatrics, child psychiatry, ophthalmology, and ENT diseases may therefore also be involved in the treatment and regular follow-up of the child. If the child also present with congenital malformations in other organ systems, such medical specialists may also be part of the specialist team.

Other health specialists on demand depending on medical issues may include the following:

(a) Eye doctor/ophthalmologist: Diagnose and treat any eye abnormalities and visual disorders (strabismus, ptosis, reduced vision) .
(b) Otolaryngologist/audiologist: Diagnose and follow-up of mechanical and neurologic hearing loss.
(c) Pediatrician: Assist the family physician in the medical treatment of the child. In the initial phase: rule out other diagnoses (genetic syndromes, ADHD, autism).

(d) Child neurologist/neurologist: Treat epilepsy and other neurological conditions like autism specter disorders.
(e) Mental health professionals (child psychiatrist and psychologist, school psychologist, behavior management specialist): Offer psychiatric and behavioral treatment.
(f) Others: Dysmorphologist, immunologist, plastic surgeon, endocrinologist, gastroenterologist, and cardiologist.

22.3 Types of Treatments and Interventions in Follow-Up

There is no cure or specific treatment for fetal alcohol spectrum disorders. The physical defects and cognitive, behavioral, and mental health deficiencies typically persist for a lifetime. However, early diagnosis and intervention services can help reduce some of the effects of FASD and hopefully prevent some of the secondary disabilities that may result. Since the clinical picture will vary between patients with FASD, the types of treatments and intervention offered will be different for each person. Although several research papers have documented that early diagnosis and intervention is beneficial for the child with FASD [3, 4], very limited research exists about which exact management techniques and intervention programs that will provide most benefits [5–9]. In addition, there is a lack of standardized follow-up programs for this patient group and their family.

Table 22.3 suggests the types of treatment and intervention that could be offered by the different professionals during the follow-up of children with FASD. Some of the main treatment options and who should be in charge are listed.

22.3.1 Medication (See Chap. 28 for More Details)

Although there is no medication that specifically treats FASD, several of the symptoms of FASD and from comorbid and secondary conditions may be relieved by different medications. This could include medications to minimize severe behavioral consequences, to give a child better control over unwanted behaviors, or to facilitate the ability to better functioning at home, in the classroom, and in the community with friends and family. The primary care physician/family practitioner could manage this either alone or in collaboration with a pediatrician or a child psychiatrist to search for the optimal, individualized medication plan.

22.3.2 Psychological and Social Intervention: Behavior Therapy and Education

The clinical psychologist, the family counselor, or the child psychiatrist should offer psychological and social intervention. Behavioral problems are frequently reported in children with FASD, and the Child Behavior Checklist (CBCL) could be

Table 22.3 Types of treatment/intervention and follow-up that could be offered by the different professionals

Type of professional	Treatment and follow-up
Primary care physician/practitioner	• Medical intervention includes medication of: – ADHD: Stimulants and non-stimulants – Behavioral problems: Neuroleptics, antipsychotics, antianxiety drugs, and antidepressants – Sleep disorders: Antihistamines, melatonin – Epilepsy: Antiepileptic drugs • Medication may be administered in collaboration with child psychiatrist or pediatrician • Regular follow-up visits with focus on ongoing medication, any growth deficiencies, eating disorders, and sleep disturbances
Clinical psychologist/ family counselor/child psychiatrist	• Psychological and social intervention – Behavioral therapy – Education intervention/counseling: Behavioral modifications, psychotherapy to work through emotions and past trauma, psychotherapy to work on early attachment disturbances, and social interventions like environmental modifications (e.g., visual cues), parent training in groups or as individual counselling – Regular support of families at follow-up • Repeat cognitive and neuropsychological testing, for instance, at 10 years • Repeat adaptive assessment at age 10 years with, for instance, the VABS to reveal any persisting discrepancy in function and cognition. Interventions should be tailored based on functional level. The VABS may also be performed before entering high school and before entering adulthood

used to screen for behavioral and/or psychiatric problems at regular intervals during follow-up. Behavior therapy and education intervention are important parts of the treatment plan for children and adolescents with FASD. Counseling and educating intervention may be more beneficial than medication in reducing behavior problems, reducing aggression, and avoiding academic failure. Families with a child with FASD will need regular support from a psychologist, a family counselor, or another therapist, for instance, a child psychiatrist. Issues for these types of intervention would include behavioral modifications, psychotherapy to work through emotions and past trauma, group/family therapy, psychotherapy to work on early attachment disturbances, and social interventions like environmental modifications (e.g., visual cues).

In addition to professional help and guidance, local support groups of parents of FASD children or access to specific web sites and discussion groups (see, for instance, http://www.fasportalen.se/ or http://www.faslink.org/) may be very helpful for families where they can discuss concerns, ask questions, get advices from people in similar situations, and thereby find solutions and gain encouragement. Parents with a child with FASD may experience that usual parenting practices do not work, and they therefore need to be taught alternative ways of parenting with

their child. Our experience is that parental strategy involving negative consequences and punishment does not work. Neither does asking and discussing with the child why he/she acted as they did. This may be related to problems with understanding cause-effect, lack of ability to learn from experience, and inability to control impulses.

Parent training by professionals has been successful in several ways: to educate parents about the different aspects of FASD, to teach them how to stimulate and encourage their child's strengths and skills and overcome the weaknesses, and to help them cope with everyday behavioral or social challenges [5, 8, 9]. Parent training can be organized in groups or as individual counseling. Although each child and family is unique, the following parenting tips can be helpful as general guidelines:

Parenting Tips

1. Concentrate on your child's strengths and talents, to further improve function.
2. Accept, but do not focus on your child's weaknesses and limitations.
3. Be consistent and thereby predictable for your child (discipline, school, and behaviors).
4. Use concrete language and examples that are familiar to the child.
5. Be stable in your daily routines, do not change daily.
6. Keep messages and language short and simple, that is, easy understandable for the child.
7. Be specific and clear in what you mean, the child will not understand "hidden messages."
8. Structure your child's world to make daily living predictable.
9. Use visual aids, multimedia elements, and practical activities to facilitate learning.
10. Use positive reinforcement (praise and incentives); punishment has no preventive effect.
11. Supervise: friends, visits, and routines.
12. Remember that your child needs several repetitions to learn.

22.3.3 Ensure a Common Understanding of the Child's Diagnosis, Deficits, and Different Needs

In our experience, the parents, kindergarten/school, and primary service providers often have diverging opinions and experience regarding the child with FASD. This is especially frequent in younger children as behavioral challenges often present at home, and this may complicate the cooperation needed to provide efficient interventions. Establishing a common understanding of FASD as a medical condition that is caused by a brain disorder/damage and the individual child's function is therefore critical.

After diagnosis, information to the family, network, and primary health care should be offered. First, a meeting with the diagnostic team should be offered to the guardians and those that referred the child for assessment. Conclusions from the multidisciplinary assessments should be explained, and a plan for treatment and follow-up should be outlined. Depending on the child's age and function, one should consider offering a feedback to the child together with the guardians.

22.4 Follow-Up Program

A child with FASD has a chronic disorder that will not disappear with age. On the contrary, impairments and deficits may be more pronounced with time and the gap between the child and healthy age-matched children will increase, especially with regard to adaptability and the need for parental support and guidance. It is therefore imperative that the child and the family are offered a follow-up program with regular visits at the family physician or a pediatrician who knows the child.

22.4.1 Focus Areas of Follow-Up

Repeated Cognitive and Neuropsychological Testing

If the child is diagnosed in preschool age, executive functions are less developed, and therefore retesting should be scheduled at a later age and conclusions drawn cautiously at time of diagnosis. It has been reported that even children with full FASD do not display the full range of cognitive problems if diagnosed at preschool age, emphasizing the need for retesting at follow-up. As such, even if the child does not obtain scores indicating neuropsychological deficits at time of diagnosis, a retesting should be performed before concluding about cognitive functioning. This retesting should be performed by a trained clinical psychologist or a neuropsychologist.

For children diagnosed in preschool age, we suggest cognitive and neuropsychological retesting at about 10 years of age. At this time point, the curriculum in school is changing in many countries from focusing on learning how to read—to be able to read to remember what you are reading. In addition, the content of the curriculum is becoming increasingly abstract with less room for visualization and experience-based learning. This represents increased demands on language as well as on executive functions. Given the high prevalence of attention/executive function deficits and language deficits, especially receptive language disorder, neuropsychological assessment is indicated.

Time Points for Follow-Up

At certain time points, a more comprehensive multidisciplinary assessment should be performed to evaluate development, functioning in daily life activities and whether enough resources have been offered to the family with regard to social benefits and relief services. Appropriate time points for more comprehensive evaluations of the child are listed below, also included are suggestions regarding methods.

(a) *Before entering school*
- IQ, language.
- Vineland Adaptive Behavior Scale, including questions regarding maladaptive behavior that may indicate the need for Child Behavior Checklist.
- Behavior Rating Executive Functions preschool (BRIEF).
- Gross and fine motor (visual motor).
- Medical (nutrition/eating, sleep).
- Parental Stress Index.

(b) *At age 10 years*—when the demands in school change from more practical teaching to more abstract thinking
- IQ, attention/executive, language, learning, and memory. Visual-motor/sensory assessment by neuropsychologist or occupational therapist/physiotherapist.
- Academic assessment, especially reading speed and comprehension, math.
- Vineland Adaptive Behavior Scales.
- Behavior Rating Executive Functions (BRIEF, guardians, and teacher).
- Child Behavior Checklist.
- Parental Stress Index.
- Medical assessment if indicated.

(c) *Before entering high school*
- IQ, selected neuropsychological tests based on results from last assessment to monitor development/stagnation.
- Academic assessment.

(d) *Before entering adulthood*
- IQ and full neuropsychological assessment.
- Vineland Adaptive Behavior Scales.
- Behavior Rating Executive Functions (BRIEF, guardians and teacher, self-report).

Table 22.4 summarizes recommended assessment methods at the different follow-up time points. The extent of the assessment is dependent on the current function of the child and the questions at hand from guardians or other members of the core team to be addressed.

Table 22.4 Cognitive/neuropsychological tests and assessment methods for follow-up of children with FASD

Domain	Methods and subtests	Age (years)
General cognitive ability (IQ)	Wechsler tests (WPPSI/WISC/WAIS)	2–18
	Nonverbal IQ (Wechsler nonverbal or the Leiter test)	2–18
Attention/executive function	NEPSY-II Animal sorting, auditory attention and response set, inhibition, and statue	2–16
	Delis-Kaplan: Trail Making Test 1–5, color-word interference test, tower test	6–18
	Conners Kiddie Continuous Performance Test second Edition™ (Conners K–CPT 2™) assesses	4–7
	Conners Continuous Performance Test third Edition™ (Conners CPT 3™)	8+
	Behavior Rating Inventory of Executive Function (BRIEF) questionnaire parent/teacher/self-report	5–18
Language function	NEPSY-II: Comprehension of instructions, phonological processing, repetition of nonsense words	2–16
	Clinical Evaluation of Language Fundamentals – Preschool-2 (CELF-Preschool-2)	3–7
	Clinical Evaluation of Language Fundamentals – Fourth edition	5–13
Visual-motor function	Beery-Buktenica developmental test of visual-motor integration, sixth edition (Beery VMI)	2–18
	NEPSY-II: Arrows, geometric puzzles, picture puzzles	2–16
	Vineland Adaptive Behavior Scales, second edition. Motor domain, questionnaire/interview	2–6
Sensory-motor function	Fingertip tapping, imitating hand position, visual-motor precision	2–16
	Sensory profile, second edition	0–15
Social perception	NEPSY-II: Affect recognition and theory of mind	2–16
	Vineland Adaptive Behavior Scales, second edition. Social function domain	2–18
Learning and memory function	NEPSY-II: Word list, memory for faces, narrative memory, sentence repetition, word list interference	2–16
	Wechsler Memory Scale-III/IV	16–18
Activity of daily life	Vineland Adaptive Behavior Scales, second edition	2–18
Supplemental assessment	Child Behavior Checklist (CBCL) Parenting Stress Index, fourth edition	1, 5–18 0–12

Practice Points

(a) The management of a child with FASD should be multidisciplinary.
(b) One should create a core team of 3–4 professionals (family physician, a child development specialist, the main educator/special education teacher in school or kindergarten, and a social worker or the public health nurse) around the child and family.
(c) A supplemental team of professionals is often necessary to establishment depending on existing resources in the core team.
(d) With regard to management and follow-up, it is important to focus on more than "pure" medicine: The cognitive, behavioral, and adaptive problems may be far more important to solve/compensate than any medical issues.

References

1. Cook JL, Green CR, Lilley CM, Anderson SM, Baldwin ME, Chudley AE, Conry JL, LeBlanc N, Loock CA, Lutke J, Mallon BF, McFarlane AA, Temple VK, Rosales T, Canada Fetal Alcohol Spectrum Disorder Research Network. Fetal alcohol spectrum disorder: a guideline for diagnosis across the lifespan. CMAJ. 2016;188(3):191–7. Epub 2015 Dec 14. Review. https://doi.org/10.1503/cmaj.141593.
2. Chudley AE, Conry J, Cook JL, Loock C, Rosales T, Le Blanc N, Public Health Agency of Canada's National Advisory Committee on Fetal Alcohol Spectrum Disorder. Fetal alcohol spectrum disorder: Canadian guidelines for diagnosis. CMAJ. 2005;172(5 Suppl):S1–S21.
3. Streissguth AP, Bookstein FL, Barr HM, Sampson PD, O'Malley K, Young JK. Risk factors for adverse life outcomes in fetal alcohol syndrome and fetal alcohol effects. J Dev Behav Pediatr. 2004;25(4):228–38.
4. Green CR, Cook J, Salmon A, Canada Fetal Alcohol Spectrum Disorder Research Network. Why is FASD diagnosis important? 2015. https://canfasd.ca/wp-content/uploads/sites/35/2016/05/FASD-and-Dx-EN.pdf.
5. Bertrand J. Interventions for Children with Fetal Alcohol Spectrum Disorders Research Consortium Interventions for children with fetal alcohol spectrum disorders (FASDs): overview of findings for five innovative research projects. Res Dev Disabil. 2009;30(5):986–1006. Epub 2009 Mar 26. https://doi.org/10.1016/j.ridd.2009.02.003.
6. Koren G, Zelner I, Nash K, Koren G. Foetal alcohol spectrum disorder: identifying the neurobehavioural phenotype and effective interventions. Curr Opin Psychiatry. 2014;27(2):98–104.
7. Paley B, O'Connor MJ. Intervention for individuals with fetal alcohol spectrum disorders: treatment approaches and case management. Dev Disabil Res Rev. 2009;15(3):258–67. https://doi.org/10.1002/ddrr.67.
8. Reid N, Dawe S, Shelton D, Harnett P, Warner J, Armstrong E, LeGros K, O'Callaghan F. Systematic review of fetal alcohol spectrum disorder interventions across the life span. Alcohol Clin Exp Res. 2015;39(12):2283–95. Epub 2015 Nov 18. https://doi.org/10.1111/acer.12903.
9. Kully-Martens K, Denys K, Treit S, Tamana S, Rasmussen C. A review of social skills deficits in individuals with fetal alcohol spectrum disorders and prenatal alcohol exposure: profiles, mechanisms, and interventions. Alcohol Clin Exp Res. 2012;36(4):568–76. Epub 2011 Oct 21. https://doi.org/10.1111/j.1530-0277.2011.01661.x.

Understanding and Improving Sleep for Children with FASD

23

Fiona Aiton and Ruth Silverman

Chapter Highlights

- Understanding sleep and sleep difficulties
- Approaches to assessing sleep difficulties
- Approaches to managing sleep difficulties with relevance to FASD.

We know that sleep problems affect 20–30% of all children during childhood and that disturbed or poor sleep affects many aspects of daytime functioning. Poor sleep is known to affect memory consolidation, cognitive function, academic achievement, daytime behaviour and even levels of obesity [1].

Sleep problems for children with neurodevelopmental disorders are well documented. Children with FASD are highly likely to have difficulties around sleep. Evidence suggests that around 85% of children aged 4–12 years with a diagnosis of FASD have a clinically significant sleep disorder [2].

Babies and toddlers with FASD may have a fragmented sleep pattern. They are prone to irritability and poor settling. Older children and adolescents are likely to present with difficulty in establishing and maintaining sleep routines, settling problems at bedtime, separation anxieties and night waking.

This section briefly describes the physiology of sleep, a guide to considerations to include when undertaking sleep work, gives information about good sleep hygiene and suggests behavioural approaches to sleep relevant for children with FASD.

F. Aiton (✉) · R. Silverman
Child Development Services, Sussex Community Foundation NHS Trust, Brighton, UK
e-mail: fiona.aiton@nhs.net; ruthsilverman@nhs.net

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_23

23.1 Defining Sleep

It is important to have a general understanding of normal sleep and its functions when working with families with children with FASD.

Sleep is 'a condition of body and mind which typically recurs for several hours every night, in which the nervous system is inactive, the eyes closed, the postural muscles relaxed and consciousness practically suspended' [3].

Sleep serves a function for:

- Restoration and repair.
- Growth.
- Evolutionary and energy conservation.
- Brain function, memory consolidation and learning.

Sleep difficulties/disorders can be classified by a subjective account of difficulty in initiating or maintaining sleep or early morning wakening with at least one related daytime impairment, such as fatigue, reduced attention issues or mood disturbance, maintained for a 3-week period over 3 months [4].

Chronic or severe sleep disturbances can contribute to a range of difficulties for both the child and the wider family leading to high levels of stress, family conflict, depression, marital breakdown and disruption for siblings. With the complex difficulties encompassed by a diagnosis of FASD, it is not surprising that these problems can become augmented with poor outcomes.

Children with FASD with sleep issues may present as increasingly hyperactive over a period of time, prone to more aggressive outbursts, have increased sensory needs and generally have less consistent executive functioning.

Mindell and Moore [5] suggest that there is a complex relationship between sleep and development and suggest that the best measures of sleep quality are likely to be a day-to-day functioning and skill development assessment within context of the family situation.

In practical terms, the three-pronged approach used by Durand [6] adapts well when discussing and planning sleep work with parents and carers and children within the context of FASD:

1. Physiological
2. Emotional
3. Behavioural

23.2 Physiological Considerations

Homeostatic sleep drive and the circadian rhythm.

The homeostatic sleep drive (process S) is our natural biologically driven need to sleep after being awake for a certain number of hours.

The circadian rhythm (process C) is also known as the 'body clock'. This is the regulation of the body's internal processes and alertness levels, governed by the

body's internal biological clock. The circadian rhythm is influenced by external stimuli, in particular, light and darkness. These are known as zeitgebers. The circadian rhythm works on a 24-h sleep/wake cycle. Research has shown [7] that without the stimulus of natural daylight, the natural cycle would be longer than this (probably around 25 h).

The Cortisol and melatonin hormonal sleep/wake cycle:

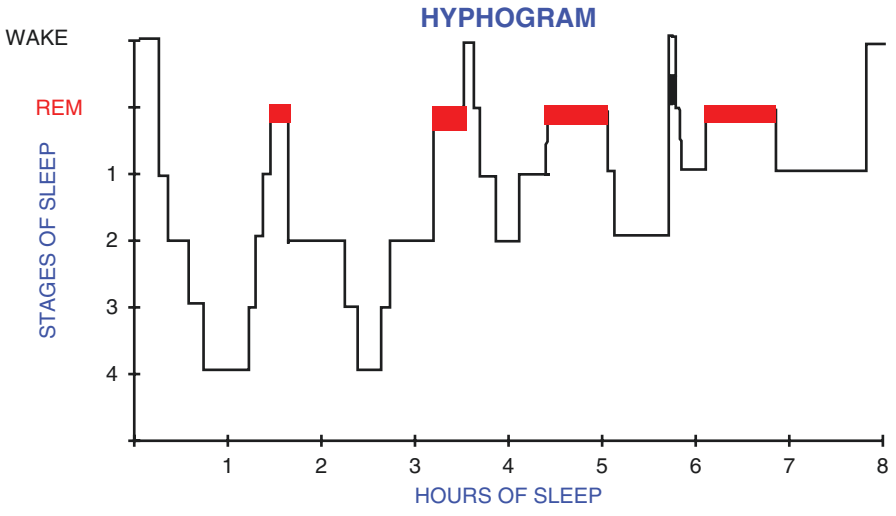
Melatonin (often described as the sleep hormone) is produced by the pineal gland located in the epithalamus. Melatonin reduces core body temperature, reduces motor activity and induces fatigue—it does *not* induce sleep. Natural melatonin production increases after 6 p.m. in the evening, peaks around midnight and then is gradually reduced over the rest of the night.

It is now well established that melatonin production is hindered by light, which also includes the blue light emitted from screens/tablets and other electronic devices.

Cortisol is glucocorticoid steroid, produced from the adrenal cortex of the adrenal gland. Cortisol presents also in a diurnal rhythm to the melatonin cycle. Generally, production is higher in the mornings than in the evenings—the lowest levels being produced from midnight to 4 a.m. Stress and exercise are also known to increase production. High levels of blood cortisol also affect immune responses and an increase in appetite cravings for high calorie foods. Core temperature drops once cortisol production is reduced. High cortisol can contribute to disrupted sleep/sleep deprivation as it induces an alert state and a surge induces the fright/fight state.

23.2.1 Hypnogram

The hypnogram is a pictorial representation of the physiological sleep phases which occur during the night. It records sleep phases in detail so that sleep patterns can be assessed and analysed. A normal hypnogram will identify 4/5 cycles of sleep per night lasting from around 90–110 min.



A hypnogram is a useful tool to use with parent/carers to visually show what sleep look likes and aids discussion about sleep patterns and night wakenings.

Sleep can be divided into rapid eye movement (REM) and non–rapid eye movement (NREM). During the first part of the night, we spend much of the time in NREM and deep sleep, and in the latter part of the night, more time is spent in lighter REM sleep. During sleep, it is common to have 'partial wakening', and these momentary wakenings are normal and usually occur between cycles. Most people are not aware of these short wakenings and return immediately to sleep.

23.2.2 Parasomnia/Dyssomnia

- Parasomnias are sleep disorders that result in abnormal activities during sleep, for example, night terrors, night arousals or sleep walking. These usually only occur in the first few hours of non-REM deep sleep. However, nightmares occur in REM sleep later in the night.
- Dyssomnia are sleep disorders where there is difficulty in falling asleep and/or staying asleep.

23.2.3 Common Dyssomnia-Related Problems in Children with FASD

1. Sleep-onset delay.
2. Night wakening.
3. Early rising/poor sleep duration.

Infants with FASD have been shown to have evidence of disturbed sleep patterns even from the early days of life. This presents as restlessness/more muscular movements, easy arousal and more awake periods.

In older children with FASD, it is more likely that fragmented sleep will gradually become more obviously recognisable, with easily disrupted sleep and likely abnormal melatonin production.

There are many ways to approach assessment of sleep in these children. Professionals need to consider the best approach for their practice. This can include the following:

- Taking a clear detailed history.
- Carrying out a baseline medical examination.
- Using sleep diaries for information.
- Using sleep assessment tools such as BEARS and Epworth Sleepiness Scale [8].
- Using a polysomnography to analyse sleep.

It is always important to consider any medical issues that could be impacting on sleep, for example: constipation, enuresis, teething or dental pain, illness, snoring/obstructive sleep difficulties, diet deficiencies and visual and hearing difficulties.

Other sleep problems such as parasomnias, rhythmic movement disorders, restless legs syndrome and sleep apnoea are not addressed in this chapter. Any unusual queries raised from the sleep assessment should be referred onto relevant professionals for further analysis. Seizure activity may also sometimes need to be ruled out.

23.3 Emotionally-Related Considerations

The emotionally-related considerations which impact on sleep can be due to the impact of anxiety/stress/familial and environmental issues.

Consider sensory issues—as discussed elsewhere in this book—this is an essential part of assessment to gain optimum opportunity for induction of sleep.

First trimester exposure to alcohol has been shown to cause damage to the central brain and limbic system. These areas of the brain help control self-regulation, arousal and emotional intensity.

Some children with FASD may be living with foster carers or other family members and may also present with complex attachment issues or have been exposed to domestic abuse or neglect. Night-time usually means a period of separation from the main care giver, and this can cause anxiety for some children. Acknowledgement of such needs should underpin any sleep work.

Increased anxiety will in turn lead to increased arousal, and it is likely to present as difficulties around sleep. This may present as bedtime resistance, night waking, delayed sleep onset and early morning rising.

Children with FASD have complex individual varied emotional needs, as discussed elsewhere in this book.

23.4 Behavioural Approaches

Behavioural approaches are often referred to as ‘sleep hygiene’. Behavioural approaches to managing sleep difficulties in children are often the most effective techniques and should usually form the initial step.

They include focusing on preparation for sleep and what works best for optimum sustainable sleep maintenance.

The key when working with children with FASD is in the detail of routines and how the impact of potentially small changes affects behaviours.

Ten Guiding Principles When Considering a Behavioural Approach

1. **Is sleep *actually* a problem**, that is, does it have a daytime impact? Is there night-time disturbance? Is sleep the issue that could change other things?

   Is it affecting cognitive functioning? Has sleep disturbance occurred for more than 3 months? Are previous behavioural strategies now not working? Is the child more hyperactive or aggressive? Have you noticed more sensory symptoms?
2. **For *whom* is it a problem?**
   For example: Child, siblings, parents, wider family, school, nursery, etc. Are night wakenings disturbing siblings? Are school commenting on tiredness levels? Do parents want to reclaim their evenings? What is the child saying?
3. **What is the evidence that the problems are *sleep* related?**
   Other behaviours that are challenging may well be sleep deprivation related, but not always.
4. **Is it the *right time* to initiate a behaviour management programme** which can be emotionally draining for parents/carers in particular and where managing change can be challenging for children with FASD?

   For Example: Recognise the need to assess parent/carer emotional resilience. What else is going on within the family? Practical issues such as housing/bedroom sharing are relevant. Procrastination may not always be helpful despite difficult home circumstances. Alternatively, sleep may be the ideal starting point—despite the presence of other problems which can't be changed at this time.
5. **Are there any other *health issues* that might need to be looked at before or alongside sleep work?**
   For example: Physiological/health problems? snoring/ENT assessment? sensory impairments or difficulties? toileting needs such as nocturnal enuresis? Do the parent/carers have any health needs of their own?
6. **How does the *family* manage behaviour issues in general?**
   For example: What are the strengths within the family? What is working well? Trying to introduce some behavioural strategies and routines at bedtime or in the night may not be fully effective if the family need help and understanding with daytime behaviour problems.
7. **What level of *understanding* does the child have?**
   Any sleep work needs to be tailored around the child—maximising their strengths, celebrating what is going well and engaging the child where appropriate.
8. **On a practical level *who* is going to support the family with the sleep programme?**
   For example: Other carers, other relatives/family members, school or nursery awareness around avoiding daytime naps, etc. if necessary. School transport is sometimes a problem where children fall asleep after a busy day at school.

9. **Is there a *timescale* involved?**
   Sleep work takes several weeks to elicit change. Is there any point starting a detailed sleep plan if the family are moving/going away/changing schools, etc. and hard work put in is likely to be severely disrupted? Or is a sleep routine going to be the fixed event in a period of change?
10. **What is the 'sleep culture' within the family unit?**
    For example: Do they have set times for going to bed and falling asleep? What is the adult sleep hygiene like? What are the parents/carer's attitudes towards sleep? How are the bedrooms used during the day? Is the bedtime too early/late? Explore cultural issues around sleep, such as bed sharing.

23.4.1 Managing Sleep Difficulties in Children with FASD

Checklist
Sleep diary—Assessment—Sleep plan—Review-adapt-review.

A sleep diary including sleep times, wake times and behaviours is a valuable tool for the professional and the family—initiating a joined-up approach, highlighting problem areas and showing the family are wanting to be involved with sleep work.

A full sleep assessment is then recommended with a detailed interview with the parents/carers and/or individual. This includes details around birth history, family health and development, communication, play and schooling. Also included is a detailed sleep history, what has been tried so far, whether sleep is altered with different carers or venues, any sleep triggers and sleep associations.

When this has been done, then compiling an individual targeted written sleep plan should be completed. Ideally, this should be achieved in partnership with the parents/carers so that they have ownership and will be 'on-board' in implementing it. It is important to discuss time scales and review, so that ongoing support is offered. When implementing changes, sleep can become worse before it improves. It is usually in the first week that parents/carers are most likely to give up as it is when challenges will occur and they are likely to become even more sleep deprived. It is known that parent/carer's beliefs about behaviour affect outcomes and the ability for change.

Always follow safe sleeping guidelines for babies and children as per Lullaby Trust [9].

23.5 General Sleep Hygiene

23.5.1 Preparation

Eating: avoid eating at least 2 h before bedtime to allow for stomach emptying, and ensure an adequate evening meal or tea to avoid later hunger/behavioural issues around food.

Drinking: milky drinks before bedtime will help sleep. Caffeine/additive drinks such as coke, chocolate, tea and coffee will be stimulating. No fluids after 6 p.m. and avoid blackcurrant drinks if enuresis is a problem.

Napping during the day: Some younger children need to 'recharge' and nap. However, prolonged or late afternoon naps could impact on night sleep by disrupting circadian rhythm. Finish any naps by 2.30 p.m.

Activity: Beware of stimulating activities 1–2 h before bed as this could keep them active all night.

23.5.2 Environment

A quiet environment helps the child to settle and recognise it is a bedtime. Consider other children who may be sharing a bedroom. Toys away, no electronic gadgets, try and keep the room calm and uncluttered. The room should be dark, ideally using blackout blinds. The temperature should be a comfortable level. Using a very low-level night light can be reassuring for children who don't like complete darkness. However, no blue lights, which includes televisions, computers and mobile phones. Switch of these gadgets at least 1 h pre-bed (preferably two) and they should preferably not be in the bedroom.

A bedroom for sleeping: A as far as possible the bedroom should be used just for sleeping. This will help promote good sleeping habits.

23.5.3 Routine

Set a regular bedtime routine. This should take no longer than about 45 min. This will regulate the body and reinforce message that sleep is about to happen. This can include:

- Bath or wash (a shower is often too energising), dental care.
- Story time/relax time in the bedroom/massage, etc. Story books can help settle and be part of a good bedtime routine.
- A bedtime mantra as you tuck them in 'Good night, we love you, see you in the morning' (repeating this each night will become a sleep onset association).
- Use visual cues and support.
- Wake up at the same time: It is important to wake the child at the same time every morning to maintain circadian rhythm.

Give consistent and clear messages. After tucking-in, parent/carers should always leave the bedroom completely. Verbal interaction, physical contact and eye contact should be minimal if parents need to return during the night. But remember that many children at some point will go through a time where they have problems with their sleep for no specifically obvious cause and returning to a good routine usually will solve it.

23.5.4 Appropriate Sleep Associations

Having good sleep associations will help a child stay asleep through the night. It is important that when a baby or child goes to sleep, that they are able to fall asleep on their own with minimum external cues. This is often referred to as self-settling. Any external cues or associations with falling asleep will need to be reproduced if they wake during the night. The aim is to help the child learn to self-settle from an early age.

Avoid

- Rocking to sleep: If a child is rocked to sleep at the start of the night, when they wake during the night they will seek to be rocked to sleep again, as they will associate the holding and rocking motion with sleep.
- Sleeping/lying next to children: If a parent lies next to a child to aid sleep onset, when the child wakes during the night this will need to be repeated.
- Dummies: Dummies are commonly used to help sleep and often become a strong sleep association. Dummies can become problematic if the baby is unable to replace it themselves. This will mean that parental intervention is needed to do this, and both baby and parent wake up fully. (See safe advice regarding dummies on Lullaby Trust website.)

Try

- Using a safe, soft toy/familial clothing to cuddle at night.
- Having a safe small nightlight.
- Having familiar pictures/photos on the walls for the child to look at as they drift to sleep.
- If using sensory approaches, for example lights/music, these need to be turned off *before* sleep and are used to calm only.

23.5.5 Sleep Hygiene Particularly Relevant to Consider for Children with FASD

- A clear routine that does not vary from day to day. Consider at least up to 2 h before bedtime, or longer if necessary. Keep consistency over schooldays and weekends. Recognise potential trigger factors in the day in advance that may excite/reduce potential settling.
- It is essential to have the co-operation of the whole family.
- Consider verbal, practical and/or visual warnings/timings for transitions between activities as to strengthen sense of time awareness.
- Consider images/photos for each stage to help with sequencing.
- Social stories can help with understanding. (A social story models appropriate social interaction by describing a situation with relevant social clues, other's perspectives and a suggested relevant reaction).

- Use concrete instructions and maintain identical language together with physical gestures if necessary for each stage/as appropriate to developmental level. Be aware that a certain word, phrase or sign might be a trigger for anxiety or anger and adjust accordingly.
- Assisting and supervising the child with each stage as developmentally appropriate.
- Addressing sensory needs (again do not underestimate the importance of this), that is, massage, relaxation techniques and calming procedures. Also assess bedding, night-time clothing, room temperatures, any potential olfactory irritants, external factors—noise levels, street lighting, etc.

It may be beneficial to involve an occupational therapist where there are significant sensory issues to enable fuller understanding for the parent/carers and where appropriate for the child.

- Acknowledge visual and auditory needs—white/pink noise has been shown to be calming and to help with sleep latency.
- Note that a pre-bed bath time may be too stimulating for a child with FASD.
- Ensure safety is addressed (i.e. window locks, door catches, stair gates and small toys removed).
- Bedroom environment—clear the clutter, use calm colours, examine bedroom sharing. Try and reduce 'visual crowding'.
- Consider what is a motivator for the child—delayed reward schemes may not be effective for children with FASD. Consider instant rewarding and adapting routine reward-based schemes to suit the individual.
- Consider use of 'special time' in the pre-bed routine or earlier in the day (a period of clear child-led calming and engaging activity for 10–15 min prior to commencing bedtime routine with a specific start and finish time).
- Enable knowledge of day/night according to the level of understanding—that is, gro-clock, digital clock, clock radio to indicate morning or simple pictures of a sun or moon. A 'gro clock' shows stars and suns on the dial and changes gradually over the night, so the child knows when morning is approaching.
- Consider the effects of any medications the child is already taking, for example for ADHD/epilepsy and the impact on sleep.
- Examine family sleep culture and beliefs around bedtime – established long-term rituals that need to change will take time to plan. Consider parental/carer's belief, barriers or resistance around making changes.
- Examine effective behavioural input in the daytime as these are likely to be helpful at night.
- Do not overpraise success—acknowledge it, praise the effort and if appropriate ask how it feels.

23.5.6 Settling Using Gradual Retreat

It involves parent/carer gradual withdrawal out of the bedroom over several days/weeks. They are transitioning from being close to the child to allowing the child to fall asleep alone. The starting point is usually the parent/carer being close to the child, lying next to them. They remain in the current 'position' for 10 min after the child has fallen asleep. Each night they move further away from the child—for example: lying in/by the bed, to sitting in a chair, then gradually moving out of the room. The aim is to change position every few days by a very small amount to eventually be outside the room. Waiting for an extra 10 min once asleep ensures commencement of deep sleep phase.

Delayed sleep phase is falling asleep very late and not waking in the morning at a suitable time.

Start the bedtime routine 45 min before sleep is naturally occurring.

Gradually move the bedtime routine time earlier by 5–10 min every few days. Maintain consistent morning waking at agreed time. Any waking before this time is treated as night settling, and the steps above are followed.

Night-time waking: remodel settling routine exactly (including stage of gradual retreat if also working on this). Examine possible cause. Occasionally, a 'scheduled awakening' at normal disturbed time may be helpful to disrupt sleep pattern (Go into child, stir them briefly and resettle before the child is fully awake). If this is not effective, adjust to 15 min earlier. This 'resets' the sleep cycle and may avoid the need for the child to rouse.

Early rising: examine what may motivate this. Set alarm for current arising time—gradual shifting of alarm by several minutes every few days. Once alarm has sounded, 'morning' or another agreed activity can occur. This may involve parent/carers commencing their day too initially. The child gradually understands to 'wait' before rising.

Shortened sleep duration has been noted in children with FASD.

23.5.7 The Problem Gets Better, Then Worsens

Behavioural work takes a significant time scale—there is also often a deterioration in progress known as the 'extinction burst' before the behaviour improves again. Revisit the plan if necessary. Unexpected external factors also affect sleep behaviours so be prepared to revisit these if necessary.

23.5.8 Allow a Suitable Timescale

Being prepared for several weeks of teaching a behaviour and then still needing to give reminders. Examine motivators for achievement and adjust these accordingly.

23.6 Medication

FASD is a complex issue, and behavioural approaches alone may not significantly improve sleep quality. However, in principle, they should always be instigated for a period of time (at minimum 6–8 weeks or more).

In such circumstances, specialist medical referral and advice is recommended and prescribed melatonin may be considered for problems associated with sleep latency/circadian rhythm difficulties. Most importantly, it must be used in conjunction with sleep behaviour work and to incorporate sensory needs.

Melatonin is a chronobiotic, which is different to a hypnotic medicine which initiates and prolongs sleep. There is evidence that melatonin can be effective in treating circadian disturbances and rhythm disorders, and this has been shown to be effective for sleep latency for children with FASD.

In the United Kingdom, melatonin is often prescribed as a shared care agreement between the paediatrician and the GP.

Melatonin is given as part of the nightly sleep hygiene—usually between half to 1 h before bedtime. The slower release long-acting melatonin tablets should not be crushed.

There are very few studies examining the use of melatonin longer term (i.e. more than 18 months). It is worth noting that if children are prone to night terrors or similar parasomnias, then these can worsen with melatonin medication. If a child is taking melatonin, regular sleep and medicine reviews are vital—to offer ongoing support and medical review.

If sleep continues to be disturbed due to issues around overactivity and stimulation rather than a circadian rhythm problem, further specialist medical advice should be sought and a stimulant medication/alpha agonist could be used.

Again, specialist advice needs to be sought for polypharmacy issues relating to comorbid diagnoses.

Practice Points

- Sleep is a complex issue.
- Empowerment of parent/carers and the strength of motivational discussion and interviews are essential to help instigate change. There are always many factors to consider.
- Changing a behaviour takes time—a lot of time. It may well take between 6 weeks or even longer to establish a minor behavioural change. Illness or unexpected events can also alter outcomes and progress. Acknowledgement and helping parent/carers/individuals with professional support are essential. If success is elusive, medication is a recognised consideration, but good sleep hygiene always remains the primary factor.
- Behaviour in FASD can change with peaks and troughs so always recognise and celebrate success.

References

1. Owens J. A classification and epidemiology of childhood sleep disorders, vol. 3. Orlando, FL: Elsevier Science; 2007.
2. Chen ML, et al. Sleep problems in children with fetal alcohol spectrum disorders. J Clin Sleep Med. 2012;8(4):421–9.
3. Oxford English Dictionary. Oxford: Oxford University Press.
4. Chung K, Yeung W, Ho FY, Yung K, Yu Y, Kwok C. Cross-cultural and comparative epidemiology of insomnia: the Diagnostic and Statistical Manual (DSM), International Classification of Diseases (ICD) and International Classification of Sleep Disorders (ICSD). Sleep Med. 2015;16(4):477–82.
5. Mindell JA, Moore M. Does sleep matter? Impact on development and functioning in infants. Paediatrics. 2018;142(6):e20182589.
6. Durand VM. A guide to improving sleep for children with special needs. Baltimore, MD; Chelsea, MI: Paul H Brookes; Sheridan Books; 2014.
7. Borbely A, Wirz-Justice A. The two process model of sleep regulation: a reappraisal. J Sleep Res. 2016;25(2):131–43.
8. Bastida-Pozuelo MF, Sánchez-Ortuño MM. Preliminary analysis of the concurrent validity of the Spanish translation of the BEARS sleep screening tool for children. J Psychiatr Ment Health Nurs. 2016;23(8):513–20.
9. www.lullabytrust.org.uk

Assessing and Managing Sensory Processing

24

Joanne Pennell

Chapter Highlights

- What is Sensory Integration.
- Understanding the Sensory System.
- Interventions Targeted at the Sensory System.

Sensory integration is an automatic neurological process that occurs throughout life. Our brain receives information from our senses and organise it so that we are able to respond adaptively to particular situations and environmental demands.

For most people, sensory integration develops through ordinary childhood activities. They are able to process information automatically and effectively; but for some people, sensory integration does not develop as efficiently as it should and this leads to difficulties in activities of daily living, academic achievement, behaviour or social participation.

Sensory integration (SI) theory offers a framework for understanding some characteristics and behaviours which people with fetal alcohol spectrum disorders are commonly reported to experience. The theory not only provides a way to understand why behaviour may occur but also guides structured assessment processes and provides a range of interventions that can be incorporated into everyday life. Aspects of SI theory can be taught to the individual and their family or carers, so that they can develop their own strategies for responding to their sensory needs or accommodating sensory challenges.

J. Pennell (✉)
Treetangle Occupational Therapy, Walton on Thames, Surrey, UK

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_24

24.1 Introduction

Interventions based on sensory integration (SI) theory are commonly used by occupational therapists working with young people. However, there is a misconception that any formulation or intervention with a sensory component can be described as 'Sensory Integration' therapy, this leads to confusion and lack of clarity about the effectiveness and quality of interventions. As difficulties with sensory integration are frequently referred to in descriptions of fetal alcohol spectrum disorders (FASD) [1–3] it is the intention here to discuss common terminology and indicate the range of assessment and intervention options that may be broadly described as 'sensory-based'.

This chapter begins by summarising current research into sensory integration and FASD. It will then describe current use of terminology, outline key aspects of sensory integration theory and review the role of sensory systems before moving on to describe assessment and intervention processes and conclude with key practice points.

24.2 What Is Sensory Integration?

Sensory Integration (SI) theory is based on the understanding that neurological processing and the integration of sensory information underpin purposeful behaviours [4, 5].

The overarching theory was developed by Dr. A Jean Ayres, an Occupational Therapist, Clinical Psychologist and Neuroscience researcher. Beginning in the 1960s, Ayres developed a theory to explain the behaviours she was observing in her clients and went on to develop tests and a therapy approach called Sensory Integration Therapy, in order to provide effective interventions for the young people she worked with. The model has continued to evolve as a result of her own research, and that of an increasingly diverse range of therapists and researchers [5, 6].

'Sensory integration' describes the neurological process of organising the information we get from our bodies and from the world around us for use in daily life. It forms an essential foundation for later more complex learning and behaviour (Fig. 24.1). For most people, sensory integration develops in the course of ordinary childhood activities in a developmental sequence [6].

Sensory integration is a theory with three key functions: describing typical sensory integration development; defining sensory integrative difficulties and guiding therapeutic intervention. Thus, a sensory integration framework can be used to develop hypotheses to explain behaviour, plan appropriate interventions and predict how the behaviour may change as a result.

Children spend, in general, the first 8 years of their life experiencing and challenging their bodies and their environment. They are fascinated with information received through their bodies, through movement, touch, taste, vision, smell and sound. This stage in their development is crucial because it lays the foundation with which a child gains skills, self-confidence and mastery of his/her body and environment [7].

We receive messages from all of our senses and respond to this information in a way that allows us to do the things we want or need to do.

For example,

In order to read a book; we screen out noises going on around us; we don't notice the feel of our clothing; we automatically adjust posture and muscle tone to maintain sitting balance; we are able to scan the page by moving our eyes without our head. Difficulties in sensory integration can affect any aspect of this activity.

Fig. 24.1 Sensory integration in everyday activity

Ayres theorised that impaired sensory integration may result in functional problems which she referred to as sensory integration dysfunction [8]. Ayres' early research consistently found clusters of difficulties in children's motor skills, their behaviour, emotions and attention, these clusters have since been validated by more recent research [4, 9, 10].

It is not possible to observe central nervous system processing, sensory integration or the process of motor planning. It is possible to see and assess behavioural difficulties; the assertion that the behaviour results from deficits in sensory integration are hypothesised, although there is a developing body of evidence which supports some premises of sensory integration [5, 11].

A sensory integration framework can be used to observe, understand and develop interventions for people experiencing behavioural and functional difficulties relating to sensory processing disorders.

Most practitioners qualified in sensory-based therapy are occupational therapists and, as such, the focus of intervention should always be facilitating the person's ability to participate in daily occupations which are meaningful and satisfying for them within their own environment [Royal College of Occupational Therapist (RCOT); American Occupational Therapy Association (AOTA)] [12, 13]. While assessment and treatment will be individually defined, it can be broadly categorised as aiming either to remediate underlying impairments or to enable participation through accommodation and adaptation.

To function in everyday life, we must interpret and respond to the world around us through our sensory systems. We can all show signs of sensory integration difficulties (for instance, if we are tired, unwell or preoccupied), but for some individuals these difficulties have a significant and on-going impact on daily life resulting in difficulties in self-care, education or work, leisure activities and social interaction.

Difficulties in sensory integration may result in unusual sensory seeking or avoiding behaviours, which interfere with the ability to engage in purposeful activities. Although

the majority of literature focuses on children, there is a small but growing body of research exploring the impact of SI difficulties in adults. They have been found to experience difficulties in managing a range of daily activities [14, 15]. In a case-matched comparative study of 28 adults, Kinnealey, Koenig and Smith (2011) [16] discovered that sensory response style was 'significantly' and differentially related to poor affective mental health and poor quality of life scores related to social functioning. While this was a small exploratory study, participants with any diagnosed health problems were excluded, reducing possible contributory factors other than SI difficulties.

Sensory integration issues can influence the style and nature of an individual's overall engagement in performance skills and patterns. Enabling the individual and those around them to understand their choices, preferences and challenges from a sensory perspective may help identify alternative strategies or accommodations to support engagement [17].

24.3 Why Should We Consider Sensory Integration When Assessing and Planning Intervention for People with FASD?

Neuro behavioural problems including sensory-motor difficulties related to coordination, balance, planning and gross-motor activities are reported in people with fetal alcohol spectrum disorders [18–20].

It is suggested that sensory integration difficulties can play a significant role in how people behave in any given situation or environment [2]. An understanding of these difficulties can help develop a better understanding of potential triggers and contribute to more informed interpretation of behaviour (Fig. 24.2) [1].

A developing body of research supports clinical and anecdotal reports of higher than typical levels of sensory integrative (SI) difficulties in the population of people with FASD. Studies have identified SI difficulties as a significant aspect of problems experienced by many children with FASD [3, 21] including difficulty interpreting and reacting accordingly to sensory input from their environment. These difficulties in processing sensory information can cause children with FASD to respond in an unusual way to their environments, and may be associated with higher-than-average behaviour problems, social and attention problems, rule breaking and problem-solving difficulties [22, 23].

In a 2008 study, Franklin et al. [3], explored sensory integration behaviours of school age children with FASD, as compared to typically developing peers, in relation to their adaptive function at home and school. They found that children with FASD were three times more likely to be categorised as having 'significant sensory processing deficit' when screened than their typically developing peers and that significant correlations between sensory integration difficulties and adaptive and academic function suggest a relationship exists between the two. Children with FASD present with prevalence of up to 90.9% showing definite differences using the Short Sensory Profile and 81.8% showed some problems or definite dysfunction on the Sensory Processing Measure [24].

Ben and Jon are playing with bricks

Antecedent:Ben moves closer to Jon to reach a piece bumping his arm
Behaviour: Jon hits Ben
Consequence: Ben runs off.
Interpretation: Jon does not want to share the bricks

Adding information about Jon's sensory difficulties

Jon is already feeling overloaded, the TV is on, his parents are talking in the background and he cannot screen any of it out to concentrate on his building, he also has poor tactile discrimination, he cannot tell how hard he has been bumped but he thinks it felt painful. Hitting his brother was an instinctive response but now he is in trouble again!

Fig. 24.2 Is it behavioural or is it sensory?

Jirikowic et al. [2] also found significant correlations between measures of sensory processing and sensory–motor performance, adaptive behaviour and some aspects of academic performance. They suggest that sensory–motor impairments should be considered when determining the developmental needs of children with FASD and claim impairments may co-occur with and contribute, at least in part, to difficulties in adaptive and school functioning.

There is currently less research specifically addressing interventions for SI difficulties in children with FASD. Clinical experience and tentative initial research [25] indicates that approaches which are effective in other populations where SI difficulties co-occur with other diagnosis (such as autism) are likely to be effective with this population. For example, in a study delivering a modified version of the Alert Program, Wells et al. [26] found a significant treatment effect on parent-reported measures of self-regulation, modulation and planning in school-aged children with FASD. This was a randomised control trial involving 78 children, who were assigned to either the intervention condition or to a control group, and received a comprehensive evaluation and community services such as speech and language therapy, occupational therapy and physical therapy. The study was adequately powered to lead to significant differences found in the intervention group versus the control group. At time of writing a research proposal has been accepted for a much larger study of the use of the Alert Program with children with FASD living in remote Australian Aboriginal communities [27].

24.4 Questions of Terminology

The terminology used in relation to sensory difficulties can be confusing, a fact which reflects the evolving nature of the field [2, 7]. As new findings are revealed through research and practice, theoretical paradigms are necessarily revised and

adapted to reflect these findings, integrate them with existing knowledge and develop models and classifications that will guide future practice and research.

It is most common for therapists trained in the model to refer to sensory integration (and related difficulties, challenges or dysfunction) while more generically the term 'sensory processing' (and disorder) is used. In this chapter, the term sensory integration will be used to refer to the brain behaviour theory proposed by Ayres [6, 8] and Ayres Sensory Integration (ASI) to describe the intervention based on the theory when it conforms with the fidelity model proposed by Smith-Roley et al. [27].

The term sensory integration is used in a variety of fields and can be applied to neurophysiological cellular processes rather than a behavioural response as proposed by Ayres. This potential confusion in terminology led Miller et al. [28] to a proposed nosology which supported the continued use of 'Sensory Integration Theory' and 'Sensory Integration Treatment and Evaluation' but proposed a new diagnostic categorisation of 'Sensory Processing Disorder' for individuals who present with sensory integration challenges. This was to distinguish the disorder from the theory as well as the cellular processes. However, this terminology has not been universally accepted. Many researchers and practitioners argue that the proposed patterns of sensory integrative dysfunction differ from those already in clinical use and supported by extensive research, and also that 'sensory processing' is itself a generic term used in a variety of fields [29, 30]. Nonetheless, this continues to be a popular proposal for diagnostic terminology to children with FASD and further research is underway to verify the clusters within it [31].

Smith Roley and Mailloux [29] noted 'Part of the controversy [about the effectiveness of the sensory integrative approach] stems from the many publications and intervention programmes that do not truly reflect the principles of Ayres's work but that nonetheless have been mistakenly associated with sensory integration '(p1). As a result, a fidelity measure has been developed with the intention of evaluating adherence to underlying principles of intervention for researchers, therapists and clients [32].

24.5 The Sensory Systems

Our senses provide the information we need to function in the world. They receive information from stimuli from outside and inside our bodies. The five senses we most commonly think of (hearing, seeing, taste, smell and touch) respond to external stimuli that come from outside our bodies. The vestibular, proprioceptive and tactile senses are the three body-centred sensory systems that give us a sense of ourselves in the world. Within our bodies, interoception detects internal regulation responses such as hunger, heart rate and the need for elimination (Table 24.1). These interconnected senses develop early and have a major impact on an individual's development. They are connected to several areas of the central nervous system and brain. Therefore, the activation of some regions of the central nervous system can influence the function and plasticity of others [2].

Table 24.1 Description and function of sensory systems

Sensory system	Description	Important for:
The sense of touch (the tactile system)	– The largest sensory system. Plays a significant role in determining physical, mental, and emotional behaviour – Receptors are located in the skin – Information about touch sensations: light touch, pressure, vibration, movement, temperature and pain. – Comprises of two components: the protective (or defensive) system and the discriminative system	– Identifying whether a stimulus is painful or dangerous (protective) – Recognising shape and texture of objects (discriminative) – Planning movements – Development of fine more skills and manipulation of objects – Emotional security and attachment – Social skills
The sense of body position and movement (the proprioceptive system)	– Proprioception is the sensory information that we receive from our joints and muscles regarding our position and orientation of the body and body segments in space – Receptors are located in muscle spindles, joints and mechanoreceptors of the skin, (these being important in detecting joint movement in the hand as a result of one's own hand movement)	– Development of an internal map of our body (body scheme) – Body awareness – Motor control and motor planning – Judging the rate and timing of our movements – Determining how much force our muscles are exerting – Regulation
Gravity, balance and movement (the vestibular system)	– The vestibular system senses head movement and influences balance and posture – Regarded as the most influential sensory system. Directly or indirectly, the vestibular system influences nearly everything we do. It is the unifying system in our brain that modifies and coordinates information received from other systems. – The two types of vestibular apparatus are located in the inner ear; the otoliths (utricle and saccule) sense static functions, the utricle horizontal movement, for example when moving forward in a car and the saccule vertical acceleration, for example, going up in a lift, and also to gravity, while the semi-circular canals respond to rotational movement of the head in space and also detect change of speed, direction, pitch, roll, rotation and transient angular head movements – Vestibular receptors are the most sensitive of all sense organs and the vestibular is one of the first sensory systems to develop in utero, providing some of the building blocks for later development of seeing and hearing and is a major organiser of varied sensory input	– Balance – Seeing clearly while moving – Eliciting postural reflexes to keep our head erect; – Facilitating spatial orientation by signalling the direction of gravity – Making sense of all other senses – Modifying arousal levels – Preparing for "fight or flight" in an emergency. – Feeling grounded and secure

(continued)

Table 24.1 (continued)

Sensory system	Description	Important for:
The sense of sight (the visual system)	– Helps us to navigate in the world and judge the speed and distance of objects and people	– Watching a moving object – Locating objects – Guiding hand movements
The sense of sound (the auditory system)	– Located in our ear and relates to the ability to receive sounds	– Locating sounds in the environment – Discriminating between sounds and words – Attending to, understanding, or remembering what is read or heard – Speaking and articulation
The sense of taste (the gustatory system) This is closely related to:	– Helps us to recognise bitter, salty, sweet and sour flavours	– Tastes are important in our selection of food or to inform us whether certain tastes might be harmful for our body
The sense of smell (the olfactory system)	– Smell travels directly to the emotional brain or the limbic system which is often why our emotions are tied to smells and foods.	– Plays an important role in establishing and receiving memories and associations that influence some of our choices and preferences – Food choices are greatly dependent upon the sense of smell. If it smells bad that sends a warning that we may not like it OR that it is dangerous for us to eat
The sense of the physiological condition of the body (Interoception)	– Responsible for detecting internal regulation responses, such as respiration, hunger, heart rate, and the need for digestive elimination – Detected through nerve endings lining the respiratory and digestive mucous membranes – Provides internal picture of how the human body is perceived, along with the vestibular and proprioceptive senses	– Recognising if we are hungry or full-up – Noticing if we need to use the toilet

24.6 Sensory Integration as a Basis for Function

A sensory integrative perspective understands the organised behaviours of self-care, learning, work or leisure to be wholly dependent on effective integration of sensory information. The relationship can be thought of as building blocks with each element being dependent upon the underpinning levels (Fig. 24.3) [2].

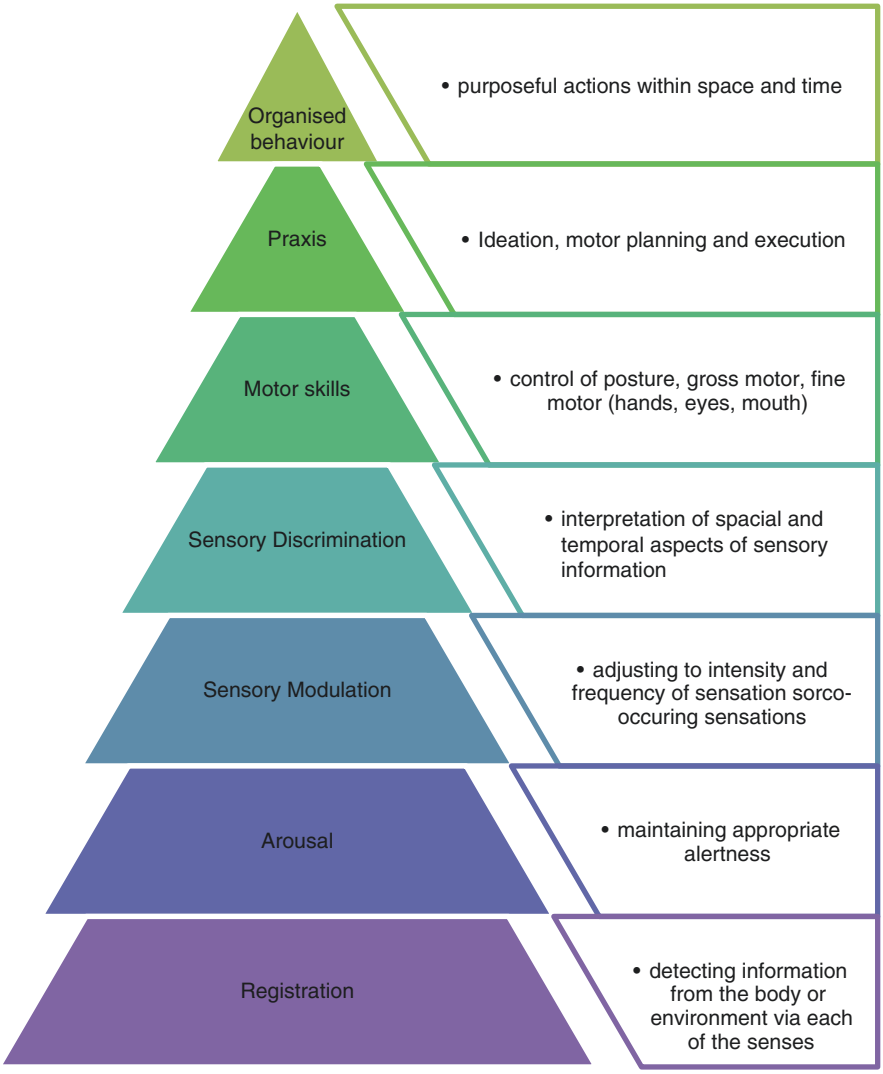


Fig. 24.3 How sensory integration functions underpin organised behaviour such as learning and daily activities

24.7 Recognising Signs of Sensory Integration Difficulties

If sensory integration is disordered, it will result in observable problems in learning, motor development or behaviour, for example:

- *Over-reactive to sensory stimulus.* Distractible, withdrawal when touched, avoidance of textures/clothes/foods. Fearful reactions to movement activities such as playground play. Fear of loud noises. Aggressive responses to unexpected sensory input.
- *Under-reactive to sensory stimulus.* Seeks out intense sensory experiences such as spinning, falling and crashing into things. Unaware of pain or to body position.
- *Unusually activity level (high/low).* Ceaselessly on the go/slow to get going and tires easily.
- *Coordination problems.* Difficulty learning a new task that requires motor coordination, appears awkward, stiff, or clumsy and poor balance.
- *Disorganised behaviour*. Impulsive, easily distracted, unable to plan how to approach a task, difficulty anticipating result of actions. Difficulty adjusting to a new situation or following directions. Appears easily frustrated, aggressive, or withdraw when unsuccessful.
- *Problems with activities of daily living*. Difficulty with handwriting, tying shoelaces, buttoning and zipping clothes. Messy eater—avoids using knife and fork, spills things, and doesn't notice food on face.

A variety of models exist for describing sensory integration problems, each of them have strengths for addressing particular areas of a complex and evolving field of practice. One of the most frequently presented is the nosology proposed by Miller et al. in 2007 [28] in an attempt to explicate current understanding of sensory processing deficits (Fig. 24.4). The intention of this work was to provide a diagnostic framework. The groups differ from the subtypes identified through the factor analytic studies conducted by Ayres and colleagues [8, 33].

Dunn [33] has developed a model which provides much greater specificity in the area of sensory modulation. Fisher, Murray and Bundy, in their 1991 text, Sensory Integration Theory and Practice, included sensory modulation disorder in a revised version of the classic Ayres sensory integration flow chart and it is perhaps this model which offers practitioners the most useful overview for assessment and treatment [34].

24.8 Attachment Difficulties and Trauma

Studies indicating high incidence of FASD in the looked after or adopted population [35] suggest that any consideration of SI difficulties should be able to include an attachment and trauma perspective where appropriate [36].

Many children coming from backgrounds of neglect suffer from both attachment disorders and sensory integration dysfunction. In practice, it can sometimes be difficult to distinguish the two, since the symptoms can overlap [37, 38].

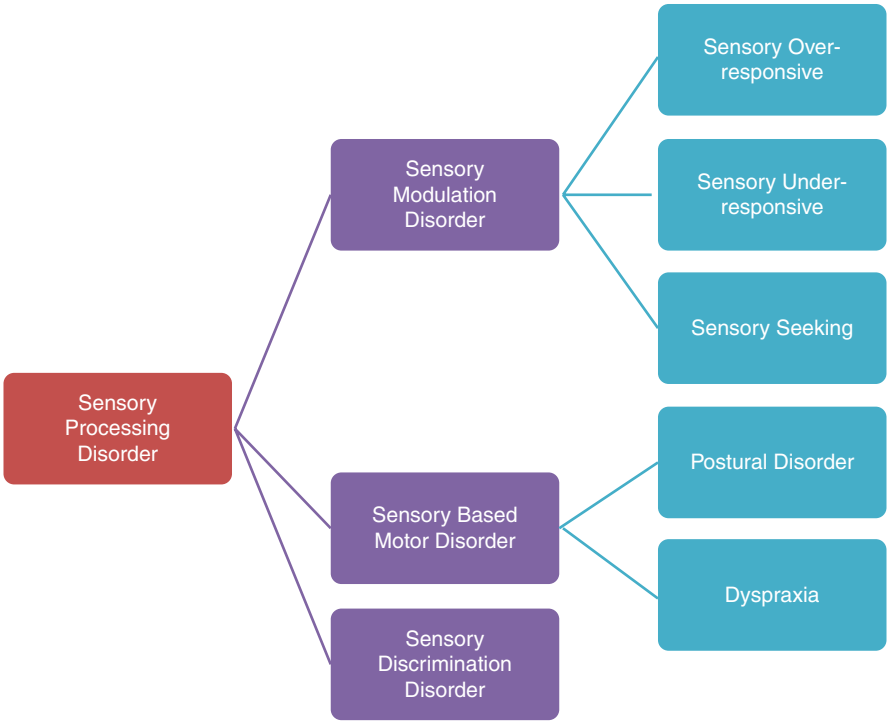


Fig. 24.4 Proposed nosology of sensory processing. (Amended from Miller et al. 2007)

Throughout the attachment cycle, the primary caregiver acts to regulate the child; eventually, through many repetitions of the cycle, the child internalises this experience and learns to modulate their own responses to fit the situation. It is suggested that this ability to manage arousal levels develops, in part because throughout the cycle, the carer is providing the infant with physiological experiences (e.g. rocking, swinging, sucking and touch) which provide the foundations for future development [39]. A child who has had poor or disrupted experiences may be neurologically disadvantaged. They are likely to have difficulty processing sensory messages from the environment in as efficient a manner as peers with more optimal early experiences. In a developing child, sensory experience helps to shape neural connections and to prime the areas of the brain which deals with emotional regulation. If those connections are absent or disrupted, the child may struggle to feel safe and secure. Instead, more primitive areas of the brain may persist in an exaggerated fight or flight response to stress [37, 38].

Increasingly, rather than focusing on differentiating between attachment and trauma difficulties and sensory processing disorders, clinicians and theorists have adopted a combined approach, it seems that treatments for attachment can help a child to overcome or respond more adaptively to sensory processing problems, and treatments for sensory processing disorder can help to secure attachment. Key models in this evolving field are Sensory Attachment Intervention (Bhreathnach 2008) [37] in the UK and SAFE PLACE (Jane Koomar2012) [38] in the USA. Both

have combined trauma and attachment-related models with sensory integration to provide assessment and intervention approaches. Neuro-Physiological Psychotherapy also builds on this work and recognises that the child needs help to repair missed developmental stages and their ability to manage the sensory environment [40].

24.9 Assessment

Sensory integration is usually assessed using a combination of standardised tests; formal and informal clinical observation and carer or self-questionnaires [2]. The data arising from these assessments form the basis of a hypothesis regarding the individual's neurobiological processing of sensory information and its impact upon behaviour and function.

There are a range of standardised tests to assess either specific aspects of sensory integration or the full range of components. The most rigorously researched and in-depth tool is a battery of 17 sub-tests called the Sensory Integration and Praxis Tests SIPT [41]. The SIPT focuses primarily on functions related to praxis, motor-free visual perception and visual-motor function. The SIPT is the most complete and flexible assessment of sensory integration available. However, it is time consuming and requires the individual to be able to engage in the formal testing process and so may not be appropriate in some situations. Data collection is currently underway to standardise a new comprehensive test of sensory integration, The Evaluation in Ayres Sensory Integration (EASI) which is expected to be available from 2020 [42].

Structured observations of specific tasks, postural responses and signs of nervous system integrity associated with sensory integrative functioning such as Blanche's Observations Based on Sensory Integration Theory [43] are used to supplement standardised tests, as are informal observation of the individual's movement, function and interactions across environments. Together they provide information about how someone responds to and organise sensory information in day-to-day life. Sensory-motor histories or questionnaires have traditionally been orientated to gathering information from parents or carers (for example the Sensory Processing Measure) [44]; however, there is an increasing number aimed at self-reporting more appropriate for older children and adults. The Sensory Profiles [45, 46] are standardised sensory history checklists which focus on sensory modulation. The profiles include a short version (SSP) which is intended for screening and research only. For a wider discussion of available assessment tools, see Schaff and Roley [7] Bundy and Lane [47], for example.

24.10 Intervention

Sensory integration theory supports a range of intervention options; however, they all adhere to a set of shared principles.

- The therapist has certified training in sensory integration which includes an extensive knowledge of the application of neuroscience to behaviour and brain-body interactions.
- The relationship between therapist and client is collaborative.
- Assessment and intervention are positioned within a wider understanding of what the client wants, needs, or is required to do and aims to enhance engagement and participation in meaningful activities.
- The intervention plan is person and family centred and based on a full assessment and interpretation of the patterns of sensory integrative difficulties [2].

24.10.1 Direct Interventions

Sensory Integration Therapy

This aims to strengthen capabilities through improving underlying neurological processing and organisation.

- Intrinsic motivation and drive are used to interact through enjoyable activities (play).
- Treatment goals are guided by setting a 'just right' challenge.
- Activities chosen are rich in sensation (especially vestibular, tactile and proprioceptive) to guide self-organisation and enable optimal arousal states.
- Direct therapy is delivered in an environment that is engaging, maximises success and ensures safety.
- The environment includes equipment that will provide vestibular, proprioceptive and tactile sensations and opportunities for praxis.
- Activities promote optimal postural control in the body, oral-motor, ocular-motor areas and bilateral motor control, including moving through space and adjusting posture in response to changes in the centre of gravity.
- Intervention strategies provide the 'just-right challenge' through the therapist, ensuring the client's success in whatever activities are attempted by altering the activities to meet the client's needs and abilities.
- Opportunities exist for the client to make adaptive responses to changing and increasingly complex environmental demands [32].

Sensory Diets

A sensory diet is a strategy that consists of a carefully planned practical programme of specific sensory activities that is scheduled according to each child's individual needs. Like a diet designed to meet an individual's nutritional needs, a sensory diet consists of specific elements designed to meet the child's sensory integration needs. The sensory diet is based on the notion that controlled sensory input can affect one's functional abilities [48]. A sensory diet can help maintain an age-appropriate level of attention for optimal function or reduce sensory defensiveness.

Teaching Cognitive Strategies

People can learn about their sensory systems, how they affect choices and preferences and strategies for addressing difficulties encountered. One of the best examples of this is the use of Sensory Ladders and Sensory Spiders or Flowers [49] to build a shared narrative and understanding of how sensory experiences affect each aspect of our lives. Originally developed for use in adult mental health, they are now widely used to promote awareness of self-states and how to use sensory strategies for self-regulation, promoting successful engagement and participation in everyday life [50]. Another example is the Alert Program for Self-Regulation.

The Alert Program

The Alert Program, [51] designed by occupational therapists, Mary Sue Williams and Sherry Shellenberger, teaches children and adults about self-regulation. It is a complementary approach that encourages cognitive awareness of alertness through the use of sensory strategies to support learning and behaviour. Self-regulation involves the sensory systems of the brain, including the vestibular and somatosensory systems. The programme uses the analogy of a car engine to describe arousal states of the body as 'high gear,' 'low gear' and 'just right gear'. Participants learn to use sensorimotor strategies and tools from five categories (look, touch, move, listen and by putting something in the mouth) to shift their arousal level into an optimal state or 'just right place' to meet the demands of a particular task or situation as a means of self-regulation. The programme is designed to be used within a range of settings and with clients of varying ages. As discussed above, evidence suggests that this approach is useful to children with FASD and further research is underway.

Sensory Attachment Intervention (SAI)

Eadaoin Bhreathnach has combined sensory integration theory with attachment and developmental trauma theories [37]. She argues that traumatised children and adults tend to maintain a state of hypervigilance. This impedes filtering out 'irrelevant' sensory experiences such as background sights and sounds and also to sensory defensiveness as the sensory systems have become sensitised to the possibility of danger. Bhreathnach developed The Just Right State Programme (JRSP) to support the regulation of arousal states in children who have experienced trauma. The programme looks at the use of sensory activities and foods, to help children (and adults) learn how to self-regulate their emotional states and behaviour. It is based on the principle that helpful somatosensory (body-based) experiences can be incorporated into routines of daily life in order to enhance the capacity for co-regulation and self-regulation. The aim of the programme is to enable individuals to remain regulated when engaged in activities that are normally challenging for them. This enables them to successfully engage in more complex functioning such as social engagement and academic learning.

24.10.2 Sensory Integration-Informed Consultation

Non-direct interventions include the following:

- Compensating by modifying the environment.
- Compensating by modifying specific tasks.
- Reframing behaviour.

Therapists use their knowledge and expertise to collaborate with the client. The collaborative process involves identifying the problem, creating possible solutions, trying solutions and altering them as necessary for greater effectiveness [52].

Integrating strategies in schools/home. Therapists work with the young person's family or carers and other people in their support network to provide intervention within naturally occurring contexts. They monitor implementation of plans and provide supervision to team members who carry out agreed strategies in the home/school environment. This can include consultation with staff, recommending teaching strategies, equipment adaptation or making modifications to environment.

Reframing behaviour through education of individual and their network. Therapist reframes behaviours and preferences using a sensory processing perspective [4, 53]. By explaining the possible links between sensory processing and behaviours that challenge, and by recommending strategies that respond to sensory needs rather than behavioural-based strategies, challenging situations can be understood differently.

Modifying environments or routines to support self-regulation can enable the individual to more fully participate in everyday activities. This may include recommending modifications to daily routines or environments such as a balance of active and quiet activities and opportunities to participate in preferred sensory experiences (e.g. swinging in the garden or an indoor hammock, climbing on a gym set, jumping on a trampoline or sitting in a rocking chair in a room with low lighting.

24.11 Summary

Sensory integration is the organisation of sensations for use. The sensory systems are inter-related and sensory information is constantly entering the brain from all eight senses. Higher than typical levels of sensory integrative (SI) difficulties are reported in the population of people with FASD.

A strong basis in sensory integration enhances development of higher level gross and fine motor skills. It also contributes to better self-esteem, self-control and improved attention span.

Sensory processing disorder (SPD) is commonly used to describe difficulties in the sensory integrative process, these terms are often used interchangeably,

resulting in confusion and lack of precision. Behaviours arising from difficulties in sensory integration are in many cases involuntary and are reactions to the individual's internal and external environments and their inability to sort out this information. Each person is unique in strengths, interests, difficulties and degrees to which deficits manifest.

Understanding a person's sensory needs can provide the basis for helping in a wide range of daily activities.

Sensory integration therapy (ASI) provides controlled sensory input with the goal being increased adaptive behaviours/responses. It is one treatment approach amongst many. Different treatment approaches address different deficit areas and different needs and also reflect available resources.

Practice Points

- Be aware of how sensory challenges can affect a person's behaviour. They may not be able to identify what is bothering them or may assume the way they experience it is the way everyone else experiences it.
- If sensory processing might be part of the picture, organise an assessment by a therapist with certified training in Ayres Sensory Integration. A full sensory profile is a useful tool for the individual and the people supporting them.
- Consider the environment, how can it be adapted to meet the individual's sensory needs? If the environment can't be changed what accommodations can be made, for example, movement breaks, ear defenders or white noise.
- Think about daily routines and how sensory overload can be cumulative—a busy, noisy morning followed by a school bus, followed by needing to organise for the school day may end in a melt-down. How could things be rearranged to reduce the load?
- Build in 'sensory nourishment'- if a person needs a lot of movement how can they ensure they get enough as part of their routine (this is particularly helpful in teens and adults when people are expected to manage their sensory needs more subtly). Fidgets, weighted vests, ball seats, etc., can all make a big difference but only if they are meeting the right need.
- Don't underestimate the degree to which sensory processing underpins everything else (remember the pyramid diagram). For people who have an easy-to-manage sensory system, it can be hard to understand how a scratchy label in a T-shirt can be so distracting that nothing else can register.
- Encourage individuals with FASD and their families or carers to become sensory detectives—in situations which they find challenging. Is there a sensory element that makes things more difficult—are things too loud/not loud enough, too bright/ dull? is there too much sensory information coming in together? what could be blocked out to help? would movement help manage things?

References

1. Jirikowic T, Olson HC, Kartin D. Sensory processing, school performance, and adaptive behavior of young school-age children with fetal alcohol spectrum disorders. Phys Occup Ther Pediatr. 2008;28(2):117–36.
2. Roley S, Blanche E, Schaaf R, editors. Understanding the nature of sensory integration with diverse populations. San Antonio, TX: Therapy Skill Builders; 2001.
3. Franklin L, Deitz J, Jirikowic T, Astley S. Children with fetal alcohol spectrum disorders: problem behaviors and sensory processing. Am J Occup Ther. 2008;62(3):265–73.
4. Bundy A, Murray E. Sensory integration: A. Jean Ayres theory revisited. In: Bundy A, Lane S, Murray E, editors. Sensory integration theory and practice. 2nd ed. Philadelphia, PA: FA Davis; 2002. p. 3–33.
5. Schaaf R, Davies P. Evolution of the sensory integration frame of reference. Am J Occup Ther. 2010;64(3):363–7.
6. Ayres AJ. In: Network PT, editor. Sensory integration and the child: understanding hidden sensory challenges. (25th Anniversary ed., rev. and updated). Los Angeles, CA: WPS; 2005.
7. Schaaf RC, Roley SS. Sensory integration: applying clinical reasoning to practice with diverse populations. Austin, TX: PRO-ED Incorporated; 2006.
8. Ayres A. Sensory integration and learning disorders. Western Psychological Services: Los Angeles; 1972.
9. van Jaarsveld A. Patterns of sensory integration dysfunction in children from South Africa. S Afr J Occup Ther. 2014;44(2):1–6.
10. Mulligan S. Patterns of sensory integration dysfunction: a confirmatory factor analysis. Am J Occup Ther. 1998;52(10):819–28.
11. Davies PL, Gavin WJ. Validating the diagnosis of sensory processing disorders using EEG technology. Am J Occup Ther. 2007;61(2):176–89.
12. College of Occupational Therapists. Practice briefing: the role of sensory integration on occupational therapy. London: RCOT; 2015.
13. American Occupational Therapy Association. Addressing sensory integration and sensory processing disorders across the lifespan: the role of occupational therapy. Bethesda, MD: AOTA; 2015.
14. Pfeiffer B, Koenig K, Kinnealey M, Sheppard M, Henderson L. Effectiveness of sensory integration interventions in children with autism spectrum disorders: a pilot study. Am J Occup Ther. 2011;65(1):76–85.
15. Dunn W. The sensations of everyday life: empirical, theoretical, and pragmatic considerations. Am J Occup Ther. 2001;55(6):608–20.
16. Kinnealey M, Patten Koenig K, Smith S. Relationships between sensory modulation and social supports and health-related quality of life. Am J Occup Ther. 2011;65(3):320–7.
17. Watling R, Patten Koenig K, Davies P, Schaaf R. Occupational therapy practice guidelines for children and adolescents with challenges in sensory processing and sensory integration. New York: AOTA Press; 2011.
18. Jirikowic T, Olson HC, Kartin D. Sensory processing, school performance, and adaptive behavior of young school-age children with fetal alcohol spectrum disorders. Phys Occup Ther Pediatr. 2008;28(2):117–36.
19. Catterick M, Curran L. Understanding fetal alcohol spectrum disorder: a guide to FASD for parents, carers and professionals. London: Jessica Kingsley; 2014.
20. Lucas B, Latimer J, Pinto R, Ferreira M, Doney R, Lau M, Jones T, Dries D, Elliott J. Gross motor deficits in children prenatally exposed to alcohol: a meta-analysis. Pediatrics. 2014;134(1):192–209. https://doi.org/10.1542/peds.2013-3733.
21. Mattson SN, Goodman AM, Caine C, Delis DC, Riley EP. Executive functioning in children with heavy prenatal alcohol exposure. Alcohol Clin Exp Res. 1999;23:1808–15. https://doi.org/10.1111/j.1530-0277.1999.tb04077.x.

22. Mattson S, Riley E. A review of the neurobehavioral deficits in children with fetal alcohol syndrome or prenatal exposure to alcohol. Alcohol Clin Exp Res. 1998;22(2):279–94.
23. Hansen KD, Jirikowic T. A comparison of the sensory profile and sensory processing measure home form for children with fetal alcohol spectrum disorders. Phys Occup Ther Pediatr. 2013;33(4):440–52. https://doi.org/10.3109/01942638.2013.791914.
24. Steinhausen H, Spohr H. Long-term outcome of children with fetal alcohol syndrome: psychopathology, behaviour, and intelligence. Alcohol Clin Exp Res. 1998;22(2):334–8.
25. Wagner B, Fitzpatrick J, Symons M, Jirikowic T, Cross D, Latimer J. The development of a culturally appropriate school based intervention for Australian aboriginal children living in remote communities: a formative evaluation of the alert program® intervention. Aust Occup Ther J. 2017;64(3):243–52.
26. Wells A, Chasnoff I, Schmidt C, Telford E, Schwartz L. Neurocognitive habilitation therapy for children with fetal alcohol spectrum disorders: an adaptation of the alert program®. Am J Occup Ther. 2012;66(1):24–34.
27. Wagner B, Fitzpatrick JP, Mazzucchelli TG. Study protocol for a self-controlled cluster randomised trial of the alert program to improve self-regulation and executive function in Australian aboriginal children with fetal alcohol spectrum disorder. BMJ Open. 2018;8:e021462. https://doi.org/10.1136/bmjopen-2017-021462.
28. Miller LJ, Anzalone ME, Lane SJ, Cermak SA, Osten ET. Concept evolution in sensory integration: a proposed nosology for diagnosis. Am J Occup Ther. 2007;61(2):135–40.
29. Smith-Roley S, Mailloux Z, Miller-Kuhaneck H, Glennon T. Understanding Ayres sensory integration. OT Pract. 2007;12(17):CE1–8.
30. Chu S. Is it sensory based or sensory integration? Clarification of concepts in clinical practice. CYPF Occup Ther J. 2016;20:1.
31. Miller LJ, Schoen S, Mulligan S, Sullivan J. Identification of sensory processing and integration symptom clusters: a preliminary study. Occup Ther Int. 2017.
32. Parham D, Smith Roley S, May-Benson T, Koomar J, Brett-Green B, Burke J, Cohn E, Mailloux Z, Miller L, Schaaf R. Development of a Fidelity measure for research on the effectiveness of the Ayres sensory integration® intervention. Am J Occup Ther. 2011;65(2):133–42. https://doi.org/10.5014/ajot.2011.000745.
33. Dunn W. The impact of sensory processing abilities on the daily lives of young children and their families: a conceptual model. Infants Young Child. 1997;9(4):23–35.
34. Fisher A, Murray E, Bundy A. Sensory integration theory and practice. Philadelphia: F.A. Davis; 1991.
35. Lange S, Shield K, Rehm J, Popova S. Prevalence of fetal alcohol spectrum disorders in child care settings: a meta-analysis. Pediatrics. 2013;132(4):e980–95. https://doi.org/10.1542/peds.2013-0066.
36. DeGangi G. Pediatric disorders of regulation in affect and behavior. 2nd ed. San Diego, CA: Academic; 2017.
37. Bhreathnach E Parent-child engagement. A co-regulation process. In: National Conference for Occupational Therapists in Child and Adolescent Mental Health; 2008.
38. May-Benson TA. A sensory integrative intervention perspective to trauma-informed care. OTA the Koomar center white paper. Newton, MA: OTA The Koomar Center; 2016.
39. Hughes D, Baylin J. The neurobiology of attachment-focused therapy: enhancing connection and trust in the treatment of children and adolescents. New York: WW Norton; 2016.
40. McCullough E, Gordon-Jones S, Last A, Vaughan J, Burnell A. An evaluation of neurophysiological psychotherapy: an integrative therapeutic approach to working with adopted children who have experienced early life trauma. Clin Child Psychol Psychiatry. 2016;21(4):582–602.
41. Ayres AJ. Sensory integration and praxis tests (SIPT). Los Angeles: Western Psychological Services; 1989.
42. Mailloux Z, Parham L, Roley S, Ruzzano L, Schaaf RC. Introduction to the Evaluation in Ayres Sensory Integration® (EASI). Am J Occup Ther. 2017;72(1):7201195030p1–7.

43. Blanche EI. Observations based on sensory integration theory. Torrance, CA: Pediatric Theapy Network, Western Psychological Services; 2002.
44. Parham LD, Ecker C, Miller Kuhaneck H, Henry DA, Glennon TJ. Sensory Processing Measure (SPM): manual. Los Angeles: western psychological services. 2007.
45. Brown C, Dunn W. Adolescent/adult sensory profile manual. San Antonio, TX: Psychological Corporation; 2002.
46. Dunn W. Sensory profile 2 manual. San Antonio, TX: Pearson; 2012.
47. Bundy A, Lane S, Murray E. Sensory integration theory and practice. 2nd ed. Philadelphia, PA: FA Davis; 2002. (3rd edn expected 30/10/2019).
48. Wilbarger J, Wilbarger P. Wilbarger approach to treating sensory defensiveness and clinical application of the sensory diet. Sections in Alternative and Complementary Programs for Intervention, Chapter 14. In: Bundy AC, Murray EA, Lane S, editors. Sensory integration: theory and practice. 2nd ed. Philadelphia, PA: F.A. Davis; 2002.
49. Smith K. 2007. https://sensoryladders.org/about/.
50. Brown S, Shankar R, Smith K. Borderline personality disorder and sensory processing impairment. Prog Neurol Psychiatry. 2009;13:10–6.
51. Williams M, Shellenberger S. How does your engine run? a leader's guide to the alert program for self-regulation. Albuquerque, NM: Therapy Works; 1994.
52. Youngstrom M. The occupational therapy practice framework: the evolution of our professional language. Am J Occup Ther. 2002;56(6):607–8.
53. Parham LD, Mailloux Z. Sensory integration. In: Case-Smith J, Allen A, Pratt P, editors. Occupational therapy for children. St, Louis, MO: Elsevier; 2010. p. 356–411.

Social Care Issues and Complex Family Relationships Intertwined with FASD

25

Joanna Buckard

Chapter Highlights

- Identifying families who need support.
- How to assess needs across ages.
- Interventions for families affected by FASD.

For social workers, FASD is one of the more complex issues of the day. There are certain areas of social work practice with a higher prevalence of clients with or suspected of having FASD such as child protection, fostering and adoption, criminal justice, disabilities and mental health [1, 2].

Social workers in most high-need service areas will be working with clients with FASD. The majority are likely to be undiagnosed. The needs of the clients may be moderate to high and there may be support needs for many years.

25.1 Is It Possible to Predict Which Families Are at a High Risk of Having Child with FASD?

The majority of women in Britain drink alcohol [3] and many pregnancies are unplanned. Women do not always find out that they are pregnant straight away and as the information around alcohol in pregnancy is ambiguous, women may be drinking alcohol in pregnancy without realising that they are causing harm. Obviously, this is a very sensitive area as a diagnosis may still carry stigma and be associated with blame.

J. Buckard (✉)
Red Baloon Training, Derbyshire, England, UK
e-mail: joanna@redballoontraining.co.uk

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_25

However, in a culture where drinking some alcohol is considered normal by the majority of the society, it is imperative that we look more closely at how we manage the information around alcohol in pregnancy.

Women in the UK are the fourth highest consumer of alcohol in pregnancy [4]. This means that many women, even unwittingly, are potentially at risk of having a baby with an FASD.

The majority of these women will not come to the attention of social services during the time that they are pregnant.

25.2 Which Women May Come to the Attention of Social Services?

Research into birth mothers of children with FASD showed that 95% of birth mothers had mental health problems, 90% experienced physical/sexual abuse, that 60% were below the poverty line and that 77% had post-traumatic stress disorder. Without the necessary support, alcohol may have been used as a form of self-medication [5, 6]. This research would have been from groups where the diagnosis was recognised and not necessarily the wider population where alcohol exposure may have occurred. It represents, however, the most common group being seen.

Alcohol problems in the child's family seem to be the most significant risk factor for a child being born with an FASD [7].

In the UK, there is little research around Birth Mothers, but one study suggested that the mothers knew their drinking was likely to be harming their baby, but felt unable to stop drinking [8].

This is significant from a social care perspective because the question must be asked, if given the appropriate support, could these women have stopped drinking and had healthy children and if so, what would have been appropriate support?

There are 94,307 children in care in the UK and 60% are in care due to abuse and neglect [9]. For a significant proportion of these families, alcohol use will be problematic.

25.3 Which Proportion of These Children Will be Alcohol Exposed?

A recent UK study showed 6–17% of children screened positive for FASD [10]. An active surveillance study in the USA found 1.1–5% of children had FASD. This makes it likely to be more prevalent than autism [11] and the leading cause of non-genetic learning disability [12].

The figure for children who are looked-after is likely to be higher due to substance misuse and neglect being a major factor in why many children are removed. A screening prevalence audit in Peterborough found that 34% of looked-after children had been exposed to alcohol prenatally and that figure rose to 75% in children who had been placed for adoption [13].

25.4 Assessment

There are varied reasons as to why a referral may be made to social services. In the case of substance misuse, there appears to be an uneven response depending on the substance. Social work involvement in families where there is alcohol misuse may come to the attention of social services later and follow a different route through the social care system than a family where there is drug misuse [14].

These delays in offering support to the family may have further impact on the development and psychological well-being of the children and on the family's ability to function.

It is important to consider that few mothers who misuse drugs do not also drink alcohol [15]. Any level of alcohol use must be recorded as it may be impossible to get the correct diagnosis without it in the future for the majority of affected children.

In families where the mother has been using drugs and/or alcohol, there are various risks to a child, including neurodevelopmental problems, domestic violence, physical and emotional abuse and neglect [16].

Mothers who use alcohol and drugs often do not care well for themselves in pregnancy and their chaotic lifestyle does not allow for optimum health [17].The effect of prenatal alcohol on an unborn baby may be worse when there has been poor nutrition and if the woman does not attend midwives' appointments, then she may be invisible to services, missing opportunities for support and intervention.

Alcohol misuse by parents damages and disrupts the lives of their children in all social classes and areas of society. It affects the lives of the whole family and harms the development of children [18].

During social work assessments, information is gathered around whether the child is suffering, or likely to suffer, from significant harm or if they are likely or not to achieve or maintain a satisfactory level of health and development and whether it would be significantly impaired without the provision of services.

Questions regarding alcohol use can tend to be about how alcohol and indeed drugs, may be affecting the parent's ability to care appropriately for their children. Questions regarding the dynamic of family life, parental mental health may be discussed, maybe also about the child's birth and whether there were any difficulties. Whether or not questions are asked about prenatal alcohol use is subjective to each social worker and whether they have had appropriate training about alcohol-exposed pregnancies.

According to Badry [19, 20] FASD cases require complex case management and the fact that FASD is not core training may have contributed to the death of a child as described in Two Tragedies [21].

This is also the view of Thomas [22] who decided that social workers were, in fact, the primary profession to work with children and young people who are affected by their family's alcohol and substance use and that they, therefore, require mandatory training in FASD.

However, without the necessary knowledge and training, a social worker may not understand the significance of asking and recording the right questions. This can

have a significant impact on decisions that are made, which referrals a child receives and can make the process of getting a diagnosis harder.

Social workers have the skills to talk about difficult subjects with their clients and this is key to getting the right information. Dialogue and true listening are an art more than a science [23] and is imperative in trying to build up a picture of life for the family before the child was conceived, during the pregnancy as well as after.

Brief screening interventions such as AUDIT-C, T-ACE and TWEAK have been shown to be effective in determining risky from non-risky drinkers [24]. However, they tend to assess dependent drinkers rather than those who are not dependent. From the FASD perspective, any level of alcohol could cause harm to the unborn baby. Therefore, these instruments may tell you when there is a dependent drinker which can make up part of an assessment. However, they may also miss alcohol-exposed pregnancies where the mother is not a dependent drinker. They do, however, offer an initial consistent approach, which may have otherwise been lacking.

Research has shown that the Single Binge Drinking question (SBD) was as effective at identifying women who were at risk of an alcohol-exposed pregnancy. 'In the last 3 months, how often did you have 4 or more drinks on one occasion' [25, 26]. The four standard drinks (US) are equivalent to seven UK units, which could be the same as two large glasses of red wine. This may give the opportunity of early intervention.

The impact of brief interventions has shown to be powerful and by giving information and having an understanding, supportive attitude which seems to be crucial for eliciting behaviour change [27].

However, in order to assess whether there is problematic alcohol use within the family, where there is no dependence, it would most likely have to be assessed through discussion.

Where there is alcohol use within the family, then the following is also relevant in the assessment of the children, asking retrospectively to inform the assessment about the development of the children including emotional, social and behavioural development.

When asking about alcohol use in pregnancy, it could be considered a loaded question and difficult for a client to answer honestly for several reasons:

1. *Memory*—The amount of time that may have passed since the pregnancy may make recall genuinely difficult. Or indeed memory can be impaired through alcohol use.
2. *Knowledge*—Many people in the UK do not understand units and how to count them. They may have been given ambiguous information, about the safety of alcohol use in pregnancy, by professionals, as well as information that they have got from the internet, media, family and friends.
3. *Measurement*—Drinking at home means people tend to free pour their alcohol and there has been an increase in popularity with over-sized glasses. This makes it very difficult to know exactly how much alcohol has been consumed.
4. *Judgement*—Although the assessment is designed to ensure the best outcomes for the child, it is likely to be difficult to move away from the parent feeling

judged by the system and fear their child will be removed. This means that alcohol use in pregnancy may be denied or downplayed.

5. *Poly-drug Use*—After admitting to using illegal substances, a parent may consider that they have admitted to the most serious drugs and may not see the importance or relevance of any alcohol that they have consumed.

The same difficulties are faced by other relevant agencies such as midwives. A study in Scotland into fatty acid ethyl esters in meconium found that the information that women had given about their alcohol use in pregnancy was downplayed compared to the amount shown to have been used in their baby's meconium [28].

This means that in such cases, the information taken and even shared by different agencies may not have been accurate nor the information that is needed in decision-making or in later, making a diagnosis.

Motivational interviewing during the pregnancy can be a relevant tool as feedback and continued discussion keep the dialogue open. They can reflect positives in behaviour change and can reinforce them. It can also provide information for assessments.

25.5 In the Case That the Mother-To-Be Is a Dependent Drinker: What Are the Options?

Social care should be working closely in conjunction with health. Referrals to alcohol support agencies may be made. However, agencies need to work together to support the client. For some women, an inpatient detoxification stay may be the most suitable option, and for some families, this may mean that care may need to be arranged for any other children.

Some clients may refuse an inpatient stay, preferring to detox in the community. For a dependent drinker, stopping drinking suddenly can be risky to both mother and baby, so medical support will be required. Not all alcohol support agencies have undertaken training on FASD, so it is important that all agencies work together to give clear, consistent messages.

If the client is continuing to drink, then it may be appropriate to have an unborn-baby—child-protection plan.

It is important to consider that drinking alcohol in pregnancy also increases the risk of:

- Miscarriage.
- Still-birth.
- Premature birth.
- Sudden infant death syndrome (cot death) [12].

In assessments regarding existing children, the questions asked tend to be about any current difficulties that are presenting within the family.

These could be about difficulties that children are having with:

- Development.
- Behaviour.
- Social relationships.
- Sleep.
- Mental health.

These may be associated with the current situation or even due to more long-standing issues such as drug and/or alcohol use. Indeed, some of these may be attributed to these issues. However, if there has been prenatal alcohol exposure, then the neurodevelopmental effect must also be taken into account.

It is important that where there has been admission of prenatal alcohol use that the response is compassionate; there may have been significant difficulties in her life such as with her mental health, addiction or domestic violence. It is also important to remember that women may have received ambiguous information from family, friends as well as professionals regarding alcohol use in pregnancy.

During the assessment, trying to ascertain whether alcohol may have affected a child is different prenatally to postnatally. Postnatally, the question is with regard to the parent's capacity to parent the child effectively and their ability to provide good enough care for their children. Whereas prenatally, there is no known safe level of alcohol for any one woman to drink. Therefore, if there has been any level of alcohol during the pregnancy, it should be considered whether it is a factor in the child's development and needs. This is complex as the child may not only have been exposed to alcohol or alcohol and drugs but may also be affected by their environment (Fig. 25.1).

If there has been exposure to prenatal alcohol, then this needs to be considered in the assessment of the child's development. A child with FASD is likely to have difficulties that show up in this section. For example, their emotional and behavioural development and self-care skills may be significantly below what would be expected for their chronological age. However, it may not be framed that way by the parent. Self-care skills such as not remembering to brush their teeth may be considered to be laziness or lack of self-esteem, but it may be about a difficulty with memory or hypersensitivity to the feel of the brush or the taste of the toothpaste.

Not following a prompt to get ready may be seen as defiance or not listening, but could be due to auditory processing difficulties as well as working memory problems. Social relationships may have some difficulties, for example, a child may be hurting their siblings or family pets. This may be seen as bad behaviour, that they do not listen or a myriad of other reasons. With a child with FASD, then consideration must also be given to whether the child is picking up on social cues, is impulsive or has sensory processing difficulties.

In the case where the child is part of a sibling group, if any of the children are displaying signs of FASD, then all siblings should be assessed or noted for follow-up as there is 77% sibling recurrence rate [30]. Brief interventions work and counselling referrals may reduce the rate of subsequent siblings being affected.

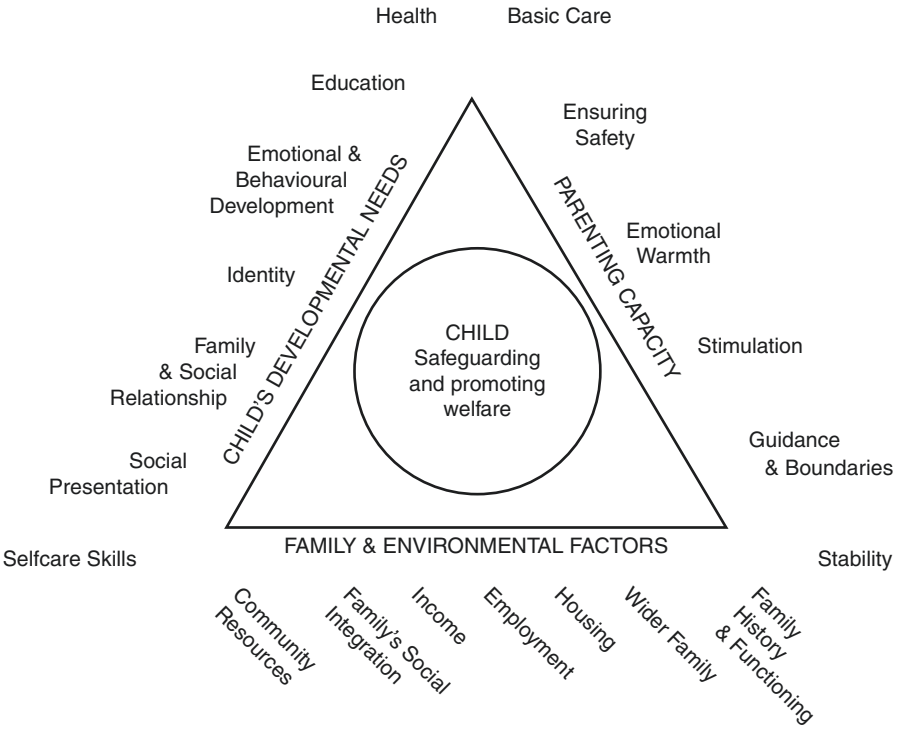


Fig. 25.1 Child safeguarding framework [29]

During assessment, the effect of living in a toxic environment will be considered on the child's outcomes. However, trauma with the presence of FASD shows greater deficits in speech and language, memory, attention, motor skills, emotional and behavioural problems than trauma alone [31].

The neurological effects of prenatal alcohol exposure appear independently of postnatal neglect and neglect does not appear to make the neurodevelopmental presentation any worse [32, 33]. This is potentially different to the non-exposed, typically developing group of children.

The assessment process will require some professional curiosity; the ability to apply critical thinking and consider different hypotheses for some of the presenting difficulties. To consider, are any behaviours or delays purely to do with the environment or is there any evidence of neurodevelopmental issues?

Therefore, it is crucial that the FASD effects are given due regard rather than focusing solely on the effects of neglect and trauma and not considering neurodevelopmental effects from prenatal alcohol exposure.

When assessing the parenting capacity, it may be relevant to conduct the Parent Assessment Manual (PAMs) [34]. This is a comprehensive assessment tool which can be used with vulnerable families, including parents with learning disabilities.

This would be suitable for a parent with FASD because it includes various observations and may inform specific areas where support is needed.

For an adult with FASD, there is a risk of there being an assumption that they cannot successfully parent children [35]. However, each adult is different, so in-depth assessments are required.

25.6 What Support May Families Need?

A family that is caring for an individual with FASD needs to be informed on FASD, so that they can effectively support and advocate for the young person. However, diagnosis is also crucial to accessing external support [36]. When a family is FASD-informed, it gives them the ability to recognise symptoms of the disability rather than simply bad behaviour and equips them with strategies to adopt at home. Beneficial family training could include brain-based techniques for emotional regulation and challenging behaviours.

When the family of a child with FASD have a limited understanding of the disorder, they may lack the ability to access the appropriate resources and not have good coping strategies causing parental/carer stress and risking family breakdown. This may also put their children at a greater risk for developing associated conditions [37].

Researchers indicate that early and accurate diagnosis is vital in the effective treatment of children with FASD [38–40].

This diagnosis can help the team around the child to understand their needs. It is essential that there is a collaborative support network that involves the families, teachers, social workers and health-care specialists [41, 42].

Discovering a child has a disability can bring a period of grief for many families—for a birth family, this may be harder to deal with. For a birth mother of a child with FASD, a referral for specific tailored support would be beneficial such as the birth mother's network.

The majority of children with FASD will not be diagnosed during infancy or even early childhood. Difficulties that are presenting may be attributed to other causes such as neglect or attachment. The child may also get other diagnoses initially, such as ADHD or as being on the autism spectrum.

A child with FASD who is brought into foster care should be placed in the care of experienced and FASD-trained foster carers. If foster carers have not had training, then their ability to manage and effectively advocate for the child may be impaired.

Some children with FASD will be placed under a special guardianship order. Whilst this may offer stability for the child, it is crucial that the family receive support in order to prevent stress risking the breakdown of the family.

Parents and carers can become frustrated when the professionals around them are not FASD-informed and it can leave them feeling lonely, isolated and often unable to cope [43].

Where children are placed for adoption, it is vital that prospective adopters are given information about whether a child has been exposed to prenatal alcohol and that they are informed that the effect may not be clear for some time. Even if they are meeting milestones at the time of adoption.

Everyone has their own genetic potential before they are born making them unique, when there has been alcohol exposure prenatally, its effect can depend on the dose, pattern and timing as well as other factors including, but not limited to [44]:

- Maternal genetics.
- Nutrition.
- Exposure to other drugs—prescribed or otherwise.
- Socio-economic status.
- Maternal age.
- Social relationships.
- Depression.
- Maternal body size.

Therefore, every individual with FASD is going to present differently. A review suggested that there are 428 conditions associated with prenatal alcohol exposure [45]. This means that their individual needs will be specific to how they have been affected.

25.6.1 Infancy

Babies affected by prenatal alcohol exposure may have cause to be referred to a paediatrician due to failure to thrive or one of the many other physiological conditions associated with prenatal alcohol use.

The family may be receiving extra input from a health visitor. For some families, the reason for faltering growth might be considered physiological such as tongue tie, poor sucking reflex etc.; but in other families, it may be considered to be because of neglect. Consideration may be given to maternal mental health as a reason for the infant having faltering growth.

In prenatally alcohol-exposed babies, some babies have poor sucking development and faltering growth.

Babies exposed to prenatal alcohol may have poor sleep/wake cycles and also be irritable and hypersensitive. This can lead to sleep deprivation and high stress levels within the house as the family try to meet the needs of the baby.

During infancy, some babies may fail to meet their milestones such as language acquisition or mobility, although many will. Again, this can be mistaken for neglect which may lead to referral to social services. Referrals to other health professionals such as speech and language therapy, physiotherapy or occupational therapy may be necessary. Research suggests that early input from professionals can improve outcomes for children with FASD.

25.6.2 Childhood

Children with FASD may have additional health needs that require input from various health professionals such as a paediatrician, occupational therapist, psychologist or speech and language therapist. Many children with FASD have sleep difficulties. Health professionals may offer medication in some cases, but it is important to consider the effect on family life when a child is not sleeping properly. Some children have difficulty falling asleep and/or waking in the night and getting up. The family may need aids in order to safeguard the child such as alarms to alert them that the child has got up.

The child may not be achieving at the expected level academically. Some children with FASD have a low IQ, but most children with FASD appear outwardly bright and have an IQ within the normal range. An EHCP is needed for most children with a formal diagnosis of FASD and social care could help to support and advocate for the family where appropriate. The child's learning may be delayed or it may be inconsistent and specific strategies will be needed both at home and in the classroom. Difficult behaviour can seem intentional if the damage to the child's brain is not taken into account. When traditional consequential strategies that are used to manage behaviour are ineffective, the blame can fall onto the family or indeed the child themselves. Understanding a child with FASD's struggle to link cause and effect can take blame out of the situation.

Many children with FASD struggle with social relationships. This means that children can feel excluded and can make it difficult to meet their social needs. If play dates have been arranged, children may need a higher level of supervision and support with their interactions and with managing their emotions than a neurotypical child. As children with FASD may be socially/emotionally younger than their chronological age, altering expectations can help. Other ways to meet their social needs could be to attend play or support groups for children with FASD or other associated disabilities such as ADHD or autism. There are increasing activities which are listed as autism-friendly and with training these may be suitable for children with FASD.

25.6.3 Common Difficulties

Confabulation is common in children with FASD. The child or young person can fabricate imaginary experiences as a compensation for memory deficits. This can be highly problematic because in an articulate child, the confabulation may sound plausible and they may be confident about their recollections, despite contradictory evidence. There is not a conscious intention to deceive. However, allegations arising from confabulation have led to instances of safeguarding investigations.

Older children who are in the care system who have FASD which has not been recognised or diagnosed are less likely to be adopted. They are also more likely to

have experienced more placement breakdown and moves. Therefore, the young people can present more complex behaviour.

25.6.4 Adolescence

Adolescence can be a very challenging time when caring for a young person with an FASD. The gap between a neurotypical adolescent and an adolescent with FASD can appear wider than ever. The immaturity and delays in social/emotional development become even more apparent.

Adolescence is typically the time during which young people start to develop independence skills. Certainly, for young people who are looked-after, this is actively pushed. However, it is essential that the functional and developmental age of the young person is taken into account, especially for a young person who presents well due to sounding age-appropriate.

As many young people with FASD present at a higher level than they typically function, it is really important that the psychological and speech and language assessments have been completed in order to show their ability in a number of domains. This can lead to a young person having what is referred to as a 'spiky profile'. This is where the person's range of abilities is not consistent with their IQ, highlighting both strengths but importantly weaknesses. They may have an IQ within the normal range, but they may perform significantly lower in other domains such as comprehension, adaptive behaviour, executive functioning tasks and social maturity. This could mean that the outwardly chatty young person who presents well may actually be highly vulnerable, as they may not perform all the independent tasks they say that they can or they may not pick up when they are being manipulated. This was identified by 92% of caregivers in a study which also reported that 87% of their participants had been victim to some form of violence and 77% of individuals had experienced physical and/or sexual abuse. Being easily manipulated can have implications for adolescents and adults with FASD involved in the legal system. For example, a vulnerable individual may be more likely to acquiesce, give a false confession or have problems understanding cautions and consents [46].

Therefore, a young person with FASD may also be more vulnerable to all types of grooming including Child Sexual Exploitation (CSE). This is because the young person may have few or no friends and seeks social relationships. They may not be able to recognise when they are being manipulated or what someone's intentions are. Consequently, the young people can be at an increased risk on social media.

For young people who are looked-after, it is essential that these assessments are completed in order to access the appropriate next steps. Living fully independently may not be realistic at 18 for many young people with FASD. Assessments conducted by social workers need to take into account that the young person may be able to explain what they will need to do but not be able to fulfil this without support (talk the talk, but not walk the walk). To overlook this would be to put the young person at risk.

25.7 Social Care Support for a Family with a Child with Prenatal Alcohol Exposure

Referrals to other agencies may be appropriate including CAMHS for support with any mental health issues since around 90% of people with FASD as they get older develop mental health problem [37].

There may be various therapies and opportunities that could be offered that may help a young person with FASD such as friendship training. This may have elements such as reading facial expressions, social cues and the use of social stories. However, therapies designed for attachment, which have not taken into account neurological issues in this group and their difficulty to understand and process emotion-focused approaches, may not work for a young person with FASD.

Referrals to parenting courses and family strengthening sessions may be useful but would need to be delivered by a practitioner who understands FASD.

Signposting for appropriate benefits such as DLA can help to support the family and help to fill out the forms.

Where services exist, referrals to executive functioning training such as working on areas such as working memory could help the young person to improve their functioning.

Family support could help a family to look at routines and how having consistency in the routine may help a child or young person with FASD. Also, how to consider environmental changes that may help such as having a visual timetable and reminders such as to wash hands. Children with FASD are likely to be calmer when the environment around the child is calm and quiet and finding options for the child to go to a safe space, such as a pop-up tent in order to regulate their mood can be useful. Sensory activities can also be helpful for a dysregulated child.

There may be issues relating to eating such as difficulty with chewing and swallowing which may need a referral to support with feeding. Children may also struggle to tell if they are feeling hungry or full which can lead to under/over-eating.

A child may be hypersensitive to certain foods such as not liking pieces of fruit in yoghurt, lumps in mashed potato, sour or spicy foods. However, they may be hyposensitive to foods and be sensory seeking, preferring foods with texture and strong flavours. If a parent did not understand why the child was being a ‘difficult eater’ again, it can lead to parental stress.

25.8 Respite

For some families, it would be beneficial to identify suitable respite carers. Ideally who will commit to learning about the needs of a child with FASD and to supporting the family by looking after the child to give a break to their main carers. By having the same respite family/carer, it provides consistency to the child, who may struggle with change, and enables the carer to better understand the child’s needs. This can also reduce parental/carer stress and reduce the risk of family/placement breakdown.

25.9 FASD Resources

There is huge variance across the UK in terms of access to resources. This is at every stage from recognition and diagnosis through to available services for those diagnosed.

Some areas have paediatricians who are able to diagnose the full range of FASD diagnoses; however, there are still areas where the FAS, rather than the wider FASD diagnosis is obtainable. Obviously, this leads to a postcode lottery. The national clinic, offering gold standard diagnosis, will take referrals from all over the country, but not all Clinical Commissioning Groups have been prepared to pay for the service. The clinic is unable to provide ongoing support to patients and their families.

FASD-specific resources that are available to families and individuals tend to be from the third sector, rather than governmental. There are limitations in access to these based on socio-economic status, access to technology (internet access) and geographic location. Support groups are held by FASD charities in several locations around the country and there is an FASD UK alliance group on Facebook which is active in bringing together those living with FASD and building a sense of community. The FASD charities also have support telephone lines and can also provide FASD-related materials. Similar support groups exist in other areas of the world.

For children who have been adopted then, in the UK, the Adoption Support Fund can be a way to access therapeutic services. This fund is now also available for children from intercountry adoption and for special guardians if the child was looked-after immediately prior to the special guardianship order. An assessment will be carried out by the local authority that placed the child for the first 3 years, then it is the responsibility of the local authority where the child resides. Obviously, there are many children with FASD who do not fit into the required categories which means they do not necessarily have the same opportunity to access support.

Children and young people who are adopted, have a special guardianship order, residence order, have been in local authority care for more than a day should be eligible for pupil premium in their education setting. The funding should not simply be absorbed into mainstream budgets but instead becarefully targeted at the designated children. This can be used for additional staffing and resources, but for the benefit of a child with FASD, it should be for FASD-specific resources and trained staff. Discussion with parents about how the money should be spent on the young person would be beneficial.

Home schooling may be a lifestyle choice for some; for others, it is evidence of a school system that is failing children with FASD. Parents and carers are faced with nearly impossible decisions and after huge amounts of stress and upset from a child being regularly excluded and misunderstood, some families choose to home educate. Social care may be able to work with and advocate for the family with schools.

The outcomes for looked-after children are significantly below that of the rest of the population with less attaining educationally and more becoming homeless, young parents and experiencing mental health problems. This is often attributed to trauma, attachment, placement instability but is there something else? If someone

has FASD and it is diagnosed early and the right support and structure is put in place might these outcomes improve? Are these outcomes actually a result of inappropriate care and support being offered to these young people and that it ends too early?

Without the right level and type of support, the risk of an individual coming into contact with the criminal justice system is high. Examples of crimes a client with FASD may commit can include shoplifting, assault, sexually harmful behaviour, handling stolen goods, possession of drugs with intent to supply. These may be because of organic brain damage leading to impulsivity, not linking cause and effect, poor memory, social immaturity, not reading facial expressions or body language, being easily led.

Young people with FASD are significantly more likely to become involved in the criminal justice system compared with neurotypical adolescents [47–50].

One study cites the rate of people in prison with FASD as 23.3% [49] and another as between 30 and 40% [51]. There are currently no known UK studies into how many people there are in prison with FASD. However, there are studies that may be relevant to this discussion.

7% of adult prisoners have a learning disability demonstrated by an IQ score of less than 70. There were 25% more found to have an IQ score of less than 80 [52]. The figures for those in prison who are under 18 were even higher, one study found 23% to have an IQ score under 70 and 36% had scores between 70 and 79 [53].

Dyslexia, a hidden disability, is three to four times more common amongst prisoners than in the general population [54, 55].

In one study of over half of prisoners had attended a special school at some point during their education, this figure increased to almost two-thirds for those who had possible learning or borderline learning disabilities; a further 10% said that whilst attending a mainstream school, they had received extra support. There were also prisoners who said that they had attended ‘bad schools’ for behavioural problems and pupil referral units [56].

There are significantly higher numbers of mental health disorders amongst the prison population with 72% of male and 70% of female sentenced prisoners suffer from two or more mental health disorders [57].

No One Knows [56] recognised that prisoners with learning disabilities and difficulties, especially where hidden, are ‘at risk of re-offending because of unidentified needs and consequent lack of support or services are unlikely to benefit from programmes designed to address offending behaviour are targeted by other prisoners when in custody’ [58].

ADHD action has estimated that 30% of prisoners could have ADHD [59]. As we do not have an accurate prevalence of prisoners with FASD, it is important to consider the data we do have and whether there are adults with undiagnosed FASD hidden within these numbers.

FASD is widely referred to as an invisible disability. It presents very differently in each individual, but in the prison population, there is likely to be a number who have never received a diagnosis of FASD, who are articulate and do not outwardly present as having a learning difficulty but who remain at a significant risk of reoffending without the right level of support. For an adult with FASD, there are

several brain-based difficulties that may increase the risk the re-offending including difficulty linking cause and effect, impulsivity, vulnerability and being easily led.

25.10 How Could Social Care Reduce the Risk of an Adult with FASD Offending?

Streissguth [37] found that diagnosis and the right level of support reduce the risk of associated conditions and could reduce the risk of the individual coming into contact with the criminal justice system.

The support needs will be subjective to the individual but during the process of diagnosis looking at the person's comprehension, executive functioning and adaptive behaviours through speech and language and psychological testing can help to inform the level of help and support the client would require.

Some clients may need to be in a residential setting, others in a semi-independent setting, some may be able to live independently with support and others can live independently. Areas of need are likely to include help with budgeting, shopping, managing their health, social relationships, help to access work and with referrals to other agencies for support.

The risk of 'Mate Crime' is high because you may have an individual who is prosocial but who lacks the necessary skills to protect themselves from being manipulated. An area for heightened concern is managing social media. Social media such as Facebook, Twitter etc. can be a good place to meet new people when you have difficulty with social relationships. However, the difficulty with filtering information, being easily manipulated and being impulsive can be particularly risky.

Diagnosis is important for recognising the functional level of the individual for them to be able to access appropriate services. Referrals can be made via health to the national FASD clinic.

In order to protect vulnerable adults, a mental capacity assessment may be undertaken. However, the Mental Capacity Act [60] lacks nuances about the degrees of incapacity the client may have. Clients with FASD may be financially vulnerable, may self-neglect and may be vulnerable to mate-crime without having true insight. There can be inconsistency in their ability. Without training, a social worker conducting the assessment may not understand the lack of insight and might mistake the client's ability to explain what they are going to do, for their ability to follow it through. They may confuse unwise decision-making with capacity to make welfare decisions. This may also be true for an Independent Mental Capacity Advocate (IMCA).

The Brain Injury Needs Indicator (BINI) has been created for use with adults who have had an acquired brain injury (ABI). Like in most cases of FASD, ABI is invisible, and therefore, difficulties are wrongly attributed to wilful behaviour and often widely misunderstood by health- and social-care services [61].

There are other crossovers between an acquired brain injury and FASD with similar difficulties in areas such as:

- Slower processing speeds.
- Attention difficulties.
- Memory problems.
- Difficulty with planning, organisation and problem-solving.

Their behaviour may be disinhibited, they may be concrete thinkers, who act impulsively or aggressively and have difficulty in predicting and understanding the consequences of their actions [62].

Therefore, creation of an FASD specific tool for social workers to use may be appropriate when conducting a mental capacity assessment with somebody suspected of having a hidden disability such as FASD. The BINI asks the same questions to the client and also a friend/relative which can show discrepancies in the client's insight into their own needs.

Summary Points

- Individuals with FASD can lead happy, fulfilled and successful lives.
- This is more likely where health, education, social care and society at large understand how FASD may affect an individual.
- Social workers have a role to implement the relevant supports in various settings.

Practice Points

- Prenatal alcohol exposure should be considered during social work assessments.
- FASD is more prevalent than previously thought and numbers likely to be significantly higher for children who are looked-after or adopted.
- FASD training should be mandatory for all social workers.
- Researchers indicate that early and accurate diagnosis is vital in the effective treatment of FASD.
- A collaborative support network that involves the families, teachers, social workers and health-care specialists is essential.

References

1. Fuchs D, Burnside L, Marchenski S, Mudry A. Children with FASD related disabilities receiving services from child welfare in Manitoba. Int J Ment Health Addict. 2010;8:232–44. https://doi.org/10.1007/s11469-009-9258-5.
2. Williams SJ. Research brief: is there justice in the juvenile justice system- examining the role of fetal alcohol Spectrum disorder. Justice Policy J. 2006;3:1–15.
3. NHS Digital. Health Survey for England—2016 Trend tables, Table 10. [Online]. https://digital.nhs.uk/media/34524/Health-Survey-for-England-2016-Adult-health-trends-tables/default/HSE2016-Adult-trends. Accessed 21 Feb 2018.
4. Popova S, Lange S, Probst C, Gmel G, Rehm J. Estimation of national, regional, and global prevalence of alcohol use during pregnancy and fetal alcohol syndrome: a systematic review

and meta-analysis. Lancet Glob Health. 2017; published online Jan 12. https://doi.org/10.1016/S2214-109X(17)30021-9.
5. Astley S, Bailey D, Talbot C, Clarren SK. Fetal Alcohol Syndrome (FAS) primary prevention through FAS diagnosis: I. Identification of high-risk birth mothers through the diagnosis of their children. Alcohol Alcohol. 2000;35(5):499–508.
6. Astley S, Bailey D, Talbot C, Clarren SK. Fetal alcohol syndrome (FAS) primary prevention through FAS diagnosis: II. A comprehensive profile of 80 birth mothers of children with FAS. Alcohol Alcohol. 2000b;35(5):509–19.
7. Ceccanti M, Fiorentino D, Coriale GO, Kalberg W, Buckley D, Hoyme E, Gossage P, Robinson LK, Manning M, Romeo M, Hasken JM, Tabachnick B, Blankenship J, May PA. Maternal risk factors for fetal alcohol spectrum disorders in a province in Italy. Drug Alcohol Depend. 2014;145:201–8.
8. Thomas R, Mukherjee RAS. Exploring the experiences of birth mothers whose children have been diagnosed with FASD. Adv Dual Diagn. 2018;12(1/2):27–35.
9. NSPCC. Children in care statistics. 2016. https://www.nspcc.org.uk/preventing-abuse/child-protection-system/children-in-care/statistics/. Accessed 11 Jul 2017.
10. McQuire C, et al. Screening prevalence of fetal alcohol spectrum disorders in a region of the United Kingdom: a population-based birth-cohort study. Prev Med. 2019;118:344–51.
11. Office of National Statistics. Mental health of children and young people in Great Britain. London: Palgrave Macmillan; 2005.
12. BMA. Fetal alcohol spectrum disorders. A guide for healthcare professionals. London: BMA Science and Education Department; 2007.
13. Gregory G, Reddy V, Young C. Identifying children who are at risk of FASD in Peterborough: working in a community clinic without access to gold standard diagnosis. Adopt Foster. 2015;39(3):225–34.
14. Adamson J, Templeton L. Silent voices: supporting children and young people affected by parental alcohol abuse. London: The Office of the Children's Commissioner; 2012.
15. Preece PM, Riley EP. Alcohol, drugs and medication in pregnancy—the long term outcomes for the child. London: Keith Mac Press; 2011.
16. Advisory Council on the Misuse of Drugs. Hidden Harm: responding to the needs of children of problem drug users. London: Home Office; 2003.
17. Guerrini I, Jackson S, Keaney F. Pregnancy and alcohol misuse. BMJ. 2009;338:829–32.
18. Turning Point. Bottling it up: the effects of alcohol misuse on children, parents and families. London: Turning Point; 2006.
19. Badry D. Fetal alcohol spectrum disorder standards: supporting children in the care of children's services. First Peoples Child Fam Rev. 2009;4(1):47–56.
20. Badry D. The FASD (Fetal Alcohol Spectrum Disorder) Community of Practice (CoP) in Alberta Human Services: Leading from Within Initiative; Alberta Centre for Child, Family and Community Research; 2013.
21. Pringle B. Two tragedies—holding systems accountable. Saskatoon: Saskatoon Advocate; 2014.
22. Thomas MM. A content analysis of literature on alcohol abuse in families, including social workers' training and education in this area. Unpublished master of social work thesis at the California State University, Long Beach; 2011.
23. Parton N, O'Byrne P. *Constructive social work: towards a new practice*; 2000. London: Palgrave Macmillan; 2000.
24. Bailey BA, Sokol RJ. Pregnancy and alcohol use: evidence and recommendations for prenatal care. Clin Obstet Gynaecol. 2008;51:436–44.
25. Johnson KE, Sobell MB, Sobell LC. Using one question to identify women at risk for an alcohol exposed pregnancy. J Am Osteopath Assoc. 2010;110(7):381–4.
26. Balachova T, Sobell LC, Agrawal S, Isurina G, Tsvetkova L, Volkova E, Bohora S. Evaluating alcohol use among Russian women at risk for an alcohol-exposed pregnancy: a comparison of three measures of alcohol use. J Ethn Subst Abuse. 2018;17(3):324–34.
27. Marais S. The effect of brief interventions on the drinking behaviour of pregnant women in a high-risk rural South African community: a cluster randomised trial. Early Child Dev Care. 2011;181:463–74.

28. Abernethy C, McCall KE, Cooper G, Favretto D, Vaiano F, Bertol E, Mactier H. Determining the pattern and prevalence of alcohol consumption in pregnancy by measuring biomarkers in meconium. Arch Dis Childhood Fetal Neonatal Ed. 2017. https://doi.org/10.1136/archdischild-2016-311686.
29. DOH. Framework for the assessment of children in need and their families. London: The Stationery Office; 2000.
30. Alberta Medical Association. Alberta Clinical Practice Guidelines. Recommendations on Prevention of Fetal Alcohol Syndrome (FAS) Alberta Partnership on Fetal Alcohol Syndrome. Alberta Medical Association, The Alberta Clinical Practice Guidelines Program; 1999.
31. Price A, Cook PA, Norgate S, Mukherjee RAS. Prenatal alcohol exposure and traumatic childhood experiences: a systematic review. Neurosci Biobehav Rev. 2017;80:89–98.
32. Henry J, Sloane M, Black-Pond C. Neurobiology and neurodevelopmental impact of childhood traumatic stress and prenatal alcohol exposure Western Michigan University, Kalamazoo. Lang Speech Hear Serv Sch. 2007;38:99–108. American Speech-Language-Hearing Association 0161–1461/07/3802–0099.
33. Mukherjee RAS, Cook PA, Norgate S, Price A. FASD and the impact of neglect. Alcohol. 2018. https://doi.org/10.1016/j.alcohol.2018.06.002
34. McGraw S. Parent Assessment Manual (PAMS 4.0). 2016. http://www.pillcreekpublishing.com. Accessed 11 Jul 2017.
35. Hacking I. The social construction of what? Cambridge, MA: Harvard University Press; 1999.
36. Streissguth AP, Brookstein FL, Barr HM, Sampson PD, O'Malley K, Young JK. Risk factors for adverse life outcomes in fetal alcohol syndrome and fetal alcohol effects. J Dev Behav Pediatrics. 2004;25(4).
37. Streissguth AP, Barr HM, Kogan J, Brookstein FL. Understanding the occurrence of Secondary Disabilities in Clients with Fetal Alcohol Syndrome (FAS) and Fetal Alcohol Effects (FAE): Final report to the centers for Disease Control and Prevention (CDC). Seattle: University of Washington; 1996.
38. Chandrasena AN, Mukherjee RAS, Turk JT. Fetal alcohol spectrum disorders: an overview of interventions for affected individuals. Child Adolesc Ment Health. 2009;14:162–7.
39. Mutch R, Peadon EM, Elliott EJ, Bower C. Need to establish a national diagnostic capacity for foetal alcohol spectrum disorders.2009. Paediatr Child Health. 2009;45(3):79–81.
40. Peadon E, Rhys-Jones B, Bower C, Elliot EJ. Systematic review of interventions for children with fetal alcohol spectrum disorders. BMC Pediatr. 2009;9:35.
41. Green J. Fetal alcohol spectrum disorders: Understanding the effects of prenatal alcohol exposure and supporting students. J Sch Health. 2007;77:103–8. https://doi.org/10.1111/j.1746-1561.2007.00178.x.
42. Streissguth A. Fetal alcohol syndrome: a guide for families and communities. East Peoria, IL: Versa Press; 1997.
43. Whitehurst T. Raising a child with foetal alcohol syndrome: hearing the parent voice. Br J Learn Disabil. 2011. Published Online. https://doi.org/10.1111/j.1468-3156.2011.00692.x.
44. May P, Gossage JP. Maternal risk factors for fetal alcohol spectrum disorders: not as simple as it might seem. Alcohol Res Health. 2011;34(1).
45. Popova S, Lange S, Shield K, Mihic A, Chudley AE, Mukherjee RAS, Bekmuradov D, Rehm J. Comorbidity of fetal alcohol spectrum disorders: a systematic review and meta-analysis. Lancet. 2016;387:978–87.
46. Clark E, Lutke J, Minnes P, Ouellette-Kuntz H. Secondary disabilities among adults with fetal alcohol spectrum disorder in British Columbia. J FAS Int. 2004;2(e13):1–12.
47. Baumbach J. Some implications of prenatal alcohol exposure for the treatment of adolescents with sexual offending behaviors. Sex Abuse J Res Treat. 2002;14:313–27.
48. Brown NN, Connor PD, Adler RS. Conduct-disordered adolescents with fetal alcohol spectrum disorder intervention in secure treatment settings. Crim Justice Behav. 2012;39(6):770–93.
49. Fast DK, Conry J, Loock C. Identifying fetal alcohol syndrome among youth in the criminal justice system. J Dev Behav Pediatr. 1999;20:370–2.

50. Moore TE, Green M. Fetal alcohol spectrum disorder (FASD): a need for closer examination by the criminal justice system. Criminal Rep. 2004;19:99–108.
51. Bower C. One in three young people in prison have FASD in Western Australia. 2017. http://www.abc.net.au/news/2017-03-03/one-in-three-young-people-in-prison-have-fasd-in-wa/8319724. Accessed 12 Jul 2017.
52. Mottram PG. HMP Liverpool, Styal and Hindley study report 2007. Liverpool: University of Liverpool; 2007.
53. Harrington R, Bailey S. Mental health needs and effectiveness of provision for young offenders in custody and in the community. London: Youth Justice Board for England and Wales; 2005.
54. Rack J. The incidence of hidden disabilities in the prison population. Egham: Dyslexia Institute; 2005.
55. DOH. Positive practice positive outcomes- a handbook for professionals in the criminal justice system working with offenders with learning disabilities. London: Department of Health; 2011.
56. Talbot J. No one knows. PRISONERS VOICES experiences of the criminal justice system by prisoners with learning disabilities and difficulties. Prison Reform Trust; 2008.
57. Bromley Briefings, Prison Factfile. Prison reform trust. London; 2008.
58. Loucks N. Identifying and supporting prisoners with learning difficulties and learning disabilities: the views of prison staff in Scotland. London: Prison Reform Trust; 2007.
59. ADHD Action. https://www.bbc.co.uk/news/uk-england-47443263. Accessed 27 Jun 2019.
60. Mental Capacity Act. 2005. http://www.legislation.gov.uk/ukpga/2005/9/contents. Accessed 15 Jul 2017.
61. Brain Injury Rehabilitation Trust. Brain injury needs indicator (BINI). The disabilities trust. http://www.thedtgroup.org/brain-injury/for-professionals/resources/the-brain-injury-needs-indicator-bini. Accessed 15 Jul 2017.
62. Holloway M. Making the abstract real: acquired brain injury and mental capacity. A report making recommendations following the House of Lords Select Committee review of the Mental Capacity Act. London: Report to Department of Health; 2014.

Transition into Adulthood and Avoiding Secondary Disabilities

26

Raja A. S. Mukherjee

Chapter Highlights

- Changing presentation of people with FASD across the ages.
- The impacts of missing an FASD diagnosis.
- Timelines when interventions can take place to support long-term needs.

The presentation and challenge of fetal alcohol spectrum disorders on individuals and their families change as the individual grows older. Complexities and difficulties become more evident as society places greater demand on individuals, to the point where these individuals quite often are insufficiently able to manage and respond. As shown in Fig. 26.1, there is a divergence that occurs from in those with FASD to normal development trajectories. That is, the expectations placed on an

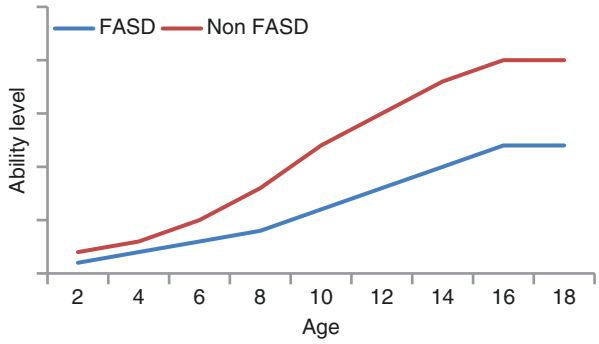


Fig. 26.1 Diagrammatic representation of developmental divergence in people with FASD

R. A. S. Mukherjee (✉)
National FASD Specialist Behaviour Clinic, Surrey, UK
e-mail: Raja.Mukherjee@sabp.nhs.uk

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_26

individual when compared to their actual developmental trajectory are closer together when younger. However, as the typical (norm referenced) group progresses more rapidly, when compared to the FASD group, who have a shallow trajectory of development, get and further apart. It is then that difficulties come more to the fore.

These difficulties may then present as disordered. In part, this is because of the level of expectation placed on the individual when compared to their ability to cope in a societal setting. As individual's progress through teenage life to adulthood, the rules, expectations and even the laws that govern them change. This means that for many, transitions can often be the most challenging time. This is not only for the individuals themselves but also for the families who have tried their best to support the individual. For example, a common difficulty is where parents and families legally can no longer offer the same level of input or influence for decision-making. This change can appear with little or no warning when the individual turns 18.

This chapter will look at some of the difficulties faced, as well considering approaches to try and minimise the impact of these difficulties.

26.1 Issues in Late Teenage Life Challenges of Changing Expectation

As a child develops into a teenager and then later into an adult, developmental maturity is seen in numerous areas. From a cognitive point of view, their primary area of development and improvement for the majority of teenagers is in executive function. This in part relates to higher level functioning and the ability to moderate and manage their own behaviours without the support of adults. Increasing responsibility is given to children in these situations. A good example of how these responsibilities change over time can be seen as a child progresses from infant school to middle schools and then later to secondary education. Early in the school life of a child, they are often supported in the same classroom by the same teacher. Later in the primary school years, it is often seen that children are encouraged to organise parts of their own daily routine. There may well be different teachers with greater expectations on self-organisation being put into place. By secondary school, children often having to walk to school on their own, organise their own routine with less reliance on the teacher or parent supporting this. More often than not, lessons will be taught by different people in the subject area, rather than by a single teacher. Organisational skills and expectations increase and responsibilities go alongside this.

The cognitive demand and the neural maturation that occurs during these processes are often more involved than many expect. In the absence of higher level executive maturity, these children struggle as they do not develop as quickly as others.

Table 26.1 below highlights anonymised audit data from the National FASD specialist clinic from the first 87 cases seen up till 2014. It shows that for an average age of 9 years and 9 months, with a range between 6 and 16, there were a range of abilities seen. In some areas, the difference between the actual average age and the

Table 26.1 Vineland Behaviour Scales adaptive age equivalence summary from 2014 UK National FASD Specialist Clinic audit $n = 87$ (consent received from all patients for anonymous auditing and distribution of any data collected)

Domain	Minimum	Maximum	Average
Age	6.10	16.00	9.93 (95%confidence interval)
Receptive language	0.9	18	3.6 (3.1–4.1)
Expressive language	0.9	22	5.7 (5.1–6.2)
Written language	–	15.3	8.2 (7.6–8.7)
Personal daily living skills	2.3	22	6.1 (5.4–6.9)
Domestic daily living skills	1	20	6.2 (5.4–6.9)
Community skills	–	18.9	7.48 (6.7–8.1)
Interpersonal skills	0.3	17.9	4.6 (4.0–5.3)
Play/leisure socialisation	1.0	13.6	4.9 (4.2–5.4)
Coping	1.1	12.6	4.8 (4.2–5.3)

functional age is not too far apart. For example, written language was near the expected level. For other areas, a wide gap is seen from expected levels. In some cases, the age is half the chronological age, for example, receptive language and coping skills. These deficits can be exacerbated and become more evident as the individual gets older.

Because societies expectation is for an older individual to be able to manage and do tasks without support, in keeping with their chronological age, when support offered as a younger child is removed the individual's ability to function deteriorates. Judging people by chronological age, as highlighted here, therefore is not something that will necessarily work in this group. This leads to some of the perceived difficulties and challenges. It is the external expectation which needs to be modified in this group, while offering ongoing support.

The above table highlights the mixed 'spiky' pattern of presentation with strengths and weaknesses. It is often the strength that masks the difficulty. The individual themselves will also, more often than not, have expectations of themselves, based upon societal norms. If these cannot be matched, this will have subsequent impact on self-esteem and function. They may want to act in a typical manner, akin to many others who have chronic illnesses who deny their issues. This can lead in some to a rejection of the difficulties and a desire for normality. When they cannot achieve what they hope, this impact on self-esteem can lead to mental health problems. Their vulnerability can lead to them being taken advantage of by others.

In these situations, identifying early the deficits and working with the individual to support them to better understand their own needs will keep them safe. Where prenatal alcohol is involved, some families may wish to not have to address the diagnosis, partly due to the difficulty this challenging conversation may involve. However, it is only when the individual themselves truly understands their own situation, and is willing to engage with help and support, that some of the later disabilities can be prevented. Where this is not seen, often it is the case that the secondary

difficulties and disabilities are exacerbated [1]. For many it is only when the individual experiences for themselves the problems that many seek to re-engage with help and seek support.

26.2 Adulthood and Changing Expectations

The transition into adulthood for individuals can be challenging but also an opportunity. In many cases, due to the difficulties faced, families may have supported individuals through many challenges. For some individuals, this may well be perceived as controlling. The changing legal rights and responsibilities as an adult, means the individuals will have the right to make their own decisions. This can lead to some individuals rejecting perceived controls and acting in a way counter to their perceived best interest. Decision-making, capacity to choose and best interest decisions all come to the fore.

As highlighted in Table 26.1, the individual's chronological age and functional age may differ. This is not always taken into account. Decision-making is a good example of where the individual with FASD may well place themselves into difficulties. In order to decide on an outcome in a capacitous way, generally the individual will need to understand what is involved in the process, weigh up the different options that exist and then decide what their preferred option, which is then communicated. This involves multiple cognitive processing, including working memory, shifting attention between pieces of information, communication and executive planning. In order to do this, a higher level of ability is often needed. It may not be necessarily always obvious that these individuals struggle. As such questions around capacity can often arise.

In abstract situations, where capacity is tested and individuals are asked direct questions, which do not require the higher level functioning as described above, individuals may well be able to answer single answers. They may not be able to do this as well if direct questions are not asked. Whilst this may be often a strategy used in childhood to see what the person knows, it is not normal in adults. This highlights that changing expectation and adhering to societal norms is as much a challenge for the adults as the underlying deficits in their cognitive function.

Table 26.2 Secondary disabilities seen in 21-year cohort of clinic patients with FASD [1]

Disability	%
Psychiatric problem	90
Disrupted school experience	60
Trouble with the law	60
Confinement	50
Inappropriate sexual behaviour	50
Alcohol/drug problems	30

The neurocognitive profiles, described elsewhere in this book, will highlight that where there is the need for multiple cognitive processing the same time, it is a particular challenge for individuals with this condition. Simple processing which involves only one task at a time is possible but not when combining them together. The individuals tend to act more in the here and now rather than considering themselves in the context of their future situation. This can cause significant challenges.

Different situations and conditions have higher levels of need for capacitous decisions to be proven. A decision around an operation or medical treatment which has permanent impact on the individual may have a higher threshold compared to simple everyday actions. Broadly some of these everyday actions, including sexual activity, which is known to have a lower threshold for capacity, may well lead in these individuals being deemed to have capacity but not really understanding many of the consequences at the time of the action. This can lead to traumatic experiences that further impact on the secondary disabilities and ability to function in broader situations.

Table 26.2 highlights the nature of the secondary disabilities that are commonly seen in this presentation. These can be wide-ranging. Increasingly it is recognised that these issues will often occur if steps are not put into place to modify early behaviours at an early stage. Even then, for many, these issues are still seen. Families who have tried hard to support individuals and therefore reduce the chance of harm can lead to feelings of rejection for the parent and family if the person chooses a different path. This for some can lead to desperation in their inability as families to intervene. Often where harm is seen and boundaries are not managed by the individual themselves, legal guardianship rules can be used, but this is reliant on capacity. As a child the individual can be protected but where an individual is deemed to be able to make a capacitous decision, then they are allowed to make unwise choices. This can be hard for the parent, especially where the capacity is called into question.

Families will often describe the transition from childhood to adulthood as daunting. In children's services, often there are professionals who have a good understanding of developmental disorders, yet in the adult world, this is often lacking. Where the individual is found to have an intellectual disability, that is, an IQ below 70, they may well have access to specialist services for the developmentally or intellectually disabled group; however, those with an IQ above 70 and no clear obvious functional deficit do not have access to any specialist services currently. Even where comorbid diagnoses such as autism and ADHD can be made, the availability of services is limited. This lack of developmental understanding, at a time where a loss of parental control is taking place, leaves many families feeling hopeless. Significant amounts of support can be undone quickly where the individual themselves wishes to experience different situations without realising or considering the potential consequences. While this can be argued is the situation for many, the naiveté and lack of forethought seen in this group as typical behaviours only exacerbates the risk and the difficulties. In some cases, this can lead to another generation of problems.

26.3 Particular Difficulties That Perpetuate into Adulthood

26.3.1 Sexuality

As with any teenager developing into adulthood, sexuality becomes part of normal life. There has been no research to suggest that individuals with FASD have less sexual desire, rather the opposite is often reported. Many descriptions would suggest that the development of sexuality causes a great deal of problems for these individuals. Whilst there is very little research on to the underlying mechanisms and processes that may moderate this, in this group of individuals, Table 26.2 above highlights that for 50% of the cohort who were followed up, sexually inappropriate behaviours were seen. In females, reports are commonly heard about exploitation, abuse and in the worst cases rape. In many males, the opposite was also seen. A lack of understanding of their own social situation, in an individual who developmentally functions at a lower level means that they may well interact with younger individuals. These chronologically younger individuals may be developmentally similar. They may also develop their sexuality, but because these individuals are older, it leads to potential safeguarding issues and in some, accusations of child exploitation. Even where the relationship is with other adults, there are reports about individuals not understanding sexual boundaries and have being accused themselves as perpetrators of abuse. As such, in this domain, individuals with this condition can be vulnerable adults but also vulnerable perpetrators. Safeguards need to be placed in terms of education and support to prevent harm.

Education, whilst important, needs to be provided with consideration. Simply providing sex education without support for the way the person may act on what has been taught can lead to issues. The use of social stories around the appropriateness and inappropriateness of situations can prevent risky behaviour, whilst simply presenting facts can lead to curiosity without understanding other people's boundaries. Where there is a lack of inhibitory control and poor empathy, this can lead to situations which are potentially dangerous. It is often seen that sex education class without the right level of background and support has led to individuals testing out their sexuality which has led to the above-described inappropriate situations. This needs to be understood, monitored and managed. To not do so risks harm.

26.3.2 Criminal Justice System Involvement

Both Tables 26.2 and 26.3 highlight that involvement with criminal justice system is very common in this population. Often the nature of the presentation is of more minor crimes, but this does not preclude more serious involvement. Some of the highest profile cases have, for example, been murder cases. Often these are linked to impulsive behaviours and not necessarily premeditated actions. Despite this when an act is perpetrated, whilst intent may not be there, that act clearly has been done. A lack of boundaries or consequence does not necessarily help the individual; however, whilst consequences may be necessary, disposal and sentencing may well be

Table 26.3 Denver Colorado, screening study for people with FASD in youth offending [2]

Number (percentage of group [% whole sample])	Seventeenth Judicial District Records Jan 2006–Aug 2010	Hennepin County Sep 2008–Sep 2010
Screened FASD	718	**148**
Positive for prenatal alcohol	183 (25)	60 (41)
Completed FASD diagnostic evaluation	79 (43 [11])	48 (82 [32])
Received FASD diagnosis	40 (50 [5.5])	46 (96 [31])
Received serviced based on FASD diagnostic evaluation	40 [100]	35 (76 [24])
Clients declining service	0	11 (24[7])

necessary to include supportive engagement and scaffolding of difficulties in order to prevent recidivism. Where this is not seen, there is often a repeat of behaviours.

Part of the difficulty with this presentation relates to the fact that in a criminal justice setting, the majority of individuals may not necessarily present with a clear diagnosis, and it is often a hidden disorder. Table 26.3 shows that when cases are screened for it is possible to identify significant populations of people with FASD. This is not something that happens often. Labels are attributed, often incorrectly, which means that rather than getting support and appropriate redirection to prevent reconviction, individual simply go through a cycle of imprisonment release and recidivist behaviour. It has been demonstrated that the costs associated with this are extensive. Where strategies are in place to identify, redirect and offer support the individuals, benefits can be seen.

It is important earlier in the life of the individual to try and identify the difficulties and the diagnostic profiling to ensure that if the individual and the court system can prevent a cycle of recidivist activity. Where appropriate help can be offered at a time where individuals often want it, can prevent subsequent re-incarceration.

26.4 Ongoing Clinical Risk Where Capacity Is in Doubt

Another area of difficulty for adults related to decision-making is about risk to self and others. Many individuals to not always act in a self-supporting manner, acting impulsively and putting themselves into difficult and dangerous situations. It is not unusual for individuals with FASD to have been involved in traumatic experiences with PTSD being a subsequent consequence. As highlighted in Table 26.1, psychiatric symptomatology is the most common secondary disability seen in this group. Anxiety and depression are the most common presentations; however, all forms of psychiatric illness can present [3, 4]. Many go on to develop their own addictions, perpetuating another generation of affected individuals, whilst others are known to self-injure and self-harm. In the worst-case scenario, the individual can act to self-harm physically or psychologically needing the individual to be admitted to hospital, often for mental health reasons or for rehabilitation and treatment. In certain extreme cases where this is repeated cycle, it may be necessary to consider legal approaches in order to support the individual and prevent harm. In the UK, for

example, mental health act allows guardianship of an individual to reside at a certain place and attend for treatment but not to take it. By residing in a certain area, however, allows engagement and support to be provided when it has been demonstrated that the individual struggles in this area. The challenge is demonstrating that the individual lacks capacity and as suggested above, is not simply acting in an unwise manner. Case law is not developed sufficiently, in the UK at least, to guide practitioners where appropriate intervention is required. Because individuals continue to develop and this is a potentially changing situation, re-evaluation is needed, but it is clear that the vulnerability of individuals may sometimes make this approach worth considering.

26.5 Conclusions

Identification of difficulties when young that predict how an individual will develop into later adulthood is still uncertain. It is clear that evidence would suggest that both physical and mental considerations continue to have impact in a lifelong manner on the individual. Where they are supported through childhood into adolescence to support the difficulties and build on their abilities, better outcomes seen. It is important that the challenges are not hidden but confronted and faced in order to prevent the complex situations becoming worse. To not do so has a significant burden on not just the individuals and their carers but also society through the financial and social challenges that they bring. Good transition planning, engagement with services through different stages individual's life can minimise the impact of this (Fig. 26.2).

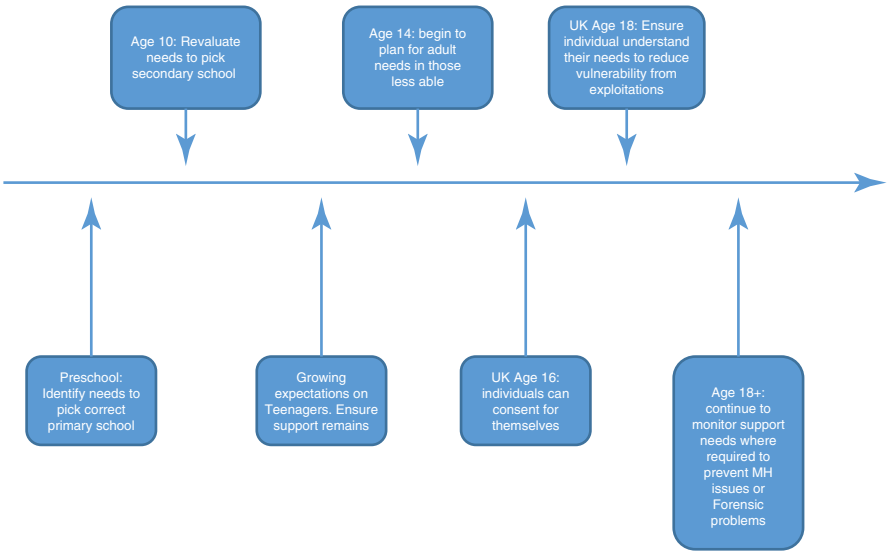


Fig. 26.2 Summary timeline for transition issues for individuals and their families from childhood to adulthood based on the clinical experience of the author

References

1. Streissguth AP, O'Malley K. Neuropsychiatric implications and long-term consequences of fetal alcohol spectrum disorders. Semin Clin Neuropsychiatry. 2000;5(3):177–90.
2. Bisgard EB, Fisher S, Adubato S, Louis M. Screening, diagnosis, and intervention with juvenile offenders. The Journal of Psychiatry & Law. 2010;38(4):475–506.
3. Famy C, Streissguth AP, Unis AS. Mental illness in adults with fetal alcohol syndrome or fetal alcohol effects. Am J Psychiatry. 1998;155:552–4.
4. Gray R, Mukherjee RAS. A psychiatrists guide to fetal alcohol spectrum disorders in mothers who drank heavily during pregnancy. Adv Mental Health Learn Disabil. 2007;1(3):19–26.

What Psychological Therapies Might Be Helpful?

27

Nicole M. Taylor

Chapter Highlights

- What psychological therapies have been shown to work in FASD.
- Different areas to focus interventions and approaches.
- Models to approach therapeutic interventions for people with FASD.

Fetal alcohol spectrum disorder (FASD) is a multifaceted disorder that has a significant impact on daily functioning and attainment of goals for those affected. Given the complexities of FASD, researchers and clinicians are tasked with identifying general principles of effective intervention and support, as well as understanding how therapies may be tailored to meet individual needs. Over the past 30 years, research has grown substantially in the field of FASD, but has lagged considerably behind for intervention studies. This chapter provides a glimpse of the psychological therapies that have been studied among FASD-affected populations, predominantly over the past 10 years and predominantly in early to middle childhood. The intervention studies that are discussed center around the following broad themes: attention, executive functioning, and self-regulation; specific cognitive or academic deficits; adaptive and social skills; and caregiver or family-based interventions. Findings suggest that the deficits in FASD are amenable to treatment, and that in some cases, treatment effects are generalizable to aspects of daily life and are maintained for periods up to 6 months. Best practice guidelines are discussed. Future research is needed to understand the effect of psychological interventions across broader age ranges (especially in adolescence and adulthood), with novel psychological therapies (e.g., mindfulness) that have not been previously studied, across systems, and over more extended follow-up periods.

N. M. Taylor (✉)
Department of Clinical Health Psychology, Max Rady College of Medicine, Rady Faculty of Health Sciences, University of Manitoba, Winnipeg, Canada
e-mail: ntaylor@hsc.mb.ca

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_27

27.1 What Psychological Therapies Might Be Helpful?

By definition, individuals affected by FASD experience a range of impairments that significantly impact daily functioning and goal attainment. Broadly, prenatal alcohol exposure (PAE) is shown to affect aspects of neuropsychological functioning (e.g., intelligence, memory, executive functioning, language, visual-perceptual skills), social and adaptive functioning, as well as self-regulation (i.e., attention, behavior, and emotion regulation). However, the effects are not the same across individuals and may also vary as a function of time, developmental stage, or environment. Furthermore, impairments may be compounding. That is, secondary impairments may arise when primary issues have not been sufficiently addressed.

A fundamental goal, of course, is to create supportive structures and psychological therapies that mitigate the effects of prenatal alcohol exposure, prevent the occurrence of secondary effects, and promote a good quality of life. While the salient aspects of a good quality of life may vary, it generally comprises factors such as economic stability (housing, food, adequate resources to meet daily needs), experience of personal safety, healthy relationships, meaningful engagement in purposeful activity, experiences of joy, and perhaps a sense of self-efficacy and self-worth.

Given the complex nature of FASD, researchers and clinicians are tasked with identifying general principles of effective intervention and support, as well as understanding how therapies may be tailored to meet individual needs.

27.2 Determinants of Positive and Negative Outcomes

One way to consider which therapeutic interventions may be suitable for the management of FASD is to examine the determinants of positive and negative outcomes. As noted above, brain-based impairments associated with FASD are bound to have a significant impact on a diverse set of outcome variables. For example, in a study of adults with FASD [1], adaptive functioning played a substantial role on the rate of secondary disabilities (i.e., alcohol and drug problems, mental health diagnosis, confinement, trouble with the law, sexually inappropriate behavior, and disruptive school experience), more so than overall intelligence (i.e., IQ < 70). Participants who fared poorly in this study, specifically with respect to diagnosed mental health problems, sexually inappropriate behavior and disruptive school experience, were also described as being very easily manipulated. While thorough neuropsychological assessments were not undertaken, it is likely that difficulties with social intelligence, executive functioning, and reasoning contributed to the preponderance of secondary difficulties among the group described as easily manipulated. Indeed, there is an association between executive functioning skills and theory of mind (a social-adaptive skill related to perceiving other's perspectives) in FASD [2]. Furthermore, executive functioning skills predict adaptive functioning among those with FASD [3]. As such, one approach to treatment planning is to develop strategies

that specifically target areas of impairment, by way of devising accommodative approaches or by honing skills related to those areas of impairment. In many cases, targets have included executive functioning skills, adaptive and social skills, and self-management skills. These will be discussed in greater detail in Sect. 27.2. Suffice it to say that targeting specific areas of neuropsychological impairment is a tenable approach to intervention planning for individuals with FASD.

However, treatment planning is not necessarily as straightforward. Clinicians and researchers in the field are well acquainted with the myriad of other risk factors that are commonly found in the lives of individuals with FASD. Unfortunately, adverse postnatal environments (e.g., exposure to trauma, poverty, lack of consistent and nurturing caregivers, lack of educational opportunities, etc.) are prominent among those exposed to PAE. To understand this better, Streissguth and colleagues [4, 5] used a prospective longitudinal approach to evaluate the impact of numerous protective and risk factors on the likelihood of adverse life outcomes and secondary disabilities. Adverse outcomes and secondary disabilities included alcohol and drug problems, trouble with the law, confinements (in mental health, alcohol/drug, or criminal facilities), inappropriate sexual behaviors, disruptions in schooling, mental health problems, employment problems, dependent living, and problems with parenting. Across studies, the authors found that stability and nurturance in the home environment, an early diagnosis, a diagnosis of FAS (rather than fetal alcohol effects, FAE), and, to a lesser degree, access to disability-based supports, served as critical protective factors. The finding that early diagnosis plays a protective role in FASD has been replicated by numerous studies (e.g., [6, 7]), and speaks to the potential that exists to alter developmental trajectories in order to produce more favorable outcomes for those affected by FASD. The benefit that early diagnosis confers on outcomes is likely associated with the opportunity it provides to access supports and interventions early on, and for caregivers to shift conceptual frameworks for understanding and managing their child's behavior and learning.

Of course, positive functional outcomes are most readily achieved in families that can provide good quality parenting and nurturing environments. The flipside is that adverse postnatal environments will have deleterious effects on functional outcomes and could seriously hamper any intervention efforts employed. Olson and colleagues [8] highlight the "double jeopardy" of FASD and psychosocial risk, and thus emphasized the need to understand the environmental context when planning interventions. Therapeutic interventions that promote family functioning and parenting, or reduce sources of family stress (e.g., economic instability, lack of housing, job instability, inadequate child care supports, etc.) may serve as critical components of an intervention. Further, these factors should be considered in treatment planning in order to limit some of the potential barriers to successful engagement with the intervention. For example, an intervention that is sensitive to the needs of some families would consider the provision of child care or bus fare to facilitate engagement with the intervention for those who need it.

Another aspect of the multidimensionality of FASD was described by Kodituwakku [9], who focused on the interplay between experience and neural development. Specifically, he outlined a model of development in FASD that is based on the theoretical framework of neuroconstructivism [10]. This theory is based on the premise that development, specifically neurodevelopment, is a complex and dynamic phenomenon that occurs in the context of interactions among multiple processes (e.g., genetically based or acquired brain injuries, learning experiences, interpersonal experiences) and neural development, vis-à-vis structural or functional changes within and between neural regions. Neural development, in turn, impacts on further experiences, which, in turn, affect neural development, and so on. In the treatment literature, evidence for the interplay between experience and neural development comes from a study of neural changes that arose in the context of metacognitive and self-regulation training [11]. Specifically, children with FASD who underwent a self-regulation intervention demonstrated changes in gray matter volumes in regions that are critical for self-regulation. Furthermore, associations were also observed between frontal regions showing intervention-related changes and improved emotional and inhibitory control.

Kodituwakku [9] argued that in order to maximize treatment efficacy in FASD, neuroconstructivistic guidelines based on knowledge gained through empirical studies in cognitive neuroscience and neuropsychology of FASD (Box 27.1) should be applied. It is clear that in order to devise and implement therapies that are effective, reliable, meaningful, and enduring, we must develop a deeper understanding of the various determinants of outcomes in FASD, including the interrelationships among them.

27.3 Current Treatment Approaches/Modalities

Over the past decade, research in interventions for FASD has grown considerably. Whereas only 3 and 12 studies met criteria for systematic review in 2007 and 2009, respectively [14, 15], 32 met inclusion criteria for systematic review in 2015 [16]. In the 2015 review, the authors incorporated study modalities that did not exist or were only underway as of 2009, such as randomized controlled trials (RCTs) and quasi-RCTs. A number of other descriptive reviews have also been undertaken over this timeframe [17–21]. In general, intervention studies have centered around the following broad themes:

Practice points:

- Neurodevelopment in FASD is a multifaceted and complex process that involves an interplay between experience, neural injury, and neural development.
- Attention, executive functioning, and self-regulation.
- Specific cognitive or academic deficits.
- Adaptive and social skills.

- Caregiver or family-based interventions.
- Pharmacological management. A discussion on the efficacy of pharmacological approaches in FASD is addressed in Management: Chap. 9.

27.3.1 Attention, Self-Regulation, and Metacognition

Given the high occurrence of difficulties with attention, self-regulation, and metacognition in an FASD-affected population [22], strategies that help modulate and improve these functions have been the focus of numerous studies. As noted in Sect. 27.1, Kodituwakku [9] posited that interventions geared toward improving attention and executive functioning skills should extend to other functions that were not expressly targeted. Thus, it is reasonable to argue that these functions ought to be a core element of any intervention program for FASD. Research interest in such interventions is certainly reflected in the literature. A larger proportion of intervention studies over the past 10–15 years either focus solely on the development of executive functions, such as self-regulation and metacognition, or include these functions as salient components of a broader intervention model.

One such intervention is the ALERT® program [23], which has shown promise among various FASD samples [11, 24–27]. See Table 27.1 for a description of this intervention. Across a number of different samples, school-aged children with FASD show improved parent-reported self-regulation and emotional regulation, as well as improved performance on a number of neuropsychological tests that tap into affect recognition, problem-solving, and inhibitory skills. Furthermore, as noted, increases in gray matter (relative to controls) have been observed following the 12-week ALERT program [11]. Follow-up measurements ranged between 1–2 weeks and 6-months post-intervention. As such, more research is needed to determine whether these improvements persist beyond these initial timeframes. Recently, a feasibility study has been conducted to identify elements of the ALERT program that are suitable for a population of indigenous children in remote communities in Australia [26]. The investigators, in conjunction with local community stakeholders, devised a modified version of the ALERT program that suited the needs of the population. In doing so, Wagner and colleagues also outlined a suitable process that other researchers and clinicians may follow for adapting intervention programs to suit the specific needs of their own communities.

The ALERT program is an example of cognitive control therapy (CCT), whereby children are explicitly taught metacognitive skills, such as self-regulation and self-monitoring. A number of other studies have been conducted under this premise, and show some promise [13, 28–30]. For example, in Pay Attention!, a manualized daily 12-session program of graded sustained attention training, improvements were made from pre to post training on measures of sustained, selective and alternating attention, as well as on a test of nonverbal reasoning [30] in a group of 20, 6–11-year-old children. No differences were observed between the treatment and control group on teacher ratings of attention postintervention; however, ratings did improve for both groups. Kerns and colleagues [13, 29] also evaluated the use of in-school

Table 27.1 Potential FASD interventions

Intervention	Description
ALERT® program	A manualized 12-week program that utilizes sensory integration and cognitive-behavioral strategies to help children monitor and regulate their behavior. The program uses the analogy of a car engine to explain self-regulation, explaining that both engines and brains can run in high-, low-, or just right gear. Children are taught to identify their engine speed(s), learn and apply strategies to modulate to the desired speed state, and to monitor sensory input in order to regulate their state of arousal
GoFAR	Promotes self-regulation and adaptive functioning in children via direct instruction, practice, and feedback with emotional and behavioral self-regulation. FAR is an acronym that stands for (1) focus and plan: The child learns to approach a problem thoughtfully and to devise a plan, rather than to respond impulsively, (2) act: The child carries out the plan, and (3) reflect: The child reflects on the plan and with guidance, considers what worked and what didn't work. The GoFAR intervention incorporates three elements: (1) a 5-week FAR computer game intervention for the child, (2) a concurrent 5-week parent-based intervention to promote parents' skills at supporting their child's behavioral regulation, and (3) a subsequent 5-week series of behavior analog therapy (BAT) during which the parent and child practice the FAR strategy in the context of real-life adaptive or behavioral problems in the home
Neurocognitive habilitation therapy	Teaches children how to recognize and accommodate their particular areas of deficit and capitalize on specific areas of strength. This program also incorporates strategies from the ALERT program to teach metacognitive and self-regulation strategies. Other elements that are addressed include memory strategies, planning skills, social and problem-solving skills, cause-and-effect reasoning skills, and self-esteem. Parent sessions run concurrently, covering some of the same topics and covering some content that is specific to the caregivers (e.g., understanding the effects of alcohol, or self-care for the caregiver)
Math interactive learning experience (MILE)	Provides individualized math instruction through interactive and experiential learning using tangible objects and tools, as well as slow-paced learning. In addition, MILE incorporates the strategies of the FAR intervention described above to provide the metacognitive tools to promote learning
Children's friendship training (CFT)	Involves 12 weeks of group-based social and friendship skills training for children, with concurrent group-based parent sessions that teach parents how to support the development of adaptive and social skills in their children
Parent-child interaction therapy (PCIT)	Aims to reduce behavioral problems among children and to lower stress among caregivers by providing parents with in-vivo practice and coaching of behavioral parenting skills. Additional goals of the intervention include promoting the parent-child relationship, as well as improving social skills and reducing misbehavior in the child

Table 27.1 (continued)

Intervention	Description
Families on track integrated preventive intervention program	Begins with a comprehensive neuropsychological and diagnostic assessment to identify the child's individual neuropsychological profile of strengths and weaknesses. This is followed by a 30-week intervention, with weekly child sessions and bi-monthly parent consultations sessions. PATHS components were designed to promote social competence in children with a view to mitigate violence, aggression, and behavioral problems
Strongest families™	Consists of 11 sessions that are designed to be completed weekly, and are complemented by a weekly telephone consultation with a trained coach. Sessions are geared to teaching caregivers skills they need to help their children with executive functioning (e.g., learning to make a plan with their child), and emotional and behavioral regulation (e.g., using calming strategies with their child). In addition, the model helps parents to foster improved relationships with their children (e.g., learning to notice good behavior), and teaches them strategies for navigating other systems (e.g., working with the school)
SEEDS-IT	A 20-session intervention that is designed to improve self-regulation in young children, and is comprised of an attachment-based parenting intervention, a music-based parent-child program, and a parent education and advocacy component

computer-based interventions for improving attention and executive functions. Both systems employed a hierarchical structure whereby task difficulty increases over the course of the program, and both interventions were supported by trained Educational Assistants (EA) who provided metacognitive strategies and support to the students. Outcomes were suggestive of improvements in target-related functions (i.e., working memory, distractibility, as well as sustained, divided, and selective attention) over the course of the intervention, as well as generalizability to other adaptive and academic functions that were not explicitly a target of intervention (i.e., reading fluency, math fluency).

Finally, the GoFAR® intervention was implemented and studied in a group of 30, 5–10-year-old children with FASD [28]. See Table 27.1 for a description of this intervention. In this pilot study, the GoFAR intervention produced reductions in disruptive behaviors among children with FASD, with reductions in disruptive behavior occurring as early as the culmination of the first 5 weeks of intervention.

27.3.2 Interventions to Target Specific Cognitive and Academic Deficits

As already mentioned, individuals with FASD demonstrate deficits across a wide variety of neuropsychological functions. Given that no single neuropsychological profile is pathognomonic to FASD, a thorough assessment is critically informative. It is important to know and understand, for the *specific individual*, what particular

patterns of strengths and deficits they face. This knowledge enables the clinician to recommend strategies that will accommodate for or target deficits, as well as to capitalize on areas of strength or intact functioning in order to maximize growth and learning. Additionally, a number of researchers have undertaken to target specific deficits that are common in FASD more formally in structured intervention programs. For example, interventions targeting deficient mathematics, language, and literacy skills have been devised [31–34]. In addition, Wells and colleagues [27] outlined a group-based neurocognitive habilitation therapy model that is intended to target individual areas of deficit. See Table 27.1 for a description of this intervention. The researchers detected improvements in executive and emotional functioning that were maintained 2–3 months after the conclusion of the intervention.

Given a high occurrence of math and executive functioning difficulties among those with FASD [35–37], Kable and colleagues [32–34] devised a math intervention program called Math Interactive Learning Experience (MILE) that focuses not only on math skills but also on fostering learning behaviors and supporting underlying issues affecting family functioning. See Table 27.1 for a description of this intervention. Findings from initial studies demonstrate increased performance on math standardized testing for the treatment group up to 6 months postintervention. The most recent study sought out to determine the feasibility and applicability of the treatment paradigm when provided by community instructors in school or tutoring settings. Findings from 60, 6–7-year-old children provided support for the viability of the MILE program in a community-based setting, with improvements in math skills and parent satisfaction observed among the intervention group. Further, satisfaction ratings regarding the training process were high among the community instructors who were provided with the training. This extension is important as strategies that increase accessibility for children with FASD are needed, with school-based interventions providing a suitable option.

Literacy and language skills have also proven amenable to intervention in FASD [31], although the improvements appear domain-specific and do not generalize to general scholastic performance. The intervention comprised 19 h each of alternating language therapy and literacy/phonological awareness training. The study was carried out over nine school-term months among 60 third-grade children across 10 schools in South Africa. Three groups were studied: (1) 20 children with FASD in the literacy and language training, (2) 20 children with FASD as FASD controls, and (3) 20 children without FASD as typically developing controls. Children in the treatment arm showed gains in complex phonological processing and early literacy (syllable manipulation, letter sound knowledge, written letters, word reading, and nonword reading and spelling), as compared to the nonintervention FASD control group. These findings suggest that, while targeted interventions on literacy and language development are valuable, a more comprehensive approach is needed to transfer gains toward more general academic learning and skills. As described above and emphasized in Kodituwakku [9], the incorporation of metacognitive training might prove useful to facilitate transfer effects.

27.3.3 Adaptive Behavior and Social Skills

Adaptive behavior and social skills represent another set of functions that are often compromised in FASD, with poorer parent and teacher ratings of social skills, impaired social cognition, and weak adaptive skills described among those with FASD [38–40]. To address social deficits in FASD, O'Connor and colleagues [41–43] adapted an evidence-based manualized social skills intervention termed Children's Friendship Training (CFT) [44] for use with children that were prenatally exposed to alcohol. See Table 27.1 for a description of this intervention. Across a series of studies, children with FASD who underwent the treatment protocol showed a number of positive outcomes, including parent-reported gains in their use and knowledge of social skills, as well as reductions in problem behaviors, improved self-concept, and decreased hostile attributions in vignettes depicting peer-entry situations (i.e., a child trying to enter a social activity). Treatment effects were observed across university-based [42, 43] and community [44] settings and were maintained in a 3-month postintervention follow-up [41, 42]. Teacher ratings of social behavior did not improve as a consequence of the intervention, and no changes were observed in hostile attributions regarding peer provocation scenarios (i.e., a child experiencing a negative outcome in a social situation in which the peer's intention is ambiguous). Thus, further work is required to promote the transfer of adaptive and social skills to classroom settings and in situations that have the potential for more negative emotional valence.

27.3.4 Family-Focused Interventions

As mentioned earlier, families and parents provide a suitable avenue for intervention in FASD. Children spend considerable amounts of time with caregivers, placing caregivers in a prime position to foster growth and learning. By intervening at the level of the family, a clinician may increase the chance that treatment gains are maintained and generalized to new situations. Additionally, children with FASD have high rates of behavior problems, and caregivers of children with FASD report high levels of parenting stress [8, 45]. Conversely, families or caregivers that are empowered and feel efficacious in their parenting have lower levels of parenting stress, and parent more effectively [8, 46, 47]. Given that stable, good-quality parenting is one of the strongest determinants of childhood outcomes in FASD [4, 5], it is not surprising that numerous studies have taken a family-based approach to intervention.

Research suggests that caregiver expectations and attributions surrounding behavior play a significant role in the parenting behaviors that follow (e.g., [48, 49]). Parents who attribute their child's behavior to willful disobedience (e.g., being stubborn, trying to get attention, lacking motivation, etc.) are either more likely to respond with negative affect and to utilize punitive parenting strategies, or less likely to use antecedent strategies (e.g., redirection, modifying the environment or their expectations) to avoid the occurrence of behavioral problems. On the other

hand, parents who have greater knowledge about FASD are more likely to view misbehavior as a consequence of skills deficits associated with neurodevelopmental impairments. They are also more likely to use strategies to prevent misbehavior (e.g., planning around the child's sensory needs) or reward positive behavior (e.g., sticker rewards, praise, etc.), to feel more confident about their parenting, and to have more success managing their child's behavior. Thus, strategies aimed at providing a neurodevelopmental framework to contextualize misbehavior in FASD are warranted. For some parents, this knowledge may be sufficient to foster a change in parenting practices, and in other cases, it may be important to challenge parental attributions more directly. One important lesson from these findings is that while parents adjusted their perspectives and adopted a less blaming stance surrounding their children's misbehavior, this did not dissuade them from implementing behavioral approaches with their children. In fact, those who used behavioral strategies reported success with the use of such strategies. This suggests that, while behavioral approaches must take the neurodevelopmental context into consideration, they can be successfully used to promote desirable behaviors and mitigate misbehavior among children with FASD.

The bulk of the interventions discussed in this section seek to reduce disruptive behavior in children via caregiver-based interventions. For example, a group-based adaptation of Parent-child Interaction Therapy (PCIT) was trialed with 57, 3–7-year-old children and their caregivers [24]. See Table 27.1 for a description of this intervention. Families in the treatment group were offered 14 weeks of PCIT intervention (12 joint parent-child sessions and 2 parent only sessions), and the active control group was offered 14 weeks of group-based, parent-only intervention that included psychoeducation about FASD as well as information and discussion about the implementation of behavioral systems. Attrition was high in both groups (54% overall) and no group differences were observed across measures of parenting stress or child behavior problems. However, improvements across all intervention targets were observed across both treatment modalities, suggesting a general positive effect of intervention. Another study evaluated the Families Moving Forward (FMF) program among 52, 5–11-year-old children and their parents [24]. The FMF program was conducted over a period of 9–11 months, and consisted of bi-weekly 90-min sessions addressing parental attributions and parenting behaviors to foster reductions in disruptive behaviors. Children assigned to the FMF group (compared to a community standard of care group) demonstrated a reduction in behavior problems immediately posttreatment, and the parents showed an increased sense of self-efficacy as well as greater engagement in self-care behaviors.

In other research, Petrenko and colleagues [50] married the FMF program with the Promoting Alternative THinking Strategies (PATHS) curriculum [51] to develop and pilot a program, called The Families on Track Integrated Preventive Intervention Program. See Table 27.1 for a description of this intervention. Thirty, 4–8-year-old children and their families underwent the pilot study. Those in the treatment arm showed gains in caregiver variables such as knowledge, attitudes, targeted parenting practices, parenting efficacy, support, and engagement in self-care. Further, children in the treatment group showed improvements in self-regulation and self-esteem.

These small-scale but promising findings provide support for further randomized trials, which reportedly are underway in this research group.

Finally, the usability of another caregiver-based intervention, the Strongest Families™ intervention [52], originally designed for children with disruptive behavior, has been piloted online with caregivers ($n = 8$) and health care professionals (i.e., social worker, occupational therapist, nurse, government consultant, program manager; $n = 10$) [53]. See Table 27.1 for a description of this intervention. The goals of the study were to identify whether the website design and content were effective from the perspectives of learnability, efficiency, and acceptability. Caregivers and health care professionals alike found the website to be user-friendly and appealing. A number of usability problems were identified and are being addressed in preparation for a Randomized Control Trial [54] comparing the Strongest Families FASD program to a static resource webpage. The findings have not yet been published.

As noted above, Olson and colleagues [8] argue for an integration of a developmental systems perspective with a family systems approach to intervention. They also argue for a lifespan perspective, noting that developmental needs and challenges change over the course of development, and may become particularly evident at certain time points (e.g., during the transition to middle school, or to young adulthood). The next section highlights special considerations regarding a number of developmental stages.

27.3.5 Special Considerations: Infants, Preschoolers, and Adults

The majority of intervention research in FASD focuses on early to middle childhood years [16–21] highlighting the lack of intervention studies at other stages of development. A few studies have investigated the efficacy of early interventions in infants and toddlers with FASD, with a focus primarily on parenting and the parent-child relationship. Of course, early interventions such as these offer the opportunity to alter developmental trajectories early and may also provide caregivers with anticipatory guidance to help keep their expectations and parenting on course as the child grows. One example is the Breaking The Cycle intervention [55], which offers a range of integrated services for mothers with a history of substance abuse to promote parent-infant relationships and provide necessary supports as needed. Compared to a community-based standard treatment, mothers that accessed the program showed decreased maternal substance use, improved mental health, and increased relational capacity. In addition, infants showed improved outcomes. The Strategies for Enhancing Early Developmental Success Program for Infants and Toddlers (SEEDS-IT) [56] is another preventative intervention for young children, but one that targets those in foster, adoptive, or kinship care. See Table 27.1 for a description of this intervention. The content of the parent education and advocacy components was developed following focus-group discussions with 19 clinicians in the field of FASD who work with families. The clinicians' perspectives were sought out regarding the challenges and needs of caregivers of children with FASD. The

themes that emerged surrounded the difficulties inherent in seeking and receiving a diagnosis, in coming to terms with the child's difficulties, in seeking knowledgeable support and being part of a community, in reframing the child's behaviors, and in becoming an educator, advocate, and expert for their child with FASD. Intervention findings have not been published.

Recognizing the need to promote functioning among young children with FASD, a neurodevelopmentally informed intervention was recently piloted with 10, 10–53-month-old adopted children with FASD who also had a history of maltreatment [57]. The premise of this intervention is based on the idea discussed earlier that neurodevelopment interacts with experience, and that the first few years of life represent a critical and sensitive period for learning and brain plasticity. Thus, the interventions were designed to strengthen developmental functions and repair deficits through a tailored combination of regulatory, somatosensory, relational, and cognitive enrichments. Treatment gains were observed in child developmental functioning, as well as parent caregiving skills and (lowered) caregiving stress after 6 months of intervention, although no control group was used for comparison and the small sample size hampers the generalizability of these findings. The authors suggest that future larger-scale studies on this intervention are warranted.

Even fewer studies are found in the FASD intervention literature addressing adolescent or adult populations. This is greatly concerning as these are developmental stages marked by dramatically increased risk for the development of secondary conditions [4]. Adolescents undergo vast changes as they mature into young adulthood. Stressors abound related to social pressures, increased demands for abstraction and for self-management of adaptive and executive functions, and increased expectations for independence and self-care. In addition, recent neuroimaging findings suggest that adolescents and young adults with FASD may experience altered neurodevelopmental trajectories over these developmental timeframes that further impact neurocognitive skills [58, 59].

Only one intervention that specifically targets adolescents with FASD is noted in the literature [60], with a specific goal of reducing substance use and abuse among youth with FASD. The intervention, termed Project Step Up, was conducted with 54 adolescents and their parents over 6 weeks of 60–min group sessions using a harm reduction approach. Adolescents were provided with psychoeducation about alcohol and taught how to respond to social pressures regarding alcohol consumption. In a concurrent but separate group session, caregivers were provided with psychoeducation about alcohol's effect on the brain and were taught parenting strategies to help their teen manage their alcohol use better. The program was successful in reducing alcohol use as well as the negative consequences of alcohol use among the participants. This was especially true with high-risk youth that reported greater alcohol use and alcohol-related problems prior to the start of the intervention. Treatment gains at the conclusion of the intervention were maintained at 3-month follow-up as compared to a control condition in which youth were provided with written materials on alcohol misuse and stress reduction.

Two adult-based intervention studies targeting parents with FASD are described [61, 62]. The first examines the impact of a 3-year home visitation program, the Parent-Child Assistance Program (PCAP), modified for women with FASD [61]. The program offers an integrated service that helps mothers parent their children, connect with appropriate community services and supports, and manage their own and their families' basic needs (e.g., housing, food, safety, stability, etc.). 19 women with a mean age of 22 years were involved in a 12-month community pilot of this program. Treatment outcomes at the completion of the 12 months included decreased alcohol and drug use, increased housing, increased use of contraceptives, and improved uptake of medical and mental health care services. While the women with FASD were not independently managing their own access to services at the completion of the program, they were making use of community services with the support of their mentor.

The step-by-step program [62] is based in part on the PCAP program in so far as it also utilizes a 3-year mentoring system to provide practical and parenting support to women with FASD who are parenting. Mentors work with families from a strength-based perspective to identify needs (e.g., housing, financial issues, mental health issues, addictions, etc.), to develop goals (e.g., parenting, management of personal skills, self-care, health), and to connect women to community supports and services that will help them address their needs and goals. At the end of the program, participants showed a significant reduction of needs (e.g., decreased experience of abuse, decreased social problems, decreased housing problems) and an overall increase in progress toward or attainment of goals.

27.4 Best Practice Guidelines and Future Directions

Despite slow beginnings, research evidence is mounting suggesting that effective interventions can indeed be designed and implemented to promote positive life outcomes among those with FASD. The sometimes-held misconception that behavior and learning in children with FASD is not amenable to intervention is simply untrue. Flexible interventions that are devised within a developmental psychopathology framework are provided early but take a lifespan approach, are attuned to the potential barriers to treatment, and are apt to be the most effective. In addition, treatments that incorporate elements of metacognitive and self-regulation training are likely to increase the likelihood that skills will transfer to daily life as well as to other functions that are not expressly targeted in the intervention.

Practice point:
Therapeutic interventions for FASD may target attention, executive functioning, and self-regulation; specific cognitive or academic deficits; adaptive and social skills; and/or caregivers and families. Interventions that include some form of metacognitive training and involve the family unit may lead to more enduring and generalizable treatment effects.

In recent years, interest has grown in gathering qualitative information from stakeholders that enables a deeper understanding of the needs of individuals with FASD and their families [63–65]. For example, Petrenko and colleagues [63] conducted a qualitative analysis of parents and service providers' opinions regarding the ideal aspects of intervention programming for those with FASD. The resulting program characteristics centered around five themes:

1. *Availability across the lifespan*: FASD is a life-long condition that requires the continuation of appropriate services across the lifespan.
2. *Focus on prevention*: A proactive or preventative approach that enables supports to be in place before an individual struggles is likely to lead to more positive outcomes for the individual and their family. Anticipatory guidance helps lay the groundwork for what difficulties may lie ahead and thus, how to reduce the likelihood of latent problems.
3. *Individualized programming to meet individual needs*: Given that FASD is a multifaceted disorder, and that an individual's strengths and challenges will intersect with their experiences, a one-size-fits-all approach is unlikely to suit the needs of this population. Suitable programming will need to be tailored to the individual's needs in consideration of their strengths, weaknesses, and family or living circumstances.
4. *Comprehensive intervention*: Stakeholders emphasized a need for comprehensive interventions that account for the complex needs and diverse experiences of people with FASD. Comprehensive services include those that involve the individual, family members, and multiple systems of care, such as other providers, teachers, and policy makers.
5. *Coordinated care across systems and developmental stages*: Services must not function as discrete entities but should be coordinated across systems and developmental stages.

These themes were echoed from stakeholders in a multi-stakeholder symposium on integrating care for individuals and families affected by FASD [65]. Together with the recommendations made by Kodituwakku [9], clinicians and researchers have strong conceptual frameworks to devise, implement, and evaluate good evidence-based programs. Petrenko [64] argues that the themes are also consistent with the positive behavioral support (PBS) framework [66, 67], which uses educational methods and environmental adaptation to enhance an individual's quality of life first and foremost, and to minimize problem behavior.

In summary, research is clearly showing that children with FASD can and do benefit from intervention, and that in some cases, treatment effects are generalizable to aspects of daily life and are maintained for periods up to 6 months postintervention. Future research is needed to understand the effect of psychological interventions across broader age ranges (especially in adolescence and adulthood), with novel psychological therapies (e.g., mindfulness) that have not been previously studied, across broad systems, and over more extended follow-up periods.

Box 27.1: Neuroconstructivistic Model for Treatment Planning

1. A child's overall cognitive or developmental level must be taken into account. As such, interventions should be planned, not around the child's chronological age, but around their developmental age
2. Scaling of treatment difficulty should take Vygotsky's zone of proximal development (ZPD) [12] into account. The ZPD represents the difference between what a child can do on their own and what they can do with support or scaffolding. As such, the difficulty level for a given child should occur within the ZPD and titrate higher as the child master's skills
3. Self-regulation and attention represent critical skills that not only are often compromised among those with FASD, but also, when improved can generalize to other areas of functioning. Thus, an emphasis on interventions that promote attention and self-regulation ought to be considered. Further, Kodituwakku asserts that self-regulation treatments should be provided as early as possible, citing evidence that long-term developmental outcomes are predicted by self-regulatory skills in the preschool years
4. Enriched environments and experiences foster growth and development. These experiences, however, should be provided in a controlled and guided manner. Indeed, research does provide support for adaptive training protocols (whereby the level of difficulty adjusts according to the child's performance) rather than nonadaptive approaches (e.g., the level of difficulty does not account for the individual's performance) for cognitive skill development [13]
5. Evidence-based pharmacotherapy and behavioral interventions should be combined where necessary

References

1. Clark E, Lutke J, Minnes P, Ouellette-Kuntz H. Secondary disabilities among adults with fetal alcohol spectrum disorder in British Columbia. J FAS Int. 2004;2:e13.
2. Rasmussen C, Wyper K, Talwar V. The relation between theory of mind and executive functions in children with fetal alcohol spectrum disorders. Can J Clin Pharmacol. 2009;16:e370–80.
3. Ware AL, Crocker N, O'Brien JW, Deweese BN, Roesch SC, Coles CD, et al. Executive function predicts adaptive behavior in children with histories of heavy prenatal alcohol exposure and attention-deficit/hyperactivity disorder. Alcohol Clin Exp Res. 2012;36:1431–41. https://doi.org/10.1111/j.1530-0277.2011.01718.x.
4. Streissguth AP, Barr HM, Kogan J, Bookstein FL. Understanding the occurrence of secondary disabilities in clients with fetal alcohol syndrome (FAS) and fetal alcohol effects (FAE). In: Final Report to the Centers for Disease Control and Prevention (CDC). Seattle: University of Washington, Fetal Alcohol & Drug Unit, Tech. Rep. No. 96–06; 1996.
5. Streissguth AP, Bookstein FL, Barr HM, Sampson PD, O'Malley K, Young JK. Risk factors for adverse life outcomes in fetal alcohol syndrome and fetal alcohol effects. J Dev Behav Pediatr. 2004;5:228–38.
6. Currie BA, Hoy J, Legge L, Temple VK, Tahir M. Adults with fetal alcohol Spectrum disorder: factors associated with positive outcomes and contact with the criminal justice system. J Popul Ther Clin Pharmacol. 2016;23:e37–52.
7. Alex K, Feldmann R. Children and adolescents with fetal alcohol syndrome (FAS): better social and emotional integration after early diagnosis. Klin Padiatr. 2012;224:66–71. https://doi.org/10.1055/s-0031-1299682.
8. Olson HC, Oti R, Gelo J, Beck S. "Family matters:" fetal alcohol spectrum disorders and the family. Dev Disabil Res Rev. 2009;15:235–49. https://doi.org/10.1002/ddrr.65.

9. Kodituwakku PW. A neurodevelopmental framework for the development of interventions for children with fetal alcohol spectrum disorders. Alcohol. 2010;44:717–28. https://doi.org/10.1016/j.alcohol.2009.10.009.
10. Sirois S, Spratling M, Thomas MS, Westermann G, Mareschal D, Johnson MH. Precis of neuroconstructivism: how the brain constructs cognition. Behav Brain Sci. 2008;31:321–31. Discussion 331–356. https://doi.org/10.1017/S0140525X0800407X.
11. Soh DW, Skocic J, Nash K, Stevens S, Turner GR, Rovet J. Self-regulation therapy increases frontal gray matter in children with fetal alcohol spectrum disorder: evaluation by voxel-based morphometry. Front Hum Neurosci. 2015;9:108. https://doi.org/10.3389/fnhum.2015.00108.
12. Vygotsky LS. Mind in society: the development of higher psychological processes. Cambridge, MA: Harvard University Press; 1978.
13. Kerns KA, Macoun S, MacSween J, Pei J, Hutchison M. Attention and working memory training: a feasibility study in children with neurodevelopmental disorders. Appl Neuropsychol Child. 2017;6:120–37. https://doi.org/10.1080/21622965.2015.1109513.
14. Premji S, Benzies K, Serrett K, Hayden KA. Research-based interventions for children and youth with a fetal alcohol spectrum disorder: revealing the gap. Child Care Health Dev. 2007;33:389–97. https://doi.org/10.1111/j.1365-2214.2006.00692.x.
15. Peadon E, Rhys-Jones B, Bower C, Elliott EJ. Systematic review of interventions for children with fetal alcohol spectrum disorders. BMC Pediatr. 2009;9:35. https://doi.org/10.1186/1471-2431-9-35.
16. Reid N, Dawe S, Shelton D, Harnett P, Warner J, Armstrong E, et al. Systematic review of fetal alcohol spectrum disorder interventions across the life span. Alcohol Clin Exp Res. 2015;39:2283–95. https://doi.org/10.1111/acer.12903.
17. Coriale G, Fiorentino D, Di Lauro F, Marchitelli R, Scalese B, Fiore M, Maviglia M, Ceccanti M. Fetal alcohol spectrum disorder (FASD): neurobehavioral profile, indications for diagnosis and treatment. Riv Psichiatr. 2013;48:359–69. https://doi.org/10.1708/1356.15062.
18. Koren G, Zelner I, Nash K, Koren G. Foetal alcohol spectrum disorder: identifying the neurobehavioural phenotype and effective interventions. Curr Opin Psychiatry. 2014;27:98–104. https://doi.org/10.1097/YCO.0000000000000038.
19. Paley B, O'Connor MJ. Behavioral interventions for children and adolescents with fetal alcohol spectrum disorders. Alcohol Res Health. 2011;34:64–75.
20. Petrenko CL, Alto ME. Interventions in fetal alcohol spectrum disorders: an international perspective. Eur J Med Genet. 2017;60:79–91. https://doi.org/10.1016/j.ejmg.2016.10.005.
21. Wilhoit LF, Scott DA, Simecka BA. Fetal alcohol spectrum disorders: characteristics, complications, and treatment. Commun Ment Health J. 2017. https://doi.org/10.1007/s10597-017-0104-0.
22. Khoury JE, Milligan K, Girard TA. Executive functioning in children and adolescents prenatally exposed to alcohol: a meta-analytic review. Neuropsychol Rev. 2015;25:149–70. https://doi.org/10.1007/s11065-015-9289-6.
23. Williams MS, Shellenberger S. How does the engine run? A leader's guide to the alert program. Albuquerque, New Mexico: Therapyworks; 1996.
24. Bertrand J. Interventions for children with fetal alcohol spectrum disorders (FASDs): overview of findings for five innovative research projects. Res Dev Disabil. 2009;30:986–1006.
25. Nash K, Stevens S, Greenbaum R, Weiner J, Koren G, Rovet J. Improving executive functioning in children with fetal alcohol spectrum disorders. Child Neuropsychol. 2015;21:191–209.
26. Wagner B, Fitzpatrick J, Symons M, Jirikowic T, Cross D, Latimer J. The development of a culturally appropriate school based intervention for Australian aboriginal children living in remote communities: a formative evaluation of the alert program® intervention. Aust Occup Ther J. 2016;64(3):243–52. https://doi.org/10.1111/1440-1630.12352.
27. Wells AM, Chasnoff IJ, Schmidt CA, Telford E, Schwartz LD. Neurocognitive habilitation therapy for children with fetal alcohol spectrum disorders: an adaptation of the alert program. Am J Occup Ther. 2012;66:24–34. https://doi.org/10.5014/ajot.111.002691.
28. Coles CD, Kable JA, Taddeo E, Strickland DC. A metacognitive strategy for reducing disruptive behavior in children with fetal alcohol spectrum disorders: GoFAR pilot. Alcohol Clin Exp Res. 2015;39:2224–33. https://doi.org/10.1111/acer.12885.

29. Kerns KA, MacSween J, Vander Wekken S, Gruppuso V. Investigating the efficacy of an attention training programme in children with foetal alcohol spectrum disorder. Dev Neurorehabil. 2010;13:413–22. https://doi.org/10.3109/17518423.2010.511421.
30. Vernescu RM. Sustained attention training in children with fetal alcohol spectrum disorder (Doctoral dissertation). Memorial University of Newfoundland. 2009; Retrieved from Proquest Dissertations & Theses International (NR55383).
31. Adnams CM, Sorour P, Kalberg WO, Kodituwakku P, Perold MD, Kotze A, et al. Language and literacy outcomes from a pilot intervention study for children with FASD in South Africa. Alcohol. 2007;41:403–14. https://doi.org/10.1016/j.alcohol.2007.07.005.
32. Coles CD, Kable JA, Taddeo E. Math performance and behavior problems in children affected by prenatal alcohol exposure: intervention and follow-up. J Dev Behav Pediatr. 2009;30:7–15. https://doi.org/10.1097/DBP.0b013e3181966780.
33. Kable JA, Coles CD, Taddeo E. Socio-cognitive habilitation using the math interactive learning experience program for alcohol-affected children. Alcohol Clin Exp Res. 2007;31:1425–34.
34. Kable JA, Taddeo E, Strickland D, Coles CD. Community translation of the math interactive learning experience program for children with FASD. Res Dev Disabil. 2015;39:1–11. https://doi.org/10.1016/j.ridd.2014.12.031.
35. Crocker N, Riley EP, Mattson SN. Visual-spatial abilities relate to mathematics achievement in children with heavy prenatal alcohol exposure. Neuropsychology. 2015;29:108–16. https://doi.org/10.1037/neu0000094.
36. Enns LN, Taylor NM. Factors predictive of a fetal alcohol spectrum disorder: neuropsychological assessment. Child Neuropsychol. 2016;10:1–23. https://doi.org/10.1080/09297049.2016.1251894.
37. Nash K, Stevens S, Rovet J, Fantus E, Nulman I, Sorbara D, et al. Towards identifying a characteristic neuropsychological profile for fetal alcohol spectrum disorders: 1. Analysis of the motherisk FASD clinic. J Popul Ther Clin Pharmacol. 2013;20:e44–52.
38. Lindinger NM, Malcolm-Smith S, Dodge NC, Molteno CD, Thomas KGF, Meintjes EM, et al. Theory of mind in children with fetal alcohol Spectrum disorders. Alcohol Clin Exp Res. 2016;40:367–76. https://doi.org/10.1111/acer.12961.
39. Olson HC, Feldman AP, Streissguth AP, Sampson PD, Bookstein FL. Neuropsychological deficits in adolescents with fetal alcohol syndrome: clinical findings. Alcohol Clin Exp Res. 1998;22:1998–2012.
40. Roebuck TM, Mattson SN, Riley EP. Behavioral and psychosocial profiles of alcohol exposed children. Alcohol Clin Exp Res. 1999;23:1070–6.
41. Keil V, Paley B, Frankel F, O'Connor MJ. Impact of a social skills intervention on the hostile attributions of children with prenatal alcohol exposure. Alcohol Clin Exp Res. 2010;34:231–41. https://doi.org/10.1111/j.1530-0277.2009.01086.x.
42. O'Connor MJ, Frankel F, Paley B, Schonfeld AM, Carpenter E, Laugeson EA, Marquardt R. A controlled social skills training for children with fetal alcohol spectrum disorders. J Consult Clin Psychol. 2006;74:639–48.
43. O'Connor MJ, Laugeson EA, Mogil C, Lowe E, Welch-Torres K, Keil V, et al. Translation of an evidence-based social skills intervention for children with prenatal alcohol exposure in a community mental health setting. Alcohol Clin Exp Res. 2012;36:141–52. https://doi.org/10.1111/j.1530-0277.2011.01591.x.
44. Frankel F, Myatt R. Children's friendship training. New York: Brunner-Routledge; 2003.
45. Paley B, O'Connor MJ, Frankel F, Marquardt R. Predictors of stress in parents of children with fetal alcohol spectrum disorders. J Dev Behav Pediatr. 2006;27:396–404.
46. Bloomfield L, Kendall S. Parenting self-efficacy, parenting stress and child behaviour before and after a parenting programme. Prim Health Care Res Dev. 2012;13:364–72.
47. Ludmer JA, Sanches M, Propp L, Andrade BF. Child Psychiatry Hum Dev. 2017. https://doi.org/10.1007/s10578-017-0732-1.
48. Miller SA. Parents' attributions of their children's behavior. Child Dev. 1995;66:1557–84.
49. Petrenko CL, Pandolfino ME, Roddenbery R. The association between parental attributions of misbehavior and parenting practices in caregivers raising children with prenatal alcohol exposure: a mixed-methods study. Res Dev Disabil. 2016;59:255–67. https://doi.org/10.1016/j.ridd.2016.09.005.

50. Petrenko CLM, Pandolfino ME, Robinson LK. Findings from the families on track intervention pilot trial for children with fetal alcohol spectrum disorders and their families. Alcohol Clin Exp Res. 2017. https://doi.org/10.1111/acer.13408.
51. Domitrovich CE, Greenberg MT, Cortes R, Kusche CA. The preschool PATHS curriculum. South Deerfield, MA: Channing-Bete; 2005.
52. McGrath PJ, Lingley-Pottie P, Thurston C, MacLean C, Cunningham C, Waschbusch DA, et al. Telephone-based mental health interventions for child disruptive behavior or anxiety disorders: randomized trials and overall analysis. J Am Acad Child Adolesc Psychiatry. 2011;50:1162–72.
53. Hundert AS, Huguet A, Green CR, Hewitt AJ, Mushquash CJ, Muhajarine N, et al. Usability testing of guided internet-based parent training for challenging behavior in children with fetal alcohol spectrum disorder (strongest families FASD). J Popul Ther Clin Pharmacol. 2016;23:e60–76.
54. Turner K, Reynolds JN, McGrath P, Lingley-Pottie P, Huguet A, Hewitt A, et al. Guided internet-based parent training for challenging behavior in children with fetal alcohol spectrum disorder (strongest families FASD): study protocol for a randomized controlled trial. JMIR Res Protocols. 2015;4:e112. https://doi.org/10.2196/resprot.4723.
55. Koren G. Breaking the cycle (BTC)-20 years of breaking records in managing addicted mothers and their young children. J Popul Ther Clin Pharmacol. 2013;21:e505–7.
56. Baskin J, Delja JR, Mogil C, Gorospe CM, Paley B. Fetal alcohol spectrum disorders and challenges faced by caregivers: clinicians' perspectives. J Popul Ther Clin Pharmacol. 2016;23:e114–30.
57. Zarnegar Z, Hambrick EP, Perry BD, Azen SP, Peterson C. Clinical improvements in adopted children with fetal alcohol spectrum disorders through neurodevelopmentally informed clinical intervention: a pilot study. Clin Child Psychol Psychiatry. 2016;21:551–67.
58. Lebel C, Mattson SN, Riley EP, Jones KL, Adnams CM, May PA, et al. A longitudinal study of the long-term consequences of drinking during pregnancy: heavy in utero alcohol exposure disrupts the normal processes of brain development. J Neurosci. 2012;32:15243–51. https://doi.org/10.1523/JNEUROSCI.1161-12.2012.
59. Treit S, Lebel C, Baugh L, Rasmussen C, Andrew G, Beaulieu C. Longitudinal MRI reveals altered trajectory of brain development during childhood and adolescence in fetal alcohol spectrum disorders. J Neurosci. 2013;33:98–109. https://doi.org/10.1523/JNEUROSCI.5004-12.2013.
60. O'Connor MJ, Quattlebaum J, Castañeda M, Dipple KM. Alcohol intervention for adolescents with fetal alcohol spectrum disorders: project step up, a treatment development study. Alcohol Clin Exp Res. 2016;40:1744–51. https://doi.org/10.1111/acer.13111.
61. Grant T, Huggins J, Connor P, Pedersen J, Whitney N, Streissguth A. A pilot community intervention for young women with fetal alcohol spectrum disorders. Community Ment Health J. 2004;40:499–511.
62. Denys K, Rasmussen C, Henneveld D. The effectiveness of a community-based intervention for parents with FASD. Community Ment Health J. 2011;47:209. https://doi.org/10.1007/s10597-009-9273-9.
63. Petrenko CLM, Tahir N, Mahoney EC, Chin NP. A qualitative assessment of program characteristics for preventing secondary conditions in individuals with fetal alcohol spectrum disorders. J Popul Ther Clin Pharmacol. 2014;21:e246–59.
64. Petrenko CLM. Positive behavioral interventions and family support for fetal alcohol spectrum disorders. Curr Dev Disord Rep. 2015;2:199–209. https://doi.org/10.1007/s40474-015-0052-8.
65. Masotti P, Longstaffe S, Gammon H, Isbister J, Maxwell B, Hanlon-Dearman A. Integrating care for individuals with FASD: results from a multi-stakeholder symposium. BMC Health Serv Res. 2015;15:457. https://doi.org/10.1186/s12913-015-1113-8.
66. Koegal LK, Koegal RL, Dunlap G. Positive behavioral support: including people with difficult behavior in the community. Baltimore: P H Brookes; 1996.
67. Carr EG, Dunlap G, Horner RH, Koegel RL, Turnbull AP, Sailor W, et al. Positive behavior support: evolution of an applied science. J Posit Behav Interv. 2002;4:4–20.

Approaches to the Medical Management of FASD

28

Raja A. S. Mukherjee

Chapter Highlights

- Approaches to assessing behaviour in a wider context.
- Principles to medical management.
- Examples of medication usage in FASD.

28.1 Introduction

Fetal alcohol spectrum disorder represents a group of condition with a wide range of presentations and comorbidities [1]. Individuals with this condition, having been exposed to differing levels of prenatal alcohol, with individual vulnerabilities, show a range of outcomes. There is commonly a profile of strengths and weaknesses in each individual. By identifying the areas of vulnerability and situations, which cause a challenge to the individual, allows appropriate support and scaffolding by people around them. By building on their strengths, the individual can help develop resilience. Unfortunately, often the case is that individual needs are missed, leading to challenging and disturbed behaviour. Often, situations are described as unprovoked or occurring in an unpredictable manner, leading to families and individuals experiencing difficulties [2].

One of the reasons why it can be complex to assess and identify individual situations as an antecedent to subsequent behaviour is that the presentation can be multifactorial. For example, an individual who has FASD may well have difficulties with cognition and emotional regulation, as described in Chap. 14, alongside sensory processing difficulties or problems with communication and language as described in Chaps. 24 and 15, respectively. Therefore, to the individual externally

R. A. S. Mukherjee (✉)
National FASD Specialist Behaviour Clinic, Surrey, UK
e-mail: Raja.Mukherjee@sabp.nhs.uk

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_28

observing the behaviour, it cannot be seen that there is often one identifiable cause leading to this presentation. It can be a combination of a different mix of these factors which interplay to lead to a difficult situation.

What also can be confusing for families, or people working with affected individuals, is that there are times where similar situations appear to have less impact than others [2]. This inconsistency then becomes in itself a puzzle. Attribution to mood or other psychiatric phenomena can be made, and whilst these vulnerabilities exist, alternative explanations may also be true. It is only by understanding how the individual exists within their environment, how that impacts on them and therefore then trying to maximise the benefits of their ability to function in that same environment becomes crucial and key. Ultimately, by understanding the different characteristics of an individual's presentation, the factors that both increase and decrease their ability to manage themselves in that environment, impacts on their state of regulation. These situations all come together to impact on managing the individual. Therefore, any medical management should be integrated as part of this overall holistic approach, rather than seen in isolation to simply sedate or manage without wider understanding and consideration.

28.2 Assessing Arousal and Regulation

Affect and mood regulation, and its counterpoint dysregulation, are terms that are used to describe an individual and how well they are managing themselves in a given situation. Unfortunately, the more stigmatised label of dysregulation is more commonly used when challenges occur. This diminishes the fact that for much of the time, the individual can regulate themselves if situations around them are optimal. For most individuals, most of their life is spent in a regulated state, not one that is dysregulated. When dysregulation occurs, this is where multiple factors can often combine to lead to a deterioration in their ability to function. As highlighted in the chapter on cognitive testing, Chap. 14, there is evidence that emotion can impact on an individual's ability to use their higher-level executive function, therefore impacting on their decision-making, impulsivity and wider ability to manage themselves. It can be described that there are stages to increasing dysregulation from normal functioning to severe problems as suggested in Fig. 28.1. Often the case is that the stages of worsening function that lead to a more acute dysregulated situation are missed or the transition between stages occur quickly and intervention is left too late. There is often a presumption that individuals, their families and those who live and work around them have the skill sets to both identify and know how to manage individuals. This is not necessarily true.

Professionals can help individuals and families in order to support a framework by which these different situations are both documented, reported and strategies put in place to support management. Figure 28.1 highlights one such approach used in the Surrey and Borders NHS Trust UK National Specialist Behaviour Clinic. It represents a method by which individuals can identify how they function at different levels of regulation and dysregulation related to their arousal, then how strategies

	Descriptions of behaviours commonly seen in this stage	Intervention the person can do themselves to help reduce level	Intervention the supporters / Carers can do to help reduce level
1 Not Agitated (The aim for regular function)			
2 Mild Agitation (Waring level to try and intervene here to prevent escalation)			
3 Moderate Agitation (Behaviours still possible to prevent escalation but some rescue strategies will be needed)			
4 Severe Agitation (Too late for prevention and rescue strategies are needed)			

Fig. 28.1 An approach to identify and manage stages of arousal and regulation used in the UK National FASD Clinic

can be put in place to manage these. It implies and highlights that the majority of an individual's life should be spent between levels one and two. These are states which recognise that no individual, whether FASD or not, functions in a continuously optimal state. It is however the case for the majority of individuals at any one given time that a level of function is maintained between the highlighted stages one and two.

It is when there is a transition to a higher level of dysregulation and stress that behaviour begins to become more challenging to those around them. By stage 3, intervention may well have benefits but by stage 4 often the individual is no longer fully in control of their own behaviours and their executive function and self-management skills are disordered. By this stage, external intervention including recue medication may be needed to recover to lower levels of arousal. At lower levels of this model, the emphasis on self-regulation is far greater than external regulation; however, the counterpoint is also true in that the higher stages of the model require external individuals to take more control. An example, as suggested from clinical practice, would include at higher levels of this model, first allowing the individual to first calm and only then when in a lower level of arousal discuss behaviours and consequences. This may be by using ways that deescalate the situation first, including the possible use of medication. Instead, often the situation can be exacerbated by the families and carers around the individual themselves becoming agitated and then only further escalating situation. This approach is often where the family needs help to learn a different way to manage these situations [3].

When considering the ability of an individual to self-regulate, understanding the factors that influence this becomes important. Every individual, whether FASD or not, exists in some form or state of arousal. The environment, emotional distress, their own internal resilience factors, and traumatic experiences, to name just a few, can influence any one given presentation. It is therefore important to individualise

the care and presentation towards supporting and managing that person. As highlighted above, these can be divided into different stages.

One method used in the national FASD UK national service in the UK with families is to consider arousal almost as a ladder, as an example shown in Fig. 28.2. As the stress levels increase, the ability to self-regulate decreases. This can be applied to almost any individual with any background or presentation. It would be true to say that by contextualising this in some form of normal behaviour, but one where those with poorer levels of resilience, often influenced by external factors, may increase and decrease their arousal more rapidly. It is however a concept that is understandable to many individuals. These are often described either as thermometers, or ladders, as a pictorial way of representing this to children and families. This can be used alongside the matrix in Fig. 28.1.

As an individual gets older and societal challenges increase on the person, often these behaviours can appear to be more akin to a developing mental health problem. Whilst protracted mental health issues, especially anxiety and depression, have been identified and are in fact quite common in people with FASD, more severe mental illnesses can also be seen [4, 5]. In most cases, however, when stress levels are not managed, acute and transient situations, including psychotic -like events as exampled in Fig. 28.3 can occur. This figure is a diagrammatic representation of how fluctuating arousal, modified and influenced by external events, can lead to people appearing to present in stressful situations and with mental health disturbance but the episodic nature of these, in those with FASD but also to some extent with wider developmental disorders, is not necessarily akin to a classic psychiatric presentation. For example, bipolar affective disorder, the grandiosity or the flight of ideas is just one example are often symptoms that are more commonly missing, therefore leading to questions about the diagnosis. Instead, it is the high arousal and the heightened dysregulation that leads to agitated behaviour. This is a far greater

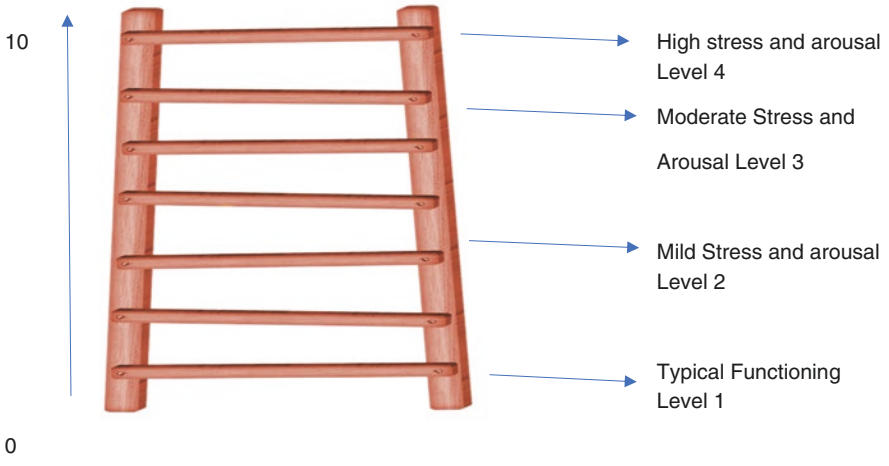


Fig. 28.2 Example of how to use a ladder of arousal

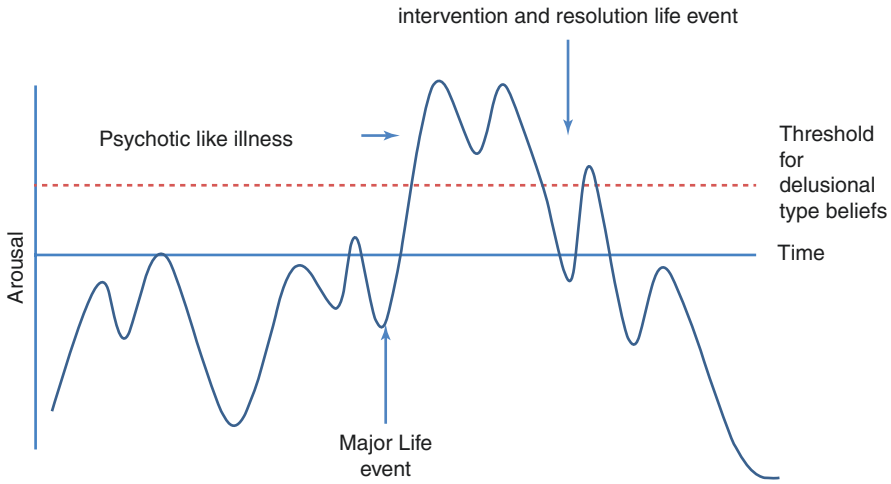


Fig. 28.3 An example of how changing arousal can lead some to develop acute and transient MH issues

presenting feature in the situations and scenarios. Unfortunately, when just agitation, the attribution of cause can be challenging. It is not that mental illness cannot also occur, and in fact is more common, however this wider issue must also be borne in mind.

28.3 Treating FASD Vs. Comorbid Presentations. Wider NDD Context

Whilst the first stage of any management strategy is to identify triggers and use environmental approaches to support management, in some scenarios, the individual's ability to function in their environment, mean they exist in a higher level of arousal much of the time. Whilst this can be in fact due to an associated psychiatric comorbid presentation, it may also be the anxiety and arousal that primarily needs managing. An individual should aim to spend the majority of their life in level 1 or 2, therefore would not need regular intervention. Should that happen, these cases can be managed solely with reactive strategies and occasional intervention with medications when higher arousal stages are reached. An example from wider typical life would be akin to occasionally taking a painkiller to help relieve a headache. For most people, headaches do not occur regularly and only occasional interventions are needed. These can help soothe and resolve what would otherwise be an uncomfortable situation. For others however, where the pain is severe and more constant, they find themselves having to take regular pain relief in order to function and manage. This would be a similar scenario here however instead of pain, it is the dysregulation and the wider arousal that is being managed. These are through approaches to reduce stress and anxiety more often than not. This should always be undertaken as

part of the wider review as described above. Further, it should always be initiated by practitioners familiar with benefits and side effects of any medication used in keeping with their practice, and national rules.

As described in Chap. 12, it is common for wider neurodevelopmental presentations to be seen in individuals with FASD. For example, ADHD is a common finding and outcome [6, 7]. In these scenarios, understanding the comorbidity of having FASD changes the management strategy. This was highlighted by Young et al. in the 2016 consensus review [8]. This highlighted approaches whereby typical strategies for individuals with ADHD are modified based on the comorbidities and the outcomes that are seen as a result of the relationship. Because FASD commonly affects not just neurological pathways linked to ADHD but wider brain pathways, comorbidities must be borne in mind as they alter the response to treatment. Also due to the understanding of the wider physical health issues that commonly are linked to FASD [1], it is worth taking more precautions in terms of baseline assessment and checks in these individuals compared to those with no clearly linked physical comorbidity.

28.4 Capacity to Consent

In the UK, generally below the age of 16, children cannot give their consent unless it is judged that they have the appropriate capacity and understanding to understand the concepts involved. Otherwise, the person with parental responsibility will be required to provide consent. Between the ages of 16 and 18, children are assumed to have the capacity to consent to treatment unless there is evidence to the contrary, in which case, the person with parental responsibility will be required to provide consent. For adults over 18, capacity is presumed but here again as described in Chap. 14 the ability to manipulate information may not be present. If that is the case no one else can consent for them and professionals should seek opinions as far as possible from others involved in the care of the person and then act in the best interest of the individual.

28.5 Specific Approaches for Medication Use

Whilst there is no specific treatment for FASD in itself, by managing these wider presentations and comorbidities, it can lead to beneficial effects for the individual and their families. Table 28.1 summarises some examples of common management approaches that can be used for different scenarios and presentations. Whilst this is not an exhaustive list, it can be used as a guide and should be used in conjunction with rules and regulations set out in each country. The treatments for conditions such as depression and psychosis do not change. It is only here that understanding the wider sensitivities and vulnerabilities can treatments however be modified. This is often in that there may be greater sensitivity to side effects. Because this population may differ in their sensitivities to medication, compared to populations where

Table 28.1 Examples of some medications used in conditions comorbid with FASD (Note any use of medication should as far be in keeping with expertise and guidance as exists in individual countries)

Areas being treated	Considerations before starting/ monitoring	Types of medication used
Preventative strategies		
ADHD [6]	• checks of height, weight, BP and where cardiac issues ECG. In some cases with wider comorbidities wider blood tests including kidney function • Side effects of agitation and behavioural disturbance more common in this group • Where wider comorbidities there should be alternate mediation used as described in Young et al.	• Dexamphetamine or its derivates often found to be more effective and may be considered first line • Methylphenidate or its derivatives can still be effective but if ineffective consider alternative soon • Third-line medications and comorbidities lead to modification of usage, e.g. mood to use Atomoxetine or ASD features and anxiety Guanfacine XL
Anxiety	• Baseline checks of height, weight and if using medication such as atypical antipsychotics metabolic blood tests and prolactin • Caution when the use of some medication where there is a noted increased risk of impulsivity or suicidality	• SSRI: Use for management of anxiety in keeping with licence • Antipsychotics: Medication at very low dose, e.g. Risperidone 0.5 mg–2 mg or Quetiapine 25-50 mg per day (high doses are not warranted) • Propranolol: Symptomatic treatment of arousal. Always begin lower doses and titrate slowly
Reactive strategies		
Short term	• Should never be used regularly and many medications have addiction potential when overused • Close monitoring of their use and regularity in keeping with the above behaviour management strategy to define when to use and never used in isolation • If there are wider impacts leading to excessive sedation should not be used • Baseline checks not usually needed due to occasional use but as above if become more regular then should be considered in the light of the need for preventative strategies and monitoring as set out there	• Lorazepam 0.5–2 mg per day in divided doses and depend on the age and size of the individual • Atypical antipsychotics such Quetiapine 25 mg

they have been initially trialled, and as very few actual randomised controlled trials undertaken in these groups specifically, treatment should be for the associated presentation as far as possible but with some cautions. For example, treating ADHD with ADHD medication within their licence. However, due to expected sensitivities,

titration should start at a lower dose and always increase slowly to balance benefits and risks. In many cases, lower doses can be effective and minimise the impact of any potential side effects.

Medication can never be the full answer and should only be used in the context of both preventative and reactive strategies in keeping with the above modelling and presentation and therefore in keeping with a broader management strategy to support the individuals' overall functioning and maintain a quality of life.

References

1. Popova S, Lange S, Shield K, Mihic A, Chudley AE, Mukherjee RAS, Bekmuradov D, Rehm J. Comorbidity of fetal alcohol spectrum disorders: a systematic review and meta-analysis. Lancet. 2016;387:978–87.
2. Mukherjee RAS, Wray E, Commers M, Hollins S, Curfs L. The impact of raising a child with FASD upon carers: findings from a mixed methodology study in the UK. J Adopt Foster. 2013;37(1):43–56.
3. Mohammed Z, Carlisle ACS, Livesey AC, Mukherjee RAS. Carers stress in FASD: implications from a national FASD service. Adopt Foster. 2020;44(3):242–54.
4. Famy C, Streissguth AP, Unis AS. Fetal alcohol exposure and adult psychiatric disorders: Dr Famy and colleagues reply. Am J Psychiatry. 1999;156(7):1128–9.
5. O'Connor MJ, Shah B, Whaley SE, Cronin P, Gunderson B, Graham J. Psychiatric illness in a clinical sample of children with prenatal alcohol exposure. Am J Drug Alcohol Abuse. 2002;28(4):743–54.
6. Mukherjee RAS. The relationship between ADHD and FASD. ADHD Pract. 2016;8:4–7.
7. Mukherjee RAS, Layton M, Yacoub E, Turk JT. Autism and autistic traits in people exposed to heavy prenatal alcohol: data from a clinical series of 21 individuals and a nested case control study. Adv Mental Health Intellect Disabil. 2011;5:43–9.
8. Young S, Absoud M, Blackburn C, Branney P, Colley B, Farrag E, et al. Guidelines for identification and treatment of individuals with attention deficit/hyperactivity disorder and associated fetal alcohol spectrum disorders based upon expert consensus. BMC Psychiatry. 2016;16(1):324.

Supporting a Child with FASD in the Classroom

29

Carolyn Blackburn

Chapter Highlights

- Strategies to manage FASD in the classroom.
- Principles of supporting children and families with FASD.
- Case examples of how to approach classroom management at different ages.

29.1 Introduction

FASD has also been described as 'The Hidden Disability', in part, because there may be no physical characteristics to show that an individual has FASD [1]. Schools may be the first environment where children with complex learning difficulties and disabilities (CLDD) are identified. CLDD describes children with coexisting conditions that can include but are not limited to attention deficit hyperactivity disorder (ADHD), autism spectrum disorder (ASD) and FASD. Evidence indicates that the population of children with CLDD has been increasing. It is suggested that meeting this need requires 'informed and reflective practitioners who are equipped with a range of observation and intervention tools to support their learning and development' [2]. In 1995, the Winnipeg School Division (WSD) established one of the first classrooms for students with FASD in Canada. This was in response to provincial surveillance data that identified 118 alcohol exposed infants born between 1993 and 1995 and the anticipation of increased identification and enrolment of children with FASD in schools. Based on early experiences in meeting, the complex needs of these children within a regular classroom, the decision was made to develop a specialised programme that recognised and addressed the unique needs of children with FASD. What occurred at that time has been referred to as a *paradigm shift* in

C. Blackburn (✉)
Birmingham City University, Birmingham, England
e-mail: Carolyn.Blackburn@bcu.ac.uk

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_29

the understanding of the needs of individuals with FASD [3]. This has been further described by Malbin who said:

> *If FASD includes changes in the structure and function of the brain, then it follows that it is by definition a brain-based physical disability. In most cases, however, it is invisible, and behaviours are typically the only symptoms. Understanding FASD as a primary physical disability with behavioural symptoms redefines problems and solutions in a manner consistent with research'* [4].

This can be supported by involving families and caregivers who provide valuable information on the curriculum and how the child is coping [5]. Assessment should be undertaken at regular intervals by interdisciplinary teams that include physical therapists, occupational therapists, speech language pathologists, psychologists, special education and regular teachers and other support staff [6, 7].

It is also recommended that educators implement strategies to address cognitive, communication, social, emotional and physical developmental delays and preparation for employment among older students [8]. Examples include strategies to improve/support: communication, literacy, abstract thinking, concept and sense of time, use of money, memory, organisation skills, understanding and following rules, sensory processing (e.g., sound, smell, movement, light, coping with overstimulation), supporting relationships and role models, understanding inappropriate behaviours, understanding danger and impulse control and adapting the physical environment to work with FASD and individual differences.

Teaching children with FASD should start with recognising that a student with FASD may not function at the same level as peers at the same age and then appropriately modifying the approach and environment to support the student's needs [9]. The objective is to identify how to teach and adapt the school environment to meet the child's strengths and interests. This requires recognising that these children process information and learn differently. However, it has been stressed that this is not a 'school only' issue or responsibility [1]. The best approach to addressing FASD in schools requires a community-wide (systems level) response (i.e., that includes: education, healthcare professionals, social services, youth justice, family and community organisations). It also helps to create a school division-wide FASD support, teacher/consultant position to support development and implementation. Initial teacher training programmes need specific and up to date training on FASD and FASD education practices. Teachers are key to understanding behaviours and learning. It is important to recognise that the behaviours are brain based. Consequently, this warrants a different approach than those typically used in regular classrooms.

29.2 Teaching Approaches

The approaches are first based upon recognising that children affected with diagnosed FASD process information differently and react to the environment differently. Following this, an assessment of the child's strengths from an interdisciplinary team should provide information to personalise learning and plan

for the child's interests. This includes adapting the teaching style, curriculum and physical environment based upon that information to develop individual plans for each child. Educators need to recognise that children may be strong in some areas such as visual hands-on learning and also more challenged through language-based learning. In addition, because of their brain differences teachers recognise that sometimes children will be unable to do some tasks that they have successfully done many times before. Educators should also make use of technology to support children with FASD. Examples include using: headphones, smart boards, calming spaces, handheld listening helpers and other learning aids.

29.3 Classroom Management Strategies

The following classroom management strategies are recommended [10]:

- Keep teaching and instruction simple and specific.
- Employ adaptive teaching techniques which focus upon the child's interests, strengths and developmental stage.
- Use consistent and predictable language to support understanding.
- Use visual cues and aids to accompany verbal instructions. (for example, visual timetables).
- Provide concrete learning resources and opportunities to support learning.
- Use pre-learning, rehearsing and practising of desirable skills and behaviours.
- Plan opportunities to repeat lessons/instructions/messages.
- Note strategies used by the student and build on these and/or teach more effective alternatives.
- Give instructions and tasks in small steps to support understanding and executive function and provide opportunities for oral or alternative methods of testing progress and competence.
- Give warning and instructions well ahead of transitions, and give support.
- Provide a structured learning environment (for example, classroom spaces, time, work, etc.)
- Have consistent and predictable routines (for example, providing visual schedules of activities).
- Make cause and effect explicit (for example, outcomes of decisions).
- Identify triggers for increased hyperactivity and inattention, such as noise and smells and reduce them.
- Reward effort rather than achievement and allow extra time for tasks and tests.
- Make use of fidget balls or stress balls to increase attention and reduce hyperactivity.
- Provide consistent language and behaviour management strategies between home and school.
- Say the student's name at the beginning of an instruction or sentence. Make sure you have the student's attention before you speak to them.

- Make sure you are facing the student so that they can see your facial expressions and gestures.
- Use simple concrete language and use consistent language across the curriculum and throughout the school. Share language for educational concepts with parents/carers.
- Think about the language used in tests/exams and whether it matches what the student is familiar with.
- Give only one instruction at a time.
- Keep instructions short; use the minimum number of words.
- Say exactly what you want the student to do (e.g., instead of saying 'Tidy up', say 'Put the scissors in the blue box'), and reinforce with pictures if necessary.
- If you are interrupted whilst giving an instruction, go back to the beginning of your sentence.
- Ensure that the student has understood by asking them to repeat an instruction back to you in their own words.
- Give the student time to think about what you have asked of them.
- Use positive communication; instead of saying 'Don't run', say 'Walk'.
- Use exaggerated facial expressions and gestures to give the student clues as to your meaning.
- Reinforce auditory input with visual aids and provide students with a visual timetable.
- Plan multisensory experiences based around the students' sensory strengths and needs including activities involving movement.
- Break tasks into small steps and be realistic about expectations.
- Use visual prompts and concrete objects such as puppets for story telling for young students and number lines for mathematics.
- Show rather than tell; demonstrate concepts so that students know exactly what is expected.
- Provide opportunities for discussion of new concepts before they introduced in the classroom and check understanding afterwards.
- Provide opportunities for new learning to be connected to existing knowledge.
- Communicate with parents/carers regularly by email/phone/home-school diary.
- Provide worksheets which have plenty of white space and do not mix mathematical concepts and operations.
- Plan around the student's strengths and interests and provide immediate, frequent praise for each achievement.
- Be flexible about how achievement is recorded, consider video, photographic evidence and provide a scriber where necessary for technical lessons such as science where the student may be overwhelmed by sensory stimulation.
- Remove as many distractions from the environment as possible to enable the student to concentrate on the teacher/task.

Busy, noisy classroom environments may be overstimulating for children with FASD. Busy walls, pictures or interactive whiteboards with writing on them, open windows, noise from the hallways, flickering from fluorescent lights and even

articles on a desk may distract and subsequently cause anxiety. In general, classroom environments need to be restructured to decrease visual, auditory and physical stimulation that can distract or overload students with FASD whenever possible (Examples include using natural light versus fluorescent light, using room dividers, covering windows and walls with pictures or signs not being used with curtains, using head phones), so the student only hears the teacher, modified desks and chairs that reduce stimulation/distractions or a special calming space/place in the room (for example, tent or enclosed space) for a sensory overload break. Other examples that sound counter intuitive include letting children chew gum or use a 'squishy ball or fidget toy' to help them relax. Children with FASD need more frequent movement breaks and might need to be able to move, wiggle, fidget, chew gum, use toys or do other physical movements in order to listen.

To support children's socio-emotional and relational skills, some specific approaches have also been recommended as discussed below.

29.3.1 Emotion Coaching

Another emerging approach to supporting children's socio-emotional development and self-regulation is Emotion Coaching. Emotion coaching is a relational and skills-based approach to supporting children's emotional competency and self-regulation and is more likely to result in decreased frustration and increased emotional well-being for children. This approach recognises that socially competent children who are able to understand and regulate their emotions are better equipped to go on to achieve higher academic success than those who lack impulse control or have poor social skills [11–13].

Emotion coaching is based on the work of Gottman and Katz and colleagues and is essentially comprised of two key elements—empathy and guidance [14]. These two elements express themselves through various processes which adults undertake whenever 'emotional moments' occur. Emotional empathy involves recognising, labelling and validating a child's emotions, regardless of the behaviour, in order to promote self-awareness and understanding of emotions. Such acceptance by the adult of the child's internal emotional state creates a context of responsiveness and security, and helps the child to engage with more reasonable solutions. The circumstances might also require setting limits on appropriate behaviour (such as stating clearly what is acceptable behaviour) and possible consequential action (such as implementing behaviour management procedures)—but key to this process is guidance: engagement with the child in problem-solving in order to support the child's ability to learn to self-regulate—the child and adult work together to seek alternative courses of action to help manage emotions and prevent future transgressions. This process is adaptable and responsive to the developmental capabilities of the child, with the adult scaffolding prosocial solutions and differentiating where necessary. By enabling children to tune in more explicitly to their emotions and problem-solve solutions that will help them to manage such feelings, and the behavioural consequences of those feelings, the child is the child is engaged in proactively

enhancing social and emotional competences. It also supports the child's development of 'meta-emotion', which refers to the organised set of feelings and cognitions about one's own emotions and the emotions of others [15]. Thus, emotion coaching helps to instil the tools that will aid children's ability to self-regulate their emotions and behaviour [16].

This approach is supported by evidence that shows how thinking and reasoning and emotional processing are fundamentally integrated in the brain at multiple levels [17, 18]. More information is available from http://www.emotioncoaching.co.uk/ and http://www.emotioncoachinguk.com/.

29.3.2 Social Stories

Social stories are used to help teach social skills to people with social communication difficulties. They involve short descriptions of a particular situation, event or activity, which include specific information about what to expect in that situation and why.

They are simple visual representations of the different levels of communication in a conversation. For example, they could show:

- The things that are actually said in a conversation.
- How people might be feeling.
- What people's intentions might be.

Comic strip conversations use symbols, stick figure drawings and colour. By seeing the different elements of a conversation presented visually, some of the more abstract aspects of social communication (such as recognising the feelings of others) are made more 'concrete' and are therefore easier to understand. Stories such as this, when designed with the child and rehearsed often, can help children with FASD understand complex social relationships.

29.4 Working with Families

Parents and carers are a child's first and most enduring educators and their role in a student's educational journey should be valued. For children with FASD, the family structure may consist of Foster or Adoptive parents as well as, or instead of biological parents and sensitivity about possible attachment difficulties is required. If the child is living with biological parents, sensitivity and understanding about how parents may be feeling about their child's disability is paramount.

Children with FASD will often present with a different set of needs in school than at home and parents/carers may have many concerns about how their child will manage through the school day. These concerns should always be taken seriously as it is important that parents/carers feel that they have been listened to and their concerns addressed. It is important that children with FASD receive consistency of approach and language in both home and school setting as this will help

them to make sense of the world and simplify the number of things they need to remember.

Working closely with families is essential and the following guide has been produced to facilitate a discussion with families about having a clear plan in place that is shared between educators and families (Table 29.1):

Table 29.1 General guidelines to supporting children and families with FASD

Approach	• Adopt a holistic approach which builds on the student's sensory strengths • Provide sequential sensory experiences (visual, kinaesthetic, or auditory) and ensure a range of sensory opportunities throughout the lesson so that the student receives a multisensory experience • Demonstrate rather than describing new techniques and be prepared to repeat demonstrations, instructions, rules and concepts often • Prepare students for new concepts by providing them with any new vocabulary beforehand to practise and learn • Provide opportunities for small group and 1:1 work where possible and construct a personalised learning plan based on the student's strengths and interests (usually in the areas of practical and artistic ability)
Communication with families	• Ensure effective communication with parents and carers to reduce anxiety and develop an ethos of partnership • Consistent language and approaches used and at home school will provide security and predictability for students who are easily overwhelmed by change and disruption
Environment	• Ensure that the environment is free from distractions as far as possible. This includes distraction from noise, smell, tactile and visual distractions • Constant supervision may be necessary to keep students who are developmentally younger safe from harm
Routine and structure	• Ensure routine is communicated to the student to reduce anxiety and enable them to organise themselves as independently as possible • Changes to routine should be communicated to the student soon as possible and the student supported through them • Structure will help the student with FASD make sense of their environment. Provide frequent breaks throughout a lesson to give the student time to refocus. It may help if they can do something physical for a few minutes between activities
Simplicity	• Ensure that instructions, directions and tasks are broken down into short achievable, easily understood steps and are delivered at a level which is developmentally appropriate to the student • Be realistic about expectations
Understanding	• Ensure that the student has understood instructions and directions. Say their name before giving instructions and directions • Ask them to repeat what you have said them back to you in their own words • Ensure that language used is simple, positive, concrete and free from jargon, sarcasm, or idioms • Provide visual aids if necessary, as students may not always respond to auditory input alone • Be specific when giving directions and provide step by step instruction

Source: Blackburn, C. (2010) Facing the challenge and shaping the future for primary and secondary aged students with Fetal Alcohol Spectrum Disorders (FAS-eD Project). Primary Framework—teaching and learning strategies to support primary aged students with FASD. London: NOFAS-UK [20]

29.5 Transitions

Transition from Primary to Secondary education can be particularly difficult for children with FASD and needs to be carefully managed to ensure that communication is efficient and services to families do not become disrupted. A full assessment of the child's needs should be undertaken at this time. For teenagers, issues around emotions, friendships and sexual behaviour, independence and achievement can compound their difficulties. A lack of understanding of the students, particular learning needs can lead to unrealistic expectations. Without sensitive support and communication between primary and secondary teachers and families, children may experience behavioural, cognitive and psychological secondary disabilities, for example, depression, self-harm, loneliness and low self-esteem, leading to disrupted schooling and trouble with the law. In addition, this is a particularly worrying time for families and they will need additional support from schools and supporting services to ensure a smooth transition [19].

29.6 Case Studies [8, 20]

29.6.1 Case Study 1: Primary Education

Background

Child 1 attends a mainstream nursery and primary school where she is placed in a class of 30 children and accesses the National Curriculum. She is supported by an Inclusion Assistant for all activities, some of which take place in the classroom alongside her peers, whilst others are delivered on a 1:1 basis according to the subject and topic. All work for this student is personalised according to her needs, including the delivery of curriculum and recording of progress.

The Child as a Learner: Strengths and Challenges

Child 1 listens well in the classroom and enjoys school. She wants to do well and be included and is willing to learn.

Her enthusiasm is coupled with a visual impairment, small stature, immaturity and inattention as well as learning difficulties. This has implications for where she is able to sit in the classroom in order to see the teacher and whiteboard, how her peers treat her in equality terms, her ability to stay on task and retain information and the length of time she is able to focus for.

For example, there is a temptation for peers to 'baby' her as it is not always easy for her to follow the rules of playground games or play them on an equal basis. Her lack of focus and attention imply that tasks must necessarily be broken down into small steps and highly personalised to her individual needs in order to engage her in learning. Her visual impairment means that she must sit near the front of the classroom so that she is able to see the teacher and whiteboard and also has implications for the use of computers and visual equipment in ICT.

The Child as a Learner: Opportunities for Inclusion

Including this child in a mainstream, secondary setting and providing access to the national curriculum have been possible through a combination of adult support and scaffolding, personalised teaching and learning and partnership with parents. For example:

- Abstract concepts such as money are taught using a range of concrete examples such as oversized laminated pictures of coins, plastic coins and games. The equipment is then sent home so that the student can practice with parents. This is then reinforced with a trip to the shop with peers to use real money and facilitate the transfer of knowledge from the classroom situation to a practical application and embed the practice of buying items in real life with peer and adult support.
- Life skills such as cooking, hygiene, peer relations, emotions, safety and life cycle issues are taught through attendance at a weekly life skills class with peers.
- Recording of achievement is appropriate to the situation and the child. For example, photographs of the child taken at various stages of the learning process demonstrate progress, without the need for her to undertake lengthy writing activities, which tire and frustrate her, leading to a sense of failure and low self-esteem.
- Pictures and symbols are displayed below the whiteboard so the student and other children with additional needs have a visible timetable of the day/lesson. The teacher/inclusion support assistant will talk through the timetable so that the student knows what's happening now and next.
- A buddy system is provided at break and lunch times to ensure that the student has peer companionship and support throughout the day.
- A home-link diary is used to keep parents informed and parents are able to use this to ensure that school is aware of issues at home that may impact on learning or emotional well-being throughout the school day.

These measures ensure that this child is able to access the national curriculum in a mainstream educational setting alongside her peers.

29.6.2 Case Study 2: Secondary Education [8, 20]

Background

Child 2 attends an Autistic Spectrum Condition Unit attached to a mainstream secondary school. Most lessons and learning take place supported in the mainstream, with individual programmes in the Unit where necessary. She also has access to music therapy and occupational therapy through her place in the unit.

Child 2 is supported in all lessons by at least one TA, although for some practical lessons, it is necessary to provide 2:1 adult support.

The Child as a Learner: Strengths and Challenges

Child 2 is a keen learner, when she is confident, she will be able to complete tasks. She enjoys school, in particular, she likes public speaking, drama, French, history,

literacy, dance and gymnastics. She takes pride in her artistic talent and her ability to write stories. She is noted for her general knowledge and politeness.

This enthusiasm is coupled with extreme impulsivity, hyperactivity and a propensity to become overstimulated by busy, noisy, tactile environments (due to a sensory processing disorder). This has particular implications for practical lessons such as science, food technology and physical education, where close supervision is required to ensure her safety and the safety of other students and staff. For example, in science lessons, she can easily become overwhelmed by equipment such as Bunsen burners, bright liquids and noisy experiments. When she is overstimulated, she may pick up or touch equipment (Bunsen burners, ovens, chemicals) and move around the room with them before a member of staff can react. When in noisy changing rooms, overstimulation can lead to her climbing on top of equipment such as lockers. This can be interpreted by uninformed staff as a behavioural issue and result in sanctions rather than understanding.

The Child as a Learner: Opportunities for Inclusion

Including this child in a mainstream secondary setting and providing access to the national curriculum have been possible through a combination of careful and thorough risk assessments, adult support and scaffolding, personalised teaching and learning and partnership with parents. For example:

- Before practical sessions such as food technology or science take place, the science teacher or ASC unit teacher (accompanied by a TA) will walk and talk the student through the equipment, providing her with clear demonstrations and explanations of the safe use of equipment. She is then in a position to attend a lesson alongside her peers armed with a basic understanding of the principles and expectations. This will always be supported by at least 1:1 and sometimes 2:1 adult supervision.
- The student has a visual timetable located in the ASC which she looks at each morning on arrival at the unit.
- A smaller version of the timetable is copied into her individual planner so that she can view it during lessons as necessary.
- TAs monitor her anxiety and arousal level throughout the day through discussions with her and liaise with her and each other, as well as the ASC unit teacher about those lessons where more support may be necessary. They can then discuss the nature of support necessary to ameliorate the effects of her Sensory Processing difficulties as far as possible.
- The ASC unit teacher (and school SENCo when appropriate) corresponds daily with parents by email regarding issues arising.

In addition, the student is provided with access to occupational therapy and music therapy to address sensory processing and social and emotional difficulties, combining a therapeutic approach with a differentiated national curriculum.

These measures ensure that this student is able to access the national curriculum in a mainstream educational setting alongside her peers.

29.7 Conclusion

This chapter has discussed the need to adapt the teaching environment for children with FASD and ways to do this as well as specific pedagogical approaches. The need to work closely with families has been stressed in order to increase consistency and predictability and reduce anxiety for children with FASD.

References

1. Millar JA, Thompson J, Schwab D, Hanlon-Dearman A, Goodman D, Koren G, Masoitti P. Educating students with FASD: linking policy, research and practice. J Res Res Spec Educ Needs. 2014;17(1):3–17.
2. Blackburn C, Carpenter B, Egerton J. Educating children and young people with fetal alcohol spectrum disorders. London: Routledge; 2012.
3. Malbin DV. Fetal alcohol spectrum disorder (FASD) and the role of family court judges in improving outcomes for children and families. Juv Fam Court J. 2004;55(2):53–63.
4. Jonsson E, Dennitt L, Littlejohn G, editors. Fetal alcohol spectrum disorder (FASD): across the lifespan. Edmonton, AB: Institute of Health Economics; 2009.
5. Blackburn C. Walking through a moonless night: FASD and early childhood intervention. In: Carpenter B, Egerton J, editors. FASD and interdisciplinary perspectives. London: Routledge; 2013. p. 102–22.
6. Green JH. Fetal alcohol spectrum disorders: understanding the effects of prenatal alcohol exposure and supporting students. J Sch Health. 2007;77(3):103–8.
7. Jirikowic T, Kartin D, Olson HC. Children with fetal alcohol spectrum disorders: a descriptive profile of adaptive function. Can J Occup Ther. 2008;75:238–48.
8. Blackburn C. Facing the challenge and shaping the future for primary and secondary aged students with fetal alcohol spectrum disorders (FAS-eD project, project toolkit including project report, literature review, case studies, podcasts and primary and secondary learning and teaching frameworks). 2010.
9. Kalberg WO, Buckley D. FASD: what types of intervention and rehabilitation are useful? Neurosci Biobehav Rev. 2007;31:278–85.
10. Blackburn C. Developing inclusive practice for young children with fetal alcohol spectrum disorders. London: Routledge; 2017.
11. Webster-Stratton C, Reid MJ. Strengthening social and emotional competence in young children—the foundation for early school readiness and success. Infants Young Child. 2004;17(2):96–113.
12. Graziano PA, Reavis RD, Keane SP, Calkins SD. Emotion regulation and Children's early academic success. J Sch Psychol. 2007;45(1):3–19.
13. Linnenbrink-Garcia L, Pekrun R. Students' emotions and academic engagement: introduction to the special issue. Contemp Educ Psychol. 2011;36:1–3.
14. Gottman J, Katz L, Hooven C. Meta-emotion—how families communicate emotionally. Mahwah, NJ: Lawrence Erlbaum; 1997.
15. Gottman J, Katz L, Hooven C. Parental meta-emotion philosophy and the emotional life of families: theoretical models and preliminary data. J Fam Psychol. 1996;10(3):243–68.
16. Shortt JW, Stoolmiller M, Smith-Shine JN, Eddy JM, Sheeber L. Maternal emotion coaching, adolescent anger regulation, and siblings' externalizing symptoms. J Child Psychol Psychiatry. 2010;51(7):799–808.
17. Goswami U. Cognitive neuroscience and learning and development. In: Moyles J, Georgeson J, Payler J, editors. Beginning teaching beginning learning in early years and primary education. Maidenhead: Open UP; 2011.

18. Goswami U. Learning difficulties: future challenges. In: Mental capital and wellbeing project. London: HMSO: Government Office for Science; 2008.
19. Blackburn C. Drinking it in. SEN Magazine September/October 2012 SEN60; 2012. p. 80–3.
20. Blackburn, C. Just one drink special world. September 2015. http://www.specialworld.net/author/dr-carolyn-blackburn/.

Managing the Transition to Adulthood: A Personal Perspective

30

Lee Harvey Heath

Chapter Highlights

- Personal reflection of an adult with FASD.
- Challenges faced by an adult with FASD.
- The positive changes that can be made with a diagnosis and understanding.

My name is Lee Harvey Heath. I was born with alcohol-related neurodevelopmental disorder. I was diagnosed at the age of 26. I founded FASD Devon & Cornwall consultancy, a growing organisation that I am going to be registering as a community interest company. We provide one-to-one consultations, advice, information, education, and raise awareness of fetal alcohol spectrum disorders, in the southwest. As well as doing public talks, we host support group and being the voice of those affected by prenatal exposure to alcohol around the UK. So far, I have met with local city councillors, local media, started one-to-one mentoring, helped individuals receive a diagnosis, educated adopters and foster carers all through my own personal experiences of living with FASD.

I do not suffer from FASD, but I am affected by it. Daily life at times can be very challenging, but I have learnt strategies to move and cope with my FASD; learning how my FASD affects me has been a huge part of the change in my life. The last 2 years have been a success story, my name is known not only in the UK but also in other countries for the work that I have done, but my story has not always been so good, I did not have the best start in life. Both my birth parents were alcoholics. I would often be left at nursery having nobody come to pick me up. I would often be left outside the pub, because my parents would be drinking and we were not allowed in. For the first 3 years of my life, I was raised by my two older sisters who were only three and six.

L. H. Heath (✉)
Plymouth, UK

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_30

At the age of 3, I was taken into foster care, and passed from foster home to foster home for the next 3 years, and one of those years I was separated from my sisters.

As a child, I was very withdrawn from the world, I was very emotional and would cry all the time; at the age of 6, myself and my sisters were put up for adoption. In 1991, we were adopted by a single woman and we were moved from our hometown of Plymouth to Norfolk.

Between the ages 6 and 16, I struggled terribly at school, I always knew I was different, but I did not know why. Life was very hard and I would constantly have melt downs, the melt downs were very different, rather than have violent outbursts and scream and shout, I would go very quiet and stopped speaking to everyone. I did not want to acknowledge anyone else. I wanted to be on my own. I would go into my own world and try to forget everything that was going on; this was the coping strategy for me and often, I would forget what happened, and that was the only way I knew how to cope. I hated school and did not want to be, I often felt nobody understood me and nobody would listen. I was always told he could not be bothered, I was lazy, and did not want to do the work that I was asked to do, did not listen, and would never concentrate. Every school report ever received always said 'lack of concentration'; but for me, this was never the case. I constantly tried my hardest in class but it was never good enough, I could not do what was being asked of me, I was too embarrassed to admit it, because I knew teachers would believe me and the other children would laugh at me, and I was already being bullied because I was an outsider and people knew I was an adopted child.

At the age of 10, my biological father passed away due to addictions to alcohol, and this was one of the hardest things I have never had to go through, because I always hoped I would see him again, then there is nothing worse than losing a parent of such a young age, the last time I saw my birth father when I was four. I still remember the game and I am told to walk home from school, I can still remember it like it was yesterday. This made life a lot harder for me at home and at school, the relationship with me and my (adoptive) mum became very difficult and I became very hard work or go into trouble at school for stealing, and I stole from home too. I stole money from my (adoptive) mum to buy sweets to take to school to give two other children so they would like me, I was buying my friends, the friends I did have at school were the special needs children, but even then, I felt different, we had a closer friendship together than I had with any of them. I always felt like the outsider and I never change.

After struggling so much in mainstream schools, my (adoptive) mum decided that I would be better off going to a private special needs school, so I went to the boarding school. I did finish school, and did do my GCSE's, but with no good grades I could use for anything. My best grade was a D, and that was in music. I always found music helped me, I could quite happily go into my own little world listening to or making music.

After returning home from boarding school, life between me and my (adoptive) mum became very hard. She did not understand me and I did not understand her and she thought that I could not be bothered and I was lazy. I began to run away from

home, at first for a day or two, and then more days at a time and the longest time was 2 weeks I spent away from home. Eventually, police realised I did not want to go back home and managed to find me a bedroom in the youth hostel. This is when I had my first alcoholic drink, and within 6 months, I was addicted. I would wake up not remembering what I had done the night before and down a couple of occasions attempted suicide, I cut my wrists, and also jumped at the window damaging my back, which still causes me chronic pain today for which I am on a pain relief. It always felt like the addiction was waiting for me, but now I know I was born addicted to alcohol.

After a year of living in a hostel, I decided to move to Plymouth to try and rebuild the relationship with my biological mother. However, after a short space of time, this did not work, and I was in Plymouth with no home, no real friends, and nowhere to go. I ended up being placed in another hostel over the next few years alcohol to hold of my life, more attempted suicide happened, more broken relationships happened, and I was unable to hold down a job. Nobody understood me and I still did not understand myself, and at the age of 19–20, people just assumed that I did not care, which was not the case.

Early in my twenties, I started a relationship, we moved in together, but I was not working and it was a financial burden to keep living in a flat. My partner was from Holland, and we ended up moving up there. She left before me that means being unable to manage money, it took me a lot longer to get a passport and follow her out there, in a few months, I was in England on my own. She found out she was pregnant, eventually ended to get out there, and after a few more months, my first daughter was born.

Unfortunately, this relationship did not last due to the problems that I had, the partner would find it very hard to deal with having to explain over and over again simple instructions such as doing laundry or cleaning, and at the end of the day, she would get home and things that she had asked me to do, did not get done because I had forgotten. This had caused problems and often ended in arguments to the point that we were both unhappy in the relationship, in the end, I had to move back to England on my own because I had nowhere else to go.

And back in with my (adoptive) mum temporarily but eventually I moved back to Plymouth again. I was not working and I was drinking very heavily and due to my drinking, I was unable to see my daughter, I had also become homeless due to my inability to manage money and did not always pay my rent on time, and my rent money would be spent on alcohol, so sometimes I spent nights on the street or I would spend nights on friend's sofas.

This was very hard because alcohol was the reason I was not allowed to see my daughter; this upset me deeply and only fuelled my drinking. In 2008, I was drinking daily, had been in and out of school for various suicide attempts, including being run over after running in front of the car, taking an overdose whilst under the influence of alcohol, and cutting myself. Being arrested for drunk, and disorderly was becoming a common thing. I would often find myself waking up in a police cell with no recollection of what had happened, but I was only ever charged with a caution.

I was hanging around with the wrong crowds and due to my undiagnosed fetal alcohol spectrum disorder, I did not have the ability to say no and was very easily led by others. In September 2008, I was arrested for robbery and I was placed on remand for 6 months, and because I was the oldest and seen as the more responsible and mature of the group, I was classed as the ringleader and I was given the sentence of 4 years. Within 2 years (spent in prison), I got sober, and I had all these great ideas of how I would get out of prison, see my daughter, stay sober and be a good father. But upon release date in 2010, having my first day of freedom, the first thing I did was to have a drink, within a matter of days, I was back on the alcohol, highly addicted, suicidal and starting to get into trouble again.

Finally, things go to the point where I had to leave Plymouth for my own safety, I had had another child by now, whom I was not seeing again because of my addiction, but yet again not seeing him only fuelled my drinking more. I went back to live with my mum who was now living in Portsmouth, and for 3 months ago sober. One weekend, my (adoptive) mum went away, had made some friends was working as a volunteer in a charity shop and ended up going out for a drink with them and relaxed. I joined because I did not want to carry on the way I was going, I did not want to end up like my (birth) father.

Living with my (adoptive) mum started to become very hard again, she found that the issue was still dealing with the same problems that I had when I was a young child, lack of concentration, short-term memory, attachment issues, and the inability to understand that thoughts and feelings. This may make life between us very hostile and she struggled to cope with me. Not long after every lapse, again after going on a 10-day drinking session, I found myself waking up in a church crying, asking for help. My (adoptive) mum came and got me, she locked me in the house for 2 weeks so I could not go out to get more alcohol, I was ill for 2 weeks and have since been told if I drink again, it would kill me, but 2 weeks when my mum locked me in the house, she saved my life.

My (adoptive) mum came home 1 day from the library, and she told me she had looked better condition called Fetal Alcohol Spectrum Disorder, she wanted me to look at the symptoms, her words were ‘I have just read this and is the profile that explains you’. When or if the symptoms, she was right it seemed to explain, I can relate to everything I was reading, from the short-term memory, anxiety, depression, low self-esteem, lack of concentration, sensory processing, everything on the symptoms list of FASD I could relate to, and we already knew my birth mum had been drinking during pregnancy.

So, eventually we went down the route of getting a diagnosis, and I was diagnosed with alcohol-related neurodevelopmental disorder. I was now 26, and I started to make sense. I finally began to realise that everything I have been through was not my fault, and all the problems I had were not because I could not be bothered, I was lazy, I did not listen, I did not want to do what was being asked of me. It was the fact that I could not do what was being asked of me, and this changed everything. It changed how my mum viewed me, it changed how I view to myself, and people around me especially my mum could begin to understand me. This made life very different.

I slowly began to understand what my problems were, and then began to be more open about what was really going on and how different I felt from the rest of the world. So, finally my life began to change.

I came to realise that the problems that I had were never going to go away. Fetal alcohol spectrum disorder is irreversible brain damage, but I also began to realise the various things that I can do to make life easier for me and for those around me, strategies that would help me to deal with day-to-day life, which would in turn make life easier for those around me.

Understanding is one of the biggest things when it comes to living with FASD and is not just understanding yourself, but having others around you be able to understand the moment that happens and life can change for the better.

I decided to look at what was on social media in terms of support for those living with FASD, because between me and my mum, we would come to realise that not many people knew what FASD was, G P's and other health professionals had not heard of it. So trying to find support is like looking for a needle in a stack. So I turned to social media, and managed to find Facebook group, with a lot of parents, carers, fosterers, and adoptive parents; this happened when I first got in contact with another adult with FASD. As I looked through this Facebook group and began to realise that there were people all up and down the country desperate for support, and not very many really understood the FASD mind. So I began to answer questions that parents have put into this group. And I found myself helping others, and it was very rewarding. It got to the point where I was getting messages asking for specific advice, so I decided to sell Facebook group, and I called the Q and A on FASD.

Up until now, I had never felt my life was worth anything, but that began to change over time, as I found myself helping parents to understand their children who were affected by prenatal exposure to alcohol. It gave my life some meaning.

Eventually, I was able to move back to Plymouth and began to rebuild the relationship with my children. My name began to spread on social media, as time passed, I would receive more and more messages and E mails asking for a specific advice on different topics, all regarding Fetal Alcohol Spectrum Disorder and the wide range of disabilities that overlapped with it. Whilst I was doing this, I was also becoming to learn more about myself and learn more about FASD as a whole.

I came to realise just how little awareness, knowledge, and support there was in the UK. And after everything I had been through, and almost did not make it back from, I did not want anyone else going through that. So I decided that I wanted to make a difference to the lives of others who either already have a diagnosis at all or have not got one, because I had witnessed first-hand what happens when you go through life within invisible disability and nobody picks up on, and when you do not receive the support you should, I know, if I had received the right support at my young age, half of what I have been through, the addiction, the suicide attempts, the homelessness, and my time spent in prison, possibly would not have happened, it could have all been avoided.

About 2 years ago, I moved into a small flat, and now I live independently, alcohol-free and I am in my children's lives. I now have a cat which helps with my anxiety and depression, and I finally have a place I can call home, my bills are paid,

and I can finally say I am happy. But it is taken a long time for me to get to where I am today. Having the diagnosis even though it came 26 years too late, changed my life for the better.

Now I am a public speaker, FASD consultant, author, and managing director of my own community interest company called FASD Devon and Cornwall consultancy, also known as FASDDAC consultancy. There are many things I cannot do, but I have gained a good network of people around me, they are able to help me with the things I can not do. It is the only organisation in Plymouth providing information, advice, and support for families affected by premium exposure to alcohol, and we now have local organisations coming to us. I host support groups, I also mentor, and I have been in national press and local press.

Part IV

Public Health and Policy

Public Health Perspectives on FASD: Prevalence, Inequalities, and Prevention

31

Kate M. Fleming and Penny A. Cook

Chapter Highlights

- How common is FASD and approaches to assess this.
- Public heath priorities related to prenatal alcohol exposure (PAE).
- Approaches to public health interventions related to FASD.

31.1 Overview

Public health is 'the art and science of preventing disease, prolonging life and promoting health through the organised efforts of society' [1] and focuses on the entire spectrum of health and well-being, not only the eradication of diseases. Public health activities can be targeted from population to individual level. Population level interventions include those applied generally, such as a health campaign to increase knowledge and awareness of drinking in pregnancy, to those aiming to address the social, economic and environmental conditions that cause ill-health such as legislation to increase the price or restrict the availability of alcohol. Individual level public health activities include personal services such as behavioural counselling and health advice. Non-medical interventions to individuals, which take place outside the clinical setting and have a positive impact on health

K. M. Fleming (✉)
Department of Public Health, Policy and Systems, Institute of Population Health, University of Liverpool, Liverpool, UK
e-mail: kate.fleming@liverpool.ac.uk

P. A. Cook
School of Health and Society, University of Salford, Salford, UK
e-mail: p.a.cook@salford.ac.uk

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_31

and well-being, also fall within the remit of public health. Modern public health emphasises reducing avoidable differences in ill-health between the most and least well-off in society [1, 2]. The reduction or elimination of Fetal Alcohol Spectrum Disorders (FASD) is a legitimate aim for public health activity because FASD is, in theory, an entirely preventable spectrum of conditions. If we were able to eradicate the drinking of alcohol in pregnancy, we would never see a child born with FASD. FASD is also a condition that leads to worse health outcomes, poorer educational outcomes, and fewer opportunities across the life course, and therefore contributes significantly to health inequalities. A significant number of affected individuals become involved in criminal activity or are exploited. People with FASD are often themselves more likely to have alcohol-use disorders leading to an increased risk of having a child with FASD, thus perpetuating the cycle of inequalities across generations.

A multi-pronged systems approach to such an intractable issue is required to see progress in reducing the number of individuals affected by FASD. We begin this chapter by reviewing what is known about the prevalence of FASD and outline why obtaining accurate prevalence estimates is challenging. We describe the inequalities in the burden of FASD in particular groups. We then introduce the public health approach from primary prevention strategies that should be applied at a population level to secondary prevention with populations at risk of drinking in pregnancy. We illustrate our examples with international case studies.

31.2 Prevalence

The prevalence of a condition is the proportion of individuals (in a specific geography at a specific time) who are affected by that condition. The absence of robust, reliable data on the prevalence of FASD is a major obstruction to providing accurate awareness in women, professionals, and society more broadly of what is a relatively common disorder. It is also a barrier to developing a public health approach to the prevention of FASD. We can approach measuring the prevalence of FASD from two ways. Firstly, obtaining accurate knowledge of the number of pregnancies that are exposed to alcohol, and secondly, looking for the outcome of that prenatal alcohol exposure in the children who are born, that is, counting the number of individuals with FASD. We first describe why gaining such estimates is a challenge.

31.2.1 Ascertaining Prevalence of Prenatal Alcohol Exposure

In 2017, a systematic review and meta-analysis estimating the prevalence of alcohol use during pregnancy provided country-specific data either from published studies (29 countries) or from modelled data (158 countries) [3]. Worldwide, prevalence of drinking in pregnancy was estimated to be 10% with countries from the European region reporting the highest prevalence, with one in four women drinking alcohol during pregnancy.

Data on alcohol consumption during pregnancy generally relies on self-reported data, which is problematic because of reporting bias [4]. Consequently, there is increasing interest in using objective measures of prenatal alcohol consumption such as biomarkers [5]. These include the measurement of a variety of alcohol metabolites, such as fatty acid ethyl ester and ethanol glucoronide taken from biological samples, including meconium, infant, or maternal blood [5]. A disadvantage with this is that most markers are only useful for determining alcohol consumption during later pregnancy, whilst the damage caused to the developing fetus occurs across the course of the pregnancy. According to a recent systematic review, the evidence base is not yet sufficiently strong for using such markers in practice [5].

Even if accurate measurements of prenatal alcohol exposure were available, this is still only half the battle. To date, there are no conclusive studies indicating a threshold level of harm to the fetus. Indeed, the most recent systematic review to examine drinking at a low level (i.e. less than 32 g alcohol/week) specifically stated that there was a paucity of evidence [6]. There are several inter-related factors which lead to a fetus being harmed from alcohol consumption, including smoking, nutrition status, frequency, timing and duration of drinking, many of which are themselves related to cultural and socioeconomic factors [7]. As some women who have consumed alcohol will not have affected babies, perhaps mediated by genetic and epigenetic factors, it is possible that active follow up of all pregnancies with any known alcohol consumption may not be a cost-effective public health approach to take.

31.2.2 Ascertaining Prevalence of Fetal Alcohol Spectrum Disorders

Owing to the limitations of both accurately identifying those pregnancies that have been exposed to alcohol and the absence of evidence of a threshold effect of alcohol harm, the best estimates of the prevalence of FASD come from studies that have actively sought to identify cases of FASD.

May and Gossage [8] summarise the common methods to assess prevalence of FASD, which include passive and active systems. Passive systems, which rely on gleaning data from existing sources, are less useful for capturing the prevalence of FASD than they are for other more recognisable conditions, because the diagnosis is not obvious [8]. Underdiagnosis occurs for a range of reasons, including: healthcare professionals lacking knowledge or specialist training; lack of established multidisciplinary teams to carry out all the necessary evaluations [9]; behavioural and developmental problems typical of FASD may not emerge until a child is at primary school, by which time evidence about whether the birth mother drank during pregnancy may be missing; difficulty with isolating FASD from the other common co-occurring disorders (such as ADHD or autism spectrum disorder); and the perception of FASD being a stigmatising label leading to an unwillingness to consider the diagnosis [10]. Moreover, since FASD rarely leads to a child being hospitalised, utilisation of hospital data sources for prevalence estimates is not reliable [11, 12]. An alternative prospective study design, following up women during and after

pregnancy, has the serious drawback that FASD is most commonly diagnosed later in the child's life leading to a long and costly follow up period [8, 12] . Prevalence estimates using active case ascertainment are considered the 'gold standard' method, and at their best, involve screening a cross section of the general population of children [13]. The substantial drawback to this method is the significant cost involved in conducting a rigorous study using active case ascertainment.

The existing high quality prevalence studies that use active case ascertainment have been systematically reviewed by Lange et al. [14]. The authors of this review went on to model the likely prevalence of FASD countries that did not have prevalence data. This was done by combining country estimates for drinking in pregnancy in each country that also had FASD prevalence estimates and deriving the relationship between the two. They then used this to derive estimates for countries without prevalence data, in order to highlight the need for FASD care for these countries. This review found that South Africa had the highest prevalence of FASD (11.1% of children estimated to have FASD), followed by Croatia (5.3%) and Ireland (4.8%). While South Africa and Croatia's data were directly obtained from FASD prevalence studies, the figure for Ireland was derived from the prediction model. Twenty countries in the European region were estimated to have a prevalence of over 2%, including the UK where the estimate was 3.2% (again, based on the prediction model). Based on active case ascertainment studies, Lange et al. estimated that the USA had a prevalence of 1.7%. However, a more recent study by May et al. [15] suggests that prevalence among the general US population could be substantially higher than previously reported: they propose a conservative estimate of 1–5%. Only two of the 222 children with FASD found in May et al.'s study had previously had an FASD diagnosis, underscoring the fact that diagnosis rates are very low, as has been reported elsewhere [11, 16].

31.3 Inequalities

As shown above, the prevalence of FASD varies widely across the globe. FASD has especially high levels among particular subgroups of the population. The best estimates of prevalence come from Australia, South Africa, and North America [3, 13, 14], where the initial work on Fetal Alcohol Syndrome (FAS) and FASD highlighted the inequality. In Australia, the indigenous populations of Aboriginal and Torres Strait Islander Australians have been highlighted in the work of Elliott [17]. In South Africa, the high level of FAS in the Western and Northern Cape Provinces has been extensively investigated [18], and in Northern America, inner city Black American populations and indigenous Canadians all exhibit high levels of FAS/FASD [19]. In fact, Jones and Smith [20], who are widely credited for the first description of FAS, used case studies of alcohol-affected native American children. Of the two active case ascertainment studies identified in aboriginal populations in Lange et al.'s systematic review [14], the prevalence of FASD was calculated to be 12% (Australia) and 19% (Canada). This has led to a concerted effort in Australia to explore the cycle of inequality to enable future generations to have the best opportunities (Fig. 31.1).

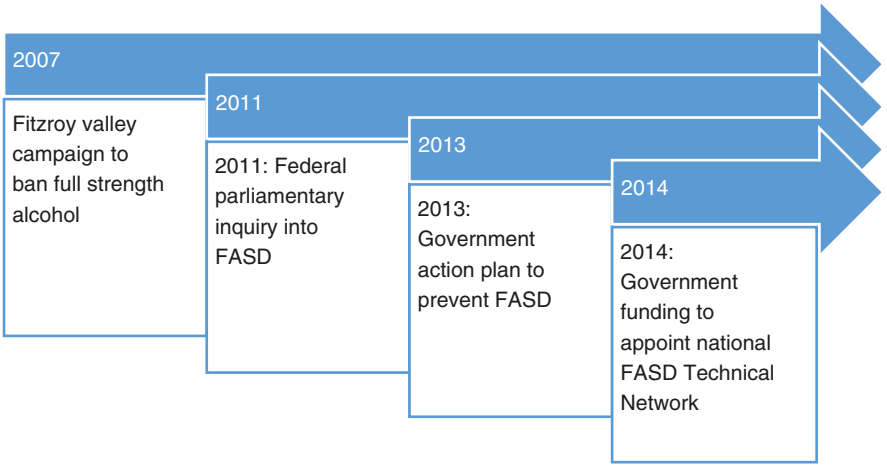


Fig. 31.1 Country profile: Australia local to national policy on prevention - timeline of major governmental and policy initiatives

It is not as simple as stating that alcohol consumption is higher in these communities than in others; rather, the patterns of drinking and the ability to reduce or cease drinking during pregnancy differ. These factors are themselves related to upstream ingrained social problems, such as poverty, poor education and housing, lack of opportunities, and racism faced by these communities [21]. These are part of the causal pathway of alcohol consumption and the interplay of complex social determinants of health [22]. For a further discussion of these social determinants of FASD, readers may wish to explore 'Fetal Alcohol Syndrome: The causal web from disadvantage to birth defect' [23].

Another special population that has been highlighted as bearing the brunt of FASD is children who are looked after in the adoption or social care system. Most of the emergent trends with 'looked after' children relate to data from international /transnational adoptions. For instance, children adopted from Central and Eastern Europe have often been reported to be prenatally exposed to alcohol. A meta-analysis published in 2013 [24] presented a pooled estimate of 17% for FASD in child care settings (for studies using active case ascertainment), ranging from 52% in a Swedish study of children adopted from Eastern Europe to 0% in two studies (USA children adopted from China and Eastern European children adopted from Romania, Ukraine and Moldova). Not included in the review was a study on mixed-race looked after children in England, which showed a prevalence of 30% [25]. Other more recent studies confirm the very high prevalence rate in this group: 29% of looked after children referred for behavioural problems in Chicago, USA [26]; 27% of looked after children referred for behavioural problems in Peterborough, UK [27]; 31% of children from Poland adopted to Dutch families [28]; and 17% children in a Brazilian orphanage [29] had FASD.

Many individuals with FASD find themselves in trouble with the law, and therefore criminal justice populations also have a high prevalence of FASD [30]. This is because the consequences of unsupported FASD include addiction, mental health problems, disengagement with education and inappropriate behaviour. A systematic review of studies carried out in the criminal justice system (e.g. prisons) in 2011 [31] found that all the studies had been carried out in either Canada (five studies) or USA (one). Studies using active case ascertainment (two Canadian studies) found prevalence of FASD substantially higher than the general population at 10.8–23.3%. A more recent systematic review to inform an analysis of the costs of FASD to the criminal justice system [32] did not reveal any more recent estimates. A recent study based in an Australian correctional facility, where three quarters of the detainees were Aboriginal, used active case ascertainment to reveal a prevalence of 36% among young detainees aged 10–17 years [33].

For individuals with FASD the differences in the prevalence among different groups are only one side of the inequalities coin. A recent systematic review identified 438 different ICD10 conditions linked to prenatal alcohol exposure [34], highlighting the significant range of physical and developmental conditions that have been attributable to the effects of alcohol on the fetus. Evidence from a 30-year cohort follow up of diagnosed individuals identified significant levels of mental health problems, criminalisation, sexual exploitation as well as addictions in affected individuals [35]. If adequate support and provision for individuals with FASD is not provided, these factors like health, education, and social disadvantages lead to an increase in health and social inequalities, which affect the individual, their existing family, and future generations. For this reason, FASD becomes entrenched and intertwined with social disadvantage in some populations.

31.4 Prevention of FASD: A Multicomponent Challenge

While prevalence studies reveal that FASD is a common disorder in the general population of many countries, and is especially concentrated among particular populations, prevention activities have not been proportional to the scale of the problem. This section gives a brief overview of the three levels of prevention, primary, secondary, and tertiary (Fig. 31.2), and how they can be applied to the prevention of FASD. At the top of the triangle in Fig. 31.2, tertiary prevention strategies aim to reduce the long-term consequences of FASD for individuals already diagnosed. Such interventions are outside the remit of this chapter (see Part III), suffice to comment that they have a public health function as they are vital to reduce costly individual and societal costs in terms of: poor mental and physical health; lack of engagement with education and consequent risk of criminal behaviour; risk of further alcohol-exposed pregnancies; and perpetuation of the cycle of alcohol-exposed pregnancies. Secondary prevention approaches are reviewed below, which include targeted interventions with women at risk of having an alcohol-exposed pregnancy and pregnant women. Primary prevention is aimed at the entire population, from which two main categories emerge: information campaigns for the general

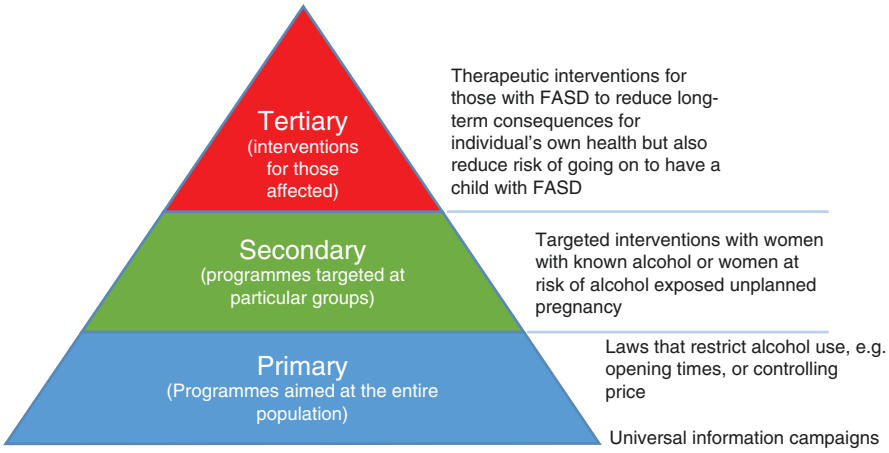


Fig. 31.2 Public health prevention pyramid with examples from FASD prevention

population and general alcohol reduction strategies, for example, the restriction of availability of alcohol and increasing the price of alcohol.

The prevention of FASD depends on the ability to reduce and ideally eradicate maternal alcohol consumption during pregnancy. The responsibility for this lies with society in general, not simply limited to women who are pregnant, or considering becoming pregnant, and their support networks, as demonstrated by the Canadian model (Fig. 31.3). The public health strategies that are likely to succeed in reducing alcohol consumption in pregnancy therefore need to consider this challenge in a multi-stage way and adopt a systems approach (see the New Zealand approach, Fig. 31.4).

31.4.1 Knowledge of the Risk of Alcohol Consumption in Pregnancy

Most countries have adopted the precautionary principle and within pregnancy promote the message 'No alcohol = No harm'. The timing of adoption of such a message has varied, with noticeably the UK lagging behind and introducing a new guideline recommending abstinence from drinking alcohol in pregnancy only in 2016 [38]. This may have contributed to uncertainty in health professionals, with recent UK research finding that fewer than 2% of professionals surveyed were 'very prepared to deal with the subject' [39]. In contrast, a study in Australia, where there has been clear guidance for some time, showed that two-thirds provided information about the effects of alcohol in pregnancy [40]. These variations in healthcare professionals' views alongside the lack of definitive guidance may have led to ambivalence by women in the UK towards drinking in pregnancy, in contrast with, for example, the strong belief that alcohol in pregnancy should be avoided held by Swedish women [41].

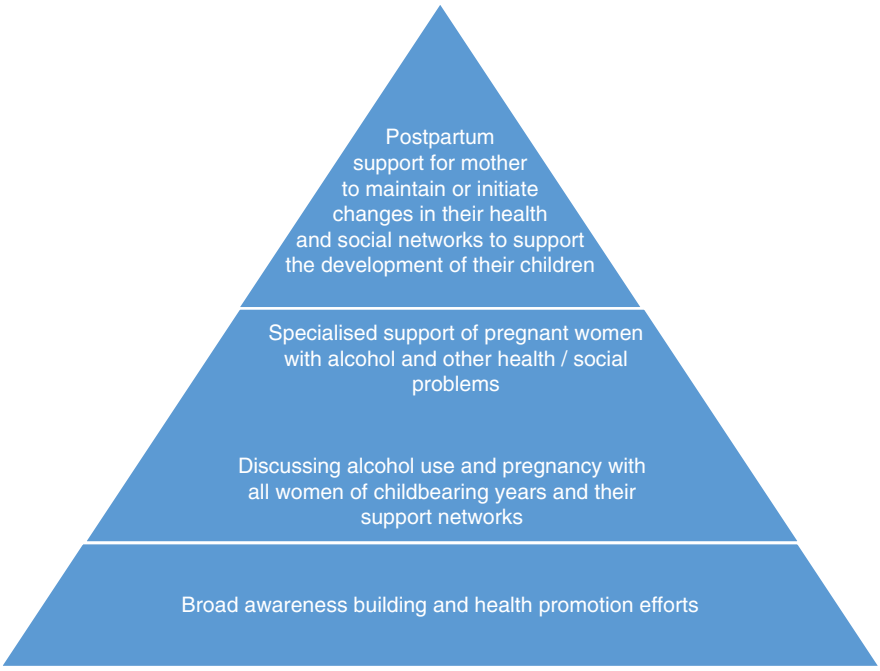


Fig. 31.3 Country profile: Canada's four-part model of prevention. Representing possibly the most developed strategy for the prevention of FASD, Canada has built on decades of work on learning about how to prevent FASD, advocating an approach which is inclusive of women and their support structures, through a multi-agency holistic approach [36]

Studies from around the world, including studies in the UK, have identified that the level of knowledge about FASD is limited. Increasingly, people have heard of the condition but, unlike conditions of arguably similar prevalence (e.g. autism), know little else about it. Professionals, public and carers of individuals with FASD all highlighted that there is a lack of knowledge and understanding broadly about FASD including appropriate care and support pathways for individuals who are affected [10, 42, 43].

31.4.2 Delivering the Right Message in the Right Way at the Right Time to the Right People

Based on the premise of adopting a universal message of 'no alcohol = no harm', it is necessary to explore the best formats for this message to be communicated and received by relevant groups of the population. The message from the public health community needs to balance its messaging for those women who are not yet pregnant, providing adequate information such that women can make their choices in an informed way against those women who discover they are pregnant and may have consumed alcohol early in their pregnancy, where there is a need to guard against

In August 2016 the New Zealand Ministry of Health set out its goals and aspirations with regard to "Taking Action on Fetal Alcohol Spectrum Disorder" for the period 2016-19(37). This action plan focusses on population based-strategies and the reduction of health inequalities. The 4 priorities set out within the action plan are
1. Prevention: to educate families and whānau*about the risk of drinking during pregnancy and support them to have healthy, alcohol-free pregnancies
2. Early identification: to identify people with neurodevelopmental impairments early and to provide timely and effective assessment from FASD capable teams
3. Support: to ensure people and their families, whānau and caregivers receive timely joined up support tailored to their needs, strengths, age and stage.
4. Evidence: to improve the New Zealand evidence base.

Taken from "Taking Action on Fetal Alcohol Spectrum Disorder: 2016-2019. An Action Plan" Wellington: Ministry of Health, New Zealand
whānau* : extended family or community of related families

Fig. 31.4 Country profile: New Zealand

undue worry and stigmatisation. In addition, there are specific groups of the population who are already known harmful drinkers who require more intensive help and support, which may come in the form of brief intervention or sustained involvement with healthcare professionals.

Pregnant Women with Known Alcohol Problems

Women with known alcohol problems, as with women with other physical or mental health problems, should ideally have a tailored package of care during pregnancy, usually delivered by a multidisciplinary, multi-agency care team. Preferably, intervention in women with known alcohol problems should begin before pregnancy, be this a first or a subsequent pregnancy. This can be akin to preconception counselling as is recommended for many other medical conditions and can be provided either in a clinical or community setting. A brief alcohol intervention is known not to work in this group of women and enhanced service involvement is required [44]. Some success has been seen when intervening in women with an affected child for support before subsequent pregnancies with research in the US and Canada providing suggested models of interventions [36]. The WHO 'Guidelines for the identification and management of substance use and substance use disorders in pregnancy' provide additional information for professionals working with women with known alcohol use disorders in the pregnancy and post-partum periods [45].

All Pregnant Women

Opportunities exist across healthcare contacts during pregnancy in which discussions about alcohol consumption could be initiated. Screening all pregnant women and brief interventions for all women who drink is recommended by the WHO 'Guidelines for the identification and management of substance use and substance use disorders in pregnancy' [45].

Few studies have examined the effectiveness of public health interventions in reducing alcohol consumption during pregnancy [46]. These interventions have ranged from universal prevention interventions, which include mass media campaigns and individual contact, through the use of text messaging and multimedia interventions, single brief interventions to multiple motivational interviewing sessions. Interventions showed varied results dependent on goal setting and messaging, with few showing statistically significant differences between intervention and control arms. Qualitative research that has examined the type of messaging that is most effective has shown the same results for both alcohol in pregnancy and smoking in pregnancy [47, 48]. Messages that were explicit in relating harm to the child and elicited high affect-arousing emotions, rather than those based on a concept of self-efficacy, were more likely to result in a change of intention.

All Women of Childbearing Age

In an ideal situation, we want to ensure that all pregnancies begin and continue without any exposure to alcohol. Mass media campaigns, often using television and radio advertisements alongside posters and brochures are commonly used in order to try and provide public health messages to a wide audience, including women of childbearing age. Studies from North America have shown that similar to interventions that were targeted to pregnant women, these types of interventions increased knowledge and awareness of alcohol in pregnancy and the associated risks in women of childbearing age [46]. Interventional methods that have been used to target non-pregnant women include brief intervention and motivational interviewing. In the pervasive culture of pro-alcohol social norms, we also need to take parallel steps to reduce the significant potential for unintentional harm by promoting policies that are aimed at reducing the number of unplanned pregnancies. With only just over half of pregnancies being classed as planned, and unplanned pregnancy being associated with low educational attainment [49], there is the potential for a further widening inequality in those pregnancies unknowingly affected by prenatal alcohol exposure. Therefore, some interventions target both alcohol use and contraception use (e.g. the Changing High Risk Alcohol Use and Improving Contraception Effectiveness Study, CHOICES) [50].

Any public health campaign should take account of the reports of scientific papers that are covered in the mainstream media. Regrettably, erroneous front page headlines such as '*Light drinking does no harm in pregnancy'* (The Times, Sept 12, 2017), become widely publicised, particularly through social media, yet their corrections are not so easily accessible '*No need for guilt over light drinking in pregnancy*' (The Times, Sept 14, 2017 (pg 34 correction)) [51].

31.4.3 Primary Prevention: Universal Public Health Strategies

The above strategies for reduction in alcohol consumption in pregnancy and prevention of FASD lie largely in the realm of individual behaviour change, supported to a

greater or lesser extent by health professionals. To facilitate this, we must also construct a wider society that can support women to make these changes. Attention needs to focus on the partners and support networks that individual women rely on. More generally, the public health community must also advocate for a society where not drinking is not perceived as being 'abnormal' such that women are protected from intrusive questions regarding their fertility choices when not drinking which may lead to both anxiety and potential 'coercion' into drinking alcohol, intentionally or otherwise.

A current mechanism for alcohol-related health promotion is on the labels of bottles. In this way, there is targeting of those who already engage in drinking. Though relatively cheap to implement, with many manufacturers engaging in voluntary labelling, it is regrettably a relatively ineffective way of changing behaviour [52]. Industry involvement in messaging can also unfortunately result in incorrect messaging being communicated. In 2018, posters funded by the Australian drinks industry-funded DrinkWise initially included the sentence 'It is not known if alcohol is safe to drink when you are pregnant' until coordinated action from the Australian Medical Association forced its removal [53]. Universal measures are also more likely to benefit those of a higher education and socioeconomic status, potentially leading to even wider inequalities in health behaviour.

Some organisations have called for the implementation of school-based education programmes to include the awareness of the risks of alcohol consumption in pregnancy [9] thereby introducing the concept of risk to an audience before they are likely to engage in sexual activity. However, the effectiveness of alcohol education programmes in changing attitudes rather than simply increasing knowledge, is variable and inconclusive [54].

31.4.4 Pushing for Real Change: Addressing Upstream Factors

All the above interventions approach the prevention of alcohol consumption in pregnancy, and therefore consequent children born with FASD, through the mediation of downstream factors. In addition to specific messages that are aimed at educating the public about the risks of alcohol in pregnancy, any public health initiatives or governmental policy successful at reducing alcohol intake more generally in society will have a knock-on effect on reducing alcohol consumption in pregnancy. Such policies include the regulation of supply, affordability, and promotion of alcohol across society, the discussion of which is beyond the scope of this chapter, but which are known to be more effective than education and messaging [52]. Therefore, as illustrated by the 'framework for public health action' (Fig. 31.5), for a concerted effort to reduce FASD, the upstream causes of alcohol consumption such as poverty, social injustice, and oppression must also be addressed (see the bottom rung of Fig. 31.5).

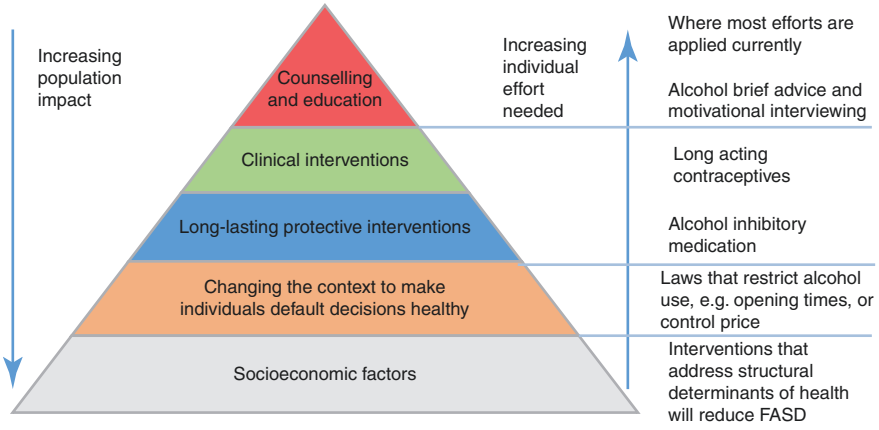


Fig. 31.5 Framework for public health action adapted from Frieden [55] combined with examples for FASD

31.5 Summary

The information and awareness raising strategies presented in this chapter are represented at the top of the framework for public health action (Fig. 31.5), where there is the lowest degree of impact and the highest degree of individual effort required to make a behaviour change. This is not to say that education efforts should be ignored. Indeed, adequate messaging to all women, partners, and society regarding the harms of alcohol in pregnancy alongside brief interventions and contraceptive provision is vital. However, to truly impact on present and future generations and prevent FASD, we need a systems approach to reducing the social inequalities of health.

References

1. Acheson D. Public health in England. The report of the Committee of Inquiry into the future development of the public health function. London: HMSO; 1988.
2. Marmot M. Fair society healthy lives. UCL Institute of Health Equity; 2010. http://www.instituteofhealthequity.org/resources-reports/fair-society-healthy-lives-the-marmot-review.
3. Popova S, Lange S, Probst C, Gmel G, Rehm J. Estimation of national, regional, and global prevalence of alcohol use during pregnancy and fetal alcohol syndrome: a systematic review and meta-analysis. Lancet Glob Health. 2017;5(3):E290–E9.
4. Muggli E, Cook B, O'Leary C, Forster D, Halliday J. Increasing accurate self-report in surveys of pregnancy alcohol use. Midwifery. 2015;31(3):E23–E8.
5. McQuire C, Paranjothy S, Hurt L, Mann M, Farewell D, Kemp A. Objective measures of prenatal alcohol exposure: a systematic review. Pediatrics. 2016;138(3).
6. Mamluk L, Edwards HB, Savovic J, Leach V, Jones T, Moore THM, et al. Low alcohol consumption and pregnancy and childhood outcomes: time to change guidelines indicating apparently 'safe' levels of alcohol during pregnancy? A systematic review and meta-analyses. BMJ Open. 2017;7(7).
7. May PA, Gossage JP. Maternal risk factors for fetal alcohol spectrum disorders not as simple as it might seem. Alcohol Res Health. 2011;34(1):15–26.

8. May PA, Gossage JP. Estimating the prevalence of fetal alcohol syndrome—a summary. Alcohol Res Health. 2001;25(3):159–67.
9. BMA. Alcohol and pregnancy: preventing and managing fetal alcohol spectrum disorders. London: British Medical Association, Board of Science; 2016. https://www.bma.org.uk/what-we-do/population-health/drivers-of-ill-health/alcohol-and-pregnancy-preventing-and-managing-fetal-alcohol-spectrum-disorders.
10. Mukherjee R, Wray E, Curfs L, Hollins S. Knowledge and opinions of professional groups concerning FASD in the UK. Adopt Foster. 2015;39(3):212–24.
11. Morleo M, Woolfall K, Dedman D, Mukherjee R, Bellis MA, Cook PA. Under-reporting of foetal alcohol spectrum disorders: an analysis of hospital episode statistics. BMC Pediatr. 2011;11:14.
12. Moberg DP, Bowser J, Burd L, et al. Fetal alcohol syndrome surveillance: age of syndrome manifestation in case ascertainment. Birth Defects Res A Clin Mol Teratol. 2014;100(9):663–9.
13. Roozen S, Peters GJY, Kok G, Townend D, Nijhuis J, Curfs L. Worldwide prevalence of fetal alcohol spectrum disorders: a systematic literature review including meta-analysis. Alcohol Clin Exp Res. 2016;40(1):18–32.
14. Lange S, Probst C, Gmel G, Rehm J, Burd L, Popova S. Global prevalence of fetal alcohol spectrum disorder among children and youth a systematic review and meta-analysis. JAMA Pediatr. 2017;171(10):948–56.
15. May PA, Chambers CD, Kalberg WO, et al. Prevalence of fetal alcohol spectrum disorders in 4 US communities. JAMA. 2018;319(5):474–82.
16. Watts M. Progress in addressing FASD in Scotland. Adopt Foster. 2015;39(3):256–62.
17. Elliott EJ. Fetal alcohol spectrum disorders: Australian perspectives. In: Carpenter B, Blackburn C, Egerton J, editors. Fetal alcohol spectrum disorders: interdisciplinary perspectives. Abingdon, Oxon: Routledge Taylor and Francis Group; 2014. p. 294–305.
18. Viljoen D. Fetal alcohol spectrum disorders: the current situation in South Africa. In: Carpenter B, Blackburn C, Egerton J, editors. Fetal alcohol spectrum disorders: interdisciplinary perspectives. Abingdon, Oxon: Routledge Taylor and Francis Group; 2014. p. 288–93.
19. Grant TM, Clarren SK. North American perspectives on fetal alcohol spectrum disorders. In: Carpenter B, Blackburn C, Egerton J, editors. Fetal alcohol spectrum disorders: interdisciplinary perspectives. Abingdon, Oxon: Routledge Taylor and Francis Group; 2014. p. 265–76.
20. Jones KL, Smith DW. Recognition of fetal alcohol syndrome in early infancy. Lancet. 1973;2(7836):999–1001.
21. Tait CL. Aboriginal identity and the construction of fetal alcohol syndrome. In: Kirmayer LJ, Macdonald ME, Brass GM, editors. The mental health of indigenous peoples: proceedings of the Advanced Study Institute, The Mental Health of Indigenous Peoples, McGill Summer Program in Social & Cultural Psychiatry and the Aboriginal Mental Health Research Team (May 29—May 31, 2000, Montréal, Québec). Division of Social & Transcultural Psychiatry, McGill University. https://www.mcgill.ca/tcpsych/files/tcpsych/Report10.pdf2001.
22. Dahlgren G, Whitehead MW. European strategies for teaching social inequalities in health: levelling up part 2 (studies on social and economic determinants of population health, no. 3). WHO Regional Office for Europe: Copenhagen; 1991.
23. Gray R. Fetal alcohol syndrome: the causal web from disadvantage to birth defect. In: Carpenter B, Blackburn C, Egerton J, editors. Fetal alcohol spectrum disorders: interdisciplinary perspectives. Abingdon, Oxon: Routledge Taylor and Francis Group; 2014. p. 27–38.
24. Lange S, Shield K, Rehm J, Popova S. Prevalence of fetal alcohol spectrum disorders in child care settings: a meta-analysis. Pediatrics. 2013;132(4):E980–E95.
25. Selwyn J, Wijedesa D. Pathways to adoption for minority ethnic children in England—reasons for entry to care. Child Fam Soc Work. 2011;16(3):276–86.
26. Chasnoff IJ, Wells AM, King L. Misdiagnosis and missed diagnoses in Foster and adopted children with prenatal alcohol exposure. Pediatrics. 2015;135(2):264–70.
27. Gregory G, Reddy V, Young C. Identifying children who are at risk of FASD in Peterborough: working in a community clinic without access to gold standard diagnosis. Adopt Foster. 2015;39(3):225–34.

28. Knuiman S, Rijk CHAM, Hoksbergen RAC, van Baar AL. Children adopted from Poland display a high risk of foetal alcohol spectrum disorders and some may go undiagnosed. Acta Paediatr. 2015;104(2):206–11.
29. Stromland K, Ventura LO, Mirzaei L, de Oliveira KF, Bandim JM, Ivo AP, et al. Fetal alcohol spectrum disorders among children in a Brazilian orphanage. Birth Defects Res Pt A Clin Mol Teratol. 2015;103(3):178–85.
30. Streissguth AP, Bookstein FL, Barr HM, Sampson PD, O'Malley K, Young JK. Risk factors for adverse life outcomes in fetal alcohol syndrome and fetal alcohol effects. J Dev Behav Pediatr. 2004;25(4):228–38.
31. Popova S, Lange S, Bekmuradov D, Mihic A, Rehm J. Fetal alcohol spectrum disorder prevalence estimates in correctional systems: a systematic literature review. Can J Publ Health. 2011;102(5):336–40.
32. Thanh NX, Jonsson E. Costs of fetal alcohol spectrum disorder in the Canadian criminal justice system. J Popul Therap Clin Pharmacol. 2015;22(1):e125–31.
33. Bower C, Watkins RE, Mutch RC, Marriott R, Freeman J, Kippin NR, et al. Fetal alcohol spectrum disorder and youth justice: a prevalence study among young people sentenced to detention in Western Australia. BMJ Open. 2018;8(2).
34. Popova S, Lange S, Shield K, Mihic A, Chudley AE, Mukherjee RAS, et al. Comorbidity of fetal alcohol spectrum disorder: a systematic review and meta-analysis. Lancet. 2016;387(10022):978–87.
35. Streissguth AP, O'Malley K. Neuropsychiatric implications and long-term consequences of fetal alcohol spectrum disorders. Semin Clin Neuropsychiatry. 2000;5(3):177–90.
36. Poole N. Fetal alcohol spectrum disorder (FASD) prevention: Canadian perspectives. Public Health Agency of Canada, Division of Childhood and Adolescence; 2008.
37. FASD Working Group. Taking action of fetal alcohol spectrum disorder: 2016–2019. an action plan. Wellington: New Zealand Ministry of Health; 2016.
38. UK Chief Medical Officers. UK Chief Medical Officers' low risk drinking guidelines, 2016. https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment_data/file/545937/UK_CMOs__report.pdf.
39. Winstone AM, Verity C. Antenatal alcohol exposure: an east Anglian study of midwifery knowledge and practice. Br J Midwifery. 2015;23(3):171–7.
40. Payne JM, Watkins RE, Jones HM, Reibel T, Mutch R, Wilkins A, et al. Midwives' knowledge, attitudes and practice about alcohol exposure and the risk of fetal alcohol spectrum disorder. BMC Pregnancy Childbirth. 2014;14:377.
41. Scholin L, Hughes K, Bellis MA, Eriksson C, Porcellato L. Exploring practices and perceptions of alcohol use during pregnancy in England and Sweden through a cross-cultural lens. Eur J Public Health. 2018;28(3):533–7.
42. Mukherjee R, Wray E, Commers M, Hollins S, Curfs L. The impact of raising a child with FASD upon carers: findings from a mixed methodology study in the UK. Adopt Foster. 2013;37(1):43–56.
43. Mukherjee R, Wray E, Hollins S, Curfs L. What does the general public in the UK know about the risk to a developing foetus if exposed to alcohol in pregnancy? Findings from a UK mixed methodology study. Child Care Health Dev. 2015;41(3):467–74.
44. Raistrick, Duncan and Heather, Nick and Godfrey, Christine (2006) Review of the effectiveness of treatment for alcohol problems. London: National Treatment Agency; 2016.
45. World Health Organisation. Guidelines for the identification and management of substance use and substance use disorders in pregnancy. Geneva: World Health Organization; 2014. http://www.who.int/substance_abuse/publications/pregnancy_guidelines/en/.
46. Crawford-Williams F, Fielder A, Mikocka-Walus A, Esterman A. A critical review of public health interventions aimed at reducing alcohol consumption and/or increasing knowledge among pregnant women. Drug Alcohol Rev. 2015;34(2):154–61.
47. Hoek J, Gifford H, Maubach N, Newcombe R. A qualitative analysis of messages to promote smoking cessation among pregnant women. BMJ Open. 2014;4(11).

48. France KE, Donovan RJ, Bower C, Elliott EJ, Payne JM, D'Antoine H, et al. Messages that increase women's intentions to abstain from alcohol during pregnancy: results from quantitative testing of advertising concepts. BMC Public Health. 2014;14:30.
49. Wellings K, Jones KG, Mercer CH, Tanton C, Clifton S, Datta J, et al. The prevalence of unplanned pregnancy and associated factors in Britain: findings from the third National Survey of sexual attitudes and lifestyles (Natsal-3). Lancet. 2013;382(9907):1807–16.
50. Project CHOICES Intervention Research Group. Reducing the risk of alcohol-exposed pregnancies: a study of a motivational intervention in community settings. Pediatrics. 2003;111(5 Pt 2):1131–5.
51. Lay K. No need for guilt over light drinking in pregnancy. The Times. 2017. https://www.thetimes.co.uk/article/no-need-for-guilt-over-light-drinking-in-pregnancy-ggsw5vwpc. Accessed 12 Sept 2017.
52. Babor TF, Caetano R, Casswell S, Edwards G, Giesbrecht N, Graham K, et al. Alcohol: no ordinary commodity: research and public policy. Oxford Scholarship Online; 2010.
53. Han E. 'Utterly wrong': what happens when the alcohol industry makes pregnancy warning posters. The Sydney Morning Herald. 2018. https://www.smh.com.au/healthcare/utterly-wrong-what-happens-when-the-alcohol-industry-makes-pregnancy-warning-posters-20180827-p50022.html.
54. Martin K, Nelson J, Lynch S. Effectiveness of school-based life-skills and alcohol education programmes: a review of the literature. National Foundation for Educational Research; 2013.
55. Frieden TR. A framework for public health action: the health impact pyramid. Am J Public Health. 2010;100(4):590–5.

Alcohol-Related Harm and Pregnancy: Public Policy, Attitudes and Recognition

32

Moira Plant

Chapter Highlights

- Drinking patterns including during pregnancy.
- Methods of identifying high-risk drinking.
- Policy context to alcohol consumption.

32.1 Prevalence

Until a short while ago the worldwide prevalence of alcohol consumption amongst pregnant women was unknown. However, recent work provides the picture [1, 2]. The recent meta-analysis of World Health Organisation (WHO) member states [2] reported that worldwide the prevalence of prenatal alcohol exposure is almost 10%. By country the highest-ranking were Russia, UK, Denmark, Belarus and Ireland. However rates of drinking during pregnancy change in countries depending on a number of factors such as raised awareness of risk and the continuing improvement in collecting alcohol consumption data from pregnant women more detailed accurate. Clearly even in countries where drinking in pregnancy is decreasing overall there are a number of women who continue to drink into their pregnancies. Some studies have focused on women at higher risk of having an alcohol-exposed pregnancy [3, 4].

M. Plant (✉)
University of the West of England, Bristol, UK
e-mail: Moira.Plant@uwe.ac.uk

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_32

32.2 Drinking Patterns

In general, alcohol consumption is measured by pattern, usually in terms of quantity of alcohol drunk, the frequency at which this occurs and—in relation to pregnancy—the timing of drinking as it equates to the stage of pregnancy. The pattern of heavy episodic drinking (HED) or what is commonly known as binge drinking is now generally accepted as the most risky for the fetus [5].

Although the term binge drinking is used commonly in many countries, there are problems with translation. Firstly, a binge is defined in different ways in different countries. Indeed, the original definition of a binge was defined as one of the clinical features of an alcohol problem and was assessed as such in most alcohol treatment units. It was defined as drinking heavily for a period of days to the detriment of all other aspects of the person's life. More recently, a binge has been defined as drinking more than a certain *amount* of alcohol *within* a set period of time. This term is a good illustration of the difficulties encountered in cross-country comparison when different counties define behaviours in different ways [6, 7]. Indeed, there is particular confusion around the terms binge drinking, heavy episodic drinking (HED) and risky single occasion drinking (RSOD) as these terms are at times used interchangeably when the harm caused may differ [8]. Furthermore, while binge drinking is the most risky pattern for the fetus, it is worth questioning whether a more complete overall picture would be found if other measures of drinking such as regular volume and special occasion drinking were to be taken into account. For example, special occasion drinking may occur at binge levels, but the woman may not define them as such [9, 10]. Special occasion drinking should now be added to any alcohol consumption history taken in clinics.

32.3 Changes in Drinking Pre-Pregnancy and During Pregnancy

The ideal may be that every woman is alcohol free throughout her pregnancy. It appears the women who drink when pregnant will be drinkers *prior* to pregnancy and women who binge drink are more likely to continue to drink while pregnant albeit at lower levels [11]. Other studies from a variety of countries has shown continuing or sporadic drinking in pregnancy [12–15]. Studies from Ireland, for example, [16] have shown that between 75% and 81% of pregnant women drank in the periconceptual period.

A recent study of Australian women [17] showed that although the heavy drinkers reduced their consumption during pregnancy, the lighter drinkers did not. The issue of personality traits being associated with continuing or stopping drinking in pregnancy has been less well explored. The idea of conscientiousness, one of the five major personality traits, is associated with conforming to social norms and a sense of personal responsibility. Scoring high on this trait may be associated with a reduction or cessation of drinking during pregnancy [18–20]. This would fit with a willingness to conform to the public health messages and societies' views of

acceptable behaviours. Further work may elucidate this and help to clarify whether this is a productive field of inquiry.

The literature shows minimal consistency around variables, such as age, with some studies identifying younger women as more likely to drink in pregnancy, particularly in early pregnancy [21]. One reason for this may be whether the pregnancy was planned or unplanned, this will be discussed next. A study carried out by Cooper et al. [5] showed differences in the alteration of drinking levels during pregnancy. Those more likely to reduce rather than stop drinking entirely were women with higher education levels and those who were older. Other studies identified older women as more likely than their younger counterparts to drink during pregnancy [22]. Women who had friend or family experiences of drinking problems or those experiencing intimate partner violence (IPV) [23] were the least likely to be able to stop drinking. This latter factor has been identified repeatedly [24, 25] and will be discussed more fully later in this chapter.

Another major problem in assessing alcohol consumption is the question "what is a drink?" This is important within individual antenatal clinics for consistency, and also to enable comparisons within and between countries. It has long been known that drink measures poured at home are larger than pub measures with home measures often containing 2–3 times more alcohol [26–30]. When people report how much they drink, they will usually underestimate the amount. In the majority of cases, this is not because they are trying to mislead the health professional, but rather that they are not aware of the amount of alcohol in the glasses from which they drink. This latter is an important point to note when it comes to screening women in antenatal clinics both for objective and subjective responses. If the health professional begins the interview believing the woman is going to lie, the stage is set for a less than productive encounter.

32.4 Screening Tools

Many of our original alcohol screening tools were designed for male drinkers. Some of these were very brief, for example the four questions CAGE [31], others took more detailed information and therefore more time such as the timeline follow back technique [32]. The majority of these tools were less sensitive in identifying alcohol problems amongst female drinkers in general, and far less in pregnant women. For example, a question such as "Have you ever missed a day's work due to your drinking" may fail to identify a woman who is at home with her children. She may not have an alcohol-related problem, but will certainly be someone the midwife or obstetrician would want to talk with further.

There are a number of alcohol screening tools available for use in pregnancy: the T-ACE [33], TWEAK [34, 35], TACER-3 [36]. The "T" standing for tolerance appears to be a useful measure for general screening of pregnant women. Anyone who drinks develops a degree of tolerance, which means, they start by feeling the effects of alcohol at very low doses but with continued drinking practice comes the development of a tolerance. In short, people have to drink more to feel the effects.

There may be a risk; these screening tools will not pick up women with serious drinking problems. This will be briefly discussed next.

In Europe, the most commonly used tool is the Alcohol Use Disorders Identification Test (AUDIT) [37]. It can be applied to many different populations including pregnant women. As with the above-mentioned tools, the AUDIT asks questions about quantity and frequency of drinking, as well as alcohol-related problems such as feeling guilty about drinking or having a drink first thing in the morning—the latter is a common question also named "eye opener". This screening tool has 10 questions and was deemed too long for many settings such as antenatal clinics. The AUDIT-C, AUDIT-3 and AUDIT-4 were therefore designed to take this into account and have been reported to be as effective as the full AUDIT screening tool [38]. A number of studies have reported using these tools [39–42]. Other short screening tools include the Fast Alcohol Screening Test (FAST) [43] and the 4P's Plus [44]. Other studies have shown some success with very brief screening [45]. A number of comparison between different tools have been carried out [46–50].

There are a number of important issues related to the screening tools used in antenatal clinic settings, for example they need to be short—antenatal clinics are busy places. The woman will be asked many questions during her first visit, so questions on alcohol should take no longer than 3–4 min. Alcohol questions should be routinely administered to *every* woman—in this way the health professionals both become used to asking questions on alcohol and are given a defence if met with hostile responses. They can honestly say "we ask everyone these questions". If the screening tool is only used when the health professional thinks the woman has a problem with alcohol, the woman will likely become defensive leading to possibly inaccurate or incomplete responses to vital questions.

Practice point: women who are older, have a family history of drinking problem and who have experienced adverse childhood life events appear to be at higher risk.

Practice point: Do not assume that drinking patterns and levels of consumption remain static throughout a pregnancy – need to ask again in some clients [51].

Screening leading to identification is not an end in itself. It is where necessary, the *beginning* of a treatment programme. Examples of these include guidelines for identification and management of alcohol and other drug use in pregnancy [52–55]. Further examples are the US Department of Health and Human services guidelines on screening, brief intervention and referral to treatment (SBIRT) (United States Department of Health and Human Services [56]) and use of the Alcohol Screening

and Assessment in Pregnancy (ASAP) tool [57]. Attractive to governments and policy makers as a quick and non-expensive treatment [58–60], the evidence on the longer term success of brief interventions is awaited.

Other measures used in screening for alcohol use/abuse in pregnancy include biomarkers such as Gamma GT (GGT) and mean corpuscular volume (MCV) [61, 62]. A recent review [63] compared biomarkers with self-report information. After searching through four of the major databases, the team found only eight studies which allowed for comparison of biomarkers with self-report data. The team concluded that at best a combination of biomarkers or biomarkers plus self-report data may be useful in assessing drinking levels. Measures such as blood alcohol levels (BAC) may also be used as a measure of alcohol consumption, but there is a risk, these types of measures can bring legal and ethical consequences. This will be discussed briefly later in this chapter.

Whatever screening tool is chosen, it is imperative to note that identification is only the first step. It must be the beginning of a relationship, which will enable the woman to feel supported throughout her pregnancy with benefit to both herself and her baby. It is not an end in itself. There is no such thing as the perfect assessment tool, or even research project, the important thing is to monitor and improve on where we are. One thing which *is* clear is that any tool not integrated into clinic practice will be less effective and more difficult to evaluate.

32.5 Problem Drinking and Liver Damage

As noted earlier, many of the most commonly used screening tools use a measure of tolerance to identify at risk drinking. For a woman with a drinking problem, the measure of tolerance can be misleading. The onset of liver damage may mean that the woman's tolerance for alcohol diminishes. Thus, she may not score high on screening tools which use tolerance as a major measure of harm.

There have been few studies on pregnant women with identified alcohol problems [64]. There is little in the literature of heavy/problem drinking pregnant women about the possible effects of alcohol-related liver damage in the mother and how this may affect the fetus [65, 66]. Indeed, many scholarly articles on liver disease in pregnancy do not even mention alcohol as a possible contributing factor. Even so, a review [67] suggested that the raised levels of acetaldehyde found in women with drinking problems would be an area worth pursuing in relation to alcohol-related fetal harm.

This population may be small, serious liver damage affects fertility and increases the likelihood that the pregnancy will not reach full term [66], but it is highly relevant to the issue. A recent review of trend data from the United States has shown an increase in numbers of pregnant women presenting with alcohol-related liver disease between 2000 and 2010. Perhaps not surprisingly, the women tended to be older and poorer and proportionately they were amongst those costing the healthcare system the most [68]. Rates of liver disease are rising in many countries [69, 70]. Women in their childbearing years are more likely to have alcohol-related liver

disease now than in the past and much of this may be undetected for a number of years. Undiagnosed liver disease in a pregnant woman who continues to drink will have a greater impact on the health of both her and her unborn child.

32.6 Staff Training Issues

The effective implementation of any screening and treatment is dependent on health professionals being trained [71]. It is also dependent on them consistently applying the protocol. If staff do not see the importance of the protocol, if they feel uncomfortable using it or if they only screen when they think there is a chance the pregnant woman is at risk [72], then any screening for prenatal alcohol exposure has a much greater chance of failure [73]. Paraphrasing a well-known saying: "It's not what you ask it's the way that you ask it". The way any sensitive questions are asked and importantly the *attitude* of the heath professional who is asking those questions can make the difference between getting honest accurate answers or answers which are guarded and perhaps misleading. It can also make the difference between the encounter leading to positive health and behaviour steps being taken during pregnancy or not.

> *Practice point*: It is important that staff are trained in more than simply taking a drinking history and other facts about prenatal alcohol exposure. They need to be willing to examine their own attitudes towards women who report drinking. If not, their ability to relate, support and encourage the woman will be seriously affected.

A number of studies have shown that a reported lack of training and fear of stigmatising the woman and her child may be one reason FASD is underdiagnosed by health professionals [74–78]. This issue of stigma, as with many issues in drinking in pregnancy, can lead to confusion for individual patients, and mixed messages in wider public health policy [79, 80].

The judgemental attitudes of some staff may have a knock-on effect on colleagues with less critical attitudes who become protective towards the mother and her unborn child. Unfortunately this may lead to accurate alcohol consumption levels not being recorded in case notes for fear of stigmatisation of the mother and, as importantly, the baby. In some cases, maternal alcohol consumption may not even be included in the baby's case notes, thus decreasing the chance of an early diagnosis of prenatal alcohol exposure. In a 2006 study of paediatricians [81], it was shown that around 12% of paediatricians would not make a diagnosis of FAS in the child for fear of stigmatising the child and their family. Although this could be viewed as showing compassion on the part of the health professional, it also may be related to the fact that giving bad news is difficult [82]. Trying to explain to a woman that something she has done when she was pregnant has caused the harm to her child is

uniquely so. Nevertheless, without a diagnosis the family will not get the support they need.

The reality for many women with drinking problems, pregnant or not, is that no amount of judging by health professionals can match the negative way these women view themselves. Professional must be adequately trained to ensure their attitudes—sympathetic or otherwise—do not compound an already difficult situation.

32.7 Adverse Life Experiences

Many women who become pregnant will report trauma in their lives [49, 83]. Whether that is recent such as intimate partner violence (IPV) [84] or historical childhood physical, emotional or sexual trauma they may well be experiencing aspects of post-traumatic stress disorder (PTSD). Women with drinking problems are even more likely to report such circumstances [85–87]. In relation to IPV, the two occasions a woman is most at risk of serious physical harm in these relationships is if she leaves her abusive partner of if she becomes pregnant [88]. Evidence supports the view that IPV negatively affects pregnancy outcomes (for review, see [89] and [90]). Contrary to what many health professionals believe, women in situations of IPV would like someone to ask them about their history of abuse [91–94].

Many studies have shown a link between pre-pregnancy drinking and adverse life events, these include:

- Stress [95, 96]
- Anxiety [96–98]
- Depression [40, 99]
- Childhood sexual abuse and neglect [100–103]
- Poverty [104, 105]

The complexity of this inter-relationship is borne out by studies [106]. This South African study found women with histories of childhood trauma or more recent experiences of IPV disclosed more drinking in pregnancy. Even those women who had been light drinkers prior to pregnancy recognition drank more heavily during their pregnancies. Heavy drinking in pregnancy is not an equal opportunity women's issue [107].

Practice point: Supporting rather than judging is not only good practice but also compassionate care.

Health professionals must always remember when they are working with a pregnant woman they are working with a dyad, both mother and baby. You cannot work with one without affecting the other. Indeed, if the pregnant woman is using alcohol to self-medicate because of trauma in childhood, the health professional may even

be working with a triad, the pregnant woman, the child she was when she was traumatised and her unborn child. The difficulties of trying to stop problem/heavy drinking at any time can never be underestimated. For a woman in this situation with a traumatic past or present and perhaps an unplanned pregnancy, it may be easier to think of stopping breathing than giving up the only thing she can depend on, the thing that never criticises, shames or blames her—alcohol.

> *Practice point*: You may not feel it is possible to "put yourself in the woman's shoes", but suspend your judgement of her behaviour and your ability to help her and her unborn child will be increased.

There is an increasing trend towards the bio-medicalisation of alcohol research [108], although the biomedical model fits well with FASD, it may also be doing the affected women a disservice. If your goal is to help women, the social psychological and behavioural aspects of drinking in pregnancy are important to understand. No matter how serious someone's drinking problem is they will still report some positive aspects of their consumption—perhaps helping dull the pain of traumatic events or just keeping withdrawal away. Yet pregnant women, indeed, any women who are drinking heavily, are rarely given the opportunity explain these positives. This is bad practice: if the health professional only focuses on the negatives, the woman will begin to feel she is not being heard or acknowledged as an individual, her life, *all* of her life, is not being seen in its entirety. Given the feelings of low self-esteem and the hypersensitivity that comes with trauma she may begin to shut down, may even lie to protect herself. This is never a good place to start to help someone.

> *Practice point:* People use alcohol as self-medication for good reasons. To effectively support them, ALL their reasons for drinking have to be heard. This will enable you to help them identify "at risk" times and places and enable them to reduce or even stop their drinking.

The idea of a more holistic approach to women who have experienced these situations, known as trauma-informed care, is now providing a more positive way of working with women who have lived with trauma [109–111].

32.8 Prevention

Prevention of alcohol-exposed pregnancies can be achieved in two ways each with different policy implications. Firstly and most obviously, by women not drinking when they are pregnant. Secondly, but surprisingly often ignored, the use of effective contraception [112, 113] exploring recent trends in unplanned pregnancies in 2012 reported that of the 213 million pregnancies which were reported 40% of these were defined as unplanned.

Evidence for the US National Survey of Family Growth indicates that in the last two decades there has been little change in how soon women become aware they are pregnant [114, 115]. According to a recent report [116], "18% of women at risk [of unintended pregnancy] who use contraceptives inconsistently or incorrectly account for 41% of all unintended pregnancies". While this is bad news, the good news is that it can actively be changed and therefore offers a real hope of prevention as shown in the recent WHO report [112].

In many countries, more focus and—significantly—money are now being spent on contraception and raising awareness of pregnancy. Countrywide databases such as the Pregnancy Risk Assessment Monitoring System (PRAMS) in the United States can be used to monitor the effectiveness of this approach. If positive results are found, then this presents a strong avenue for future public health policy. Such data could, for example, be used to encourage health authorities to increase funding for family planning or planned parenthood clinics [117].

32.9 Policy

Countries differ in their recognition and understanding of prenatal alcohol exposure. In 2007, O'Leary and her colleagues wrote a review of alcohol policies in pregnancy in seven English-speaking countries noting that even *within* countries different health and governmental bodies give different advice [118, 119]. A number of countries have recently lowered the level of alcohol consumption in pregnancy that is defined as low risk. The United Kingdom is an example of this where the Chief Medical Officers of England, Scotland and Wales [120] brought out a joint statement stating the no drinking in pregnancy is the best choice for the baby. Difference in religious practices and cultures may also have an impact on whether the focus is on policies for prevention or intervention—even which stage of prevention: primary, secondary or tertiary is deemed most relevant.

The issue of alcohol-exposed pregnancies can be viewed under the banner of alcohol-related harm to others and both the World Health Organisation and the European Union are now addressing the issue through this lens [112].

The questions of public health policy making in this field has led to some interesting debates [121–127]. In general, policies designed for pregnant women can be categorised as enabling or punitive [128]. There are strengths and weaknesses in each approach. Examples of positive policies could be ensuring that health clinics, not only antenatal clinics but also clinics for women, have information available on drinking in pregnancy. It has been suggested that even this may push women into choosing a termination because of their anxiety about drinking before they knew they were pregnant, but there is little evidence to support this [129, 130]. However, evidence for the effectiveness of these health campaign leaflets has rarely been shown to be effective. They can change knowledge and attitudes, but rarely lead to changes in behaviour [87].

Ensuring that screening to aid with identification and the option to fast-track women into treatment services if this is required can be very helpful. Pregnancy is

not a time when delays and waiting lists are acceptable. Some policies suggest warning labels on bottles or at the point of sale [131–136].

Other policies are more questionable in their ability to encourage the pregnant woman to acknowledge her drinking, particularly if she is drinking heavily or reporting alcohol-related problems. Examples of how this might manifest in policy include making it a legal requirement that health professionals report a pregnant woman who has been drinking heavily. Perhaps the most extreme examples of a punitive policy would be making it illegal for a woman to drink while pregnant and enabling the state to imprison her for the duration of her pregnancy. As noted earlier, the use of breathalysers may be used to "prove" alcohol consumption in court cases. Few people would agree that imprisoning a pregnant woman is an effective way to support her.

Another example would be removing her children or her baby at birth and placing them in the foster care system. The latter example relates to women with serious alcohol problems and usually a multitude of other complex needs. For these women and their children there are no simple solutions. However, dictating that health professionals become reporting agents will place them in a very difficult situation in terms of their ability to develop a trusting relationship with women who desperately need their help and support. As noted earlier [81], if a diagnosis of FASD is not made, the true prevalence rates of FASD will not be identified and the vitally needed monies and services cannot be put in place to help with impacts far beyond the affected individuals.

32.10 Conclusions

Over the past three decades women's drinking has been increasing in a number of countries. Although some countries have seen a levelling off or decrease in per capita consumption, women's drinking overall is higher than it has been for many years [137]. While progress has been made, there remains a pressing need for governments to work with health organisations and to provide funding in the areas of contraception and prevention, screening and staff training for pregnant women who drink, particularly pregnant women with alcohol-related problems and complex needs. Attitudes towards the women themselves make this harder. While there is some genuine concern about women's drinking in pregnancy many of the attitudes: sometimes cultural, sometimes religious, sometimes even driven by the media, are judgemental and condemning. Society plays a role in helping or hindering how a pregnant woman who drinks is treated and how she perceives herself [138, 139]. This in turn affects her baby.

The attitudes of the general public have an impact on midwives and other health professionals. The clearer a country's government health department are on no drinking being the safest option in pregnancy, the more confident health professionals will be in consistently asking more effective questions on consumption and then providing clear messages.

In many countries, the present situation is a vicious circle—evidence is (sometimes) based on research, money for research is (sometimes) based on the need to provide evidence and policy decisions are (sometimes) based on this evidence. Looking from this standpoint, it is easy to see how if research is needs driven, and a need is not identified, then the eventual outcome is that policies will not be put in place.

The reality is we still have a long way to go in convincing many women that abstinence is the best course for themselves or their unborn children [140, 141].

For all women who become pregnant, encouraging them to stop drinking is agreed to be the best message. Even so, as noted earlier, the reasons why women drink or drink heavily are many and varied. If women who are drinking only come into focus when they are pregnant and only *in relation to* that pregnancy, then we are seeing women purely as vehicles. Clinically this is ineffective; ethically it is unacceptable.

References

1. Petersen I, McCrea RL, Lupattelli A, Nordeng H. Women's perception of risks of adverse fetal pregnancy outcomes: a large-scale multinational survey. BMJ Open. 2015;5:6.
2. Popova S, Lange S, Probst C, Gmel G, Rhem J. Estimation of national, regional, and global prevalence of alcohol use during pregnancy and fetal alcohol syndrome: a systematic review and meta-analysis. Lancet Glob Health. 2017;5(3):e290–9.
3. Cannon MJ, Guo J, Denny CH, Green PP, Miracle H, Sniezek JE, et al. Prevalence and characteristics of women at risk for an alcohol-exposed pregnancy (AEP) in the United States: estimates from the National Survey of family growth. Matern Child Health J. 2015;19(4):776–82.
4. Kitsantas P, Gaffney KF, Wu H. Identifying high-risk subgroups for alcohol consumption among younger and older pregnant women. J Perinat Med. 2015;43(1):43–52.
5. Cooper DL, Petherick ES, Wright J. The association between binge drinking and birth outcomes: results from the born in Bradford cohort study. J Epidemiol Community Health. 2013;67(10):821–8.
6. Stahre M, Naimi T, Brewer R, Holt J. Measuring average alcohol consumption: the impact of including binge drinks in quantity–frequency calculations. Addiction. 2006;101:1711–8.
7. Herring R, Berridge V, Thom B. Binge drinking: an exploration of a confused concept. J Epidemiol Community Health. 2008;62:476–9.
8. Gmel G, Kuntsche E, Rehm J. Risky single-occasion drinking: bingeing is not bingeing. Addiction. 2010;106:1037–45.
9. Muggli E, O'Leary C, Donath S, Orsini F, Forster D, Anderson P, et al. Did you ever drink more? A detailed description of pregnant women's drinking patterns. BMC Public Health. 2016;16:683–96.
10. Tan CH, Denny CH, Cheal NE, Sniezek JE, Kanny D. Alcohol use and binge drinking among women of childbearing age—United States, 2011–2013. MMWR. 2015;64:1042–6.
11. Strandberg-Larsen K, Nielsen NR, Andersen A-MN, Olsen J, Gronbaek M. Characteristics of women who binge drink before and after they become aware of their pregnancy. Eur J Epidemiol. 2008;23:565–72.
12. Ethen MK, Ramadhani TA, Scheuerle AE, et al. Alcohol consumption by women before and during pregnancy. Matern Child Health J. 2009;13:274.
13. Skagerström J, Chang G, Nilsen P. Predictors of drinking during pregnancy: a systematic review. J Womens Health. 2011;20(6):901–13.

14. Mallard SR, Connor JL, Houghton LA. Maternal factors associated with heavy periconceptional alcohol intake and drinking following pregnancy recognition: a post-partum survey of New Zealand women. Drug Alcohol Rev. 2013;32(4):389–97.
15. Symon A, Rankin J, Butcher G, Smith L, Cochrane L. Evaluation of a retrospective diary for peri-conceptual and mid-pregnancy drinking in Scotland: a cross-sectional study. Acta Obstet Gynecol Scand. 2017;96:53–60.
16. Mullally A, Cleary B, Barry J, Fahey TP, Murphy DJ. Prevalence, predictors and perinatal outcomes of peri-conceptional alcohol exposure—retrospective cohort study in an urban obstetric population in Ireland. BMC Pregnancy Childbirth. 2011;11:27.
17. Tran NT, Najman JM, Hayatbakhsh R. Predictors of maternal drinking trajectories before and after pregnancy: evidence from a longitudinal study. Aust N Z J Obstet Gynaecol. 2015;55(2):123–30.
18. Magnusson A, Goransson M, Heilig M. Hazardous alcohol users during pregnancy: psychiatric health and personality traits. Drug Alcohol Depend. 2007;89(2–3):275–81.
19. Ystrom E, Vollrath ME, Nordeng H. Effects of personality on use of medications, alcohol, and cigarettes during pregnancy. Eur J Clin Pharmacol. 2012;68:845–51.
20. Beijers C, Burger H, Verbeek T, Bockting CLH, Ormel J. Continued smoking and continued alcohol consumption during early pregnancy distinctively associated with personality. Addict Behav. 2014;39(5):980–6.
21. Parackal SM, Parackal MK, Harraway JA. Prevalence and correlates of drinking in early pregnancy among women who stopped drinking on pregnancy recognition. Matern Child Health J. 2013;17(3):520–9.
22. Meschke LL, Holl J, Messelt S. Older not wiser: risk of prenatal alcohol use by maternal age. Matern Child Health J. 2013;17(1):147–55.
23. Hellmuth JC, Gordon KC, Stuart GL, Moore TM. Risk factors for intimate partner violence during pregnancy and postpartum. Arch Womens Ment Health. 2013;16(1):19–27.
24. Ceccanti M, Fiorentino D, Coriale G, Kalberg WO, Buckley D, Hoyme HE, et al. Maternal risk factors for fetal alcohol spectrum disorders in a province in Italy. Drug Alcohol Depend. 2014;145:201–8.
25. Powers JR, McDermott LJ, Loxton DJ, Chojenta CL. A prospective study of prevalence and predictors of concurrent alcohol and tobacco use during pregnancy. Matern Child Health J. 2013;17(1):76–84.
26. Lemmens PH. The alcohol content of self-report and 'standard' drinks. Addiction. 1994;89:593–601.
27. Kerr WC, Greenfield TK, Tujague J, Brown SE. A drink is a drink? Variation in the amount of alcohol contained in beer, wine and spirits drinks in a US methodological sample. Alcohol Clin Exp Res. 2005;29(11):2015–21.
28. Kerr WC, Stockwell T. Understanding standard drinks and drinking guidelines. Drug Alcohol Rev. 2012;31(2):200–5.
29. Mukherjee R, Wray E, Curfs L, Hollins S. Estimation of alcohol content of wine, beer and spirits to evaluate exposure risk in pregnancy: pilot study using a questionnaire and pouring task in England. International Journal of Alcohol and Drug Research. 2013;2:71–8.
30. "Wikipedia Standard Drink". https://en.m.wikipedia.org/wiki/standarddrinks.
31. Ewing JA. Detecting alcoholism. The CAGE questionnaire. JAMA. 1984;252(14):1905–7.
32. Sobell L, Sobell M. Timeline follow back: a technique for assessing self-reported alcohol consumption. In: Litten R, Allen J, editors. Measuring alcohol consumption. Totowa, NJ: Humana Press; 1992. p. 41–72.
33. Sokol R, Martier SS, Ager JW. The T-ACE Questions: practical prenatal detection of risk-drinking. Am J Obstet Gynecol. 1989;160:863–70.
34. Russell M. New assessment tools for risk drinking during pregnancy. Alcohol Health Res World. 1994;18:55–61.
35. Russell M, Martier S, Sokol RJ, Mudar P, Jacobson S, Jacobson J. Detecting risk drinking during pregnancy: a comparison of four screening questionnaires. Am J Public Health. 1996;86(10):1435–9.

36. Chiodo LM, Delaney-Black V, Sokol RJ, Janisse J, Pardo Y, Hannigan JH. Increased cut-point of the TACER-3 screen reduces false positives without losing sensitivity in predicting risk alcohol drinking in pregnancy. Alcohol Clin Exp Res. 2014;38(5):1401–8.
37. Saunders JB, Aasland OG, Babor T, De La Fuente JR, Grant M. Development of the alcohol use disorders identification test (AUDIT). In: WHO Collaborative Project on early detection of persons with harmful alcohol consumption-II, vol. 88; 1993. p. 791–804.
38. Gual A, Segura L, Contel M, Heather N, Colom J. AUDIT-3 and AUDIT-4: effectiveness of two short forms of the alcohol use disorders identification test. Alcohol Alcohol. 2002;37(6):591–6.
39. Savage C, Wray J, Ritchey PN, Sommers M, Dyehouse J, Fulmer M. Current screening instruments related to alcohol consumption in pregnancy and a proposed alternative method. J Obstet Gynecol Neonatal Nurs. 2003;32(4):437–46.
40. Hicks M, Tough S, Johnston D, Siever J, Clarke M, Sauve R, et al. T-ACE and predictors of self-reported alcohol use during pregnancy in a large, population-based urban cohort. Int J Alcohol Drug Res. 2014;3(1):51–61.
41. Smith L, Savory J, Couves J, Burns E. Alcohol consumption during pregnancy: cross-sectional survey. Midwifery. 2014;30(12):1173–8.
42. Doi L, Cheyne H, Jepson R. Alcohol brief interventions in Scottish antenatal care: a qualitative study of midwives' attitudes and practices. BMC Pregnancy Childbirth. 2014;14(1):1–27.
43. Hodgson R, Alwyn T, John B, Thom B, Smith A. The FAST Alcohol screening test. Alcohol Alcohol. 2002;37(1):61–6.
44. Chasnoff IJ, Wells AM, McGourty RF, Bailey LK. Validation of the 4P's plus screen for substance use in pregnancy validation of the 4P's plus. J Perinatol. 2007;27:744–8.
45. Göransson M, Magnusson Å, Bergman H, Rydberg U, Heilig M. Fetus at risk: prevalence of alcohol consumption during pregnancy estimated with a simple screening method in Swedish antenatal clinics. Addiction. 2003;98:1513–20.
46. Chang G. Alcohol-screening instruments for pregnant women. National Institute on Alcohol Abuse and Alcoholism. Alcohol Res Health. 2001;25:204–9.
47. Dawson DA, Das A, Faden VB, Bhaskar B, Krulewitch KJ, Wesley B, et al. Screening for high and moderate risk drinking during pregnancy: a comparison of several TWEAK based screeners. Alcohol Clin Exp Res. 2001;25(9):1342–9.
48. Kesmodel U, Olsen SF. Self-reported alcohol intake in pregnancy: comparison between four methods. J Epidemiol Community Health. 2001;55(10):738–45.
49. Parkes T, Poole N, Salmon A, Greaves L, Urquhart C. Double exposure: a better practices review on alcohol interventions during pregnancy. Vancouver, BC: British Columbia Centre of Excellence for Women's Health; 2008.
50. Burns E, Gray R, Smith LA. Brief screening questionnaires to identify problem drinking during pregnancy: a systematic review. Addiction. 2010;105(4):601–14.
51. Strandberg-Larsen K, Andersen A-MN, Olsen J, Nielsen NR, Grønbæk M. Do women give the same information on binge drinking during pregnancy when asked repeatedly? Eur J Clin Nutr. 2006;60(11):1294–8.
52. United States Dept of Health and Human Services. Guidelines for the identification and management of substance use and substance use disorders in pregnancy. Rockville, MD: Substance Abuse and Mental Health Services Administration; 2014.
53. World Health Organisation. Guidelines for the identification and management of substance use and substance use disorders in pregnancy. Geneva: World Health Organization; 2014.
54. Wong S, Ordean A, Kahan M, et al. Substance use in pregnancy: Society of Obstetricians and Gynaecologists Canada Clinical Practice Guidelines. J Obstet Gynaecol Can. 2011;33(4):367–84.
55. Substance Abuse and Mental Health Services Administration. Treatment improvement protocol pregnant, substance-using women: treatment improvement protocol (TIP) series 2. HHS publication 13–4801. Rockville, MD: Substance Abuse and Mental Health Services Administration; 1995.

56. United States Department of Health and Human Services. Substance Abuse and Mental Health Services Administration. White Paper on Screening, Brief Intervention and Referral to Treatment (SBIRT) in Behavioral Healthcare. Rockville, MD: Substance Abuse and Mental Health Services Administration; 2011.
57. Kennedy C, Finelstein N, Hutchins E, Mahoney J. Improving screening for alcohol use during pregnancy. Matern Child Health J. 2004;8(3):137–47.
58. O'Donnell A, Anderson P, Newbury-Birch D, Schulte B, Schmidt C, Reimer J, Kaner E. The impact of brief alcohol interventions in primary healthcare: a systematic review of reviews. Alcohol Alcohol. 2014;49(1):66–78.
59. O'Brien P. Performance measurement: a proposal to increase use of SBIRT and decrease alcohol consumption during pregnancy. Matern Child Health J. 2014;18(1):1–9.
60. Moyer A, Finney JW, Swearingen CE, Vergun P. Brief interventions for alcohol problems: a meta-analytical review of controlled investigations in treatment-seeking and non-treatment-seeking populations. Addiction. 2002;97:279–92.
61. Bakhireva LN, Savage DD. Focus on biomarkers of fetal alcohol exposure and fetal alcohol effects. Alcohol Res Health. 2011;34(1):56–63.
62. Yang JY, Kwak HS, Han JY, Choi J-S, Ahnn H-K, Oh YJ, et al. Could a first-trimester blood phosphatidylethanol concentration ≥4 nM be useful to identify women with moderate to-heavy prenatal alcohol exposure who are at high risk of adverse pregnancy outcomes? Med Hypotheses. 2015;85:965–8.
63. Howlett H, Abernethy S, Brown NW, Rankin J, Gray WK. How strong is the evidence for using blood biomarkers alone to screen for alcohol consumption during pregnancy? A systematic review. Eur J Obstet Gynecol Reprod Biol. 2017;213:45–52.
64. Halmesmaki E, Roine R, Salaspuro M. Gamma-glutamyltransferase, aspartate and alanine aminotransferases and their ratio, mean cell volume and urinary dolichol in pregnant alcohol abusers. Br J Obstet Gynaecol. 1992;99:287–91.
65. Esposito SD. Pregnancy in patients with advanced chronic liver disease. Clin Liver Dis. 2014;3:62–8.
66. Italian Association for the Study of the Liver. AISF position paper on liver disease and pregnancy. Dig Liver Dis. 2016;48:120–37.
67. Hard M, Einarson T, Koren G. The role of acetaldehyde in pregnancy outcome after prenatal alcohol exposure. Ther Drug Monit. 2001;23(4):427–34.
68. Ellington SR, Flowers L, Legardy-Williams JK, Jamieson DJ, Kourtis AP, et al. Recent trends in hepatic diseases during pregnancy in the United States, 2002–2010. Am J Obstet Gynecol. 2015;212:524.e1–7.
69. Wellings K, Jones KG, Mercer CH, Tanton C, Clifton S, Datta J, et al. The prevalence of unplanned pregnancy and associated factors in Britain: findings from the third National Survey of sexual attitudes and lifestyles (NATSAL-3). Lancet. 2013;382(9907):1807–16.
70. Sheron N. Alcohol and liver disease in Europe—simple measures have the potential to prevent tens of thousands of premature deaths. J Hepatol. 2016;64:957–67.
71. Zoorob RJ, Durkin KM, Gonzalez SJ, Adams S. Training nurses and nursing students about prevention, diagnoses, and treatment of fetal alcohol spectrum disorders. Nurse Educ Pract. 2014;14(4):338–44.
72. Loxton D, Chojenta C, Anderson AE, Powers JR, Shakeshaft A, Burns L. Acquisition and utilization of information about alcohol use in pregnancy among Australian pregnant women and service providers. J Midwifery Womens Health. 2013;58(5):523–30.
73. van der Wulp NY, Hoving C, de Vries H. Dutch midwives' experiences with implementing health counselling to prevent prenatal alcohol use. J Clin Nurs. 2014;23(21–22):3286–9.
74. Room R. Stigma, social inequality and alcohol and drug use. Drug Alcohol Rev. 2005;24:143–55.
75. Broom D. Hazardous good intentions? Unintended consequences of the project of prevention. Health Sociol Rev. 2008;17(2):129–40.
76. Stengel C. The risk of being 'too honest': drug use, stigma and pregnancy. Health Risk Soc. 2014;16:1. Risk, Pregnancy and Childbirth.

77. Bell E, Andrew G, Di Pietro N, Chudley AE, Reynolds JN, Racine E. It's a shame! Stigma against fetal alcohol spectrum disorder: examining the ethical implications for public health practices and policies. Public Health Ethics. 2015:1–13.
78. Laslett A-M, Catalan P, Chikritzhs Y, Dale C, Doran C, Ferris J, et al. The range and magnitude of Alcohol's harm to others. Fitzroy, Victoria AER Centre for Alcohol Policy Research, Turning Point Alcohol and Drug Centre, Eastern Health; 2010.
79. Eggertson L. Stigma a major barrier to treatment for pregnant women with addictions. Can Med Assoc J. 2013;185:1562.
80. Eguiagaray I, Scholz B, Giorgi C. Sympathy, shame, and few solutions news media portrayals of fetal alcohol spectrum disorders. Midwifery. 2016;40:49–54.
81. Elliot EJ, Payne J, Bower C. Diagnosis of foetal alcohol syndrome and alcohol use in pregnancy: a survey of paediatricians' knowledge, attitudes and practice. J Paediatr Child Health. 2006;1(42):698–703.
82. Gill I, Sharif F. Out of sight, out of mind? A national survey of paediatricians in Ireland regarding Fetal Alcohol Spectrum Disorders. Ir Med J. 2017;110(3):528.
83. Frankenberger DJ, Clements-Nolle K, Yang W. The association between adverse childhood experiences and drinking during pregnancy in a population-based sample of women. Womens Health Issues. 2015;25(6):688–95.
84. Chang JC, Gordon K, Stuart GL, Moore TM. Risk factors for intimate partner violence during pregnancy postpartum. Arch Women Mental Health. 2013;16:19–27.
85. Krug EG. World report on violence and health. Geneva: World Health Organization; 2002.
86. Campbell JC. Abuse during pregnancy: A quintessential threat to maternal and child ... so when do we start to act? Can Med Assoc J. 2001;164(11):1578–9.
87. Plant ML. The role of alcohol in women's lives: a review of issues and responses. J Subst Abuse. 2008;13(3):155–91.
88. McFarlane J, Campbell JC, Sharps P, Watson K. Abuse during pregnancy and femicide: urgent implications for women's health. Obstet Gynecol. 2002;100(1):27–36.
89. Taillieu TL, Brownridge DA. Violence against pregnant women: prevalence, patterns, risk factors, theories, and directions for future research. Aggress Violent Behav. 2010;15:14–35.
90. Murphy CC, Schei B, Myhr TL, Du Mont J. Abuse: a risk factor for low birth weight? A systematic review and meta-analysis. Can Med Assoc J. 2001;164(11):1567–72.
91. Feder G, Hutson M, Ramsay J, Taket AR. Women exposed to intimate partner violence: expectations and experiences when they encounter health care professionals: a meta-analysis of qualitative studies. Arch Intern Med. 2006;166(1):22–37.
92. Bacchus L, Mezey G, Bewley S, Haworth A. Prevalence of domestic violence when midwives routinely enquire in pregnancy. BJOG. 2004;111(5):441–5.
93. Bacchus L, Mezey G, Bewley S. Domestic violence: prevalence in pregnant women and associations with physical and psychological health. Eur J Obstet Gynecol. 2004;113(1):6–11.
94. Lutz KF. Abused pregnant women's interactions with health care providers during the childbearing year. J Obstet Gynecol Neonatal Nurs. 2005;34(2):151–62.
95. Austin MP, Leader L. Maternal stress and obstetric and infant outcomes: epidemiological findings and neuroendocrine mechanisms. Aust N Z J Psychiatry. 2000;40(3):331–7.
96. Agrati D, Browne D, Jonas W, Meaney M, Atkinson L, Steiner M, et al. Maternal anxiety from pregnancy to 2 years postpartum: transactional patterns of maternal early adversity and child temperament. Arch Womens Ment Health. 2015;18(5):693–705.
97. Buist A, Gotman N, Yonkers KA. Generalized anxiety disorder: course and risk factors in pregnancy. J Affect Disord. 2011;131(1):277–83.
98. Arch JJ. Pregnancy-specific anxiety: which women are highest and what are the alcohol-related risks? Compr Psychiatry. 2013;54(3):217–28.
99. Szegda K, Markenson G, Bertone-Johnson ER, Chasan-Taber L. Depression during pregnancy: a risk factor for adverse neonatal outcomes? A critical review of the literature. J Matern Fetal Neonatal Med. 2014;27(9):960–7.

100. Dong M, Anda RF, Felitti VJ, Dube SR, Williamson DF, Thompson TJ, et al. The interrelatedness of multiple forms of childhood abuse, neglect, and household dysfunction. Child Abuse Negl. 2004;28(7):771–84.
101. Hillis SD, Anda RF, Dube SR, Felitti VJ, Marchbanks PA, Marks JS. The association between adverse childhood experiences and adolescent pregnancy, long-term psychosocial consequences, and fetal death. Pediatrics. 2004;113(2):320–7.
102. Plant ML, Miller P, Plant MA. Childhood and adult sexual abuse: relationships with alcohol and other drug use. Child Abuse Rev. 2004;13:200–14.
103. LoGiudice JA. A systematic literature review of the childbearing cycle as experienced by survivors of sexual abuse. Nurs Womens Health. 2012;20(6):582–94.
104. Larson CP. Poverty during pregnancy: its effects on child health outcomes. Paediatr Child Health. 2007;12(8):673–7.
105. Holmila M, Raitasalo K. Mothers who abuse alcohol and drugs: health and social harms among substance-abusing mothers of small children. Nordic Stud Alcohol Drugs. 2013;30(5):361–73.
106. Choi KW, Abler LA, Watt MH, Eaton LA, Kalichman SC, Skinner D, et al. Drinking before and after pregnancy recognition among South African women: the moderating role of traumatic experiences. BMC Pregnancy Childbirth. 2014;14:97.
107. Abel E. An update on incidence of FAS: FAS is not an equal opportunity birth defect. Neurotoxicol Teratol. 1995;17(4):437–43.
108. Midanik L. Biomedicalization of alcohol studies: ideological shifts and institutional challenges. Piscataway: Transaction; 2006.
109. Poole N, Isaac B. Apprehensions: barriers to treatment for substance-using mothers. Vancouver, BC: British Columbia Centre of Excellence for Women's Health; 2001.
110. Poole N, Greaves L. Becoming trauma informed. Toronto: Centre for Addiction and Mental Health; 2012.
111. Substance Abuse and Mental Health Services Administration. Trauma-informed Care in Behavioral Health Services. Treatment improvement protocol (TIP) series 57. HHS publication 13–4801. Rockville, MD: Substance Abuse and Mental Health Services Administration; 2014.
112. Schölin L. Prevention of harm caused by alcohol exposure in pregnancy: rapid review and case studies from member states. Copenhagen: World Health Organisation Regional Office for Europe; 2016.
113. Sedgh G, Singh S, Hussain R. Intended and unintended pregnancies worldwide in 2012 and recent trends. Stud Fam Plann. 2014;45(3):301–14.
114. Branum AM, Ahrens KA. Trends in timing of pregnancy awareness among us women. Matern Child Health J. 2017;21(4):715–26.
115. Adejoke AB. Late recognition of unintended pregnancies. Public Health Nurs. 2015;32(5):462–70.
116. Guttmacher Institute. 2016. https://www.guttmacher.org/fact-sheet/unintended-pregnancy-united-states.
117. Melvin CL, Rogers M, Gilbert BC, et al. Pregnancy intention: how PRAMS data can inform programs and policy. Matern Child Health J. 2000;4:197–201.
118. O'Leary CM, Heuzenroeder L, Elliott EJ, Bower C. A review of policies on alcohol use during pregnancy in Australia and other English-speaking countries, 2006. Med J Aust. 2007;186(9):466–71.
119. Drabble LA, Poole N, Magri R, Tumwesigye NM, Li Q, Plant M. Conceiving risk, divergent responses: perspectives on the construction of risk of FASD in six countries. Subst Use Misuse. 2011;46(8):943–58.
120. UK Chief Medical Officers. Low risk drinking guidelines. London: Crown Copyright; 2016.
121. Armstrong EM, Abel EL. Fetal alcohol syndrome: the origins of a moral panic. Alcohol Alcohol. 2000;35(3):276–82.
122. Warren KR, Hewitt BG. Fetal alcohol spectrum disorders: when science, medicine, public policy, and Laws collide. Dev Disabil Res Rev. 2009;15:170–5.

123. Gavaghan C. "You can't handle the truth"; medical paternalism and prenatal alcohol use. J Med Ethics. 2009;35:300–3.
124. Potter DA. Drawing the line at drinking for two: governmentality, biopolitics, and risk in state legislation on fetal alcohol spectrum disorders. Adv Med Sociol. 2012;14:129–53.
125. Leppo A. The emergence of the foetus: discourses on foetal alcohol syndrome prevention and compulsory treatment in Finland. Crit Public Health. 2012;22:179–91.
126. Racine E, Bell E, Zizzo N, Green C. Public discourse on the biology of alcohol addiction: implications for stigma, self-control, essentialism, and coercive policies in pregnancy. Neuroethics. 2015;8(2):177–86.
127. Crawford-Williams F, Fielder A, Mikocka-Walus A, Esterman A. A critical review of public health interventions aimed at reducing alcohol consumption and/or increasing knowledge among pregnant women. Drug Alcohol Rev. 2015;34(2):154–61.
128. Drabble L, Thomas S, O'Connor L, Roberts SCM. State responses to alcohol use and pregnancy: findings from the alcohol policy information system (APIS). J Soc Work Pract Addict. 2014;14(2):191–206.
129. Roberts LW, Dunn LB. Ethical considerations in caring for women with substance use disorders. Obstet Gynecol Clin North Am. 2003;30:559–82.
130. Roberts S, Lyndsay A, Sinkford D, et al. Alcohol, tobacco, and drug use as reasons for abortion. Alcohol Alcohol. 2012;47:640–8.
131. Kaskutas L, Greenfield TK. First effects of warning labels on alcoholic beverage containers. Drug Alcohol Depend. 1992;31(1):1–14.
132. Kaskutas LA. Interpretations of risk: the use of scientific information in the development of the alcohol warning label policy. Subst Use Misuse. 1995;30:1519–48.
133. Greenfield T, Graves K, Kaskutas L. Long-term effects of alcohol warning labels: findings from a comparison of the United States and Ontario, Canada. Psychol Mark. 1999;16:261–82.
134. Parackal S, Parackal M, Harraway J. Warning labels on alcohol containers as a source of information on alcohol consumption in pregnancy among New Zealand women. Int J Drug Policy. 2010;21:302–5.
135. Martin-Moreno JM, Harris M, Breda J, Møller L, Alfonso-Sanchez JL, Gorgojo L. Enhanced labelling on alcoholic drinks: reviewing the evidence to guide alcohol policy. Eur J Public Health. 2013;23(6):1082–7.
136. Bell E, Zizzo N, Racine E. Caution: warning labels about alcohol and pregnancy. Am J Bioeth. 2015;15:3.
137. Wilsnack SC. The GENACIS Project: a review of findings and some implications for global needs in women-focused substance abuse prevention and intervention. Subst Abuse Rehabil. 2012;3(Suppl I):5–15.
138. Jacobs L, Jacobs J. 'Bad' mothers have alcohol use disorder: moral panic or brief intervention? Gender Behav. 2014;123:5971–9.
139. Cherry AL. Shifting our focus from retribution to social justice: an alternative vision for the treatment of pregnant women who harm their fetuses. J Law Health. 2015;28(1):5–61.
140. Powers JR, Loxton DJ, Burns LA, Shakeshaft A, Elliott EJ, Dunlop AJ. Assessing pregnant women's compliance with different alcohol guidelines: an 11-year prospective study. Med J Aust. 2010;192:690–69.
141. Stockwell T, Room R. Constructing and responding to low-risk drinking guidelines: conceptualisation, evidence and reception. Drug Alcohol Rev. 2012;31:121–5.

Supporting Women and Their Families: A Mother's Perspective

33

Phillipa Williams and Samantha Marchant

Chapter Highlights

- A birth mother's perspective of FASD.
- Difficulties and areas to support getting help.
- Case studies and personal reflections.

In this chapter, we are going to look at why women may continue to drink, and three case histories of women whose children have been diagnosed or still seeking diagnosis. Reflecting on what happened and recommending what could have been done differently.

What we do know is that drinking in pregnancy is increasing and more babies are being born with FAS and FASD. There is growing evidence of high rates of FASD in both North America and Western European, with some estimates being as high as 2–5% of the population.[1]

There have been many public health campaigns and you would think they would make a difference, but US researchers have found despite all the millions spent on public health awareness campaigns for FASD, they have not worked. Women continue to drink despite all the science and warnings. Research shows pregnant UK and Irish women have some of the highest rates of drinking in the world.

So why are women continuing to drink during pregnancy? Recent studies claim that more women are drinking during pregnancy, so why would they be given all the guidelines and information?

Let us take a look at six of the reasons why women continue to drink [1–3]:

[1] Prevalence and characteristics of fetal alcohol spectrum disorders.

This chapter is dedicated to the memory of Pip Williams who unfortunately died prior to publication of this book, but who did so much to draw attention to FASD and support other birth mothers.

P. Williams (Deceased) · S. Marchant (✉)
London, UK

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_33

1. Women are unaware they are pregnant.
2. Women are unaware of the extent of damage alcohol can cause to the fetus.
3. Women underestimate the harms alcohol consumption can cause because they know other women who drank during pregnancy and their children appear healthy.
4. Alcohol use is the norm in their social group and abstaining may therefore be difficult.
5. Women may be using alcohol to cope with difficult life situations such as violence, depression, poverty or isolation.
6. Women may struggle with alcohol addiction.

For the latter two reasons, there are more challenges, and women are scared to disclose. The current system is not set up or encouraging for women to come forward to discuss their drinking or any other issues they have during pregnancy.

Barriers to seeking support and accessing treatment may be:

- Shame and guilt.
- Fear of child welfare involvement and/or having a child removed from their care.
- Feelings of depression and low self-esteem.
- Belief or hope that they can change without help.
- Unsupportive or controlling partner.
- Not having enough information about available services.

There could be many reasons a woman may drink when she is pregnant, but evidence shows that generally she does not drink alcohol because she wants to harm her baby.

Let us look at three women's experiences of being pregnant; delivery and support and seeking a diagnosis. (The Names are not the real names.)

33.1 Pregnancy: *Joanna*

This is my experience and my truth and I hope that it will enlighten and be of benefit to you.

I drank alcoholically during the most part of my first pregnancy, but by the grace of God, I have been sober many years since.

I found out I was pregnant at nearly 4 months gone right in the throes of my alcoholism. Finding out I was pregnant actually accelerated my drinking, I was so full of fear and in such mental and emotional pain that the only solutions I felt I had was to drink myself into oblivion or commit suicide. At this time in my life, I couldn't help myself, I felt so ashamed and alone. I desperately needed support but I hid the majority of my drinking from those who loved me and from those I wanted to please.

At the time of finding out I was pregnant I was living away from home, I was a student mental health nurse in a high security hospital and I didn't tell anyone I was

pregnant. When they did eventually find out they were not supportive at all and I don't blame them really; however, I really needed their understanding but I was stigmatized and dealt with harshly.

Thankfully, the OT staff from my university was amazing. They were really loving and supportive and did not judge me. I went in and talked to them regularly. They were professional and caring and they helped me to get out of the violent relationship I was in with the father of my unborn baby. They made a real difference to me holistically and I am very grateful to them.

It was about 5 months when I started going for scans and my baby was small so we had to be scanned often, I did tell the nursing staff that I was an alcoholic and I was still drinking, although through fear and shame I told them I was drinking half of what I actually was, but still confessing that I was drinking more than any recommended or 'safe' amount for myself, let alone for my unborn baby.

This was really an unheard cry for help. I was told social services would be involved. This again filled me with yet more fear and mistrust in the service I was receiving. If my baby was taken from me, I would not have wanted to live. All across my maternity notes is written Iong-term history of depression and alcoholism. I used to go in with tears in my eyes but was received in a clinical and judgemental manner.

The evidence that myself and baby were high risk was all in my notes. I needed someone to come beside me and work out a positive package of care. I believe the best solution for me would have been to be offered a rehab. I didn't think of this solution at the time myself, but I wish someone had suggested it. I was referred to the local drug and alcohol clinic where I was ludicrously asked to fill out a drinks diary.

33.1.1 Recommendations

- Provide her with the correct information and discuss about drinking in pregnancy so she can make an informed choice.
- Inform her that in order for you to provide the best care possible for her child; you must know all the facts about the pregnancy and possible exposures.
- When talking with a mother reinforce that, they are obviously a good mother and that there is a lot of confusion about how much alcohol is safe to drink while pregnant (or before they knew, they were pregnant). Make an assumption that they are a good person. (KM).
- If you believe, a woman needs help to stop drinking/using drugs, please take the time to suggest or refer her to an addiction professional or treatment centre.
- Alcohol dependency is not a choice. Women may be using alcohol for many reasons such as coping strategies. Be supportive and compassionate.
- There are many local alcohol services and community projects where the women would not need to go into residential treatment and detox. Think about wrap around care from her local community without having to greatly disrupt hers or children's routines.

- Contact ELEN or your local women's centre who may be able to support and provide peer support for the women.

33.2 Perinatal, Postnatal and Social Services: *Harriet*

33.2.1 First Trimester

I found out I was pregnant at 12 weeks. One of the female GP's at the surgery asked me if I drank alcohol, nervously I said 'yes'. The response came in a very disapproving stern tone 'you have to stop drinking now' but was given no reason why.

Although I came clean to the GP that I was 3 months pregnant and had been drinking I do not recall being asked how much I drank and was never advised of the potential harm I'd already put the baby.

My scans revealed that my baby was very small for her gestational period, had a small head, was breach and had heart irregularities.

33.2.2 Second Trimester

All possible tests including an amniocentesis were carried out. No causation was identified but I was advised, on two occasions, that I could abort. In addition I was advised that the baby would require a lot of input from the medical teams and would be "a lot of 'work. "Yet, no mention of fetal alcohol syndrome and why my baby would be a lot of work!

33.2.3 Third Trimester

A date for the elective Caesarean was agreed with the midwife in the community. All through the pregnancy I was trying to cut down and stop the drinking. Being inexperienced with alcohol rehabilitation I failed every time and the more I tried the worse I became. Eventually my body became dependent.

One of the things I tried in order to cut down the alcohol intake so I changed to Cider. I read the bottle and thought it was safer as it had less alcohol content. It actually was worse than wine.

I then tried to go cold Turkey in late December which unknowingly, was dangerous.

Lucky for me my mother was in town to support me. When I suffered withdrawal symptoms and had fits I was admitted into hospital.

33.2.4 In Hospital

When I regained consciousness the Doctor who was doing rounds in the maternity ward told me my baby was going to die….I was devastated and still there was no counselling or explanation offered.

During the time I was in maternity ward I recall a nurse sarcastically commenting on how 'rough' I looked and how my belly was small for a woman due in 2 weeks. It hurt but I endured. Still I did not understand what was happening to my body.

In the days following a surgeon came to discuss dates for the elective and what to expect. It was only then that I was told, in a professional manner, that my daughter may be still born due to the fits I suffered that were caused by alcohol withdrawal. I have no recollection of being told the impact the alcohol may have had on the unborn baby.

After the caesarean, my daughter was taken to NICU. I only saw her for the first time on day 3. All I recall being told was that they were waiting for DNA tests but she cried a lot.

The social worker came a few days later and told me they would take my daughter away due to the harm I had caused and potential harm I could cause. I told her if they took my daughter they would have to take me. At that point I was sectioned under the mental health act.

When I was discharged, I continued to visit my daughter daily. During the same time I had to find a lawyer and prepare for court. It was only at court that I fully understood the impact of alcohol on my daughter and the term FAS. I broke down and all the social worker could do was to keep asking if I wanted to discuss a way forward with the case. I was devastated.

I attended a place called CRI alcohol and drug service which supports people with substance misuse. I was referred by a lady who had come into hospital to talk to me about my drinking and how it affected me. She proceeded to tell me how love her life was, that she was getting paid less money than she had been but was happy and was able to go on holidays.

At the 6-week review with my GP, I told her that I was feeling very low. She referred me to the hospital counselling team and they could offer me nothing as I was not severe enough. I should go to IAPT in the Community,

IAPT was slow to respond. When they responded, they said I needed to be at least 6 months alcohol free and they could not help.

33.2.5 Support

*On my own steam I found out about services in the community who could help me with my own mental health issues. In addition, I did research on my own and found the UK & European Birth Mother network—FASD whom I contacted. They then fully explained to me, on the phone, the effects of alcohol on my baby and what to expect as she g*rows up. *They have been by my side since that day.*

33.2.6 Recommendations

- Staff must be non-judgemental, compassionate and supportive to mother who has given birth to child suspected of having FASD. Mother needs support and encouragement, not abuse as some mothers have experienced on giving birth to a child with FAS in hospital from midwifes and other staff.
- Use encouraging language; focus on positive for future outcomes with support pathways discussed. Many children born with FASD go on to have successful and fulfilling lives. This starts now with the early intervention supports put in place.
- Resources and contact details for ELEN (UK & European Birth Mothers – FASD) to be given to mother and staff to contact if mother was too distressed and needs support.
- Contacting disability social workers for support to be put in place, not to report to child protection. Mother and child need lots of support at this time.

33.3 Seeking a Diagnosis*: Bethanny*

I knew from a very young age, around 1 year old, that my son Freddy had a massive temper and that he wouldn't eat a lot of foods. I just thought at the time that this was just him. At Pre-school, they picked up a few things that gave them cause for concern. Freddy's speech wasn't developing, he found playing with other's difficult, he wouldn't share, they couldn't get him to join in group activities, he spent most of his time at pre-school on his own, and he acted strangely around certain people. It wasn't for another year before I realized that Freddy really struggled with life as a 5-year-old. His school again picked up on lots of behavioural difficulties.

I started researching the behavioural issues that had come up and a few disabilities cropped up. I read up on each one and the minute I read about FASD, I knew in my heart that this was what Freddy had. Everything fitted. I went to the Dr. with my concerns, I admitted to him the amount and how often I had drunk during my pregnancy. The Dr. informed me that they wouldn't do anything without a letter of concern from the school. I immediately went to the Head Teacher, who I had full support from, she sent through the letter of concern.

Our Dr. sent a referral to CAMHS regarding my concerns. We waited about 9 months for our first appointment. Once there they spoke with me and Freddy, they decided that we would need a second appointment. CAMHS also sent a referral to our local paediatrician. We waited for the appointments to come through. CAMHS was a minimum of 18 months for a second appointment and we waited months for a paediatric appointment. We went to see our paediatrician who again went through everything with me that I had already disclosed to my Dr. and CAMHS. They also spoke with Freddy. I had got information that we needed an array CGH blood test and that we needed to see geneticist. The paediatrician was very unhelpful stating that Freddy didn't have facial features so he wouldn't have FASD. I explained that with FASD/ARND, there were no facial features but she insisted that they still

wouldn't diagnose. I felt like it was a completely wasted trip. I went back to our Dr. to ask for a referral to a geneticist for an array test. Eventually, the appointment came through. Again, I felt like the trip was wasted, the geneticist told us there would be no diagnosis without facial features. I once again explained that there were no facial features with FASD/ARND. He told me that my son could have all the neurological disability and no facial features but they still wouldn't diagnose.

I was absolutely gobsmacked. I felt that I was banging my head against a brick wall and that no one was listening to me. I was telling these professionals that due to my consumption of alcohol during my pregnancy that my son needed their help and no one cared. I felt so alone, so ashamed and still no one helped. We were still waiting for our second appointment at CAHMS, which actually took our Head Teacher to call them and once again express her concerns for an appointment to come through. We eventually saw the ASD team within CAMHS. We saw them a few times, they did a few different things with Freddy and spoke with myself. They came to the conclusion that Freddy didn't have ASD and that CAMHS could do no more and closed Freddy's file.

Freddy's Array CGH test came back fine. So after about 2 and half years, I was back to square one. I researched doctors in my area who had diagnosed FASD and therefore competent where this disability was concerned. I went back to my Dr. and asked for a referral to one of these Drs, to which we were refused. I felt lost and alone and didn't know where to turn.

Through friends of friends, I got an appointment with a reputable private Dr. who said he would diagnose ARND and that we needed paediatrics and CAMHS on board. I came away feeling like we were getting somewhere, someone had listened and I was so grateful. The private Dr. sent through a letter about our appointment but because of the wording on the letter neither paediatrics nor CAMHS would see us.

Once again, I was back to square one. Today I'm back to the point where I'm trying to get people to see us. We now have an appointment with a private geneticist. My Dr. has asked for referrals to CAMHS and paediatrics once again which we are waiting to hear about. My son needs the diagnosis, he needs the help from the professionals, and he not only needs it but deserves it.

Both I and my son have been pushed from pillar to post. I have felt like I'm going mad, no one ever listened. Why would I lie about drinking alcohol during pregnancy? I have felt so ashamed and so guilty on a daily basis for what I have done and the professionals I have seen have made me feel 100 times worse.

I believe that the doctors, paediatricians and CAMHS need re-training in this disability, they need to listen to us mothers that have key information, who live with our children every single day. My son is now looking at a diagnosis of ARND, ADHD, SPD and VT and at the moment, his school is the only one supporting him.

33.3.1 Recommendations

- Attend up-to-date training and train up all staff about fetal alcohol spectrum disorder.

- Women do not seek a diagnosis of FASD lightly. Ninety per cent of the mothers we surveyed knew early on that their child struggled in certain areas and that it may be down to their alcohol use during pregnancy. Many birth mothers sought a referral but were told that they do not want to label the child or they did not want to stigmatize the mother.
- Women and children want labels so they can best support themselves and their children. Post-diagnostic support is critical for early interventions.
- Be compassionate and non-judgemental. Birth mothers reported watching the face of paediatrician changing when realizing they were the birth mother not the adopted parent or carer.
- Being judged and stigmatized has prevented many from seeking a diagnosis for FASD for their child for fear of what statutory services may do.
- Support her and make necessary referrals to a geneticist and paediatrician who know about FASD (too many dismiss FASD because there are no facial features).
- Be positive, there are lots of support available for families and there are many very successful individuals living with FASD.
- Good idea is to have a leaflet or photocopy list of support resources they can access, and offer to write any letters that may be needed to get the services and benefits the child may require.
- Give information and contact details of ELEN—UK & European Birth Mother network—FASD—women want to speak to other women who have had same experiences and feel better understood.

References

1. Cismaru M, et al. Preventing fetal alcohol spectrum disorders: the role of protection motivation theory. Health Mark Q. 2010;27(1):66–85.
2. Skagerström J, Chang G, Nilsen P. Predictors of drinking during pregnancy: a systematic review. J Womens Health. 2011;20(6):901–13.
3. Walker M, Al-Sahab B, Islam F, Tamim H. The epidemiology of alcohol utilization during pregnancy: an analysis of the Canadian maternity experiences survey (MES). BMC Pregnancy Childbirth. 2011;11(1):52. https://doi.org/10.1186/1471-2393-11-52.

Working to Develop the Evidence for Policy Change

34

Maggie Watts

Chapter Highlights

- Models of care and approaches to intervention.
- The range and complexity of FASD presentation and how to approach these.
- Models to identify and intervene.

34.1 Introduction

Fetal alcohol harm has become a well-recognised issue internationally, and fetal alcohol spectrum disorder (FASD), resulting from prenatal alcohol exposure, is considered to be the commonest preventable cause of mental retardation and birth defects [1]. Worldwide, it is estimated that FASD affects at least 1 in every 100 births [2] with an increasing number of studies over the years demonstrating much higher levels in some communities [3–6]. The World Health Organisation has identified alcohol-related harm to others and FASD as priorities for action by member countries [7]. However, the UK lags behind in this recognition and in its actions to reduce such harm [8].

Public health approaches focus work at population rather than individual levels and, hence, provide a structured way through which to identify and address the breadth of factors important to FASD. Such approaches are inclusive, non-judgemental and seek to ensure that no sections of communities are stigmatised.

Prenatal alcohol exposure is linked to a range of presentations—from the physical and physiological to cognitive and processing deficits, and to deficient social and educational skills. These presentations are related to the primary damage caused by

M. Watts (✉)
NHS Western Isles, Stornoway, Isle of Lewis, UK
e-mail: maggie.watts@nhs.scot

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_34

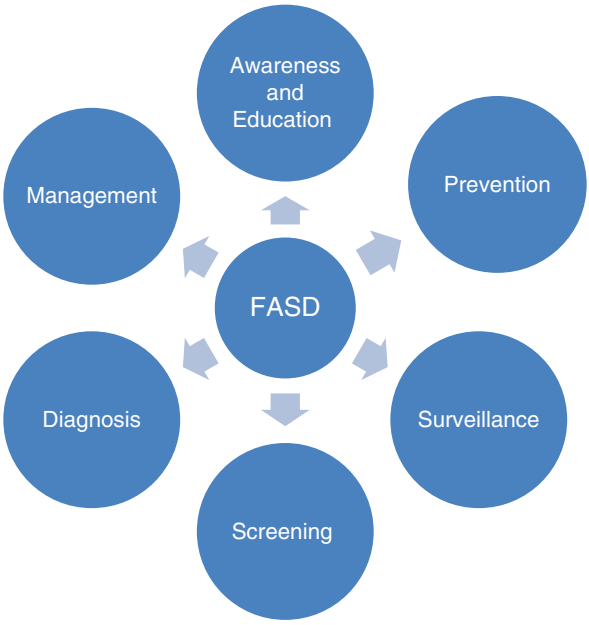


Fig. 34.1 Public health approaches to addressing fetal alcohol harm

alcohol on the developing fetus, most notably the fetal brain, and to the complications (secondary disabilities) arising from failures to recognise and manage the impacts of this damage on the individual.

FASD is therefore composed of a myriad of presentations and these require many and varied responses. These responses range from preventative ones at the widest population level, such as advice on alcohol consumption, to those for specific populations, such as pregnant women or women with addiction issues, and from generalised messages to specific actions relating to professional groups.

Population-based (public health) responses can be considered across the topics of awareness raising and education, prevention, active and passive surveillance, screening, diagnosis and management.

A summary of the different aspects to be considered in such public health approaches to FASD is set out in Fig. 34.1. As other chapters provide in-depth coverage of diagnosis and management, these aspects will not be addressed further here.

34.2 A Brief Mention of Costs of FASD

Alongside these aspects are those relating to the cost-effectiveness evidence for action in prevention, identification and early management of fetal alcohol harm, which has been studied extensively out with the UK. The myriad of agencies which may be involved in providing care and support to children and families affected by fetal alcohol harm is outlined in Fig. 34.2. Premature mortality and morbidity are

Fig. 34.2 Support services required for individuals with FASD

the greatest factors in FASD-attributable costs and the preventable nature of the condition together with these costs can be helpful levers in discussion on prioritisation of FASD prevention activities. However, low recognition of FASD across the health, social care, education and justice systems can impact on progress. With worldwide prevalence of FASD considered to exceed 1% [2], the economic consequences of fetal alcohol harm are extensive [9] and actions to prevent its occurrence, rather than manage the consequences, are warranted on economic grounds alone.

34.3 Awareness and Education

Alcohol use is ubiquitous in the UK and alcohol consumption across all population groups, including women of reproductive age, has risen considerably over the past 40 years during which FASD has been recognised and systematically researched. Knowledge of the adverse effects of alcohol is widespread—most people will

recognise harmful effects of alcohol in all aspects of life, from the effects of acute intoxication, alcohol-related crimes, road traffic accidents, the calorific effects of alcohol contributing to obesity, alcohol-related liver and brain damage, to alcohol dependence and addiction [7]. Alcohol contributes to depression and other psychiatric morbidity. Many of these effects involve harm to others such as family problems, intimate partner and stranger violence, accidents due to alcohol-impaired drivers and harm to children of alcohol-dependent parents [7].

However, knowledge and awareness are much more limited in relation to the teratogenic effects of alcohol that interfere with normal fetal development and which may result in lifelong disabilities and impact on the ability to live an independent life. Whilst many people may indicate an awareness of fetal alcohol harm, such knowledge is superficial and lacking in detail [10, 11]. Similarly, awareness of FASD amongst health and care professionals is limited [12–14].

Alcohol awareness education and public media campaigns tend to focus on aspects of harm other than that to the fetus, with little evidence of effectiveness [15]. Alcohol, its use and potential for damage are covered in a variety of ways in the education of children from primary years onwards [16]. However, there is little or no reference to the effects of alcohol in pregnancy and limited evidence of effectiveness in influencing behaviours [17]. Specifically addressing the education needs of pregnant women, there is very limited evidence that public health interventions delivered through mass media affect the alcohol consumption of pregnant women [18].

The knowledge and understanding of the effects of alcohol in pregnancy by professionals, across health services, education, psychology and social care, is important but known to be limited [19]. In the UK, there is no consistent approach across undergraduate or postgraduate curricula to alcohol in pregnancy and its potential consequences. Although some courses include reference to FASD in their undergraduate curricula; others do not. Considerable reliance is placed on 'champions'—people with interest and enthusiasm—to increase awareness among professionals and the general public. However, it is with such professional groups that education can have a positive effect on knowledge and behaviours [20].

One way in which knowledge can be enhanced, reach a wide audience and maximise training opportunities is through the use of digital technology and e-learning. In Scotland, a universally accessible FASD awareness e-learning module has been developed by NHS Education for Scotland supported by the Scottish Government [21]. This and similar e-learning modules allow learners to develop their knowledge and understanding of the topic at their own pace and to consider how to apply this knowledge into their everyday life and work.

34.4 Prevention

The classic public health approach to prevention is to consider primary (reduce the incidence of the condition), secondary (detect the condition and treat early) and tertiary (reduce the impact of the condition) elements. For FASD, whilst this

approach is appropriate, it is also restricting as it struggles to recognise the complexities of prevention in terms of women, their babies and the mother-baby dyad. Population segmentation allows different interventions to be developed and targeted at different population groups. The four-level model for prevention used by Public Health Canada [22] provides for multiple approaches, from general awareness at Level 1 to highly specific work with new mothers affected by alcohol use in pregnancy (Fig. 34.3).

At the broadest level, **Level 1** (primary prevention) addresses the whole population. It is concerned with awareness raising through the provision of information across a wide variety of formats and media, in relation to alcohol use in pregnancy. As indicated above, the quality of evidence of effectiveness across the range of universal prevention initiatives is relatively poor—amongst other reasons because it is very difficult to carry out the necessary population-based random controlled trials with the wealth of potential confounding factors.

Knowledge of the patterns of alcohol use across population groups is helpful in identifying target groups for whom prevention activities will differ—for example, the messages for people who tend to have high levels of alcohol consumption spread across the week may differ from those for the people who binge drink at weekends. Alcohol use is very common and self-reported levels of use tend to underestimate consumption when compared to alcohol sales [23]. Across the UK, harmful and hazardous patterns of drinking are common and women's alcohol use has increased across the past 40 years [24, 25].

UK Government advice regarding alcohol consumption in pregnancy has changed in recent years following a major review of the evidence, with all UK

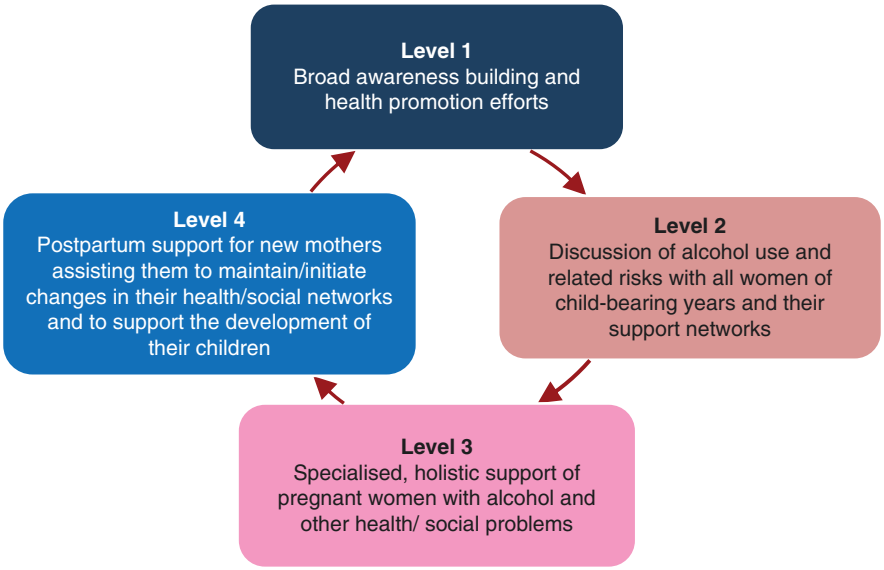


Fig. 34.3 Canadian model of FASD prevention. (Poole 2008 [22])

Health Departments now stating that no alcohol should be consumed during pregnancy [26]. Notwithstanding that, there is a paucity of evidence for human fetal alcohol harm at low levels of drinking, although the use of animal models is able to demonstrate impact of small amounts of alcohol on fetal development. The UK has joined many other countries in adopting a precautionary approach since there is no known safe limit of alcohol consumption in pregnancy. Whilst this consistency of message is helpful and many feel that this is the right message, there will remain those who will be sceptical of the lack of evidence base, despite the ethics of undertaking randomised control studies in human populations, and who will continue to advise women that small amounts of alcohol in pregnancy are not harmful [27]. It is here perhaps that the difference between a population risk-based approach (the best for all) and an individual risk-based approach by a clinician who is aware of the totality of their patient's life—such as their socio-economic status, nutritional status and pre-pregnancy alcohol consumption patterns—is most apparent.

The environment within which alcohol is available, and the controls on alcohol use and sales, can be seen as important factors in addressing fetal alcohol harm. In some countries where alcohol sales are controlled by the state, there may be a requirement to include publicity with advice on drinking in pregnancy and on the effects of prenatal alcohol exposure. Caution is needed with such an approach since this may not necessarily have the intended effect of reducing alcohol use in pregnancy but work to polarise and stigmatise women by failing to recognise the social and wider factors that may influence women's use of alcohol in pregnancy [28].

In the UK, Licensing Boards, which are required to consider the harmful effects of alcohol and may include a public health objective as one of their areas of practice, are able to encourage on- and off-sales to include promotional materials about fetal alcohol harm as part of their licensing conditions. However, it is not common for Licensing Boards to do so, which is likely to be a reflection of the low level of awareness of fetal alcohol harm amongst regulatory bodies, professionals and the wider public in the UK.

Alongside licensing powers at local level, the UK Government has the power to legislate on a range of aspects of alcohol. There is currently a voluntary code of alcohol labelling agreed by the UK Government with the alcohol industry that includes a statement to 'Avoid alcohol when pregnant or trying to conceive' or the internationally recognised 'no alcohol in pregnancy' logo. Although the required proportion (80% by the end of 2013) of on-the-shelf alcohol products carried this labelling, far fewer complied with the size and visibility recommendations, with the smallest and least visible 'pregnancy logo' available on wine, the product most likely to be consumed by women [29]. Evidence indicates that whilst such labelling is supported by the general public, and is recognised as raising awareness, it has little impact on behaviours [30].

As with alcohol labelling, general public awareness campaigns, on their own, are not effective in reducing alcohol consumption [15]. Current UK and Scottish Government policy of reducing whole population alcohol consumption is valid but requires targeting or stratifying for different recommendations so that there is a

consistent message for everyone to support women in avoiding alcohol when contemplating and during pregnancy.

In the UK and its component countries, whilst there have been mass media campaigns to raise awareness of the effects of alcohol, there are very few aimed at reducing drinking in pregnancy—the 'No Alcohol No Risk'campaign in Scotland being the most recent. This was part of a wider Scottish strategy to bring to public attention the importance of avoiding alcohol use in pregnancy, with public-facing materials (posters and leaflets) alongside training for health, social care and education professionals about fetal alcohol spectrum disorders. Although there is an inherent risk that these types of programmes will be considered to be targeted at individual women rather than for the population as a whole, such combined strategies are more likely to be successful than those with a single focus.

Specific mass media work demonstrates an increase in knowledge and awareness of messages but is unlikely, on its own, to produce behaviour change in the target groups [31]. However, whilst such campaigns have limited impact, they can nevertheless form part of wider programmes of activities and actions to prevent and reduce fetal alcohol harm.

International activity can also raise awareness, as part of wider programmes. Around the world, September ninth has become promoted increasingly as International FASD Awareness Day. This serves as a functional platform for celebrating the achievements of people with FASD as well as promoting prevention of prenatal alcohol exposure. The development of a pack or toolkit to direct such awareness raising efforts aids consistency of approach across a nation and has been used in Scotland [32] to support action at local level. This toolkit provides a range of activities, factsheets and media messages. Such a multipronged approach assists in awareness raising and in the provision of consistent simple messages supporting the avoidance of alcohol during pregnancy aimed at a whole population level.

Level 2 prevention is focused on discussing alcohol use with women of childbearing age, regardless of pregnancy status, and with those who provide care and support for women, with the aim of supporting women to reduce or stop alcohol use when trying to conceive as well as during pregnancy.

The importance of preconception advice is increasingly recognised as critical to the health and well-being of both mother-to-be and baby but is often overlooked or neglected [33]. Pre-pregnancy is an ideal time to prepare for a healthy baby and for professionals working with women to identify and address risk factors for a successful outcome. However, more than 40% of pregnancies worldwide are unplanned [34] so there is a need to work with all women of reproductive age to enhance their knowledge and understanding of, and control over, their bodies and for them to consider their choices carefully.

Prevention of fetal alcohol harm is supported by consistent messages and advice on avoiding alcohol when pregnant or thinking about becoming pregnant. Alongside consideration of alcohol use are discussions on reproductive health. The range of interventions focuses around aspects of reproductive health including pregnancy planning such as:

- The use of effective contraception for those who do not yet wish to become pregnant.

- Alcohol brief interventions and specialist alcohol services to reduce or eliminate alcohol use during pregnancy.
- Wider aspects of parenting including addressing parental support effectively, especially for the most vulnerable women.

Sexual health services and substance use treatment services together have important roles to play in supporting women to prepare for a healthy pregnancy. Sexual health and family planning services can provide advice and support with long-acting reversible contraception (LARC) to encourage pregnancy planning and enable high-quality pre-conception care for a healthy pregnancy at a time to suit the woman's circumstances and preferences.

Targeted services that seek to reduce alcohol use together with postponing pregnancy, such as Project CHOICES, integrate elements of contraception including LARC with pregnancy planning and alcohol-free pregnancies. Based on the principles of motivational interviewing, programmes have been shown to be effective across a range of settings [35]. However, they require face-to-face contacts through the course of a number of sessions, and rely heavily on women's commitment to continue attending. More recently, the efficacy of delivery of similar programmes through the Internet has been explored successfully, extending access to larger numbers of women than were previously addressed through in-person contacts [36].

For women of childbearing age who have previously accessed addiction services with alcohol use in combination with other drug use or on its own, these services are well placed to address contraceptive and reproductive healthcare early, supporting recovery by offering control for the woman of some aspects of her life. National targets for sexual health in Scotland encompass the provision of long-acting reversible contraception (LARC) to support planned pregnancy and allow healthcare interventions to improve health and well-being.

Once a woman recognises that she is pregnant and approaches maternity services, additional opportunities for prevention of prenatal alcohol exposure are available, although evidence of effectiveness of interventions in pregnancy is limited by the paucity of published studies [18]. The professional midwifery services that provide antenatal care for the vast majority of UK women are ideally placed to screen women early in pregnancy for their alcohol use from pre-conception using a standardised approach and screening tool, and to provide a brief intervention, or referral to addiction services if warranted, when any alcohol has been used. Midwives are uniquely well placed to develop a positive relationship over time with each pregnant woman and to provide a non-judgemental and supportive approach to screening for alcohol use at any point during the pregnancy. A number of screening tools are validated for use at this time, including T-ACE, TWEAK and AUDIT. In Scotland, alcohol brief interventions in pregnancy—using motivational interviewing techniques to assess the alcohol use of the individual, support the woman to review this use and to develop strategies to avoid alcohol during the pregnancy—form part of a performance standard for maternity care, although uptake across the country varies and evidence of effectiveness is limited [37, 38].

Level 3 interventions relate to more specialised support of pregnant women with alcohol and other social and health problems. It is here that specialist services, such as sexual health, addictions and specialist midwifery roles, can work together to best effect [39]. In the UK, the midwifery role takes on a particular significance for women at high risk of an alcohol-exposed pregnancy (AEP), in co-ordinating the specialist addiction support the woman may require and linking to postnatal services for follow up of the child.

Adverse life experiences, including those of childhood trauma or neglect, domestic violence and emotional or sexual abuse, are important contributors to the use of alcohol in pregnancy [40]. It is, therefore, important that staff are trained in trauma-informed practice so that women who use alcohol are encouraged to come forward and be supported in non-judgemental services. Services should recognise that women's lives may be complex and complicated, and that the choices they have and their autonomy and control of decision-making may be limited by circumstances such as adverse childhood experiences, domestic violence, sexual and emotional abuse and wider indicators of socio-economic deprivations such as educational attainment, poverty, poor housing and employment status. For women who are living with alcohol and drug use, their previous experiences of pregnancy may be impacted by involvement of children's services and placement of their children in alternative forms of care. Service approaches and attitudes towards women with alcohol or substance use problems are more effective when they are gender and trauma informed.

Contraception is one of the first areas where women with alcohol problems can be supported to gain control of their lives. One of the best mechanisms for FASD prevention in women who have alcohol use/substance use problems is the ready availability of effective long-acting reversible contraception offering protection from an unwanted or alcohol-exposed pregnancy, delivered in services that women access by staff who are non-judgemental, supportive and approachable [41]. Nevertheless, women may be reluctant to access services because of beliefs or fears about stigma, feeling judged or otherwise adversely considered. Stigma has a major impact on reducing alcohol consumption in pregnancy; it generates blame towards mothers, ignores social factors leading women to drink in pregnancy and increases reluctance to disclose [28].

Whilst many consider that fetal alcohol spectrum disorders are entirely preventable, such a view fails to recognise the deeper causal factors, which may be cultural, familial, social or economic, that contribute to women's decisions to drink during pregnancy and to society's attitude towards such alcohol consumption [28]. The presumption that drinking in pregnancy rests solely with the individual fails to recognise the pressures of society and the role that healthcare professionals can play in supporting women on issues such as childhood trauma, sexual abuse, intimate partner violence, sexual slavery, controlling behaviours and other forms of abuse and loss of control over their lives are important considerations. Indirectly related issues such as adequacy and security of housing, financial instability and mental health issues impact on the lived experiences and the choices made by women. Considering decisions through a trauma-informed lens will assist professionals in understanding

the needs of women with backgrounds that contribute to their alcohol use. The combination of addressing alcohol use and encouraging improved contraception can result in reduced risk of future alcohol-exposed pregnancies as well as better use of family planning.

The final level of prevention, **level 4,** is around postpartum support for mothers with alcohol (with or without other drug use) problems to assist them in initiating or maintaining changes in their health and social networks and to support the development of their children who may or may not have FASD. It is recognised that some of these women will also have FASD themselves, and their lived experience will affect the decisions they make with regard to their own substance use and the relationships with their child.

Evidence supports the use of personalised management and care such as happens with the Parent-Child Assistance Program (PCAP) [42, 43]. Mothers participating in such programs are supported through nurse home visiting and longer-term mentorship or advocacy interventions. These studies demonstrate the effectiveness of more intensive approaches in not only reducing alcohol use and improving treatment completion rates but also the importance of wider life circumstances in success, with stable housing a strong consideration.

From the child's perspective, level 4 interventions look to promote parenting, creating positive parent-child emotional bonding. Prenatal alcohol exposure increases the risk of neurodevelopmental and behavioural difficulties, which in turn may be exacerbated by disordered attachment as may occur with parental substance use. Success in improving relationships between child and parent resulting in less behavioural dysregulation in the child and more supportive and positive parenting has been shown to result from tailored support for such families [44].

34.5 Surveillance

Surveillance, defined as the structured monitoring of the occurrence of a condition in a defined population in a planned way by appropriately trained professionals working to a common definition, helps to identify the public health burden of disease associated with fetal alcohol harm. Such surveillance in relation to FASD is very challenging and initial efforts tended to focus on children with FAS with its more easily recognised physical features. Criteria for identification of affected children include growth (both intrapartum and postnatal), facial features and brain development together with a clear history of prenatal alcohol exposure. It is very often this history of prenatal alcohol exposure that is the most difficult to secure for a number of reasons—children may not be with their birth mother, the birth mother may not disclose their alcohol use for fear of stigma or judgement, maternity services records may not include this information or be accessible, or such information may be anecdotal and acquired from others rather than evidenced.

Surveillance methodology may be passive or active, with active case ascertainment providing the highest levels of the condition, and also with the greatest

resource utilisation. Passive surveillance through clinic reporting or record reviews is considerably less expensive.

In Scotland, a passive surveillance programme was introduced in 2010, supported by the Scottish Government and Chief Scientist's Office and operated through the Royal College of Paediatrics and Child Health in Scotland. A 4-year study carried out with paediatricians throughout Scotland (the FAS Passive Surveillance Study) showed 41 children under the age of 6 were diagnosed with FAS, which is likely to be a significant underestimate of the potential number of affected children when compared to a majority of studies identified in the literature [45]. Similar work in other parts of the world produces lower than expected estimates from passive surveillance with lack of experience and expertise in making the diagnosis together with a reluctance to diagnose or stigmatise the family identified as common features [46–48].

Active surveillance programmes have taken place in an increasing number of countries [2–4]. For these, children with indicators such as small head circumference, short stature or with learning or behavioural problems are, with parental consent, assessed for possible FASD. Since FASD is a spectrum of disorders, many studies address FAS as the most readily identifiable and recognisable condition. Such studies are providing rates of diagnoses on the FASD continuum considerably higher than previous estimates of these conditions.

Meta-analyses indicate that rates of FAS and FASD vary greatly across countries, for example, South Africa having a high pooled prevalence and New Zealand a much lower one [46]. However, there are also wide variation in study designs identified, such as the use of sampling frames of targeted populations or the general population, the age of diagnosis, lack of consistency in diagnostic criteria across studies, differences in guidelines used for FASD diagnosis (such as Canadian guidelines, Institute of Medicine guidelines or Washington 4-digit code) and methods of case identification. Despite these variations, it is apparent that FASD is a global public health issue.

34.6 Screening

Screening relates to the structured and routine assessment of a defined population for a condition that is amenable to intervention. Specific tests are applied to identified populations in order to provide a rapid assessment of individuals, with those most likely to have characteristics of the condition selected for further assessment. Usually screening for a health condition requires certain parameters to be present—for example, screening should provide for early detection and treatment, so needs to have a diagnostic test available, ideally one that is simple, non-invasive and acceptable to the population to be screened. It should also target those with previously undetected disease. In addition, the natural history of the condition should be known and there should be effective and accepted treatment available, with facilities for assessment, diagnosis and rehabilitation [49]. Screening also needs to be cost effective—the cost of screening must be proportionate to the costs of caring for

affected individuals. With the above in mind, it may be argued that screening for FASD does not meet all the required criteria, with no single diagnostic test or treatment. However, screening options are relatively simple and non-invasive and there is good evidence that early intervention reduces morbidity. In addition, there is increasing evidence of the considerable costs of caring for an individual with FASD [50].

If screening is to be used, it can be applied in respect of prenatal alcohol use, or in respect of populations of children or adults for FASD. In pregnancy, maternal screening for alcohol use is designed to prevent and manage further prenatal alcohol exposure, but can also highlight pregnancies where alcohol use would benefit from referral to treatment services and where indicative prenatal alcohol exposure means that neonatal follow up is recommended.

In the case of FASD, screening is about finding those with an undiagnosed condition, and avoiding harm to people who do not need treatment—although it is recognised that screening in itself can identify other conditions and raise concerns that would not otherwise have been noted. Notwithstanding that, screening for specific aspects related to FASD may be of value as long as the benefits for the individual outweigh the drawbacks.

In Canada, the CAPHC screening toolkit includes, amongst other elements, maternal alcohol and drug screening tools, meconium FAEEs testing and checklists and screening tools for children and young people in different settings [51]. Use of the toolkit enables services to consider points at which identification and intervention can have a positive impact. The toolkit emphasises that screening does not provide a diagnosis but raises issues that could benefit from further enquiry and investigation.

The early detection of alcohol use in pregnancy should be conducted through the use of a standardised structured screening tool, such as T-ACE, TWEAK and AUDIT, as previously mentioned. The scoring from this screen will then guide the use of an alcohol brief intervention to help the woman consider her alcohol use and how to reduce or stop it. Some women, who admit excess alcohol use, will need access to further services to help them reduce or cease their alcohol use; such access should be provided in a non-judgemental and trauma-informed way, recognising that stigma and fear are major barriers to women being able to access services.

The value of testing meconium (the first baby bowel movement) for biomarkers of alcohol use such as fatty acid ethyl esters (FAEEs) and ethyl glucuronide (EtG) lies in having an objective measure of prenatal alcohol exposure in the latter months of pregnancy. Women are not always good at recalling and reporting their alcohol use in pregnancy and biomarkers can help to identify babies at highest risk of prenatal alcohol exposure, and arrange suitable follow up. Such studies have consistently demonstrated higher levels of prenatal alcohol exposure than can be achieved through maternal recall [52–54].

Screening of populations is most commonly carried out with children of school age, when characteristic features of face and growth are more evident and when more structured testing of neurocognitive abilities is possible. As with all screening programmes, participants are invited to be included. Such populations are initially

assessed for indicators of potential FASD such as small head circumference and short stature. Those who are identified through this initial or pre-screening can then be offered more specific and sensitive screening tests of growth and neurodevelopment which may lead to referral for more in-depth assessment by multi-disciplinary teams with FASD expertise. This is often the structure for active case ascertainment studies around the world [55]. It is important to remember that the rationale for screening is to identify individuals within populations who are able to benefit from detection and possible diagnosis of FASD.

34.7 Conclusion

Since the rediscovery of the damaging effects of prenatal alcohol exposure in the 1970s, the nature of fetal alcohol harm has been examined and explored. From a small core, there are now increasing numbers of clinicians, researchers and practitioners around the world working to develop the evidence further, with studies ranging from biochemical interactions in the brain to cellular level, using animal models and up to individual and cohorts of babies, children and adults. Consistency and reproducibility of research is recognised as vital to truly demonstrating the nature, effects, impacts, prevention and management of prenatal alcohol exposure. The production of consistent internationally agreed guidelines for prevention, ascertainment, diagnosis and management is highly desirable. National clinical guidelines recognise the impact of prenatal alcohol exposure and seek to improve the outcomes for those affected [56]. We have travelled a long way in developing our understanding of FASD but still have much to do to encourage every pregnancy to be alcohol free and ensure that any affected baby is able to grow to maximise its full potential.

References

1. Riley EP, Clarren S, Weinberg J, Jonsson E. Fetal alcohol spectrum disorder: management and policy perspectives of FASD. Hoboken, NJ: Wiley; 2011.
2. Lange S, Probst C, Gmel G, Rehm J, Burd L, Popova S. Global prevalence of fetal alcohol spectrum disorder among children and youth: a systematic review and meta-analysis. JAMA Pediatr. 2017;171(10):948–56.
3. May PA, Chambers CD, Kalberg WO, Zellner J, Feldman H, Buckley D, Kopald D, Hasken JM, Xu R, Honerkamp-Smith G, Taras H. Prevalence of fetal alcohol spectrum disorders in 4 US communities. JAMA. 2018;319(5):474–82.
4. May PA, Brooke L, Gossage JP, Croxford J, Adnams C, Jones KL, Robinson L, Viljoen D. Epidemiology of fetal alcohol syndrome in a south African community in the Western Cape Province. Am J Public Health. 2000;90(12):1905–12.
5. May P, Fiorentino D, Coriale G, Kalberg W, Hoyme HE, Aragón A, Buckley D, Stellavato C, Gossage JP, Robinson L, Jones KL. Prevalence of children with severe fetal alcohol spectrum disorders in communities near Rome, Italy: new estimated rates are higher than previous estimates. Int J Environ Res Public Health. 2011;8(6):2331–51.

6. McQuire C, Mukherjee R, Hurt L, Higgins A, Greene G, Farewell D, Kemp A, Paranjothy S. Screening prevalence of fetal alcohol spectrum disorders in a region of the United Kingdom: a population-based birth-cohort study. Prev Med. 2019;118:344–51.
7. World Health Organization Management of Substance Abuse Unit. Global status report on alcohol and health, 2014. Geneva: World Health Organization; 2014.
8. British Medical Association Board of Science. Alcohol and pregnancy: preventing and managing fetal alcohol spectrum disorders. London: British Medical Association; 2016.
9. Popova S, Lange S, Burd L, Rehm J. Burden and social cost of fetal alcohol spectrum disorders. New York: Oxford Handbooks Online; 2016. https://doi.org/10.1093/oxfordhb/9780199935291.001.0001/oxfordhb-9780199935291-e-78.
10. Mukherjee R, Wray E, Hollins S, Curfs L. What does the general public in the UK know about the risk to a developing foetus if exposed to alcohol in pregnancy? Findings from a UK mixed methodology study. Child Care Health Dev. 2015;41(3):467–74.
11. Scottish Government. Omnibus survey (unpublished); 2011.
12. Carswell T. A study of the knowledge, behaviours and understanding of Scottish paediatricians in relation to alcohol and fetal alcohol syndrome. MPH thesis. University of Glasgow; 2007.
13. Birch SM, Carpenter HA, Marsh AM, McClung KA, Doll JD. The knowledge of rehabilitation professionals concerning fetal alcohol spectrum disorders. Occup Ther Health Care. 2016;30(1):69–79.
14. Mukherjee R, Wray E, Curfs L, Hollins S. Knowledge and opinions of professional groups concerning FASD in the UK. Adopt Foster. 2015;39(3):212–24.
15. Anderson P, Chisholm D, Fuhr DC. Effectiveness and cost-effectiveness of policies and programmes to reduce the harm caused by alcohol. Lancet. 2009;373(9682):2234–46.
16. Foxcroft DR, Tsertsvadze A. Universal alcohol misuse prevention programmes for children and adolescents: Cochrane systematic reviews. Perspect Public Health. 2012;132(3):128–34.
17. Harvey SA, McKay MT, Sumnall HR. Adolescents' reflections on school-based alcohol education in the United Kingdom: education as usual compared with a structured harm reduction intervention. J Subst Abuse. 2016;21(6):640–5.
18. Crawford-Williams F, Fielder A, Mikocka-Walus A, Esterman A. A critical review of public health interventions aimed at reducing alcohol consumption and/or increasing knowledge among pregnant women. Drug Alcohol Rev. 2015;34(2):154–61.
19. Elliott EJ, Payne J, Haan E, Bower C. Diagnosis of foetal alcohol syndrome and alcohol use in pregnancy: a survey of paediatricians' knowledge, attitudes and practice. J Paediatr Child Health. 2006;42(11):698–703.
20. Payne JM, France KE, Henley N, D'antoine HA, Bartu AE, Mutch RC, Elliott EJ, Bower C. Paediatricians' knowledge, attitudes and practice following provision of educational resources about prevention of prenatal alcohol exposure and fetal alcohol spectrum disorder. J Paediatr Child Health. 2011;47(10):704–10.
21. NHS Education for Scotland. Fetal alcohol harm e-learning resource [Internet]. 2012. http://www.knowledge.scot.nhs.uk/home/learning-and-cpd/learning-spaces/fasd.aspx. Accessed 30 May 2017.
22. Poole N. Fetal alcohol spectrum disorder (FASD) prevention: Canadian perspectives. Ottawa: Public Health Agency of Canada; 2008.
23. Robinson M, Thorpe R, Beeston C, McCartney G. A review of the validity and reliability of alcohol retail sales data for monitoring population levels of alcohol consumption: a Scottish perspective. Alcohol Alcohol. 2012;48(2):231–40.
24. Public Health England. Local Alcohol Consumption Survey, 2016. UK Data Service. 2017. https://doi.org/10.5255/UKDA-SN-8181-1.
25. Scottish Health Survey 2017. https://www.gov.scot/publications/scottish-health-survey-2017-volume-1-main-report/. Accessed 23 Feb 2019.
26. Department of Health. Alcohol guidelines review—report from the guidelines development group to the UK chief medical officers. London: Department of Health; 2016.
27. Gavaghan C. "You can't handle the truth"; medical paternalism and prenatal alcohol use. J Med Ethics. 2009;35(5):300–3.

28. Bell E, Andrew G, Di Pietro N, Chudley AE, Reynolds J, Racine E. It's a shame! Stigma against fetal alcohol spectrum disorder: examining the ethical implications for public health practices and policies. Public Health Ethics. 2015;9(1):65–77.
29. Petticrew M, Douglas N, Knai C, Durand MA, Eastmure E, Mays N. Health information on alcoholic beverage containers: has the alcohol industry's pledge in England to improve labelling been met? Addiction. 2016;111(1):51–5.
30. Wilkinson C, Room R. Warnings on alcohol containers and advertisements: international experience and evidence on effects. Drug Alcohol Rev. 2009;28(4):426–35.
31. Bazzo S, Battistella G, Riscica P, Moino G, Marini F, Geromel M, Czerwinsky L. Evaluation of the impact of the image used in a communication campaign to raise awareness about the effects of alcohol use during pregnancy. Alcohol Alcohol. 2012;47(6):657–62.
32. Scottish Government Child and Maternal Health Division. Fetal Alcohol Spectrum Disorder Awareness Toolkit. Edinburgh: The Scottish Government; 2013. http://www.gov.scot/Publications/2013/10/3881. Accessed 14 Dec 2016.
33. Sher J, Missed Periods—Scotland's opportunities for better pregnancies, healthier parents and thriving babies the first time… and every time: an independent report commissioned by NHS Greater Glasgow and Clyde (Public Health) [Internet]. 2016. http://www.nhsggc.org.uk/media/237840/missed-periods-j-sher-may-2016.pdf. Accessed 15 Jun 2017.
34. Bearak J, Popinchalk A, Alkema L, Sedgh G. Global, regional, and subregional trends in unintended pregnancy and its outcomes from 1990 to 2014: estimates from a Bayesian hierarchical model. Lancet Glob Health. 2018;6(4):e380–9.
35. Floyd R, Ebrahim S, Tsai J, O'Connor M, Sokol R. Strategies to reduce alcohol-exposed pregnancies. Matern Child Health J. 2006;10:149–51.
36. Ingersoll K, Frederick C, MacDonnell K, Ritterband L, Lord H, Jones B, Truwit L. A pilot RCT of an internet intervention to reduce the risk of alcohol-exposed pregnancy. Alcohol Clin Exp Res. 2018;42(6):1132–44.
37. Doi L, Cheyne H, Jepson R. Alcohol brief interventions in Scottish antenatal care: a qualitative study of midwives' attitudes and practices. BMC Pregnancy Childbirth. 2014;14(1):170.
38. Scobie G, Woodman K. Interventions to reduce alcohol consumption during pregnancy. Edinburgh: NHS Health Scotland; 2017.
39. Sweeney PJ, Schwartz RM, Mattis NG, Vohr B. The effect of integrating substance abuse treatment with prenatal care on birth outcome. J Perinatol. 2000;20(4):219.
40. Frankenberger DJ, Clements-Nolle K, Yang W. The association between adverse childhood experiences and alcohol use during pregnancy in a representative sample of adult women. Womens Health Issues. 2015;25(6):688–95.
41. Black KI, Day CA. Improving access to long-acting contraceptive methods and reducing unplanned pregnancy among women with substance use disorders. Subst Abuse Res Treat. 2016;10:SART-34555.
42. Thanh NX, Jonsson E, Moffatt J, Dennett L, Chuck AW, Birchard S. An economic evaluation of the parent–child assistance program for preventing fetal alcohol spectrum disorder in Alberta, Canada. Adm Policy Ment Health Ment Health Serv Res. 2015;42(1):10–8.
43. Poole N, Schmidt RA, Green C, Hemsing N. Prevention of fetal alcohol spectrum disorder: current Canadian efforts and analysis of gaps. Subst Abuse Res Treat. 2016;10:SART-34545.
44. Hanlon-Dearman A, Malik S, Wellwood J, Johnston K, Gammon H, Andrew KN, Maxwell B, Longstaffe S. A descriptive study of a community-based home visiting program with preschool children prenatally exposed to alcohol. J Popul Ther Clin Pharmacol. 2017;24(2):361–e71.
45. Steer C. Fetal alcohol spectrum disorder—what are the social care needs? The picture of FAS in Scotland—surveillance and vignettes. In: Proceedings of the Scottish Government; 2013.
46. Roozen S, Peters GJ, Kok G, Townend D, Nijhuis J, Curfs L. Worldwide prevalence of fetal alcohol spectrum disorders: a systematic literature review including meta-analysis. Alcohol Clin Exp Res. 2016;40(1):18–32.
47. O'Leary LA, Ortiz L, Montgomery A, Fox DJ, Cunniff C, Ruttenber M, Breen A, Pettygrove S, Klumb D, Druschel C, Frías JL. Methods for surveillance of fetal alcohol syndrome: the

fetal alcohol syndrome surveillance network II (FASSNetII)–Arizona, Colorado, New York, 2009–2014. Birth Defects Res A Clin Mol Teratol. 2015;103(3):196–202.
48. Burns L, Breen C, Bower C, O'Leary C, Elliott EJ. Counting fetal alcohol spectrum disorder in Australia: the evidence and the challenges. Drug Alcohol Rev. 2013;32(5):461–7.
49. Andermann A, Blancquaert I, Beauchamp S, Déry V. Revisiting Wilson and Jungner in the genomic age: a review of screening criteria over the past 40 years. Bull World Health Organ. 2008;86:317–9.
50. Greenmyer JR, Klug MG, Kambeitz C, Popova S, Burd L. A multicountry updated assessment of the economic impact of fetal alcohol spectrum disorder: costs for children and adults. J Addict Med. 2018;12(6):466–73.
51. Canadian Association of Paediatric Health Centres. National screening toolkit for children and youth identified and potentially affected by FASD [Internet]. 2012. www.ken.caphc.org/fasd/fasd-national-screening-tool-kit. Accessed 20 Dec 2016.
52. Abernethy C, McCall KE, Cooper G, Favretto D, Vaiano F, Bertol E, Mactier H. Determining the pattern and prevalence of alcohol consumption in pregnancy by measuring biomarkers in meconium. Arch Dis Child Fetal Neonatal Ed. 2018;103(3):F216–20.
53. Bryanton J, Gareri J, Boswall D, McCarthy MJ, Fraser B, Walsh D, Freeman B, Koren G, Bigsby K. Incidence of prenatal alcohol exposure in Prince Edward Island: a population-based descriptive study. CMAJ Open. 2014;2(2):E121.
54. Delano K, Koren G, Zack M, Kapur BM. Prevalence of fetal alcohol exposure by analysis of meconium fatty acid ethyl esters; A National Canadian study. Sci Rep. 2019;9(1):2298.
55. Popova S, Lange S, Chudley AE, Reynolds JN, Rehm J. World Health Organization International Study on the prevalence of fetal alcohol spectrum disorder (FASD). 2018.
56. Scottish Intercollegiate Guidelines Network (SIGN). Children and young people exposed prenatally to alcohol. Edinburgh: SIGN; 2019. (SIGN publication no. 156). http://www.sign.ac.uk.

Approaches to National Support Groups: Advocating for Resources at a National Level

35

Sandra Butcher

Chapter Highlights

- Stakeholders view on FASD policies and practice.
- Approaches to reducing harm.
- National and regional approaches to influence strategy.

In a book by experts for experts, what does a layperson, a mum, have to contribute? Many readers will be wondering this, as they consider whether or not to skip this chapter.

But there is a different question that needs asking.

Given the lack of official attention to FASD, is there not a special need to listen to the voices of those whose lives are most impacted? And then there are related questions: How do non-experts get 'heard' on a medical issue that has no traction? What choice does a mum like me have, what options do laypeople and families have when the medical profession with all of its weight and glory refuses to engage? In short, how does national change come about?

When it comes to FASD, the answer is, nobody really knows. True systemic change on this issue has not yet happened anywhere. While a few countries like Australia, Canada and the USA are ahead of others, FASD has not yet broken through the wall of indifference in the way other medical conditions have. I am an American who has lived in the UK for most of my parenting journey. While we often look to the North America, for example, as being so far ahead in this area, the reality is, there is not one country in this world that has put its full weight behind reducing incidence of FASD and supporting those who have it.

S. Butcher (✉)
The National Organisation for FASD (formerly NOFAS-UK), Ware, Hertfordshire, UK
e-mail: sandra.butcher@nationalfasd.org.uk

R. A. S. Mukherjee, N. Aiton (eds.), *Prevention, Recognition and Management of Fetal Alcohol Spectrum Disorders*, https://doi.org/10.1007/978-3-030-73966-9_35

Imagine the national campaigns that have been endorsed and encouraged regarding heart disease, lung cancer or diabetes. Like FASD, these are conditions that have a preventive element as well as the need to support those with the conditions. Or consider the widespread attention and increasing awareness regarding autism, a similar neurodevelopmental disorder. Imagine the public safety efforts like wearing seatbelts, helmets on bicycles—preventive messaging designed to protect against head injury.

Even in those countries that are leading the way, FASD is still a whisper in the wind compared to these other truly national campaigns. Even in those countries with what are currently considered the 'gold-standard' of research, diagnosis and care for those with FASD, the reality is that most people seeking services and support still need to explain what FASD is to the majority of professionals, family and friends with whom they engage. They meet a wall when seeking diagnosis and support – the 'coalface' as Pip Williams (founder of the UK and EU Birthmothers Network-FASD) called it.

35.1 The Role of Stakeholders

This is where the charitable/non-profit community chips away, comprised of and standing side by side with other stakeholders. These include individuals with FASD, parents, carers, relatives, guardians, friends, supporters and a few professionals who stand with them seeking change.

I would argue that while advocacy by those most affected is a necessity, it is an undue burden. At their moments of highest need, individuals with FASD and their families and carers have to bash away at the stigma and professional indifference they face looking for answers that are not yet found in 'evidence-based' studies. They seek diagnosis, care and support that are not yet detailed in official guidance, and strategies that are not yet taught in medical schools. These 'non-experts' have to scan the existing literature, dig out the information needed and pass it along to any and all who might hear. Not because they want to do this, but because they have no choice. And they often do this in the face of dismissal and sadly sometimes open derision or hostility from those with degrees on the walls. The stigma runs deep, as does professional defensiveness.

Meanwhile, our children are suffering. Individuals with diagnosed and undiagnosed FASD are unseen, slipping into lives of desperation and vulnerability. Their trajectories could be changed if only this condition were 'seen' and addressed, if only political will existed to bring about the change that is needed, the change our loved ones deserve. To society's shame, disability rights have not yet been extended fully to cover those with organic brain damage resulting from exposure to alcohol in utero. This is appalling as there can be no purer innocence than having one's life so drastically affected before ever seeing the sunlight or taking a first breath.

With a neurodevelopmental disorder like FASD, the realities of this condition cannot be ignored or swept under the carpets in our homes. FASD can affect every

aspect of a person's life. It impacts families in countless ways. Despite the many and varied strengths of those with FASD, without diagnosis and support, most of the experience can be quite negative and detrimental—behaviours and secondary conditions can become so extreme and immediate that families are at breaking point.

So, while 'experts' have a luxury to debate these issues at a snail's pace, those 'lay-people' who do engage are often driven to it out of desperation. We become 'those' parents, 'those' patients, the kind for which many busy clinicians have little time in systems that focus on pre-packaged responses and answers to most medical conditions. The pathways are not found in most computer systems that a busy GP might seek for clues. Most official bodies do not yet supply the answers.

With FASD, as the saying goes, 'the path is made by walking it'. We need clinicians and policy makers to forge ahead even when there is a sparsity of data. We end up pleading.

It is incredibly frustrating. Even we laypeople know that a lack of 'proof' does not mean the evidence would not exist if researchers did the studies, if governments and funders properly resourced the research and if the academic posts and departments incentivised research in this field. We know with certainty this condition exists and it needs to be addressed. And we believe that clinicians will come to see this too.

As this book shows, this tide is now turning and what research does exist can no longer be ignored as more and more studies in various countries replicate similar results.

Traditional advocacy in the face of official and professional indifference is not easy for anyone to do, especially in cultures like the UK where doctors, teachers and other experts are not used to being questioned or challenged and where the pub culture runs deep.

Leaving it to non-professionals to bring about the change can be an impossible ask for those families on the edge with little resilience, capacity and sometimes with complicated profiles involving foster care, adoption, vulnerable adults, sometimes addictions or trauma. Sometimes these are intergenerational challenges. And yet, for decades, that is exactly where this field has been, with some notable exceptions from leading experts (many of whom are contributing to this book) who have seen the need to engage with stakeholders and who have opened up the field to other professionals. Even though the change is slow, the transformation is underway.

There is a hurdle that must be overcome. Many physicians, scientists and others with related degrees believe it is not their role to get involved in the policy realm. They have been led to believe science and medicine is somehow 'pure' and that becoming involved in policy or indeed politics weakens their professional authority or objectivity as scientists or clinicians.

It is a false dichotomy.

Dr. Frank Von Hippel, a leading physicist and one of my mentors, referred to the 'citizen scientist' in a book he devoted to this issue [1]. He argues that scientists live in society and have a responsibility to engage in policy.

Some of the greatest societal advances come when there is a synergy between those with academic, scientific and professional expertise and stakeholders and policy makers. I had a first-hand view of this in my prior involvement with scientists who received the 1995 Nobel Peace Prize for their work informing policy makers and the public on the implications of developments in weapons of mass destruction [2]. Many of those scientists had to first overcome the qualms about stepping away from the ivory tower and rolling up their sleeves to join in the messy policy world.

35.2 The UK Example

So what does this all say about FASD policy change? Let us use the UK as an example.

Modern references to fetal alcohol syndrome start to appear in Hansard in January 1979. This roughly coincides with some of the first diagnoses in North America in the early 1970s. Discussion in the UK at the time focused on heavy maternal drinking and other compounding factors.

The consistent response over decades from Government and the scientific community was that there was no proof or too little proof as to how many were affected in the UK and, therefore, it was not possible to determine changes to policy. But from the very start, there was an awareness both that studies were needed and that the UK was behind in research being done elsewhere.

For example, in January 1979, Mr. Moyle, Secretary of State for Social Services, said, 'My Department's consultant adviser on alcoholism…concluded that in this country FAS [Fetal Alcohol Syndrome] was unlikely to be a great problem numerically; that it should neither be ignored nor seen out of perspective' [3]. Sir George Young, Secretary of State for Social Services, said in February 1980, 'There is as yet no clear professional consensus on the fetal alcohol syndrome (FAS) in this country' [4]. Lord Cullen of Ashbourne said in January 1981, '[R]esearch studies abroad have pointed to the existence of a fetal alcohol syndrome—a range of mental and physical abnormalities observed in a number of children born to heavy drinking mothers. Others suggest that minor degrees of handicap may also affect the offspring of more moderate drinkers. There is no consensus of clinical opinion in this country. The DHSS has urged the Medical Research Council to consider as a priority the need for fundamental research into the relationship between drinking in pregnancy and fetal harm' [5].

A similar warning was echoed in a 1981 publication by the UK Office of Health Economics, "[T]he prevalence of this syndrome is unknown although three large epidemiological studies are underway in the United States. It would at present be irresponsible to overstate the danger of drinking in pregnancy. Kessell (1977) has commented that at the moment the evidence 'is not strong enough to justify any statement that women who drink moderately during pregnancy are harming their unborn children'. However, concern is now mounting that limited drinking may have damaging consequences (Dickerson et al. 1981)" [6].

This failure to fund the studies coupled with the refusal to act because of a 'lack of proof' became the consistent and ongoing Catch 22 that defaulted UK policy to inaction and stasis. While other countries went ahead and sought the data, the UK only had its first ever large-scale screening prevalence study [7] conducted by a team of researchers led by Dr. Cheryl McQuire in late 2018.

Think about that—this study came out nearly 40 years after the issue was first raised in Parliament and the UK Government identified a need for research on this topic. As of this writing, the UK has yet to put its resources into funding studies and has failed to ensure that surveillance systems appropriately track FASD cases. In contrast, the USA funded a major active case ascertainment study in four different communities that took place from 2010 to 2016 [8]. There are some additional UK prevalence studies being done at the time of this writing, but none are at the scale needed.

Perhaps this discussion is not what you were expecting from the contribution of a layperson? I would argue that it is the single-most central point.

This lack of official and academic attention is why the opinions of mums, families, carers and individuals living with FASD are salient, urgent and demand respect.

The official wheels have moved too slowly on this topic. Advocates had no choice but to step in. Local support networks developed. Regional groups formed in some places. Some national voices emerged. Emerging technology and social media helped to amplify voices. The system, this 'third sector' has grown organically. It was (and is) messy and inchoate. But it has promoted and pushed for change over decades when so-called experts failed to step in.

A series of charities over time have addressed FASD in the UK using different models for change: FASAwareUK (2004–2016), National Organisation for FASD (formerly NOFAS-UK) [9] (2003-), FASD Network-UK (2011-), FASD Trust (2007–2018), UK & European Birth Mother Network-FASD (2010-) and FASD Scotland (2012-) are some of the larger groups who work or have worked alongside other support groups spread throughout the UK.

The FASD UK Facebook support group was started by Maria Catterick (FASD Network UK) and Pip Williams (Birth Mothers Network) in February 2012. As of November 2020, it has roughly 2900 members. According to Catterick, 'FASD UK was [started by] UK based groups and individuals choosing to come together as a test to see whether social media technology could provide effective online support to families affected by FASD in the UK. The FASD UK Alliance (Birth Mothers, FASD Network UK and NOFAS-UK) was established in 2014 to support the developments of FASD work in the UK and provide support to emerging groups and coordinated responses to advance awareness of FASD and promote social change' [10]. The FASD UK Alliance has since grown and feeds the voices of stakeholders into national debates.

These third-sector groups are involved in awareness raising. They advise locally, regionally and nationally on FASD-related issues. Perhaps most importantly, they provide support to individuals and families that is so desperately needed given the complete gap in service provision.

These charities have worked when possible with professionals to seek policy change. Some bodies such as the British Medical Association [11] and the Royal College of Midwives have taken a particular and powerful interest in the field. A handful of exceptional clinicians and experts have helped to call for change. Baroness Professor Sheila Hollins, Dr. Raja Mukherjee, Dr. Moira Plant, Dr. Kieran O'Malley, Dr. Kate Ward, Dr. Helen Palmer, Dr. Cassie Jackson, Dr. Neil Aiton among others and a handful of other medical professionals have led the way in seeking to raise awareness among colleagues in the medical profession. An increasing number of younger researchers are starting to engage on these issues.

However, it has widely become recognised that science gains credibility when it teams with stakeholders. This is not a comfortable twinning for some experts, especially if those stakeholders are linked in with advocacy groups.

And yet, in the UK, as elsewhere, there is a trend for all projects to include a component involving stakeholder consultation. Indeed, in England, for example, it is a requirement 'Under the National Health Service Act 2006 (as amended by the Health and Social Care Act 2012), CCGs and NHS England have duties to involve the public in commissioning, (under sections 14Z2 and 13Q respectively)' [12].

And yet too often that is a cosmetic add-on and stakeholders are not engaged in the framing of studies, in building research from the ground up or in identifying the questions that need to be asked. Despite their 'lived experience', 'advocates' are not always taken seriously when attempting to change consensus on a medical issue.

From the patient/parent perspective, there is nothing more irritating or infuriating when trying to get diagnosis and/or support than to be looked down upon by someone with a degree who thinks they know more about it than you do. When it comes to FASD, sadly that is in the vast majority of times simply not the case.

While this chapter focuses on national policy, it must be acknowledged first that some of the most significant and far-reaching change happens on a purely individual level, in those one-on-one meetings with various professionals, especially the rare but appreciated clinicians who view engagement on a 'new' issue to be an opportunity to learn and to grow.

So, the first way national groups and other charities help change national policy is by providing stakeholders bashing away at that 'coal-face' with the knowledge, resources, confidence and resilience to have those quiet, one-on-one discussions with their doctors, teachers, social workers and so on. Armed with the latest information, individuals and families affected by FASD can at times push across that line of professional indifference or ignorance and, thus, expand the circle of awareness.

The second way these groups can have impact is to start to link people up to each other, to develop a shared sense of community and foster a sense of identity. Whether via informal coffee mornings or in regional and national conferences, through social media or in people-to-people gatherings, these groups open up topics of discussion and break through the isolation that too many experience. This is especially

important with a neurodevelopmental disorder like FASD, when the popular perception is that it is rare and the individuals and families grappling with it are isolated.

The third way is via media, seeking a wider spotlight on the challenges. This provides its own challenges given the tendency of the media to sensationalise the issue.

But those three things on their own (individual empowerment, developing a sense of community and publicising the issue via the media) do not bring about systemic change. A concerted approach to revise policy is needed.

In the UK to date there have been some policy successes and some signs that things are changing. But the biggest gains have not yet necessarily been on a national level. Some believe that that the more local progress is the most long-lasting and powerful because when local areas train up and sort out pathways this can provide the most immediate support. I would argue there needs to be both, bottom-up and top-down change.

35.2.1 Alcohol-Free Pregnancy Guidance

Despite the fact that some 40 years ago people in the UK were already raising concerns about risks of lower levels of alcohol in pregnancy, it took until 2016 for the Chief Medical Officers who revised the UK advice on alcohol in pregnancy [13] to firmly state, 'If you are pregnant or think you could become pregnant, the safest approach is not to drink alcohol at all, to keep risks to your baby to a minimum'. This change came about as a combined effort by the third sector working in cooperation with experts. Even then, due to incomprehensible bureaucracy, it took an additional 2 years until 2018 for the guiding body, NICE (the National Institute for Health Care and Excellence), to change its information on this topic. The 'alcohol-free pregnancy' message remains as of yet muted. There has not been a concerted national effort by the UK bodies to focus a truly national educational campaign on the risks of alcohol in pregnancy and issues related to FASD. This guidance was buried in wider 'low-risk' drinking guidelines. It needs to make its way into school PHSE curriculum. It needs to be included in large-scale ad campaigns.

35.2.2 Labelling

Another area of limited success in the UK has been in pushing for labelling. Charities and experts were involved in providing background and impetus for these changes. A Private Members Bill by Lord Mitchell was passed on 1 July 2008. The Bill requires alcohol companies to voluntarily put warning labels on alcohol containers. To date this labelling has not been made mandatory. That said, a report by the Alcohol Health Alliance in August 2020 said that 'Almost all (97%) labels displayed a pregnancy warning logo, but only 15% of labels included written

information about the risks of drinking during pregnancy' [14]. In some other countries, groups have advocated and pushed for point-of-sale warnings in local areas. The 2004 Sandy's Law [15] in Ontario is one example. As of the time of writing, a similar regulation was being considered in Michigan in the USA. While it has been explored on a small scale in some areas in the UK, some advocates see warnings at the point of sale as a possible national step in the UK.

35.2.3 Regional Progress

The most successful example of widespread change so far has been regional, specifically in the North East where Maria Catterick has through the FASD Network UK built a model integrated regional approach [16] resulting in innovations in prevention, diagnosis, education and support pathways for FASD for which she received the Big Society Award in 2013. The FASD Network UK now supports more than 1000 families and works strategically with all levels of the public health and care sector to support increased awareness of FASD and has conducted regional research. Greater Manchester committed £1.6 m in transformation funding to a 2-year 'proof of concept' programme to reduce alcohol-exposed pregnancies and progress towards and ambition of ending new cases of Fetal Alcohol Spectrum Disorder. The project implementation commenced in June 2018 and is engaging with stakeholders in creating materials and in its meetings. This initiative will also include regional research. Other areas have tried to implement FASD pathways and awareness training, with more limited success.

35.2.4 Westminster

In Westminster, an All-Party Parliamentary Group on FASD was formed in 2015 by Bill Esterson, MP—a Member of Parliament who also FASD. The FASD Trust served as the first secretariat of the group, a role that NOFAS-UK took over in 2019. The APPG on FASD has served as a link with policy makers, promoting awareness and action on FASD.

In 2018, then Deputy Chief Medical Officer Gina Radford convened what are believed to be the first government-initiated roundtables on FASD. One pivotal meeting with stakeholders and leads from several governmental departments took place in May 2018—the core of this meeting involved six teens and adults with FASD sharing their experiences with seeking diagnosis and support in their daily lives. Perspectives of birth mothers, adoptive and foster parents also informed the discussion. NOFAS-UK was commended by then Health Minister Steve Brine for helping with this process. A public health registrar was for the first time seconded by the Department for Health and Social Care to work for up to a year exploring

possible ways forward on FASD policy. Following this meeting, the UK Government issued some of its strongest statements yet on FASD, with the then Health Minister Steve Brine stating that there is 'is not an ounce of complacency in us…We will continue to work towards improvements in the area' [17]. It remains to be seen if these promising steps translate into policy change.

35.3 National Guidance on FASD Diagnosis and Care

Another area of change is with regards to national diagnostic and care pathways.

In partnership with leading clinicians, charities have promoted this in various ways. For example, in 2011, a team of leading experts created a 'consensus document' to define 'a model for service delivery and development in the UK' [18]. The groundbreaking exercise was organised in cooperation with the FASD Trust and included input from stakeholders.

Scotland has conducted a multi-year process that resulted in publication in 2018 of SIGN 156, 'Children and young people exposed prenatally to alcohol' [19]. Eileen Calder of FASD Scotland was involved in the process. NICE has been requested by Government to look into ways in which the SIGN Guidance can be applied in England [20]. The hope and expectation would be that stakeholders are engaged in this decision-making process.

Such national guidance is needed in an area like FASD where there is so little training and expertise available. In England, current Government policy states that 'Responsibility for commissioning Fetal Alcohol Spectrum Disorders (FASD) services lies with clinical commissioning groups working together across all sectors' [20]. And yet, charities and support groups regularly receive feedback from families and individuals that this is simply not happening. Families and individuals are consistently turned away due to lack of commissioned services and lack of funding. They have been channelling anecdotal evidence into policy streams, seeking to encourage a parliamentary inquiry or a Green Paper on this.

NOFAS-UK released in April 2019 results from freedom of information requests sent out to all CCGs and NHS trusts to test what they are doing in implementing government policy. The results indicate that very few are meeting this need. The report, 'A Crisis of Commissioning: CCGs Are Failing Government Policy on FASD' (2019), had a big impact. Its finding showed:

- Not one CCG said they have a policy on commissioning services for fetal alcohol spectrum disorders.
- Nearly 80% said they do not provide diagnosis for children with FASD.
- Ninety-two per cent said they do not provide diagnosis for adults.
- Only 19% of Trusts and Health Boards said that they provide post-diagnostic services for those with FASD [21].

On the day this report was featured at an All-party Parliamentary Group meeting in Parliament (9 May 2019), the Department of Health announced that the National Institute for Health and Care Excellence (NICE) would develop a quality standard on FASD based on the Scottish SIGN 156 Guideline [22]. The specialist committee for the quality standard includes stakeholders (including this author as a layperson) and FASD experts. Now due to be finalised in 2021, the NICE quality standard will be one of the single-most important changes to UK policy. The process is a good example of stakeholders' voices being included. Many of the FASD UK Alliance groups were registered stakeholders and sent feedback via the consultation process. The groups also collectively gathered stakeholder input from two different questionnaires including feedback from 330 and 230 people, respectively.

As of the time of writing, the five areas covered in the draft quality standard are as follows (these may change after the consultation process): advice to pregnant women, record keeping, referrals, neurodevelopmental assessment and management plans. [fetal alcohol spectrum disorder quality standards: www.nice.org.uk]

While quality standards are not mandatory, CCGs are supposed to 'have regard' for them and demonstrate how they are improving quality of care.

35.4 Department of Health and Social Care Funding

The Department of Health notably also released funding for 'Fetal Alcohol Spectrum Disorder (FASD) Interventions', which were awarded in the financial year 2020–2021 through the Children of Alcohol Dependent Parents (CADeP) Programme 'to support initiatives in the voluntary sector to support those living with FASD, and to reduce the number of alcohol exposed pregnancies to prevent new cases of FASD'. The funds were allocated through Section 64 of the Health Services and Public Health Act 1968.

It remains to be seen if funding will be provided in future. While a welcome sign, this initial grant programme is only scratching the surface.

35.5 Promoting Joined Up Support for Pregnant Women

Groups have focused attention on the need for better support for pregnant women. Pip Williams, founder of the UK EU Birth Mothers Network-FASD, emphasised that 'Birth mums of those with FASD are not getting the same consistency of care other women are due to stigma and due to the lack of training of professionals on how to have these discussions. Support needs to be joined up—statutory, community organisations, women's centres, peer support and online support should all be running together. Women need to be more included in decision making around their

child' [23]. Some progress was made on a local level. The UK and EU Birth Mothers Network-FASD was part of a 2-year research project working with maternity commissioners in London via Birth Companions and Revolving Doors. A report is now available [24].

35.6 The Adoption and Foster Care System

Another national avenue for change is promoting greater attention to the disproportionate impact FASD has on looked-after children and those who are adopted. Dr. Ges Gregory (Peterborough's Looked After and Safeguarding Designated Doctor) and former Virtual School Head Brian Roberts have drawn attention to figures associated with Peterborough's looked after children. According to Roberts, '34% of the children undertaking their annual medicals in the year prior to publication had a birth mother who we knew drank during pregnancy and showed signs of FASD…. More significantly the figure that we came around to for those children undertaking adoption medicals was considerably higher. 75% of those assessed had a birth mother who drank during pregnancy. In training that I co presented on in 2011 the LAC Doctor in Brighton was quoting a figure of 90% for children adopted under the age of two' [25].

Over the past decade there have been many meetings with the Department for Education on this topic. There has been increasing attention paid to this issue, but little research done and too little concrete action. Families and charities have worked hard to try to ensure that relevant professionals understand the importance of noting in files any exposure to alcohol during pregnancy, seeking to help provide training to foster carers and prospective adopters and to ensure that post-adoption support includes FASD. Despite the fact that in the care system there has been statutory guidance for those undertaking medicals of children in care since 2009 (Department for Education), this is widely misunderstood. The guidance says (paragraph 44) 'That Health Assessments should … pay particular attention to health conditions that may be more prevalent in looked-after children (such as fetal alcohol syndrome or attachment difficulties) and which may otherwise have been misdiagnosed' [26]. And yet, even with a recent restating of this from the Health Minister in February 2019 [20], too often families are denied access to FASD diagnosis and support because FASD is not recognised as a neurodevelopmental disability.

35.7 Training Professionals

The third sector has stepped in to provide trainings and continuing professional development courses for a range of professionals including medical, educational and social care professionals. Again, this has developed on an ad hoc basis and needs further attention to how the needs can be met on a wide-scale model given the

possibility that FASD may affect more than 6% of the population according to the November 2018 Bristol/Cardiff study.

FASD urgently needs to be included in training for midwives and GPs. For example, in 2017, NOFAS-UK conducted a limited poll of GPs [27]. Given lack of resources, the charity acknowledges that the results are of limited value statistically but stated, 'the answers we received strongly suggest that the powers-that-be should look more comprehensively into GP training and awareness of issues related to FASD and the risks of alcohol in pregnancy'. This is an urgent need and something that hopefully can be taken up further by national bodies that look at training of doctors, such as the General Medical Council and the Royal Colleges.

The NOFAS-UK GP survey was conducted by OnePoll between August 22nd and September first, 2017. It polled 150 GPs who work in England. Principal outcomes were as follows:

- Only 31% has in-depth education regarding FASD.
- Forty-one per cent have not received clear guidance regarding a pathway for diagnosis and support of FASD.
- Only 23% strongly agreed that they feel confident that all those with FASD are being diagnosed properly.

The NICE quality standard on FASD should lead to increased focus on training of professionals.

35.8 Hear Our Voices

A final point.

There is a social media meme that says, 'Be my microphone not my voice'. This to my mind is the primary role of national organisations seeking to influence FASD policy, to amplify the experiences of those who have FASD and their families and supporters.

With FASD, there is a double stigma to overcome. There is a misplaced perception that this issue does not affect all sectors of society. Most people are surprised to learn that it is older, more educated women who are most likely to drink in pregnancy, for example [28]. There also is a belief, including within the medical community, that those with FASD cannot be helped. This reflects old-school thinking that must be overcome. Many with FASD are not diagnosed or are misdiagnosed. There are very few adults in the UK with diagnoses so the numbers of those who are self-advocating is small, though increasingly powerful. Changing this public perception is key to the fundamental national mind-shift that is needed here in the UK. Empowering younger people with FASD by helping them to learn early on that they are not alone, that there are role models for them, is key.

Box 35.1 highlights what Georgia Roberts, a teen with FASD, summarised as a list of priorities [29]:

Box 35.1: Georgia Roberts' Summary
What I want is:

- People who know about FASD and can make a difference.
- People who listen to those who struggle. I know why I am different, and I know what works to help me to achieve.
- People who are committed to working with parents / guardians and carers to support them.
- People who understand the effects of FASD on minds and mental health and will support not make things worse. And mental health services who recognise and have services for those affected.
- People who will not give up on affected children and will support them growing up and especially at transitions and into adulthood.
- Real support for those of us with care experience as it adds to the challenges that we face. Despite the steps and progress made for children in care and adopted kids I have not been able to access the support.
- And finally, not to be blamed for my conditions, especially when people have been told how these conditions affect me.

There are still too many ignorant people. You wouldn't tell someone off who is blind because they can't see, but I get told off most days for things I can't help.

Samantha Hutt, a birth mother, summarised the challenges she faced seeking diagnosis and support for her child, 'My son was diagnosed in 2016 after many years of referrals to paediatricians, OTs, therapists, speech and language, CAMHS, school support workers, children's services and more. The list goes on. It's only when you look back over the years that you realise just how many professionals it took, to finally get to the point of diagnosis and support. Yet, I have had to explain what FASD is to almost every single one of those professionals' [30]. She and other birth mothers call for clear guidance to all women of childbearing years (indeed to all people, male and female), 'I'd like to think that had I been told about FASD and told not to drink, I would have done so, or at least asked for help.…Every pregnant woman has the right to be given correct guidance on alcohol in pregnancy. They have the right to be informed, warned about FASD. To be given the chance to be able to choose a different path, to safeguard their child. More needs to be done'.

This can start with increasing professional knowledge, with which this book will hopefully play a role. Box 35.2 highlights findings from a recent informal poll of 451 parent/carers in February/March 2019 written by Ali McCormick (with some input from me) for the FASD UK Alliance that identified what three things would most help caregiver's well-being. The overwhelming answer was 'professionals who know about FASD'.

Box 35.2: Findings from an Online Poll by the FASD UK Alliance

Respite care so I can have a break	39.37%
Professionals who know about FASD & know how to help my loved one	70.02%
Funding to make home safer/more sensory-friendly	17.67%
Appropriate support in school/workplace for my loved one	53.24%
Greater understanding of FASD among family/friends	29.53%
More information on strategies to help my loved one	30.20%
Knowing my loved one will have access to support/benefits as an adult	54.14%
Help with my own mental health/depression/relationship strains	18.79%
Being able to keep outside employment while caring for someone with FASD	17.00%

Which brings us back to the opening of this chapter.

What is a parent/carer/person with FASD to do in the face of professional indifference? The answer is educate, organise, disseminate and advocate on as many levels as you can until someone, some coalition of people within 'The System' takes note. And then, push harder until the topic breaks through the myriad bureaucratic and systemic hurdles. To do this, we need to find fellow travellers, including experts and clinicians who accept their role as citizen scientists who can help push for change.

This book is in and of itself proof that change is happening. But I would be failing in my role as leader of a national organisation if anyone felt complacent after reading this chapter that the job is anywhere near complete.

As Rachel Jackson, a young woman with FASD said in Parliament recently, 'I have had some help but there needs to be more, like for autism' [31].

So, why should an expert listen to a non-expert on these issues? Because, achieving truly national-level and transformative change requires that we work in partnership.

I write this as mum to a teen who has FASD. He is a bright light in a world that too often fails to understand him or his needs. Put simply, without experts and laypeople working together, he will not get the future he deserves. It is our synergy hat will create a brighter future for him and so many others.

References

1. von Hippel F. Citizen Scientist. Masters of modern physics. American Institute of Physics; 1991.
2. The Nobel Peace Prize 1995 was awarded jointly to Jospeh Rotblat and the Pugwash Conferences on Science and World Affairs. Prior to becoming chief executive of NOFAS-UK I served as the international executive director for the Pugwash Conferences. https://www.nobelprize.org/prizes/peace/1995/summary/.
3. Moyle. Secretary of state for social services. Hansard. 23 Jan 1979;961. https://hansard.parliament.uk/Commons/1979-01-23/debates/ccd1c7bf-d01e-448b-86a5-03a8b9da5c1e/FoetalAlcoholSyndrome?highlight=foetal%20alcohol#contribution-cedc3b85-4f7c-46c0-8d2e-5f96fd9e7462.
4. Young SG. Secretary of state for social services. Hansard. 12 Feb 1980;978. https://hansard.parliament.uk/Commons/1980-02-12/debates/14553bfd-7ab0-43eb-97b8-19c8b508397b/Pregnancy(AlcoholDangers)?highlight=foetal%20alcohol#contribution-51df8a54-df1c-4951-b0af-95180c1408b2.
5. Lord Cullen of Ashbourne. Hansard. 19 Jan 1981;416. https://hansard.parliament.uk/Lords/1981-01-19/debates/f724c694-afe5-432d-a3c3-185575209355/PregnancySmokingAndAlcoholConsumption?highlight=foetal%20alcohol#contribution-f0e33dab-adf9-4128-a6e4-67d2d05ecf30.
6. Taylor D. Alcohol: reducing the harm. Office of Health Economics;1981. https://www.ohe.org/publications/alcohol-reducing-harm.
7. First UK estimates of children who could have conditions caused by drinking in pregnancy revealed. University of Bristol Press Release. 30 Nov. 2018. http://bristol.ac.uk/news/2018/november/first-uk-prevalence-estimate-fasd.html. McQuire C, Mukherjee R, Hurt L, Higgins A, Greene G, Farewell D, Kemp A, Paranjothy S. Screening prevalence of fetal alcohol spectrum disorders in a region of the United Kingdom: a population-based birth-cohort study. Prevent Med. 2019;118:344–51. https://www.sciencedirect.com/science/article/pii/S0091743518303323?via%3Dihub.
8. May PA, et al. Prevalence of fetal alcohol spectrum disorders in 4 US communities. JAMA. 2018;319(5):474–82. https://jamanetwork.com/journals/jama/fullarticle/2671465.
9. Note: NOFAS-UK changed its name to The National Organisation for FASD in 2020.
10. Catterick M. Private correspondence with the author. 29 Mar 2019.
11. Alcohol and pregnancy: preventing and managing fetal alcohol spectrum disorders. First published in 2007, updated in 2016. https://www.bma.org.uk/collective-voice/policy-and-research/public-and-population-health/alcohol/alcohol-and-pregnancy.
12. Patient and public participation in commissioning health and care: Statutory guidance for clinical commissioning groups and NHS England. NHS England. 6 Apr 2017. https://www.england.nhs.uk/publication/patient-and-public-participation-in-commissioning-health-and-care-statutory-guidance-for-ccgs-and-nhs-england/. Accessed 28 Mar 2019.
13. UK Chief Medical Officers' low risk drinking guidelines. 2016. p. 8. https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment_data/file/545937/UK_CMOs__report.pdf.
14. Drinking in the dark: how alcohol labelling fails consumers. Alcohol Health Alliance;2020. https://ahauk.org/drinking-in-the-dark/. Accessed 27 Aug 2020.
15. Sandy's law. An act to amend the Liquor Licence Act by requiring signage cautioning pregnant women that the consumption of alcohol while pregnant is the cause of Fetal Alcohol Spectrum Disorder, Assented to 24 Jun 2004. https://www.ontario.ca/laws/statute/S04012.
16. Greaves Z. Healthcare needs assessment: fetal alcohol spectrum disorders in the North East of England. Public Health England;2016. Final report of the health scrutiny panel: foetal alcohol

spectrum disorder (FASD). Middleborough. http://democracy.middlesbrough.gov.uk/aksmiddlesbrough/images/att1012499.pdf.
17. https://hansard.parliament.uk/commons/2019-01-17/debates/19011751000002/FoetalAlcoholSpectrumDisorder
18. Consensus statement regarding the recognition and diagnosis of foetal alcohol spectrum disorders across the lifespan in the UK: development of proposed UK clinical pathways. Report on the findings from the first National Medical and Healthcare Professionals Conference held on the 12th and 13th October 2011. https://www.researchgate.net/publication/259558397_Consensus_Statement_Regarding_the_Recognition_and_Diagnosis_of_Foetal_Alcohol_Spectrum_Disorders_Across_the_Lifespan_in_the_UK_Development_of_Proposed_UK_Clinical_Pathways.
19. SIGN 156. Children and young people exposed prenatally to alcohol. 2019. https://www.sign.ac.uk/sign-156-children-and-young-people-exposed-prenatally-to-alcohol.html.
20. Brine S. Written answer. 19 Feb 2019. https://www.parliament.uk/business/publications/written-questions-answers-statements/written-question/Commons/2019-02-11/219508/?fbclid=IwAR2OQfn8jzCarfRVAd07ULjGmLaomkwyzoyW5lS6hD6bcx63gwXFXB-GBfE.
21. A crisis of commissioning: CCGs are failing government policy on FASD. 2019. www.NationalFASD.org.uk.
22. NICE plans new standard for fetal alcohol disorders. BMJ. 2019;365. https://doi.org/10.1136/bmj.l2186. (Published 13 May 2019).
23. Williams P. Presentation, foetal alcohol spectrum disorders: future policy development on issues relating to FASD. A roundtable discussion with FASD stakeholders convened and chaired by Professor Gina Radford, Deputy Chief Medical Officer National Liberal Club, London. 22 Oct 2018. www.NationalFASD.org.uk.
24. Cardwell V, Wainwright L, et al. Making better births a reality for women with multiple disadvantages: a qualitative peer research study exploring perinatal women's experiences of care and services in north-east London. Birth Companions and Revolving Doors Agency; 2018. http://www.revolving-doors.org.uk/file/2333/download?token=P2z9dlAR.
25. Roberts B. Presentation, quoted in foetal alcohol spectrum disorders: future policy development on issues relating to FASD. A roundtable discussion with FASD stakeholders convened and chaired by Professor Gina Radford, Deputy Chief Medical Officer National Liberal Club, London, 22 Oct 2018. www.NationalFASD.org.uk.
26. Promoting the health and well-being of looked-after children: statutory guidance for local authorities, clinical commissioning groups and NHS England. Mar 2015. https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment_data/file/413368/Promoting_the_health_and_well-being_of_looked-after_children.pdf. Accessed 27 Aug 2020.
27. NOFAS-UK GP Survey. Summary of key findings. Sept 2017. Participants were recruited online and were paid. www.NationalFASD.org.uk.
28. https://www.reuters.com/article/us-usa-health-pregnancy/older-u-s-women-more-likely-to-drink-while-pregnant-study-shows-idUSBRE86J03B20120720
29. Roberts G. Presentation, first UK-based prevalence study on FASD: implications for policy makers report of the all-party parliamentary group on FASD roundtable discussion. 13 Dec 2019. www.NationalFASD.org.uk.
30. Hutt S. Presentation, first UK-based prevalence study on FASD: implications for policy makers report of the all-party parliamentary group on FASD roundtable discussion. 13 Dec 2019. www.NationalFASD.org.uk.
31. Our forgotten children: the urgency of aligning policy with guidance on the effects of antenatal exposure to alcohol. A roundtable discussion with FASD stakeholders co-chaired by Professor Sheila the Baroness Hollins and Mr. Bill Esterson, MP Houses of Parliament. 23 May 2018. www.NationalFASD.org.uk.