Children with Cerebral Palsy: The impact of ankle-foot orthoses on gait function after lower limb surgery

PhD Thesis

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Summary

Background

Children with cerebral palsy (CP) frequently undergo orthopaedic lower limb surgery to improve and maintain functional gait. In the post-operative period ankle-foot orthoses (AFOs) are considered important to preserve the effect of the surgery (1-3). Such orthoses are typically constructed according to biomechanical guidelines specified in preoperative three-dimensional (3D) gait analyses and treatment plans. The AFOs are usually worn all day and combined with intensive physiotherapy and training.

The results of the surgery, exercises and orthotic regime are routinely evaluated by 3D gait analysis one year postoperatively. At this point, patients and carers often expect that AFOs are no longer necessary and could be discontinued (4). Nevertheless, continued use of AFOs is often recommended due to residual gait problems or risk of recurrence (5-7). There was however a lack of evidence and few existing studies evaluating the impact of AFOs on gait function after lower limb surgery in children with CP.

Aims

The aim of this PhD project was to investigate impacts of AFOs on gait quality, kinematic, kinetic and temporal spatial gait variables at the time of routine 3DGA one year after lower limb surgery in children and adolescents with spastic CP. Our objectives were to identify whether walking with AFOs provided additional improvement, the predictors for improved gait with AFOs, and indications for continued use of AFOs after the one-year follow-up. We hypothesized that many children have residual gait problems after surgery and that AFOs provide mechanical support to enhance gait function one year postoperatively.

Methods

In a cohort of ambulating children with bilateral (n=34) and unilateral (n=33) spastic CP we used an observational, repeated measures design to evaluate changes in gait after orthopaedic lower limb surgery, and additional changes walking with AFOs. Outcome was evaluated using the gait profile score (GPS)(8), ground reaction forces (GRF), sagittal plane kinematics and kinetics, walking speed, step length and cadence, with paired sample comparisons and logistic regression (Paper I), linear mixed models (Paper II and III), and functional mixed effects analysis (Paper III).

Results

Paper I: In the cohort with bilateral CP (n=34) major improvements postoperatively included the GPS, kinematic and kinetic variables, whereas walking speed and step length declined. Walking with AFOs walking speed and step length was enhanced, and additional improvements were seen in the GPS, max ankle dorsiflexion and knee extension moment. Crouch was reduced in children who used ground reaction AFOs (n=14), whereas more moderate changes were seen in the children who used hinged AFOs (n=20). A high preoperative GPS was the strongest predictor of clinically important improvement with AFOs one year postoperatively.

Paper II: In the cohort with unilateral CP gait patterns mainly changed from true equinus to drop-foot walking barefoot postoperatively. The GPS, most kinematic, and all kinetic variables improved, whereas cadence was reduced. The main impacts walking with AFOs in this group was improved walking speed and step length, prepositioning of the foot and heel initial contact due to reduced plantarflexion and knee flexion in the AFOs, corroborated by non-significant improvement of the GPS. Effect of AFO type confirmed increased stance and swing phase ankle dorsiflexion with hinged AFOs versus solid AFOs.

Paper III: Functional curve analysis of vertical GRF components (vGRF) showed improvements postoperatively. With AFOs increased vGRF in weight acceptance and increased late stance forces equivalent to bodyweight indicated clinically important improvement of CoM support and stance stability compared to barefoot one year postoperatively. This additional improvement with AFOs was most pronounced in children with unilateral CP.

Conclusion

Major improvements were found between pre- and postoperative barefoot walking. Residual gait problems such as remaining crouch and dynamic ankle equinus/drop-foot were the main indications for continued use of AFOs one year postoperatively. We demonstrated the mechanical-functional efficacy of AFOs to further improve gait function one year postoperatively at activity, body functions and structures levels (9).

List of Papers

This thesis is based on the following Papers. They are referred to in the text by their Roman numerals.

I. Skaaret I., Steen H., Terjesen T., Holm I. **”Impact of ankle-foot orthoses on gait 1 year after lower-limb surgery in children with bilateral cerebral palsy”** *Prosthetics and Orthotics International. Epub August 2018/ 2019 43:1,12-20 DOI:10.1177/0309364618791615*

II. Skaaret I., Steen H., Huse AB., Holm I. **”Comparison of gait with and without ankle-foot orthoses after lower-limb surgery in children with unilateral cerebral palsy”** *Journal of children's orthopaedics. 2019 13:2,180-189 DOI:10.1302/1863-2548.13.180146*

III. Skaaret I., Steen H., Niratisairak S., Swanson D., Holm I**. ”Postoperative changes in vertical ground reaction forces walking barefoot and with ankle-foot orthoses in children with cerebral palsy”** Submitted to Clinical Biomechanics July 2020. Accepted for publication March 2021

Abbreviations

AFO	Ankle-foot orthosis
BoNT-A	Botulinum neurotoxin A
CoM	Centre of Mass
CP	Cerebral Palsy
CPOP	Norwegian Cerebral Palsy Follow-up program
CPRN	Cerebral Palsy Registry of Norway
ICF	International classification of functioning and disability
3D	Three-dimensional
2D	Two-dimensional
FZ1	First peak of the vertical ground reaction force
FZ2	Second peak of the vertical ground reaction force
GPS	Gait profile score
GRF	Ground reaction force
vGRF	Vertical ground reaction force
GVS	Gait variable score
MAP	Motion analysis profile
ND	Non-dimensional
PreBF	Preoperatively walking barefoot
PostBF	Postoperatively walking barefoot
PostAFO	Postoperatively walking with ankle-foot orthoses

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1 Background

1.1 International Classification of Functioning, Disability and Health (ICF)

The ICF is a universal framework developed by the World Health Organization to help describe how body structure and function, activity, participation, environmental and personal factors are related and how disability influences health and functioning (9). The term functioning denotes all body functions, activities and participation, whereas disability refers to impairments, restrictions in activity and in societal participation. The ICF represents a shift in focus, from treating the disability to emphasizing health and asking what is important for each person to improve their functioning with regards to meaningful activity and participation. The ICF framework is intended to help identify both the level of capacity, i.e. what persons can do in a standardized environment, and their performance in usual environments (10).

Within the ICF, products and technology for personal use and mobility such as orthoses, and health services such as orthopedic surgery, are coded and defined as contextual environmental factors that can influence body functions and structure, and the individual’s ability to execute actions or tasks. In our study cohort consisting of children with CP, dysfunction in body functions (reflex, muscle tone, strength) and structures (range of motion, joint mobility) impaired their ability to walk and stand, which affected participation in life situations requiring ambulation and adequate body alignment. Outcomes were reported in terms of walking under the body functions and structures, and the activity domain of the ICF. This included summary measures of gait quality, 3D kinematic, kinetic and temporal-spatial variables. Since the children were walking in a standardized gait lab environment, functioning was evaluated with regards to *capacity*, rather than performance during their activities of daily living.

1.2 What is cerebral palsy? Prevalence, classification and motor function

Cerebral palsy (CP) is an umbrella term covering a heterogeneous group of motor impairment disorders secondary to brain injuries that occur before, during or immediately after birth (11). With a worldwide prevalence of 1.5-3.3 in 1000 live births, it is the most common cause of physical disability in children (12). In Norway, 2.1 in 1000 live born children were diagnosed with CP during years 1996 -98 (13). Between 1999 and 2010 the prevalence decreased significantly from 2.62 to 1.89 per 1000 live births (14). Of the registered cases in Norway per 2018, 58% were male and 42% female.

Children who are diagnosed with CP in Norway are enrolled in the CP registry (CPRN) and the CP follow-up program (CPOP) to monitor prevalence, motor development, habilitation processes, deformity, health services and interventions. CPRN was recognized as a national medical quality registry for children with CP in 2006 to collect essential data at the time of diagnosis, at 5 and 15-17 years. CPOP started in 2006 as a project involving the south-east region of Norway, and was implemented in all regions in 2010, with systematic yearly examinations and longitudinal registration of hand and gross motor function, joint range of motion, spasticity, and interventions towards motor function. X-ray imaging of the hips and spine is performed at intervals specified by age and functional level (15). CPRN is situated in Vestfold Hospital and CPOP in Oslo University Hospital.

Symptoms and clinical manifestations of CP may take long to identify, particularly in the lower levels of severity, and the average age of diagnosis for children in the Norwegian CP registry is currently 25 months (15). The diagnosis may be observed as a delay in reaching developmental milestones; however, diagnostic tools such as magnetic resonance imaging to map the extent of the brain injury have improved early diagnosis and complement clinical-neurological tests in evaluating the likely motor development and function.

The diagnosis is commonly classified as *spastic*, characterized by hyperactive stretch reflexes and hypertonicity; *dyskinetic*, observed by involuntary movements and velocity-independent rigidity (extrapyramidal, athetoid/dystonic); or *ataxic*, recognized by instability and coordination failure (extrapyramidal); according to what type of abnormal tone is more dominant. In Norway, 86% of children in the follow-up program are classified with spastic CP, 7% with dyskinetic, 4% with ataxic, whereas 3% are unclassified (15). In this thesis, we focus on ambulatory children and adolescents classified with spastic CP.

Spastic CP is divided into topographical subtypes according to the anatomical distribution of involvement. Persons with unilateral, spastic CP, or hemiplegia, are affected in the arm and leg on one side. Bilateral spastic CP includes diplegia and quadriplegia. Diplegia implies that the lower limbs are more strongly affected, while quadriplegia is more severe as it affects both upper and lower limbs. Of the children with spastic CP in Norway, 40% are categorised with unilateral and 46% with bilateral involvement (15).

Spasticity is initiated by injury to the cortical areas of the brain and damage to the upper motor neurons. Initial flaccid weakness is followed by reduced inhibition of motor neurons, with velocity-dependent, increased stretch reflex excitability and abnormal muscle tone which may subside with time (16). Dyskinetic movement disorders such as hypotonia and dystonia may occur in combination with spasticity. Spasticity is often accompanied by persistence of primitive motor patterns, reduced selective motor control, impaired balance, and postural changes. Muscle tone and co-contraction may be task-dependent and increase with activities such as walking. Other, non-motor, conditions such as impaired cognition, vision and communication are common and could also affect the level of functioning and disability. These impairments may be classified as the primary effects of the brain injury, affecting body functions.

Spasticity may cause muscular agonistic and antagonistic imbalance, leading to lower extremity joint contractures and secondary musculoskeletal impairment (17). Besides hypertonicity, deviation may also be seen in muscular morphology, as increased stiffness and less contractile tissue. The functional/contractile muscle fibre unit sarcomere is composed by actin and myosin proteins and contraction occur when the two slide past each other. Bundled together, the fibres form a fascicle, which is formed to individual muscles by connective tissue. Friden and Lieber tested the elastic properties of sarcomeres from human muscle biopsies and found systematically shorter sarcomere resting lengths and increased stiffness in spastic versus normal muscle fibres (18). Meanwhile, studies using magnetic resonance imaging and ultrasound have shown reduced lower limb skeletal muscle volumes by up to 50% in children with CP compared to typically developing peers (19, 20). There are also higher contents of intramuscular fat which for a given muscle volume reduces the contractile tissue content and contribute to muscular weakness (21).

The skeleton provides the lever arms for internal muscular and external ground reaction forces to generate movement around skeletal joints. Skeletal deformity thus contributes to secondary effects of the diagnosis and the musculoskeletal impairment. Persistent foetal alignment with excessive femoral anteversion and internal tibial torsion occurs when there is failure of skeletal remodeling after birth. In CP, this may be caused by developmental delay of physical milestones such as walking until the structures are more ossified and less malleable (22, 23). External tibial torsion is believed to develop secondary to femoral anteversion (24). The extent of musculoskeletal deformity in children with CP varies considerably and increases with age and decreasing motor function.

From the age of four, motor function in persons with CP is commonly measured using the gross motor function classification system, GMFCS (25). The classification system has five levels with clinically identifiable distinctions between levels of severity referring to usual performance in daily living. Children in Level I ambulate in all terrain, can run, jump, and climb stairs without support. In Level II, children walk without support, require support in stairs, and have difficulties to run, jump and walk in uneven terrain. Children in level III usually walk with assistive devices such as crutches or walkers. In level IV children may have limited ambulation with walkers but usually ambulate using electric or manual wheelchairs. In level V children have extensive gross motor function problems and are dependent on wheelchairs and are assistance-dependent. In Norway, the CP follow-up program report that the majority (69%) of the 1415 children registered in years 2002-2017 are classified within the higher functioning GMFCS levels I (52%) and II (17%); 6% are in level III, 9% in level IV and 13% in level V, whereas 3% are unclassified (15). In the current thesis the children were ambulatory within levels I-III.

The functional mobility scale (FMS) (26) complements the GMFCS in rating gait performance and mobility over 5, 50, and 500 meters (m). The need and use of assistive devices on the respective distances is scored from 1-6; the highest score imply that the person performs walking on all surfaces with no support, whereas the lowest score means that wheelchair is used for ambulation. Children who are classified in GMFCS levels I and II usually score in FMS levels 5 or 6 over 5 or 50m. Over 500m distance the range of scores is higher, with generally lower scores in children with reduced motor function (27). FMS scores were not used in the present thesis.

1.3 Evaluation of Gait

Normal gait

Walking is a complex function that can reveal much information relevant to diagnosis and treatment of musculoskeletal disorders. Clinical gait analysis usually involves comparison of impaired or pathologic gait with normative gait curves from typically-developing peers. Therefore, it is necessary to understand what characterises the typical function and walking patterns in persons without gait impairments.

Human bipedal gait is dependent on several factors whereby the skeleton functions as the body frame and motion is enabled by balanced muscular activity initiated and modified by the central nervous system. These factors evolve through childhood and with normal development a mature gait pattern is established by the age of four to five, when kinematic and kinetic gait variables are within adult ranges (28). Temporal- spatial parameters such as step and stride length, cadence and gait speed are influenced by stature; i.e. body height and leg length and will therefore not reach fully mature values before growth has been completed (28, 29).

The study of walking is multi-factorial and involves measurements of *kinematics*; the study of movements between body segments, *kinetics*; the study of forces and moments producing the movements, *time and distance/temporal-spatial parameters;* such as walking speed, step length, step frequency, support time, and step width, *electromyography*; describing the phasic activity of muscles, and *energy-expenditure assessment* to measure the energy cost of walking. Comprehensive gait analysis also includes physical examination, with assessment of joint range of movement, muscle strength, tone, joint contractures, and skeletal deformities.

In this thesis we describe kinematics using the terms flexion/extension for motion in the sagittal plane, varus/valgus and ab/adduction for motion in the coronal plane, and rotation in the transverse plane. For description of foot motion, inversion refers to inwards hindfoot rotation towards the anatomical midline, and eversion refers to outwards hindfoot rotation away from the midline. Supination involve triplanar motion with plantarflexion, inversion and adduction, whereas pronation refer to dorsiflexion, eversion and abduction (1). The terms anterior/posterior tilt, obliquity, and rotation are used to describe motions of the pelvis respective to the lab axis system, whereas foot progression is used to describe internal/external placement of the foot relative to the direction of walking. The measured angular motions between joint segments and joint angle motion versus time are expressed in degrees and degrees/s (sec), respectively, even if the correct SI unit for degree is radian.

Kinetics comprise internal forces in muscles, joints and ligaments; and external forces such as the ground reaction force (GRF) with vertical components due to gravity, and horizontal shear forces caused by friction with the ground. When a force (N=Newton) acts with a distance (m=meter) to the joint axis of rotation (fulcrum), the perpendicular distance is called the lever arm, and the product is the joint moment (Nm). In this thesis we refer to external joint moments, i.e. an external knee extension moment pushes the knee into extension. According to Newton's 2nd law, force is the product of mass (g) and acceleration (m/s^2). A force of 1 N is required to accelerate a body mass of one kilogram (kg) 1 m/s^2, i.e. 1 N=1 kg · m/s^2, and a force of 1 N with a 1 m perpendicular distance from the force to fulcrum executes a turning moment (torque) of 1 Nm around the pivoting axis. Power (Watt) is the product of joint moment and joint angular velocity, and is interpreted as power generation or absorption by the muscle, implying concentric, eccentric or isometric contraction (30).

Gait variables are typically analysed using the gait cycle as measurement unit (Fig.1) A gait cycle starts with ground initial contact of one limb, lasts through stance (60%) and swing periods (40%) and ends with subsequent initial contact with the same limb (100%). One gait cycle include two successive steps with right and left limbs. The step length is the distance from the toe of the trailing foot to the point of contact with the opposite leading foot. The number of steps or gait cycles made per time unit is referred to as the cadence or frequency, in this thesis reported as steps per minute.

During stance, there is a period of about 20% with double support where both feet are on the ground simultaneously, which is what characterises walking versus running. With increased gait velocity, double support and stance time decreases. Walking ends and running begins the moment when there is no period of double support and stance duration is 50% of the gait cycle.

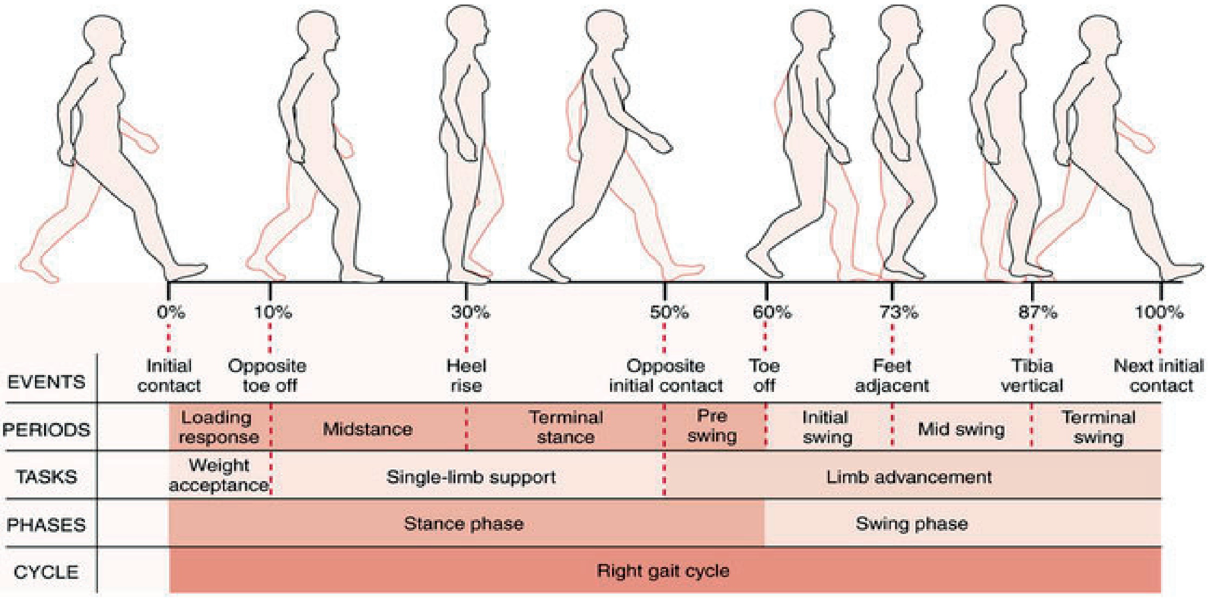


Figure 1. The gait cycle; phases, tasks, periods and events. By permission from Neumann DA: Kinesiology of the musculoskeletal system; foundations for physical rehabilitation. Ed 2, St Louis, 2010, Mosby, Figure 15-6

Major tasks have been defined that should be accomplished during the stance and swing phases of the gait cycle (31). The first task after initial contact is weight acceptance or loading response, which involves shock absorption through the heel pad and dorsiflexor controlled ankle plantarflexion until the foot is flat on the ground. This is also called the 1st (heel) rocker and is accompanied by slight knee flexion. The second task is single limb support during mid- to terminal/late stance. The ankle dorsiflexes in 2nd (ankle) rocker; while hindfoot inversion brings the foot axes into a stable, locked position that provides a solid lever for the plantarflexors to push against the floor. This brings the GRF point of application to move distally on the foot, increasing the lever arm to the ankle joint and thus increasing the ankle dorsiflexion moment. A substantial moment is generated that brings the GRF line of action anterior to the knee, while the knee moves into full extension. This stabilising feature is called the *plantarflexion-knee extension couple* and is considered the major mechanism responsible for bodyweight support and stability during stance (32, 33). In terminal stance, ankle plantarflexion with the 3rd (forefoot) rocker counteracts a large dorsiflexion moment created by the GRF at the metatarsal joints, to generate a power burst and assist forwards propulsion during push off. In the swing phase, limb advancement is the main assignment (31)**.** This requires propulsion, momentum, and sufficient ankle dorsal and knee flexion for foot clearance. In terminal swing, the foot remains plantigrade while the knee extends and the hip flexes to pre-position the foot for heel initial contact and complete the gait cycle.

During normal walking, the body center of mass (CoM) is continuously shifting upwards, downwards, and sideways. It is at its lowest during double support of stance and rising to its highest point in single support. The magnitude of the vertical GRF component varies about bodyweight due to CoM acceleration or deceleration and takes on a typical M-shape with two peaks, each normally above bodyweight (34-36). The resultant GRF vector changes direction according to horizontal shear forces (Fig. 2). Anterior-posterior forces are related to braking after initial contact, late stance push-off and pre-swing propulsion.

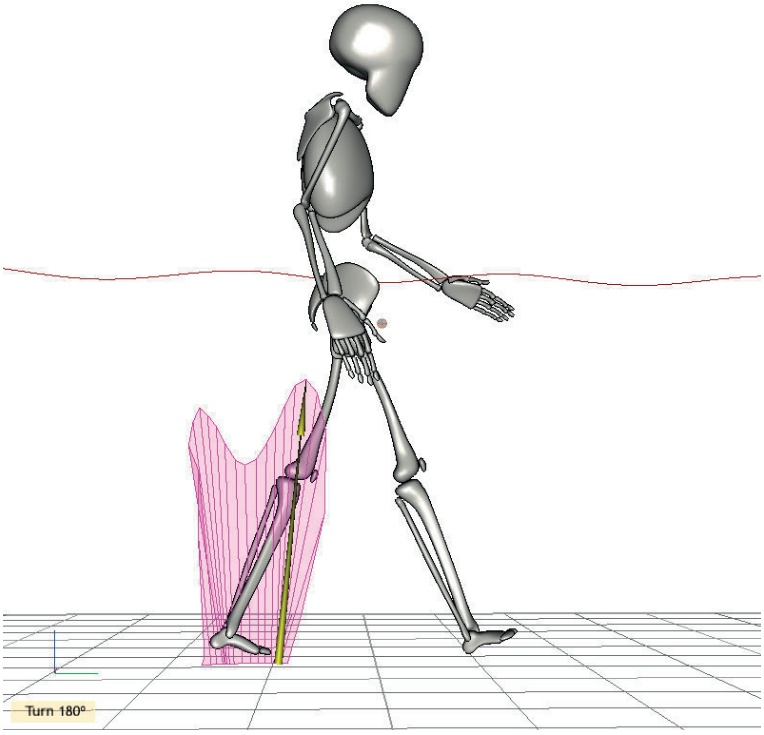


Figure 2. Visualisation of CoM trajectory (red line), the resultant GRF (yellow arrow) and butterfly diagram.(pink) during normal gait, based on data collected in the Oslo Gait Laboratory.

Saunders JB, Inman VT and Eberhardt HD defined the major determinants in normal and pathological gait. Their theory was that the determinants; 1) pelvic rotation, 2) pelvic list/obliquity, 3) knee flexion in stance, 4) foot rockers, 5) foot locking/unlocking mechanisms, and 6) knee valgus are features of human bipedal gait that serve the specific purposes to minimize the vertical excursion of the CoM and to help produce forward progression with less energy consumption. *"fundamentally locomotion is the translation of the center of gravity through space along a pathway requiring the least expenditure of energy"*(37). The determinants are still considered important as an interpretation of why we walk the way we walk. Nevertheless, some of these conclusions have in the last two decades been subject to discussion and in part refuted through clinical trials, observing that several determinants were not actually timed to reduce CoM movements (38, 39). An alternative and opposing theory, the inverted pendulum analogy*,* assumes that the human body need some degree of vertical CoM excursion while walking to increase the exchange between potential and kinetic energy and thereby enhance energy conservation (40). External work in normal gait is mainly required during step-to step transition and acquired with extension and heel-lift/push-off in the trailing limb (38) before initial contact and weight acceptance with the leading limb (41, 42).

The 'five priorities of normal gait' summarize the main tasks of functional human walking as: 1. Stability in stance; that the limb must be sufficiently stable to support bodyweight during loading, 2. Foot clearance in swing; the leading limb must swing free of the ground to avoid tripping, 3. Pre-positioning of the foot; in terminal swing the foot should be positioned and the limb prepared for heel initial contact, 4. Adequate step length and 5. Energy conservation (31, 43). These priorities may be helpful in identifying deviations and the main challenges in impaired gait.

Gait patterns in CP

Gait in CP can be classified into characteristic gait patterns using classifications that combine visual observations and quantitative kinematic 3D gait analysis data (3, 44-46). Such gait patterns are most clearly seen and defined in the sagittal plane during mid and terminal stance and refer to postural patterns based on positions of the ankle, knee, and hip. A description of the most common deviations and patterns that characterise gait in CP follows in this section.

Skeletal growth combined with triceps surae spasticity and muscle fibre stiffness, insufficient stretch, and ineffective dorsiflexor antagonists could result in dynamic or static plantarflexion contracture of the hindfoot relative to the ankle; namely *ankle equinus*. During stance, ankle equinus increases the plantarflexion-knee extension couple, moving the ground reaction force anterior to the knee joint with enlarged knee extension moment. This could effectively over-stabilise and push the knee into *hyperextension,* or *recurvatum*. Ankle equinus may also be combined with insufficient knee and hip extension during stance. A distinction should be made between *'true'* or *'apparent equinus'*, in which the latter describes toe-walking due to excessive knee flexion, but with normal ankle range of motion (3, 45).

Inadequate activation of the dorsiflexors (m. tibialis anterior) due to impaired selective motor control and/or weakness could lead to *drop-foot* during swing, causing tripping and foot clearance problems, and characterised as a 'type I' pattern in unilateral CP (Fig. 3) (3, 46, 47). Drop-foot occurs, especially in unilateral CP (27) although most regularly following surgical correction of equinus (3, 48).

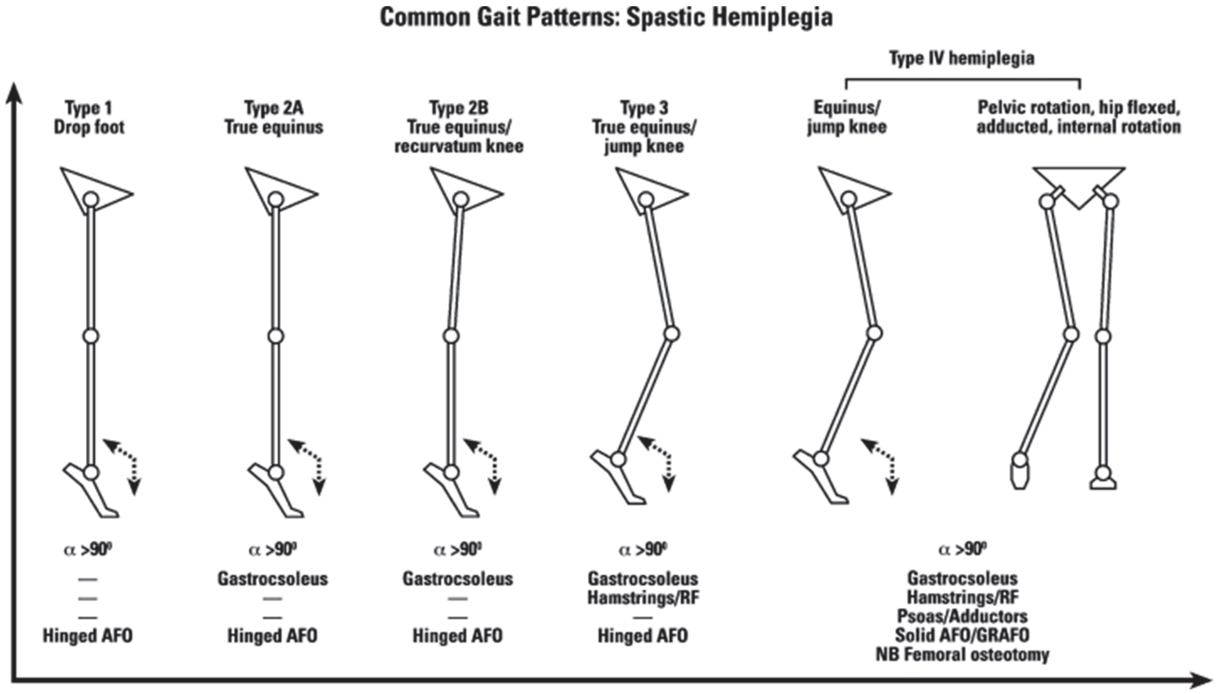


Figure 3. Common gait patterns in unilateral (hemiplegic) CP. By permission from Rodda J & Graham HK, 'Classification of gait patterns in spastic hemiplegia and spastic diplegia: a basis for a management algorithm'. Eur J Neurol. 2001;8 Suppl 5:98-108.

Ankle equinus is most frequently seen in unilateral spastic CP (27, 49), but is also common in bilaterally affected children. The condition regularly diminish with time, due to reduced spasticity, weakness and/or surgical overlengthening of the triceps surae (50). This could result in *calcaneal gait*, with increased ankle dorsiflexion, insufficient plantarflexion-knee extension couple, diminished stance stability and development of *'crouch'*, characterised by an inability to achieve appropriate knee extension during the stance period. Spasticity and contracture in the structures flexing the hip and knee are other components involved in a crouched gait pattern. (43, 51-54). Excessive knee flexion at initial contact is a common feature which is related to hamstrings and/or rectus femoris spasticity. Some subjects manage to achieve full, or near full knee extension during stance despite the increased knee flexion at initial contact. This pattern would typically be labelled as *jump knee* gait (45).

Crouch mainly develops in bilateral CP and occurs with varying levels of severity. Severe crouch has been described as knee flexion >30° throughout the stance phase of gait, combined with excessive ankle dorsiflexion and incomplete maximum hip extension (6, 55).

A feature which frequently occurs in combination with crouch is *stiff knee gait* due to tightness or spasticity in the rectus femoris, blocking the knee flexion required for adequate swing clearance. Stiff knee gait may be seen in the sagittal plane kinematic gait curves as diminished knee range of motion through the gait cycle, with reduced and delayed peak knee flexion in swing (3, 44, 45, 56).

Inadequate skeletal remodelling may cause transverse plane gait deviations with internal hip rotation, in-toeing (49, 57) and asymmetric pelvis rotation (58). External tibial torsion may cause excessive out-toeing and a malrotation of the foot, resulting in a functionally shorter foot lever-arm that affects the location and magnitude of external GRF forces (59). Excessive tibial torsion has been found to alter the line of action, and reduce the moment generating and extension capacity of the ankle plantarflexors, namely soleus (60, 61) with adverse proximal impacts on several muscles crossing the hip and knee (60). Combined femoral anteversion with hip internal rotation, external tibial torsion with out-toeing, and sometimes foot and ankle instability are referred to as the *malignant malalignment syndrome* (43) which impairs the plantarflexion-knee extension couple, introduces valgus and rotational moments about the lower limb joints, and diminished input to the vertical acceleration of the body CoM. Rotational gait deficits may be difficult to evaluate by visual observation, are very often masked by compensatory movements (ex. external pelvis rotation to offset internal hip rotation secondary to femoral anteversion), and are difficult to diagnose based on computer tomography or physical examination (62).

The obvious difference in gait patterns between topographical types of CP is only one affected side in unilateral CP. Even though the clinical manifestation in unilateral CP vary widely, there is generally a higher level of gross motor function and lower level of impairment in this group (27). The non-affected side is more capable to work against gravity and could maintain an extended posture, which is probably why this group is less predisposed to develop crouch (6). Gait deviations are common even in the non-affected side, mostly to compensate for deficits on the affected side. One example is *vaulting*, i.e. limb extension with early heel-rise and plantar flexion during stance to aid foot clearance of the opposite side in swing.

Pattern recognition has also been suggested that involve evaluation of ground reaction forces. Previous studies found that vertical GRF patterns in CP children diverge from typically developing children, often demonstrating excessive forces in weight acceptance, and reduced forces in late stance below bodyweight (Fig. 4) (36, 63). The magnitude of the vertical peaks have been used to describe stability in stance and ability to support bodyweight, and could be used to assess results of clinical interventions related to weight-bearing and CoM support in children with CP (32, 36, 63).

CoM displacement can be computed from the vertical GRF component and is related to the mechanical and muscle work production that explains the energy cost of walking. Children with CP have shown 1.3 times higher vertical CoM displacements than normal, which was associated with increased total positive mechanical work performed by the muscles, and mainly due to an equinus gait pattern (64, 65). Walking takes more energy in children with CP than typically-developing peers (66, 67), and the differences in energy cost increases with age and according to GMFCS levels (68).

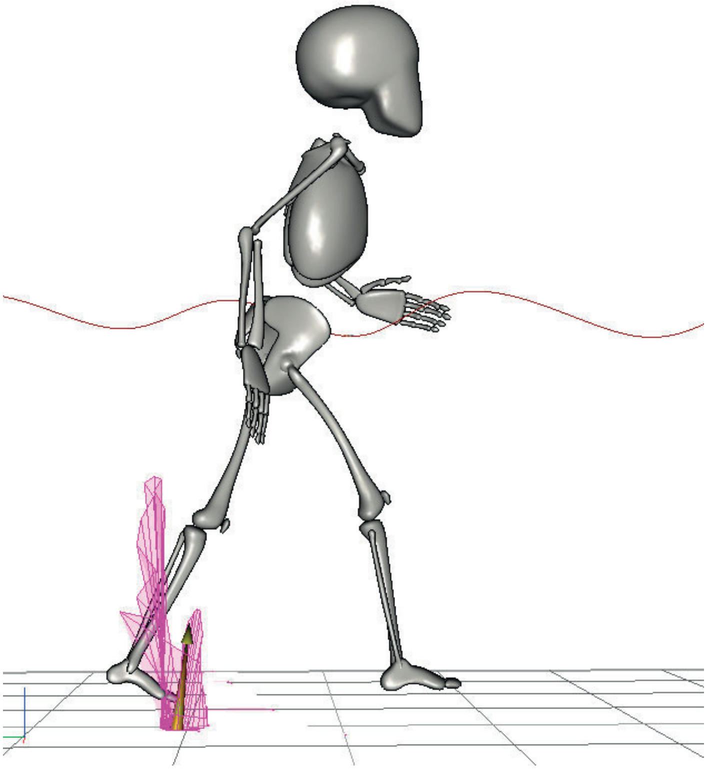


Figure 4. Visualisation of CoM trajectory (red line) with increased vertical excursion, diminished late stance GRF (yellow arrow), and butterfly diagram.(pink) during gait in a seven year old child with bilateral CP, based on data collected in the Oslo Gait Laboratory.

Measuring gait

In his *De Motu Animalium* Aristotle (384-322 BC) wrote down the first known observations and thoughts regarding human gait. Visual observations of patients’ gait are still vital for many clinicians and professions where the most important functional outcome is to help the patient stand upright and achieve ambulation in the most efficient way. Several observational gait assessment tools have been validated and designed for use in clinical settings which does not involve specialized equipment or location (69). Such tools can enhance the quality of assessment, often by filming patients with cameras positioned to observe sagittal and coronal plane 2D gait.

The theoretical basis required for 3D measurements of human gait engrosses a multitude of scientific disciplines such as mathematics, physics, anatomy, physiology, and biomechanics that were mainly founded during the European renaissance and Enlightenment period. The French philosopher and mathematician René Descartes (1596-1650), defined the *Cartesian* coordinate system with x, y and z orthogonal axes for measuring positions of objects in space. A clinical gait laboratory that measures 3D motion would typically consist of several such coordinate systems. The room in which people are being measured is the *Euclidean space*, a concept invented by the ancient Greek mathematician Euclid (325-265 BC), as defined by Cartesian x, y and z coordinates in a 'global' coordinate system. The global coordinates define the origin, and axes are aligned with the anatomical planes; usually y with the sagittal plane in the direction of walking, x with the coronal plane, and z vertical through the transverse plane. Each high-speed motion capture camera, each video camera, each force plate, and the person being measured also have coordinate axes. In kinematics, Cartesian axes are not only used to estimate *positions* of objects in space, but also applied as *dynamic axes* about which rotations take place. Leonhard Euler (1707-1783) first introduced the three elemental rotations about the xyz axes, now known as *Euler angles* (70, 71).

The methodology used for 3D gait measurements today was at large demonstrated as early as the 1890's by the German mathematician Otto Fischer (1861-1917) and professor of anatomy Wilhelm Braune (1831-1892)(70). Developments during the 20th century, particularly in optical camera and computer science, meant a revolution with respect to utility and effectiveness of movement analysis for clinical purposes (71). In our time, human gait is measured using physical markers attached over anatomical landmarks; a 3D model based on a theoretical system of xyz axes imbedded in each body segment and aligned with joint movement axes. Position of the markers in Euclidean space is calculated with *triangulation*, using the angle and time delay between signals from high-speed cameras that emits infra-red light at a specific frequency, calibrated with exact distances according to each other and the global coordinates of the gait laboratory.

Sir Isaac Newton (1643-1727) published *Philosophiae naturalis principia mathematica* in 1687 and his three laws describing the interaction between forces and their effect on movement are an inescapable part of the kinetics in gait analysis (72). The 3rd law: that for every action there is an equal and opposite reaction, is elementary: While standing still, the body acts on the ground with a force equivalent of its mass or weight. From the ground there is an equally large and oppositely directed reaction force. In modern clinical gait laboratories, the size, direction, and torque of the GRF are measured by force plates along x, y and z axes. Joint moments are calculated using the resultant GRF multiplied by the perpendicular distance to the axis of rotation and with inverse dynamics this information is combined with 3D kinematics, segment acceleration and inertial characteristics to derive more comprehensive moments related to each single joint (73).

Orthopedic surgeon David Sutherland (1923-2006) appreciated the importance of understanding gait mechanisms and deficiency caused by conditions such as CP, and was a pioneer in implementing 3D gait analysis for clinical purposes, and description of normal development (28, 74) . Through nerve block experiments he identified the stabilizing role of the plantar flexors during gait (75). Colleagues Jacquelin Perry (1918-2013) and James Randolph Gage (1933-) were strong contributors in developing 3D gait analysis, to measure and identify gait dysfunctions in neuromuscular diagnoses, improve clinical decision making, refine interventions and evaluate treatment outcome (71, 75).

It has been suggested that Archimedes (287-212 BC) defined the laws of leverage that are elemental in biomechanics. However, from a clinical gait analysis perspective Dr. Gage will be known as the person who defined the ‘Star Wars principle of gait’, stressing that “the everyday forces imposed on the child’s muscles and bones play a large part in governing normal growth”. Gage increased awareness regarding skeletal and muscular *lever arm dysfunction*; the different types and how to balance the forces about the joints with clinical interventions, such as surgery, muscular strengthening and orthoses (43).

In assessing the mobility of persons with CP and other neuro-muscular diagnoses 3D gait analysis has become a widely used and recognised tool, particularly as rotational, out-of-plane motion is common and difficult to evaluate based only on 2D frontal and sagittal plane observations (62). Meanwhile, the 3D variables that are used to assess rotational deformity, e.g. hip rotation, are also those that exhibit the greatest measurement errors (76). Although the analyses are based on analytical calculations and objective measurements, interpretations, hypotheses, and treatment decisions are at large subjective and relying on the background, clinical experience, and knowledge of people in the multidisciplinary gait lab team (77). Nonetheless, results from 3D gait analysis have become essential for multi-disciplinary teams in making clinical recommendations concerning treatment of gait problems and in evaluating outcome after interventions (43, 78-80).

Deterioration of Gait and Functioning in CP

CP has been defined as a non-progressive, but often changing condition (11). While few infants have musculoskeletal problems, it is common that secondary pathology develops through childhood. In ambulating persons with CP, a change in the level of impairment commonly materializes as a decline in gait function over time. Eventually many individuals seize walking in adult age, experiencing reduced activity and restricted participation. The ability to maintain long-term functional gait is most likely in the lower ambulatory GMFCS levels. Factors contributing to gait deterioration are multifaceted, including abnormal muscle tone, weakness, fatigue and contractures as well as skeletal rotational malalignment and lever arm dysfunctions (43, 53, 81-83) .

The natural deterioration of gait function in bilaterally affected children is primarily linked to the development towards increased crouch (6, 49, 53, 54, 82, 84)**.** Increased age, weight, and insufficiency of muscles crossing the hip, knee, and ankle to work against gravity contribute to the difficulties. Rodda & Graham point to the transition from dynamic equinus to crouch gait as the normal progression and the most commonly observed change with age in bilateral spastic CP (Fig. 5) (3). Knee flexion during stance of ≥20° and loss of plantar flexion strength are factors found predictive of progressive crouch problems (51, 52, 85) .

Rethlefsen and colleagues evaluated the prevalence of gait deviations in more than 1000 patients with bilateral and unilateral spastic CP and GMFCS levels I to IV. The study confirmed that with increased age, the risk of crouch increased, primarily in children who could walk independently (levels I-III) and only in children with bilateral CP, while the prevalence of equinus and in-toeing decreased with age and higher GMFCS level. The authors recommended special caution when making treatment decisions in younger children, due to the potential for equinus and in-toeing to decrease naturally with age (49).

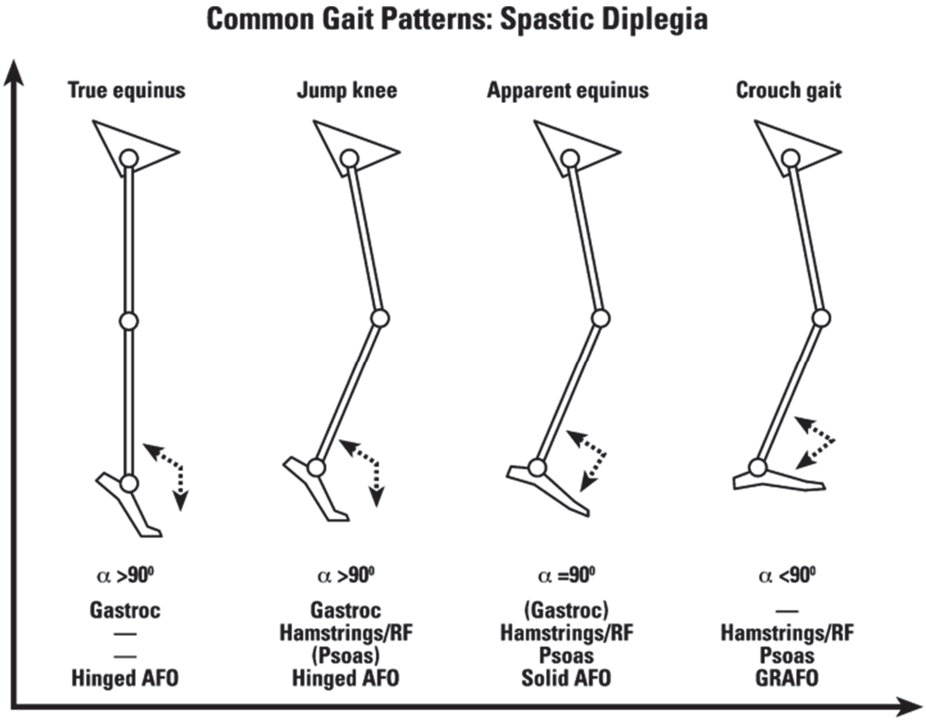


Figure 5. Common gait patterns in bilateral (diplegic) CP. By permission from Rodda J & Graham HK, 'Classification of gait patterns in spastic hemiplegia and spastic diplegia: a basis for a management algorithm'. Eur J Neurol. 2001;8 Suppl 5:98-108.

1.4 Treating gait problems in CP

Preserving optimal muscle length is important for maximising function and is the primary objective of conservative and medical intervention in the growing child with CP. Early treatment to prevent gait deterioration in CP usually comprise a combination of AFOs, physiotherapy, serial casting and spasticity-reducing treatment, with the objective to control spasticity, muscular imbalance, flexible deformities, and to suspend development of static contractures.

Physiotherapy for the growing child with CP often involve manual stretching to increase and maintain joint range of motion, with short-term outcome identified at the ankle joint (86). Recently, stretching of hamstrings and progressive resistance exercises gave non-significant improvements in popliteal angle and muscle strength (87), although the combined program did not cause differences in gait function (88).

Spasticity treatment includes intra-muscular injections of botulinum neurotoxin type A (BoNT-A) to reduce hypertonicity and preserve range of joint motion by providing temporary paralysis of spastic muscle fibres. It is usually injected in one to four large muscle groups in one session and is considered a focal intervention since there is little spread outside the muscle (89, 90). In Norway 50% of children with spastic CP had been treated with BoNT-A by the age of 5 years, increasing only slightly to 58% at ages 15-17 years (15). In ambulatory children with CP BoNT-A is most frequently utilized in the treatment of ankle equinus. A recent exploratory study found increased gastrocnemius length and reduced stretch excitability two weeks after treatment with BoNT-A and AFOs as usual, whereas a two-week casting period increased ankle range of motion due to increased Achilles tendon length (91).

Surgical release of spastic nerve roots with selective dorsal rhizotomy is a regional, invasive spasticity treatment, usually restricted to children who meet specified criteria with regards to muscle tone and age; mainly young children with bilateral spastic CP, good selective motor control and no symptoms of dyskinetic CP (43). A meta-analysis of three randomized controlled trials confirmed selective dorsal rhizotomy is effective to reduce spasticity and improve motor function, especially when combined with physiotherapy (92). Norwegian patients who could benefit from the treatment were until 2019 referred to Great Ormond Street Hospital for children in England, and by 2018, 13 patients had been treated (15). In 2019 implementation of the methodology and surgery started in Oslo University Hospital, in collaboration with the UK hospital.

A systematic review of interventions for children with CP established a traffic light system based on the quality of evidence. Green-light ‘do it’ interventions that had proved effective towards ambulation included BoNT-A, casting, physical therapy with fitness and goal-directed training, and selective dorsal rhizotomy. Orthotic and surgical interventions were graded yellow-light interventions due to a lack of randomized controlled trials (93).

Lower limb orthopedic surgery

Persons with CP who experience impaired ambulation due to fixed deformities frequently undergo lower limb surgery, with muscle tendon lengthening or transfer, rotational osteotomies and joint stabilising procedures (43, 55, 94). Equinus has been described as the most common deformity in CP (95). Indications for operation are usually failure of conservative treatment, and fixed equinus which interferes with balance, walking or wearing an AFO (6, 50, 96). Associated conditions, namely equinoplanovalgus; hindfoot equinus combined with mid- and forefoot pronation, and equinocavovarus; hindfoot equinus with mid- and forefoot supination deformity, may require tendon transfers and bony surgery to stabilise the foot lever arm and improve the base of support. An extensive body of literature describes increased passive and dynamic ankle range of motion following surgical lengthening at various levels of the triceps surae. However, a systematic review (97), a population-based study (6) and long-term follow-up (50) revealed concerns regarding calcaneal gait and crouch due to loss of effective plantarflexion- knee extension coupling after triceps surae lengthening with tendo-achilles lengthening in children with bilateral CP. Isolated tendo-achilles lengthening has a stronger indication in unilateral CP with fixed gastrocsoleus contracture and a 'true equinus' gait pattern (3, 6). Recurrent equinus is common in this group and repeated surgery may be required, particularly with young age at initial surgery (50, 98).

Since the 1990s, a change evolved in the standard of orthopaedic care, from performing single procedures at repeated occasions, to *single-event multi-level surgery* guided by comprehensive 3D gait analyses. Multi-level surgery implies operating all combined problems, such as skeletal malrotation and soft tissue contracture at different levels in one procedure and with one rehabilitation period. Surgery that addresses muscle tightness typically involve psoas lengthening in cases of hip flexion contracture, hamstrings lengthening to improve knee extension, rectus femoris transfer to correct stiff knee gait (99) and triceps surae lengthening to treat ankle equinus. More recently, distal femoral extension osteotomy combined with patellar tendon advancement have gained approval in the treatment of crouch gait with knee flexion contracture (100, 101). To a certain degree the procedure has replaced hamstrings lengthening, which has been associated with excessive anterior pelvic tilt and genu recurvatum. However, a systematic review evaluating surgical and non-surgical treatments of crouch summarized that hamstrings lengthening remained the single intervention which was most effective, as supported by clinical evidence (102).

A systematic review evaluating effects of single-event multilevel surgery in children with CP found enhancements in the ICF domain of body structure and functioning. Passive range of motion, gait-related kinematics and kinetics, summary statistics of gait quality and energy efficiency improved, whereas results concerning temporal-spatial, strength and muscle tone variables were less consistent (103). Several long-term evaluations reported sustained gait improvements 5-10 years postoperatively (55, 80, 104-107). However, a need for repeated or additional surgery after the initial surgery was reported in many studies.

Records from the Norwegian CP population confirm a relatively low rate (16%) of children who underwent orthopaedic surgery before the age of five. At 15-17 years the rate increased to 64%. Repeated surgical procedures also occurs; among the 310 children and adolescents who were operated between years 2006-17, 72 had been operated twice and 23 had repeated surgery up to 3-4 times (15). These data apply to all types of orthopaedic surgery at single or multiple levels.

A comprehensive postoperative rehabilitation program, individually tailored to optimise and maintain the surgical corrections is an important component of orthopaedic surgery in CP. This involves intensive training with physiotherapist-guided exercises 2-3 times or more per week with focus to regain and improve strength, maintain joint mobility, and automate gait patterns according to the priorities of normal gait (see section 1.3). An integral part of the postoperative regimen is the use of postoperative AFOs which are designed in accordance with gait pattern and treatment algorithms (2, 3, 55), as outlined in the preoperative 3D gait analysis.

1.4 Ankle-foot orthoses

Orthoses are “externally applied device(s) used to modify the structural and functional characteristics of the neuro-muscular and skeletal systems” (International organisation of standardisation (ISO) 8549, 1989). The devices are commonly named according to which joints are encompassed in the orthosis; they can be employed for all body segments and have a wide variety and range of purposes. In CP, interventions with AFOs are prescribed for all functional levels, and all types of CP (Fig. 6).

In ambulating children with CP, i.e. GMFCS levels I-III, AFOs are typically prescribed with the purposes to maintain range of motion, prevent or reduce development of deformity, manage functional limitations, and improve base of support, standing posture and functional walking. Some children require AFOs to facilitate skills exercises, for enhanced positioning and function during running and sports activities. With reference to the priorities of normal gait important mechanical purposes of AFOs during gait in CP would usually be to improve stability in stance, provide foot clearance and prepositioning for initial contact (2).

AFOs are considered an important tool to treat gait problems conservatively, in conjunction with spasticity-reducing medicine and after serial casting, and to prevent deterioration following surgical correction. The Norwegian CP follow-up program (2018) confirms widespread application, reporting that 56% (521 of 929) of the children use AFOs during the day, with effect in self-reported goals concerning prevention of contractures/deformities, improved function, and/or stability/balance in 84% (15). In 2011, 80% of the children with AFOs reported use for more than 5 hours per day.

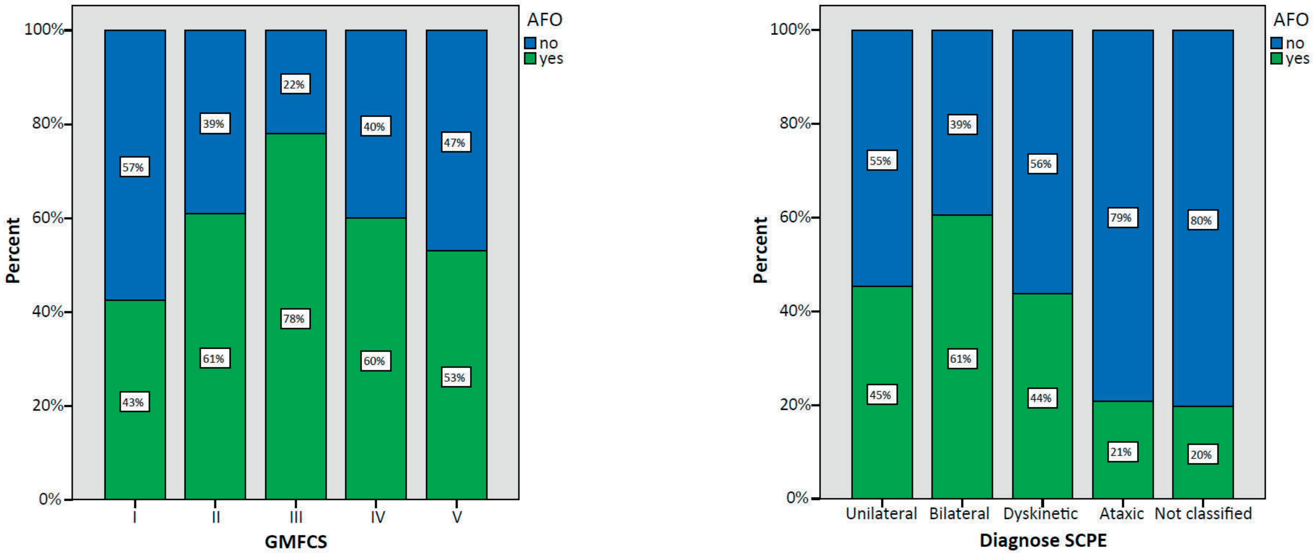


Figure 6. AFO use according to gross motor function and CP subtype. GMFCS; gross motor function classification system, SCPE; classification of CP subtype. From Myklebust et al, 2011. By permission.

Biomechanical function of AFOs

AFOs provide direct control of the anatomic joints integrated in the orthosis, i.e. the ankle and foot joints. The extent of controlling these joints depends on the orthotic design and mechanical properties such as material stiffness, durability, and range of motion, as allowed by integrated joints. The main AFO types are often referred to as solid, with no ankle motion or hinged; allowing ankle motion with a joint or material flexibility.

Using biomechanical terms, the lower limbs are kinetic chains, i.e. a system of linked rigid bodies. External force can be applied with orthoses in open- or closed linked chains (108). An example of an *open-chain* AFO force system is a three-point force distribution over the ankle during swing phase to prevent dynamic equinus/drop-foot; the limb is unloaded, allowing adjoining joints to move freely while ankle motion is controlled. A *closed-chain* force distribution relies on external segment load, for example from fixing the foot against the ground and 'indirectly' affecting motion in adjoining joints which are outside the orthosis (109). This is the mechanical idea behind a ground reaction AFO, where force is applied during stance and the distal end of the kinetic chain, the foot, is fixed against the ground. Ankle dorsiflexion and thus tibial progression over the foot is restricted by the orthosis during 2nd rocker, and through material stiffness, alignment, long distal and proximal lever arms the point of GRF application is brought distal on the foot. The perpendicular distance of the GRF anterior to the ankle and knee joint increases; resulting in enhanced external dorsiflexion moment, knee extension moment and potential reduction of crouch, as illustrated in Rodda et al (Fig. 8)(55). Similarly, AFOs that restricts plantar flexion have the potential to control knee hyperextension during stance by inhibiting the plantar flexion-knee extension couple (110). Both solid and hinged AFOs could possess similar mechanical properties of a ground reaction AFO, depending on material stiffness, the allowed range of movement and resistance in integrated joints or springs (111-114).

AFO measurement and fabrication

Prefabricated, off-the-shelf AFOs have proved effective in cases where light support is required, such as drop-foot (115). However, cases with dynamic or static contractures and deformity require individual measurement that entails "acquisition and recording of all information required to construct the orthosis by means that may include the preparation of diagrams, tracings, measurements and negative casts of the body segments" (ISO 8549 2.3.7). During casting/measurement, the ankle and foot is placed in the position where it functions optimally, usually determined by inspecting the available range of ankle dorsiflexion with the knee flexed and extended (Silverskiolds test). The length of lever arms with which the orthosis should act on the body part is determined along with correction of flexible deformities, positioning of the joints and sole of the foot for appropriate load and alignment of the body during standing and walking. The positive model is then modified to obtain a shape which indicate the form of the final orthosis *(*ISO 8549 2.3.8) and entails force distribution to pressure-tolerant and relief to sensitive areas, defining trimlines to influence mechanical properties and facilitate 'donning and doffing' of the device.

In fabricating orthoses, traditional metal and leather constructions have undergone a development which allows reduced weight applied to distal segments. Pre-impregnated carbon fibres provide similar stiffness to steel, but with less weight i.e. higher specific stiffness. Combined with other composites, stiffness and elasticity in the orthoses can be tailored to meet individual demands. Lightweight, vacuum-moulded thermoplastics or 3D printed nylon used with profiles or circular designs provide stability while allowing dynamic flexibility (116). Integrated joints include flexible plastic/rubber joints or metal joints which can be grinded to allow a certain ankle range of motion. In recent years, optimised AFO joint components are available that allow both plantar- and dorsiflexion with adjustment possibilities and elastic spring-controlled resistance (114, 117), carbon-fibre springs with energy-storage and return (111, 112, 118). However, active push-off with plantar-flexion beyond neutral ankle angle has so far only been possible with powered AFO-designs, which however have limitations for clinical use due to bulkiness and excessive weight of engines and energy sources/battery (119).

Assembly and alignment of the components should take place in accordance with patient characteristics and acquired data. Initial bench alignment would typically be refined while the orthosis is worn by the patient in standing and optimized by observing the patients' movement pattern (ISO 2.3.12-13).

Evaluation of AFO efficiency

To assess the efficacy of an orthosis it is important to identify the specific intention for using it. To enhance compliance with the device the outcome should be evaluated from a user perspective, using collaborative goal-setting that involves the patient, specifying the biomechanical rationale for prescription to match the child's needs and what one wishes to achieve with regards to body function and structure, activity and participation. Regular monitoring is desirable as there is often a need to revise treatment goals and ensure that orthotic intervention meets the purpose for prescription. When the objective is to improve gait, pattern recognition and remembering the five priorities of normal gait may be useful to determine the purpose for AFO prescription, and to specify orthotic function and design. Filming patients, preferably in both sagittal and frontal planes, is a valuable aid for visual observation and evaluation. Video vector systems that are marker-less and show the projection of ground reaction vectors from force plates on the walking subject is a convenient tool for fine tuning of AFOs, to check dynamic alignment and visualise the mechanical impacts of orthoses on gait to the user and caretakers (120). However, in many cases it may be difficult to determine AFO efficacy and refine orthotic prescription without assessing the child in a gait lab, comparing walking with and without orthoses. For research purposes 3D gait analysis is the gold standard assessment tool to objectively evaluate mechanical-functional impacts of AFOs (121).

Impacts of AFOs on gait in CP

The functional influence of orthoses in CP has been reviewed and the results confirm multiple positive effects in kinematic, kinetic, and temporal-spatial gait variables (122-125). However, some areas were identified that could benefit from further investigation and comprising higher standards of quality. This implies conducting studies with larger subject numbers, longer durations of follow-up and improved homogeneity regarding gait patterns and GMFCS levels.

Even if evidence that meet scientific standards do demonstrate the immediate biomechanical outcome of using orthoses in walking children with CP, further research is needed to document long-term implications of wearing orthoses and development of deformities over time (125). There is also a need for more complete and transparent research reports particularly when it comes to details concerning the construction, materials, stiffness and properties of the orthoses (122, 125-127).

Great variation has been found when it comes to clinical practice in orthotic prescription and management in children with CP (128). Most likely this is related to uncertainty about how different types of AFO's best enhance function, but local rehabilitation traditions and practice as well as cost-efficiency demands could be alternative factors.

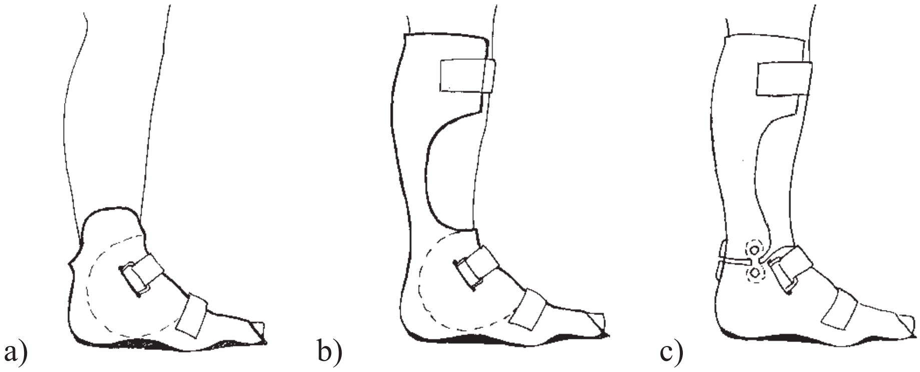


Figure 7: Examples of polypropylene AFO designs that are common in children with CP; a) supramalleolar, b) solid, and c) hinged AFOs. These types are usually made with 2.5-3.2mm flexible polypropylene-butylene, a circular total-contact fit which enables good stability and support of the structures in the foot. Motion is enabled with flexure joints, trim-lines and material reinforcements. Illustrations by Skaaret, I.

Within the activity domain of the ICF, research in children with CP has found consistent evidence that wearing AFO's helps improve their gait efficiency through increased stride and step length, maintaining walking speed whilst reducing cadence (129-133). Several studies found increased walking speed and step length, while cadence was unchanged (114, 131, 134). Significant decreases in energy cost (mLO2/kg/m) and oxygen consumption found when wearing AFO's compared to barefoot walking and/or shoes substantiate these findings (129, 130, 134-136). With regards to construction, research using simulation models has found that variation of rotational stiffness around the ankle in AFOs affects energy storage, push-off enhancement and energy cost during walking (137).

AFO’s have been found effective in controlling dynamic equinus by preventing plantar flexion. This leads to improved foot clearance and pre-positioning during swing to achieve heel instead of toe initial contact (123, 129, 130, 133, 134, 138-141).

A common indication to use AFOs is to preserve muscle length during skeletal growth. Positioning with orthoses may help maintain optimal muscle lengths for force production; however, the elongating stretching effect of AFOs to treat static contracture has limited documentation. Buckon and colleagues found that using AFOs over a one-year study period helped *maintain* passive and active ankle range of motion in patients with spastic CP (129). Hösl et al found improved passive ankle range of motion after short-term AFO use (16 weeks, SD 4 weeks), but with adverse effect on gastrocnemius muscle morphology seen as shortened fascicles and decreased volume (142).

Foot deformities such as mid foot break is common due to spasticity and altered loading on the CP foot. The impact of AFOs to protect and stabilise the foot joints against such deformities has been measured by radiography, revealing a non-significant reduction of static foot misalignment wearing AFO’s compared to barefoot (143). Dynamic motion and immobilisation of the foot and ankle during walking with different AFO types has also been documented, although as far as we know only in normal adult subjects (144).

Walking with AFOs in children with unilateral CP may alter lower limb muscle activity. Improved pre-positioning for initial contact reduced tibialis anterior activity substantially in pre- and terminal swing, and reduced hamstring activity in terminal swing (140). Similarly, a recent study comparing two AFO designs with EMG found swing phase inactivation of tibialis anterior, particularly with contoured footplates, while both flat and contoured types diminished spastic medial gastrocnemius activity in weight acceptance due to reduced forefoot loading with AFOs (116).

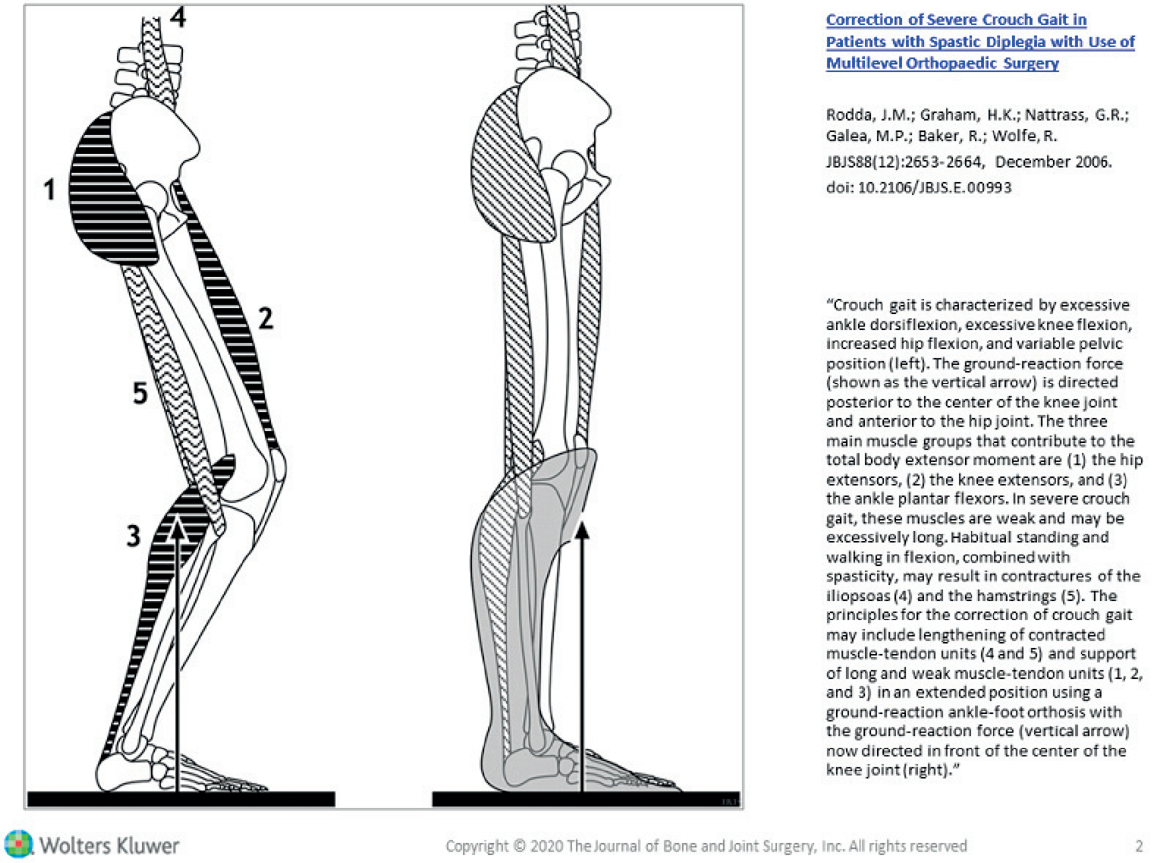
With regards to closed-chain, indirect control of proximal joints, distal stabilization with ground reaction and solid AFOs has been found efficient to provide immediate mechanical correction of knee flexion and a crouched posture (132, 145, 146), and reduction of knee and hip flexion in barefoot walking following a 12-week therapy program (147).

Restraint of ankle dorsiflexion and forward progression of the tibia during stance with ground reaction AFOs may produce knee hyperextension, anterior trunk lean, early heel lift and toe walking. Therefore, small changes in AFO and foot-wear alignment, so-called tuning which brings the tibia in slight inclination, could be necessary to improve proximal body posture, standing and walking balance (148, 149).

Certain impairments, such as knee and hip flexion contractures are significant limiting factors that compromise the efficiency and may override the mechanical impacts of AFOs. Knee and hip flexion contractures more than 15° were found contradictory to effective use of ground reaction AFOs (132), and presence of knee flexion contracture inhibited the efficacy of solid and ground reaction AFOs (146). Also, rotational dysfunction can cause excessive in- or out-toeing, resulting in functionally shorter foot lever arm, decreased sagittal plane- and increased coronal plane moments with AFOs (43, 59). In such cases it could be challenging to achieve a good result with orthoses. Surgery that addresses sagittal and transverse plane malalignment may have a bearing on AFO efficiency and necessary in cases where long-term AFO-wear is indicated (1).

AFOs Postoperatively

It is generally accepted that AFOs are an integral part of the postoperative regimen to improve and maintain effects of the surgery by applying adequate mechanical support in open- and closed-chains (3, 43, 55). Dr Gage emphasized the role of AFOs after lower limb surgery, explaining that "Appropriate orthotics contribute a great deal to the result". Therefore postoperative 3D gait analysis should not only compare pre- and postoperative barefoot walking, but include evaluation of gait with AFOs in the final outcome assessment. Furthermore, continued use of orthoses may be necessary for a prolonged time after surgery and should be continued "As long as they fulfil the purpose for which they were prescribed." (2). Davids and colleagues advised that adequate orthoses are required to ensure a functional outcome after surgery, particularly in children with more significant gait dysfunction (1). Rodda and Graham introduced treatment algorithms for spasticity and contracture in children with unilateral and bilateral spastic CP according to gait pattern and motor problem. AFO types with specified mechanical properties were described as part of the management algorithms for all gait patterns, recommending that type of AFO should be defined before surgical intervention, depending on pattern and the integrity of the plantarflexion-knee extension couple (3). Ground reaction AFOs were described as part of the intervention regimen to treat severe crouch (Fig. 8) (55).



Correction of Severe Crouch Gait in Patients with Spastic Diplegia with Use of Multilevel Orthopaedic Surgery

Rodda, J.M.; Graham, H.K.; Nattrass, G.R.; Galea, M.P.; Baker, R.; Wolfe, R.
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"Crouch gait is characterized by excessive ankle dorsiflexion, excessive knee flexion, increased hip flexion, and variable pelvic position (left). The ground-reaction force (shown as the vertical arrow) is directed posterior to the center of the knee joint and anterior to the hip joint. The three main muscle groups that contribute to the total body extensor moment are (1) the hip extensors, (2) the knee extensors, and (3) the ankle plantar flexors. In severe crouch gait, these muscles are weak and may be excessively long. Habitual standing and walking in flexion, combined with spasticity, may result in contractures of the iliopsoas (4) and the hamstrings (5). The principles for the correction of crouch gait may include lengthening of contracted muscle-tendon units (4 and 5) and support of long and weak muscle-tendon units (1, 2, and 3) in an extended position using a ground-reaction ankle-foot orthosis with the ground-reaction force (vertical arrow) now directed in front of the center of the knee joint (right)."

Wolters Kluwer

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Figure 8. The mechanical impact of ground reaction AFOs in correction of severe crouch. Rodda et al 2006. By permission.

In the1990s, the standard AFO used for CP children postoperatively in our orthopedic hospital Sophies Minde, now Oslo University Hospital, were prefabricated 'Blue Hakupa' AFOs used at night, and usually subsequent to Achilles tendon lengthening. However, night-braces worn when children could sleep with their knees flexed did not take care of gastrocnemius length and could not provide positioning during gait. After the introduction of 3D gait analysis and multi-level surgery based on multi-disciplinary team decisions in Oslo University Hospital in 2001, the use and type of postoperative AFOs were defined individually for each child following preoperative 3D gait analysis, according to gait pattern and in line with Rodda and Grahams treatment algorithms (3).

Comprehensive surgery and postoperative rehabilitation including the use of orthoses can be demanding. Capjon & Bjørk explored children and parents' experiences with the rehabilitation after multi-level surgery. Their qualitative survey confirmed that most families found the rehabilitation period hard because of pain, complex and intense training programs postoperatively. The use of postoperative ground reaction AFOs was particularly strenuous and many children conducted a countdown of the days remaining until the one-year postoperative 3D gait analysis after which they expected that the orthoses could be discarded (4). Nevertheless, the postoperative control with 3D gait analysis often results in recommendations for continued use of orthoses beyond the one-year period, additional surgery, and physiotherapy (7).

A population-based study from Australia found that before 1994, hinged AFOs were common postoperatively because ankle motion was perceived as being of functional benefit. Following introduction of routine 3D gait analysis and single-event multi-level surgery, a change in AFO prescription followed and solid AFOs were prescribed routinely for a minimum of 12 months post-operatively. *"The transition to a hinged, or no AFO, was allowed only when gait data indicated satisfactory plantarflexion-knee extension coupling and minimum risk of crouch gait"*. Indication for prolonged use of solid AFOs was to control over-lengthening of the gastrocsoleus, and remaining crouch gait postoperatively (6). Control of drop-foot and dynamic equinus has also been described as indications for recommending continued use of AFOs after the postoperative 3D gait analysis (150).

Most studies comparing gait pre- and postoperatively in children with CP focus on the surgical effects, without acknowledging the role of other elements such as AFOs. While immediate postoperative rehabilitation and care including casting usually is accounted for, specification of AFO types and utilisation vary greatly and is often poorly described.

A gap in knowledge was found when it came to the efficacy of AFOs to improve gait and indications for continued use of AFOs after the one-year postoperative follow-up with 3D gait analysis. Information from the patients and families and from the hospital rehabilitation facility in Stavern gave the impression that compliance improved when AFOs had a comfortable fit and adequate alignment. Satisfaction with the orthoses was generally better in children who experienced benefit of using them. However, in line with Capjon and Bjørks findings (4) many children and parents who came to the gait laboratory for postoperative follow-up anticipated that the AFOs were no longer needed, since they underwent the recommended surgery and had followed the guidelines for optimal postoperative rehabilitation, including use of AFOs as prescribed. Some children had also been told that the orthoses should only be used for the one-year rehabilitation. Nevertheless, we experienced that the postoperative 3D gait analysis frequently resulted in recommendations to continue using AFOs for longer durations due to remaining gait deficiencies one year postoperatively. In line with Kay et al (7), we found that postoperative 3D gait analyses serve not only as measures of treatment outcome, but are also used for planning further on-going care (7).

The current project was initiated following implementation of new routines regarding surgery and postoperative care in our hospital to enhance compliance and implementation of the suggested treatment. This involved preoperative multidisciplinary consultations where the children and families received information about the surgical procedures, including the postoperative regimen with use of AFOs. A six months postoperative orthopaedic consultation should include video-vector analysis for control of orthotic function in children who used ground reaction or solid AFOs. Furthermore, the routine 3D gait analysis one year postoperatively was extended to measure both barefoot walking and walking with AFOs whereas earlier evaluation of orthotic function was usually restricted to 2D film.

A survey of the relevant literature yielded no studies that evaluated the short- or long-term impacts of AFOs following lower-limb surgery. Major clinical indications for continued use after orthopaedic lower-limb surgery had been described, but without evidence of AFO efficacy that substantiated the recommendations (1-3, 55). Quantitative evidence was required to evaluate the impact of AFOs on gait one year postoperatively, to observe which children benefit most from using AFOs after surgery and the indications for continued use. This is important to provide realistic expectations and clearer guidelines for patients and health professionals.

2 Aims

The aim of this PhD project was to quantify the impact of AFOs on gait quality, kinematic, kinetic and temporal spatial gait variables at the time of routine 3D gait analysis ≥one year post single or multiple level surgeries in children and adolescents with unilateral and bilateral spastic CP. We hypothesized that many children have residual gait problems after surgery and that orthoses provide mechanical support to enhance gait function one year postoperatively.

The project should address the following specific aims

Paper I

Evaluate the impact of AFOs on gait one year postoperatively, and identify predictors for clinically important improvements walking with AFOs in children with spastic bilateral CP.

Paper II

Investigate whether gait problems were corrected after surgery; whether there were further changes walking with AFOs compared to barefoot and the indications for continued use of orthoses beyond the first postoperative year in children with spastic unilateral CP.

Paper III

Explore changes in the vertical component of the GRF after triceps surae lengthening and whether addition of AFOs gave improved bodyweight support and stability in stance.

3 Material and Methods

This chapter describes the materials and methods used to answer the research questions, the demographics of the participants, study design, instrumentation, procedures during data collection, outcome measures and data analysis.

3.1 Study design

The three Papers included in this thesis consists of two cohort studies [Paper I and II], and one study [Paper III] based on a sub-selection of participants in Paper I and II. The research was observational, using a repeated measures design and based on 3D gait analysis in the Oslo Movement Laboratory, including demographics and clinical advice in the patients' postoperative gait reports, quantitative kinematic, kinetic, and temporal-spatial measurements. Gait data was collected from all participants in three conditions:

1. Preoperatively walking barefoot (PreBF)
2. Postoperatively walking barefoot (PostBF)
3. Postoperatively walking with AFOs (PostAFO)

3.2 Participants

Ethical considerations

3D gait analysis is a non-invasive technique that can be performed in vivo and is appropriate for bipedal walking persons that can cooperate and understand the instructions offered during data capture. It measures how structures and bodies act dynamically without exposing the patient to radiation. All the procedures performed in the project that involved human participants were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. The study was approved by the South-East Regional Ethics committee (REC;2013/1242) and the Patient Ombudsman at Oslo University Hospital.

Power calculation

Prior to the study we calculated the number of participants required to detect a change of more than 1 SD in a relevant variable, defined as a 5° change in minimum knee flexion during stance. If a statistical method was used such as analysis of variance (ANOVA) for repeated measures, an estimated minimum of 17 participants was required in each study group with up to four repeated measures and 80% power. The number of participants available for the study depended on patients who attended the gait laboratory for postoperative gait analysis during the inclusion period, who used and were equipped with adequate AFOs. To obtain the required number for the respective subgroup participants were sampled consecutively over a four-year period.

Inclusion

We included all patients who had been seen in our gait analysis laboratory with a diagnosis of spastic unilateral and bilateral CP, GMFCS I-III, who performed postoperative 3D gait analysis walking with and without AFO's. The primary reason for referral to gait analysis had been for pre-treatment or baseline assessment of gait function.

The children who met inclusion criteria and their parents were informed about the study in the context of their one-year postoperative follow-up with 3D gait analysis. Information about the study was provided in two versions; one for the parents, and a version which was more reader-friendly and intended for the children, both made using the recommended template provided with the application for ethical approval.

Consent was required from the parents of children aged <12 years, children between 12 and 16 could sign with one parent, and youths 16 years or older signed for themselves. According to the given inclusion and exclusion criteria, participants were assigned to the study following written informed consent to use the preoperative baseline data with the children walking barefoot, and postoperative data with children walking barefoot and with AFOs.

Paper I

In the cohort with spastic bilateral CP, consecutive sampling during the inclusion period resulted in 55 patients who were eligible for inclusion and received written information about the study. Thirty-four children (62%), including 12 girls and 22 boys, consented to participate. Their mean age at surgery was 11 years (range 6–17). Eight children were operated with single level, and the remaining 26 were operated with multi-level surgery. With regards to motor function seven children were categorised as GMFCS level I, 19 were in level II and eight in level III. Many children in GMFCS level III used ambulatory devices (crutches, sticks or rollators) for activities of daily living, but had the capacity to walk independently indoors on even surfaces. Even so, we had to exclude kinetic data from four children who walked with devices that touched the force plates in one or more conditions.

Paper II

In the cohort with spastic unilateral CP, consecutive sampling during the inclusion period resulted in 43 patients who received written information and 33 (17 girls and 16 boys) who consented to participate in the study. Both affected and non-affected limbs were examined. Mean age at time of surgery was 9.2 years (range 5-16.5) Twenty-three children underwent tendo-achilles lengthening, ten children had gastrocnemius recession, and concomitant surgical procedures were performed in ten children. Twenty-two children were classified as GMFCS level I and 11 children as level II.

Paper III

The study included a subsample from Paper I and II, and was limited to children with spastic CP, who underwent surgery with triceps surae lengthening to treat ankle equinus, who used hinged or solid AFOs at the one-year postoperative 3D gait analysis, were independent ambulators in level I-II of the GMFCS and had trials with valid force plate data from both legs. According to criteria we included 32 children with unilateral and 24 children with bilateral spastic CP.

3.3 Postoperative AFOs

Types of AFOs were defined and prescribed after preoperative 3D gait analysis, guided by each participants' gait pattern and the treatment algorithms proposed by Rodda and Graham (3). This implied that ground reaction AFOs were made for children who walked with crouch and solid AFOs when there was less severe crouch or jump knee patterns. Both types fixed the ankle in neutral angle although ground reaction AFOs were made stiffer, with longer proximal lever arms. In Paper I and II solid and ground reaction AFOs were both categorized as ground reaction AFOs. In Paper III solid and ground reaction AFOs were categorized as solid AFOs. In cases with ankle equinus and single-level triceps surae surgery the children were usually prescribed hinged AFOs with integrated joints (Tamarack, Blaine Washington), free ankle dorsiflexion and restrained ankle plantarflexion past neutral angle. All AFO types had long soles past the toes. With AFOs and shoes an alignment with 0-10° shank-to-vertical inclination was used. Solid and ground reaction AFOs were generally inclined with 5-10° shank-to vertical angle to align the GRF neutral to the hip joint centre in standing and walking (Fig. 9) (120, 149).

Figure 9. Example of bench alignment of a solid AFO with shoes; the shank/tibia inclined with a shank-to vertical angle of around 7°.

According to routine protocol for lower limb surgery in CP, casts for the postoperative AFOs were made per-operatively by certified prosthetist orthotists (CPO) working at the hospital. Fitting of the fabricated orthoses took place during a one-week in-house rehabilitation, after removal of below-knee cast/splints which were mandatory in cases of triceps surae lengthening, foot, and ankle surgery. Knee immobilisers were used at night in patients who underwent either lengthening of the hamstrings, combined hamstrings lengthening with rectus femoris transfer to the semitendinosus, patellar-tendon advancement, and/or femoral extension osteotomy. Children who underwent multilevel surgery were transferred to a specialized centre for a four-week training program in a rehabilitation centre to regain strength, maintain range of motion, and automate an optimised gait pattern, before transfer to community-based physiotherapy.

The children were prescribed all-day use of the postoperative AFOs for a minimum of 12 months. Many children were assigned to the gait laboratory for control of AFO function and alignment with 2D video-vector analysis six-months postoperatively. The AFOs were sometimes exchanged during the rehabilitation period due to growth, pain or altered need for mechanical support.

3.4 Data Collection

Personnel and examination

Pre- and postoperative 3D gait analysis was performed by a multidisciplinary team and according to routine procedures. One child neurologist was present during anamnesis and neurological testing. Two experienced testers, usually one physiotherapist and one CPO took charge of the physical examination, marker placement, data capture, processing, analysis and reports. AFO type was described and noted by the CPO, who also measured alignment, i.e. tibial inclination and shank-to-vertical angle with and without shoes using a goniometer, and build-up on AFOs, shoes, and shoe heel height/drop/ with an outside calliper.

Patient anamnesis and physical examination took place prior to the gait analysis, Participants and parents responded to questions regarding motor function (GMFCS) (25), performance over 5, 50- and 500-meters walking distance (FMS) (26), pain using the faces pain scale (151), use and compliance with AFOs, experience with the postoperative training regimen and self-reported satisfaction with their function one year postoperatively. Anthropometric measures that were required for the 3D lower-body model included subject height and mass, leg length, pelvic, knee and ankle widths and were obtained with scaled weight, tape measure and calliper. Physical examination followed standard protocol where one tester was responsible for patient examination and rating and the other assisted with goniometer measurements and writing of scores into standardised forms. The protocol involved measuring passive and active angular ranges of motion across hips, knees and ankles using a goniometer, manual muscle testing of muscle strength (152) and assessment of selective motor control in ankle dorsiflexors. Muscle tone, spasticity and rigidity was assessed using the Modified Ashworth Scale (153) and Tardieu Scale (154). The physical examination parameters are only cited to describe the routine 3D gait analysis protocol and the results that contributed to the clinical recommendations.

Final decisions and recommendations concerning clinical interventions in individual cases were made in structured meetings with team orthopaedic surgeons, child neurologists, physiotherapist and CPOs and based on the comprehensive gait analysis reports as displayed with Vicon Polygon software comprising physical examination, anamnesis, 2D video film, temporal-spatial, 3D kinematic and kinetic variables. Team recommendations were written with bullet-points in the individual gait reports.

Instrumentation

Kinematic and temporal-spatial data was obtained in each of the three compared conditions, using a Vicon MX system (Vicon Motion Systems Ltd., Oxford, United Kingdom) with six cameras (MXF40) mounted on the walls of a rectangular room. The cameras were interlaced with diodes emitting infrared light at a frequency of 100Hz to allow tracking and triangulation of circular reflective markers by at least two cameras.

Sixteen 14 mm markers were fixed with double-sided tape and elastic straps on the participants. The team of two testers reached agreement on marker placement following the lower extremity marker protocol and 3D model provided with the Vicon system: The Plug-in-Gait model. This biomechanical model, also called the Helen Hayes model, is based on the work of two individual teams of researchers (155, 156). Subject anthropometric measurements, external and internal markers define each body segment by the orientation of embedded orthogonal coordinate axes, assuming that the segments articulate around fixed joint centres. Using Euler angles the rotations have hierarchical order, starting with:

1. Rotation of the pelvis segment with respect to the global laboratory axis system
2. Rotation of the thigh segment with respect to the pelvis segment (hip angles)
3. Rotation of the shank segment with respect to the thigh segment (knee angles)
4. Rotation of the foot with respect to the shank segment (ankle sagittal plane angle)
5. Rotation of the foot with respect to the global laboratory axis system

This enables sagittal, coronal and transverse plane ranges of motion to be recorded and displayed simultaneously through the gait cycle (155, 156).

Kinetic data was collected with three AMTI force plates (AMTI OR$^{6-7}$, Advanced Mechanical Technology Inc., Watertown Massachusetts, USA), to measure the vertical and horizontal ground reaction force components exerted between the body and the ground. AMTI force plates use a right-hand coordinate system with the positive z axis oriented downward in the vertical direction. Six channel analogue outputs correspond to the three orthogonal forces Fx, Fy, Fz and the three orthogonal moments Mx, My, Mz respectively. Calibration ensured that the positions of the force plates relative to the Vicon cameras were known, enabling the system to calculate joint moments, forces and powers by inverse dynamics.

A central control box collected the camera images, synchronised with the three force plates and two digital video cameras (Basler, Ahrensburg, Germany) used to film the participants in sagittal and frontal planes. Data was captured and processed with Vicon Nexus software, involving system and subject calibration; reconstruction and labelling to produce 3D trajectories from raw marker data; noise-reducing filtering; calculation of 3D model outputs such as joint angles, forces and moments. Events, i.e. initial contact and foot-off were automatically detected from the force plates and added throughout the trial based on height positions of the foot markers and visual inspection, whereby temporal-spatial variables such as step and stride lengths, step width, and cadence could be calculated. Polygon software was employed to display and interpret the results in individual gait reports, and for inspection of consistency in kinematic and kinetic curves.

Following marker placement, data collection commenced with a three second static trial and subject calibration of the patient standing, to determine joint axis centres, local coordinates within each body segment and with respect to the global orthogonal coordinate system. With AFOs, heel and foot markers were placed at estimated equal distances to the sole of the foot based on measures of heel height/drop and build-ups, and not assumed horizontal during static processing (157). When AFOs covered the lateral malleoli, markers had to be placed on the device and ankle widths were adjusted accordingly. Dynamic capture continued with children walking in their self-selected comfortable speed across the 12-metre walkway; repeatedly in 10-15 trials, and usually until at least three trials containing clean force plate strikes from right and left sides were obtained. In the preoperative condition, the children were measured walking barefoot. The priority sequence of measurement conditions postoperatively was barefoot walking first to ensure that the children had physical endurance to achieve data for comparison with the preoperative barefoot data, and followed by AFO walking

3.5 Outcome measures

Outcome measures were categorized within the ICF body functions and structures, and activity domains. The outcome measures for Paper I-III are presented in Table 1.

Table 1. Outcome measurements Papers I-III.

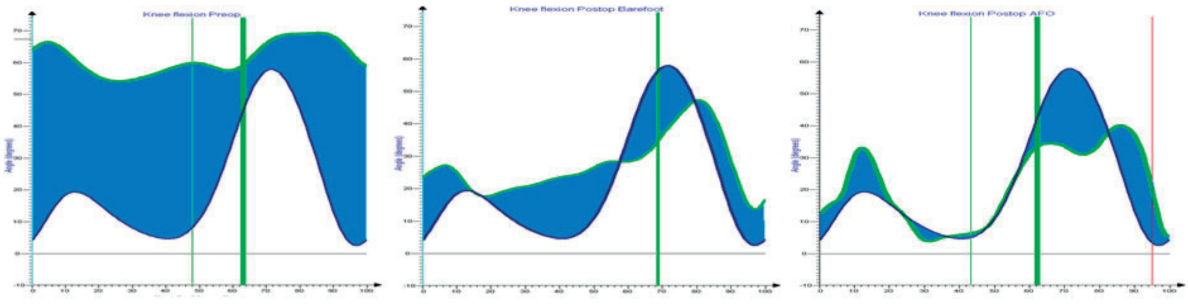
Paper I-III	**Outcome measurements**
Paper I Impact of AFOs on gait 1 year after lower limb surgery in children with bilateral CP	Gait index: • GPS • Grouping by MCID of the GPS Spatial-temporal: • ND Walking speed • ND Step length • ND Cadence Kinematic: • Ankle angle at initial contact • Stance max ankle dorsiflexion • Stance min knee flexion Kinetic variables • Stance max dorsiflexion moment • Late stance max knee moment Predictors: • GMFCS level, Gender, Age at surgery, Preop GPS, Postop GPS
Paper II Comparison of gait with and without AFOs after lower limb surgery in children with unilateral CP	Gait Index • GPS Spatial-temporal: • ND Walking speed • ND Step length • ND Cadence Kinematic: • Ankle angle at initial contact • Stance max ankle dorsiflexion • Swing max ankle dorsiflexion • Knee angle at initial contact • Stance min knee flexion • Stance min hip flexion Kinetic: • Mean ankle moment 0-10% of the gait cycle • Stance max dorsiflexion moment • Stance max ankle power generation Covariates: • Gender, GMFCS level, AFO type
Paper III Postoperative changes in vertical ground reaction forces walking barefoot and with AFOs in children with cerebral palsy	Vertical GRF (vGRF) components: • vGRF 0-100% stance • vGRF 15-35% stance • vGRF 65-85% stance • FZ_1 and FZ_2 Kinematic: • Stance max ankle dorsiflexion Temporal-spatial: • ND Walking speed Covariates • Condition (PreBF vs PostBF, PostAFO vs PostBF) • CP type (bilateral vs unilateral)

Temporal-spatial variables represented outcome measures at activity level, with walking speed as the main functional outcome measure of walking economy (121). Walking speed was evaluated in Papers I-III, step length and cadence in Paper I and II. To account for changes in body stature between pre- and postoperative conditions comparisons were made with non-dimensional velocity, step length and cadence, normalised using subject body height as the growth parameter (158).

Kadaba et al used coefficients of multiple correlations (CMC) to determine similarity of waveforms and found excellent repeatability in the sagittal plane kinematics with CMC > 0.90 for hip, knee and ankle within and between test days. For joint kinematics in the coronal and transverse plane repeatability was excellent within a test day (CMC > 0.86), but more strongly influenced by variability in marker placement between test days (159). Similar variability and sources of error have been identified by other research teams (76, 160). Most variables have reported error estimates (SD or SE) of less than 5°, which is within clinically acceptable limits. Hip and knee transverse plane values are more susceptible to marker placement error and should be interpreted with greater caution (76, 161). In our gait lab at Rikshospitalet, inter- and intra-tester reliability and repeatability of kinematic gait data was evaluated within and between days using functional limits of agreement (162). The results confirmed variability within clinically acceptable limits of 5° in sagittal plane pelvis, hip, knee and ankle variables, whereas high variability was confirmed in some transverse and coronal plane variables relating to marker placement error and thigh rotation offset (163).

Assessing variability of kinetic data, vertical and anterior-posterior ground reaction forces have been found more repeatable than the medio-lateral shear force component, whereas sagittal plane joint moments were more repeatable than frontal or transverse plane moments (159). In typically developing children and children with CP little variability was found in the vertical and anterior-posterior GRF components, and particularly the second vertical peak had high repeatability (36). Medio-lateral force components have high variability, known lack of reliability, and have been found less suitable for use in clinical assessment (34, 36, 164).

Gait indices summarize how tasks such as walking are executed and completed within the body functions and structures domain of the ICF (121). A main outcome measure in Paper I and II was the gait index Gait Profile Score (GPS) which is a summary measure of gait quality (8). The GPS is the total score based on nine kinematic Gait Variable Scores (GVS), each calculated as the root mean square difference between sagittal, transverse, and coronal plane gait curves of the patient versus gait curves from children with no gait pathology. The output is expressed in degrees where a reduction in GVS and GPS values, for example after intervention, indicate curves closer to normal and an improvement. Results can be displayed in a Motion Analysis Profile (MAP). Excellent reliability and acceptable agreement was demonstrated for the GPS in children with CP, supporting its use in research and clinical practice. Meanwhile, large variability for some of the GVS indicated caution in interpretation of outcome measures (165). A reduction of the GPS ≥1.6° has been defined as the minimal clinically important difference (MCID)(166).



PreBF PostBF PostAFO

Figure 10. Sagittal plane knee curves (green) from one participant with bilateral CP in preoperative barefoot (PreBF), postoperative barefoot (PostBF) and postoperative AFOs (PostAFO) conditions, walking with ground reaction AFOs in the latter. The blue area illustrate the difference between normal average knee curves (dark blue) and participants curves which formed the basis for GVS, GPS and MAP calculations.

In our study GVS, GPS and MAP were derived using data from three trials in each test condition. The normative gait curves were averaged kinematics from our reference database of 24 typically developing children (11 girls, 13 boys) with a mean age of 9.8 years (range 5-15 years). Paper I report GPS and MCID on individual and group level. Paper II report GPS only at group level. On group level averaged GVS in each of the three conditions are displayed in motion analysis profiles for the bilateral (Paper I) and unilateral (Paper II) subgroups respectively. Paper I used MCID for GPS to categorize the participants as ‘Improved’ or ‘Not Improved’ walking with AFOs versus barefoot one year postoperatively.

In Paper I single kinematic variables included ankle angle at initial contact, stance maximum ankle dorsiflexion and stance minimum knee flexion, whereas kinetic variables included stance maximum external dorsiflexion moment and late stance maximum external knee moment.

In Paper II gait pattern were defined pre- and postoperatively using the classification by Winters, Gage and Hicks (46). Descriptive analysis was linked to AFO type and recommendations for further AFO use after the one-year postoperative follow-up with 3D gait analysis (Paper I and II). Kinematic variables included ankle and knee angle at initial contact, maximum ankle dorsiflexion during stance and swing phases, stance minimum knee and hip flexion. Kinetic variables were ankle mean moment during loading response; in 0-10% of the gait cycle, maximum external ankle dorsiflexion moment and maximum ankle power generation in terminal stance.

In Paper III the vertical component of the GRF (vGRF) was the main outcome to evaluate stance stability and CoM support after clinical intervention with surgery and with AFOs. Force magnitudes were studied using functional curve analysis and peak values in the periods of weight acceptance, late stance and during the entire stance phase of gait. We also included stance maximum ankle dorsiflexion as a kinematic variable to describe changes in ankle range of movement following interventions, whereas non-dimensional walking speed represented changes at activity level.

3.6 Data analysis

All kinematic, kinetic, and temporal-spatial variables were calculated based on averaged data from three trials in each condition and participant. One exception was made in Paper III where functional curve analysis was based on one single vGRF curve in each condition. One gait cycle per trial was used for averaging of kinematic and kinetic data whereas all available gait cycles within each trial formed the basis for averaging and analysis of temporal-spatial variables.

Right and left limbs move and are dependent and correlated within the individual. To reduce dependency in the data our main choice was therefore to analyse one limb per participant. In Paper I which included children with bilateral CP this implied that we used data from the most affected side, defined as the side which received most surgery and/or the side where AFOs were used. In cases where there was no difference in AFO use or type of surgery we chose the left limbs for analyses. Paper II included children with unilateral CP and according to the aim of the study data from both affected and non-affected limbs was included and analysed separately. For analysis of ground reaction forces (vGRF) in Paper III data from one limb per participant involved the affected side in children with unilateral CP and the most affected side in bilateral CP.

Statistical analysis

Descriptive statistics were presented as group means and SDs, graphs, ranges, etc. Distribution of the outcome variable change scores and model residuals were tested for normality using Kolmogorov-Smirnov test. Statistical Package for Social Sciences (SPSS 21 for Windows; IBM corp., USA) was used for all analyses in Paper I and II and for parts of analyses in Paper III. R (167, 168) was used for functional curve analysis in Paper III. Differences were considered significant with level of significance set at p= 0.05.

Statistical methods were selected that accounted for correlation between repeated measures within participants. Comparisons included PreBF versus PostBF to evaluate changes following surgery, and PostAFO versus PostBF to evaluate additional changes walking with AFOs one year postoperatively. Neither analysis compared PreBF with PostAFO, since this was a comparison which was not considered relevant, and to minimise the amount of repeated comparisons that potentially could have increased the risk of Type I error.

Paper I

To evaluate changes in all outcome variables comparisons of PreBF versus PostBF and PostAFO versus PostBF conditions were made using paired, two-tailed T-tests. Paired t-test comparisons between PostAFO and PostBF conditions were also performed in subgroups who used Ground reaction AFOs and Hinged AFOs respectively. A reduction of the GPS ≥1.6° was used as an indication to categorise children with clinically important benefit of AFOs as 'Improved', and children with a GPS reduction <1.6° as 'Not improved'. GMFCS level, sex, age at surgery, preoperative and postoperative GPS were tested as predictors of clinically important improvement walking with AFOs one year postoperatively, first in univariable and subsequently in multivariable logistic regression using Wald test.

Paper II

The study design resembled Paper I, with repeated measures and a single sample design, but studying affected and non-affected limbs of the children with unilateral CP in separate analyses. Changes in outcome variables between conditions were tested using linear mixed model analyses in which the PostBF condition was the reference category, which was compared against PreBF and PostAFO conditions, respectively. The individual was termed as the random effect with a random intercept; meaning participants represented a random sample from a larger population (169, 170). In addition, continuous and categorical explanatory fixed effects included gender, GMFCS level and AFO type (hinged versus ground reaction AFOs), and interactions with each condition.

Paper III

To analyse changes in the entire vGRF curves, the vGRF data were normalized to bodyweight (N/kg) and time-normalized to 0-100% of stance phase. To study the vGRF as a dependent functional variable the normalized data was transformed using generalised additive models and basis splines to fit the vGRF curves(168, 171) and a functional F test to assess statistically significant differences between conditions (172). For descriptive analysis of peak vGRF variables FZ_1 and FZ_2, non-dimensional walking speed, and maximum ankle dorsiflexion during stance, we used linear mixed model analysis as described for Paper II. Changes in the two outcome variables were investigated with PostBF as the reference category; tested against PreBF and PostAFO conditions and using subject-specific random effects to test individual deviations from the average population trend.

4 Summary of Results

Paper I

Major improvements between pre- and postoperative barefoot conditions (PreBF versus PostBF) included the GPS, ankle angle at initial contact, stance maximum ankle dorsiflexion, stance minimum knee flexion, and late stance maximum external ankle and knee moments. Walking speed and step length were both reduced walking barefoot postoperatively.

Walking with AFOs at follow-up, all temporal-spatial variables improved with highly significant increases in walking speed and step length, whereas cadence decreased. There were also further improvements in stance maximum ankle dorsiflexion and late stance maximum knee moment compared with barefoot postoperatively. Fourteen children used ground reaction AFOs and 20 children used hinged AFOs at the one-year postoperative 3D gait analysis. Comparing relevant variables in the PostBF versus PostAFO conditions improvements were more pronounced in the subgroup using ground reaction AFOs with significantly decreased GPS, stance maximum ankle dorsiflexion and minimum knee flexion.

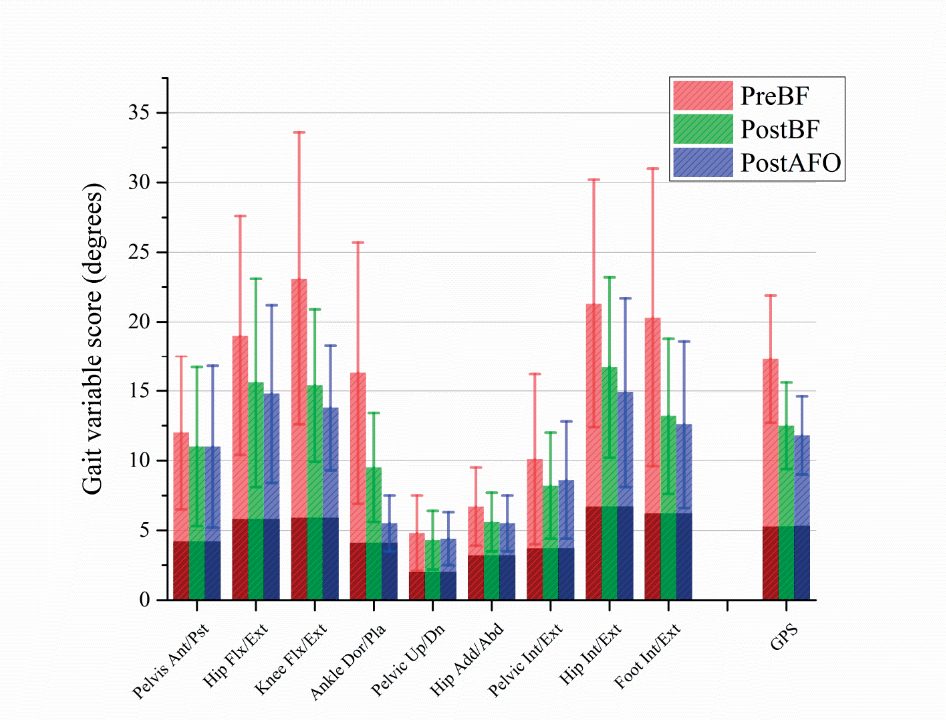


Figure 11. Motion analysis profile (MAP) of the changes in gait variable scores and total gait profile score. Each column shows root mean square differences between the bilateral cohort (n=34) and averaged scores from our normal reference data (n=24), shown as the darker are in the base of each column.

Additional impact of AFOs on overall gait quality was seen as significantly reduced GPS. A clinically important GPS improvement ≥1.6° was found in 12 of the 34 participants (35%). Multivariable logistic regression revealed that a high preoperative GPS was the only significant predictor of clinically important improvement walking with AFOs, implying that children with more serious gait problems preoperatively were the ones who had strongest impact of AFOs on gait at the one-year postoperative follow-up.

Paper II

The major change in gait pattern was from a pattern with true equinus and triceps surae contracture preoperatively, to less severe gait patterns with drop-foot postoperatively. Some children did not change, remained in true equinus or deteriorated to more severe gait patterns after surgery. In the affected limbs, the GPS, most kinematic, and all kinetic variables changed towards improvement, whereas cadence was reduced walking barefoot postoperatively.

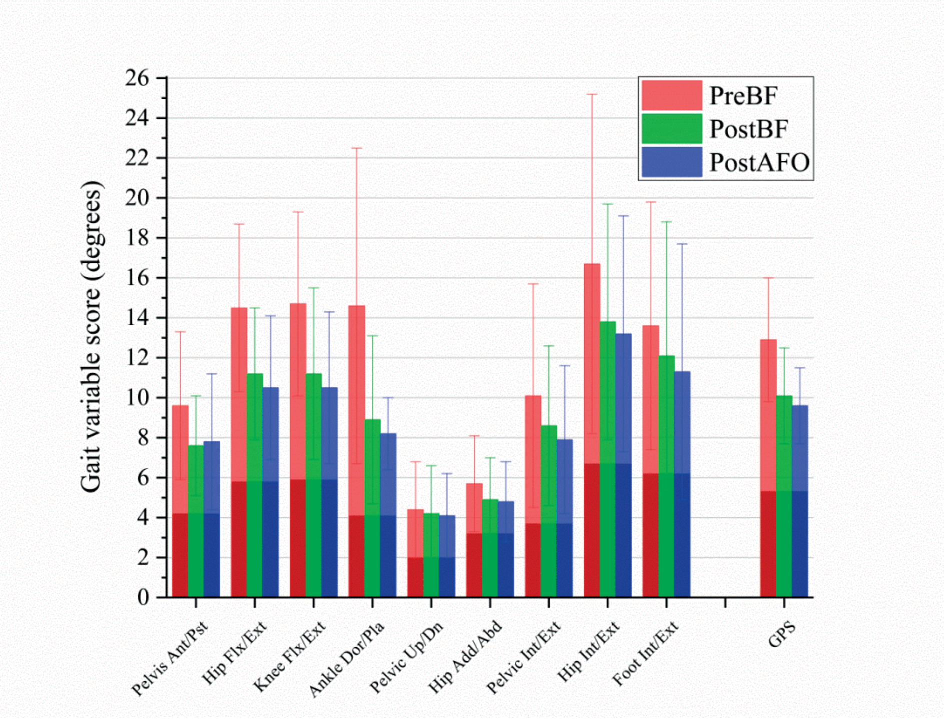


Figure 12. Motion analysis profile (MAP) of the changes in gait variable scores and total gait profile score. Each column shows root mean square differences between the unilateral cohort (n=33) and averaged scores from our normal reference data (n=24), shown as the darker are in the base of each column.

Major impacts walking with AFOs versus barefoot were seen at initial contact with significantly reduced plantarflexion and knee flexion. These changes gave improved prepositioning of the foot and heel initial contact in PostAFO, versus forefoot initial contact in PostBF, as confirmed by significant change in ankle kinetics during loading response/1st rocker. We saw an increase in walking speed due to increased step length, while cadence was reduced. Reduced GPS indicated additional non-significant improvements in PostAFO versus PostBF. In the cohort 23 children used hinged AFOs, whereas ten children used ground reaction AFOs. Significant interaction effects entailed increased stance maximum ankle dorsiflexion in children walking with hinged versus ground reaction AFOs, and that children using ground reaction AFOs had significantly more stance knee flexion preoperatively. In the cohort, 32 of 33 children were recommended continued use of AFOs after the one-year follow-up.

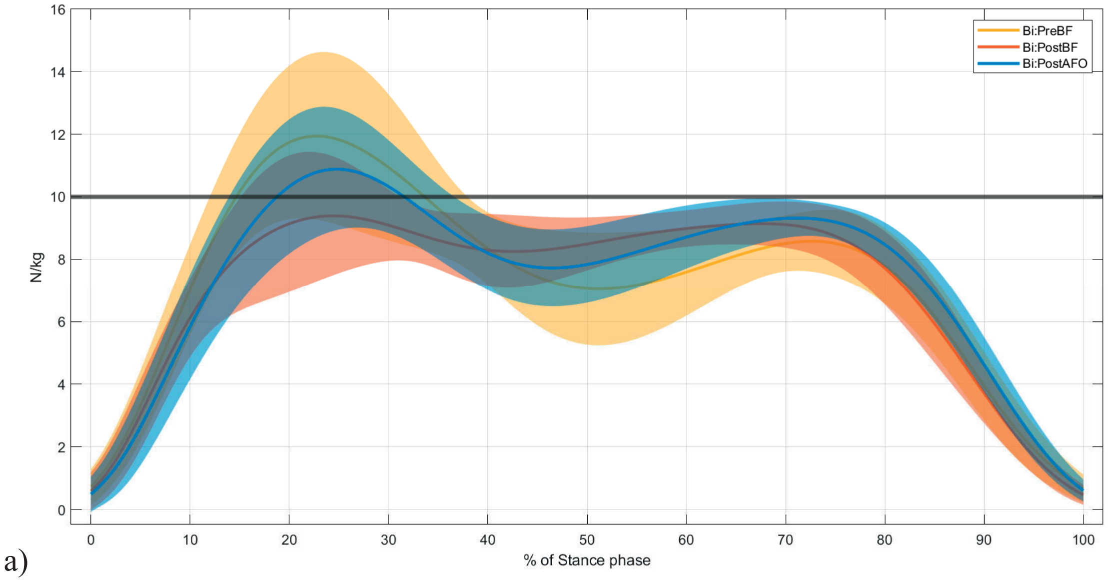
Significant changes were also found in the non-affected limbs during the stance phase, both walking barefoot and with AFOs one year postoperatively. These changes indicated that compensatory movements to improve foot clearance in the opposite, affected limbs were common preoperatively, reduced after deviations were corrected with surgery and further reduced with AFOs on the affected limb postoperatively.

Paper III

Analyzing the vertical component of the ground reaction force (vGRF) with functional curve analysis, significant group effect was found between children with bilateral and unilateral CP in each of the PreBF, PostBF and PostAFO conditions. All comparisons between conditions to test impacts of triceps surae surgery and AFOs were therefore analyzed separately for the topographical CP types.

After triceps surae surgery (PostBF versus PreBF), kinematics confirmed a change from ankle equinus to increased ankle dorsiflexion in both groups, whereas reduced walking speed was most distinct in the bilateral group. Significant changes in the entire vGRF through stance, with decreased forces in weight acceptance and increased forces in late stance implied that a vGRF pattern with so-called Ben Lomonding was less pronounced postoperatively. However, modelled curve-estimates and descriptive peak values indicated late stance support below bodyweight and remaining CoM deceleration deficit walking barefoot one year postoperatively.

Walking with AFOs postoperatively (PostAFO versus PostBF) ankle dorsiflexion was restrained in the bilateral and not changed in the unilateral group. Walking speed increased significantly in both groups. The main impacts of AFOs was additional and significantly increased vGRF in weight acceptance and increased forces equivalent to bodyweight in late stance that indicated clinically important improvement of CoM support and stance stability with AFOs. This additional improvement with AFOs was most pronounced in children with unilateral CP.



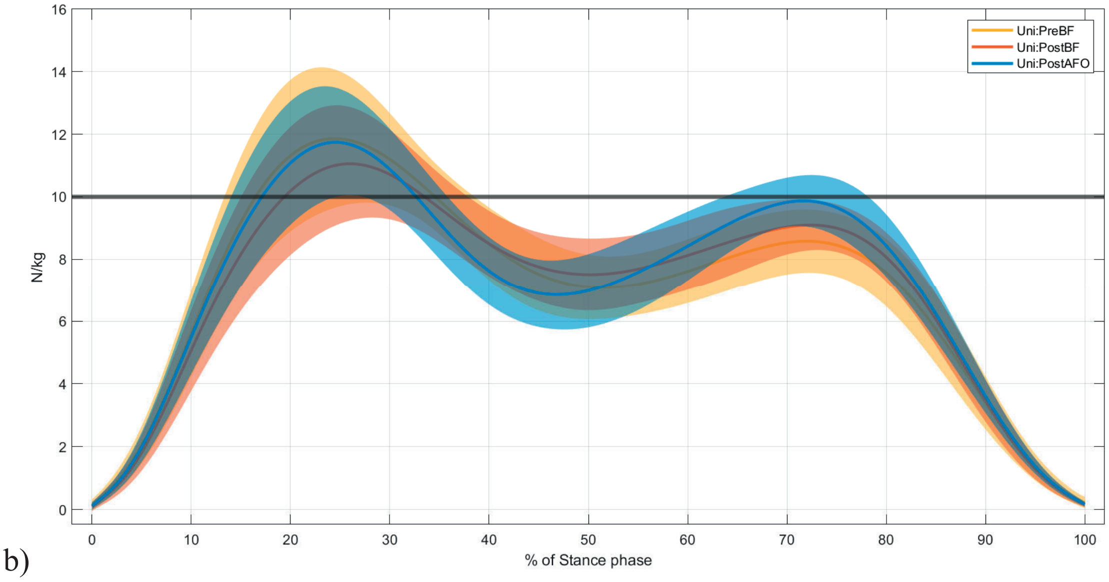


Figure 13. Graphs illustrating vGRF (mean, 1SD) in PreBF (yellow), PostBF (orange) and PostAFO (blue) conditions for a) bilateral and b) unilateral group. Forces are normalised to bodyweight (N/kg) and time-normalized from 0-100% of stance phase**.** The horizontal line at 10 N/kg illustrates 100% bodyweight.

The vGRF was found responsive to evaluate impacts of clinical interventions with surgery and AFOs in children with CP. Functional curve analysis ensured optimal representations of the entire vGRF curves and evaluation of both shape and force magnitudes in the studied time intervals.

5 Discussion

The overall aim of this thesis was to evaluate impacts of AFOs on gait function one year after lower limb surgery in children with spastic CP, with emphasis on additional changes and indications for continued use of AFOs after the one-year follow up with 3D gait analysis. What follows is a discussion regarding the methodology, the study design, the results and conclusions.

5.1 Methodological considerations

Study Design

To study the impact of AFOs at the one-year follow-up and indication for continued use we planned the current study with an observational, repeated measures design in a cohort of children who had preoperative baseline data, and were followed prospectively to compare postoperative changes walking barefoot and with AFOs. Repeated measures are widely accepted in the clinic and in research to evaluate gait pre- post intervention (5, 78), long-term results (80, 105, 106), gait with and without AFOs (132, 134, 145), comparison of different AFO types (114, 129, 130, 146) and configurations (113). The strength of a repeated measures design is that comparisons between conditions are based on changes within subject. Variability between subjects is usually considerable, especially in CP, but with repeated measures the human variation factor is diminished (173). Furthermore, statistical inference may be enabled with fewer study subjects since the variance of estimates of treatment effects is reduced (173, 174).

A prospective cohort in clinical research is defined as a group of subjects who are followed forwards in time, from baseline, during follow-up and to an endpoint; observing exposures and interventions over time as they take place from the time of inclusion (175, 176). In our study, all data were collected prospectively in conformity with routine gait laboratory protocol, using the same testers, equipment and procedures. Measurements; pre- and postoperative 3D gait analysis, and interventions; lower limb surgery and rehabilitation using AFOs, occurred with a temporal sequence and in line with the requirements of a prospective cohort study (175). Identification of the cohort was however made at the time of postoperative follow-up, and analysis of pre- and postoperative data was therefore retrospective. Enrolment to the study was highly related to exposure, since the children underwent surgery and used AFOs. Since participation was based on informed consent it is possible that there was selection bias with inclusion of the children and adolescents who had more positive experience, and found the AFOs useful. The study was conducted in the hospital and gait laboratory which serves the largest habilitation units and the largest number of children diagnosed with CP in Norway (15). We thus assumed that the cohort was representative with good external validity and applicability of the results to the target population.

The study by Novak et al questioned the efficacy of orthopedic surgery and orthoses due to the lack of experimental, randomized control studies (93). The review was later criticized for the use of meta-analysis to evaluate a wide range of interventions in a heterogeneous diagnosis such as CP (177). Randomized controlled trials are thought of as the highest form of evidence but may have limitations in trials with small numbers and large variation due to participant characteristics, such as the multifactorial CP population.

Postoperative AFOs may be viewed as a co-intervention, and a confounder which (purposely) affects the results of lower-limb surgery (175, 176). To determine the relative effect of postoperative AFOs as an adjunct to surgery, an experimental design with a control group who did not receive AFO intervention would probably be the most suitable design. It would however be challenging to conduct a study with higher level evidence due to the ethical implication of randomising children to control groups when evidence indicate the intervention is most effective, or a defined part of the treatment regime, such as postoperative AFOs (43, 55). A possible solution could be to employ a cross-over design where participants were assigned to two different groups, each receiving postoperative alternate AFO types in defined sequence.

For our research purpose, an observational study with a repeated measures design was adequate to investigate the impact of AFOs at the time of one-year postoperative follow-up, the main indications for continued AFO use and predictors for improvement with AFOs. Group effects due to gender, GMFCS level, and different AFO types were tested. When testing the effect of AFO type, there were however systematic differences in gait patterns and topographical CP type between children who used hinged and solid/ground reaction AFOs. This could have introduced confounding and alternative explanations to changes in the outcome, which may have reduced the internal validity of some results.

Use of shoes-only instead of a barefoot control condition has been recommended in best-practice guidelines to evaluate impacts of AFOs on gait (127). We chose to use barefoot as control condition postoperatively, mainly because this was required for comparison with the preoperative condition, and because more than two measurement conditions were difficult to complete for many of the children due to lack of physical tolerance and endurance in the postoperative session. Since few differences have been found between barefoot and shoes-only conditions (112, 145) we believe barefoot was an adequate control condition for our purpose. A randomized order of testing to avoid bias and fatigue in the last performed test condition has been recommended (127). In the last performed AFO condition walking speed and step length significantly increased and we therefore assumed results were not biased by fatigue.

Participants

Participants were enrolled and asked to participate at the time of follow-up with 3D gait analysis approximately one-year postoperatively. Information and consent forms were partly distributed by mail, after completion of the postoperative 3DGA, or provided when the children and parents arrived in the gait laboratory for follow-up, in which case the consent forms should be returned by mail. Few actively refused to participate, but lack of response occurred, even after a second invitation letter was sent. In the bilateral group 62% (34 of 55) consented and in the unilateral group 77% (33 of 43) consented to participate. It is possible that the number of respondents/participants might have been higher if the information and purpose of the study was communicated in advance of the postoperative gait laboratory sessions. Consecutive sampling continued within a defined period, with the objective to achieve a minimum of 17 participants within each subgroup for up to four repeated measures, as estimated with power analysis. For sub-group analyses and in testing covariates there were however few participants in some groups to detect effects with sufficient power (178).

The cohort shared defined inclusion and exclusion criteria. Nevertheless, participants varied in age, gross motor function, gait patterns, gait dysfunction, and the degree of neurological involvement. Time from surgery to postoperative gait analysis varied from 12-24 months in the children with bilateral CP and 11-27 months in children with unilateral CP. In addition, there was heterogeneity with respect to AFO type and extent of surgery. Heterogeneity was most pronounced in the cohort with bilateral CP which included GMFCS levels I-III, disproportionate gender distribution (65% boys), variability in limb involvement and extent of surgical procedures. In each study the main steps to control for heterogeneity and variability between subjects involved a repeated measures design.

Referring to best practice reporting guidelines for AFO intervention studies in CP (127), details and transparency about the participants were complete with regards to ages, gender, diagnosis and GMFCS level (25), timing of postoperative 3DGA and type of surgery, as presented in general tables (Paper I and II). Gait patterns were described with reference to published gait classification systems (3, 46, 47) which gave better transparency re sample heterogeneity whereas AFO type, GMFCS levels and clinically important improvement of the GPS were used as grouping variables (166).

Postoperative AFOs

Postoperative AFOs were fabricated by the same team and using standardized methods for hinged, solid and ground reaction AFOs with regards to material thickness, shape/design and use of Tamarack (Blaine, USA) flexible joints in the hinged AFOs. However, the type had sometimes been altered in children who were examined with video vector- analysis at the six-month postoperative control according to growth and need for support. Although not explicitly stated in our criteria, we excluded children who used AFOs that were thought inadequate in influencing sagittal plane ankle joint motion, such as supramalleolar AFOs.

Aims of the devices were described according to previously defined management algorithms (3), and further details were in line with best-practice reporting guidelines (127) including categorisation of AFO type with regards to mechanical function, the range of movement allowed, assisted or prevented in the orthosis, AFO ankle angle, toe plate length, material type, thickness, stiffness, design (ventral or dorsal shell etc), manufacture, tuning and shank-to-vertical angle.

In all papers solid and ground reaction AFOs were categorized as one AFO type because the mechanical properties were sufficiently similar. This approach was recently supported in a study by Ries & Schwartz who found the two AFO types were equally efficient in correcting crouch gait (146).

Outcome measures

It has been recommended that any AFO research project should follow a two-level approach where evidence should represent ICF activity, body functions and structures domains (121). At ICF activity level we measured walking capacity in the gait laboratory with self-selected walking speed, step length and step frequency across a 12-meter walkway.

At the ICF body functions and structures level single kinematic and kinetic variables from 3D gait analysis were selected with focus on those which were thought most relevant to report changes after surgery and impact of AFOs on gait in children with CP at the one-year follow-up. Our focus was on sagittal plane variables, to provide mechanical evidence according to the intended purpose of AFO types in the current study. Transverse and coronal plane variables were however contained within the GPS (8). The use of the latter comprehensive gait index was an important outcome to quantify general improvement as changes towards normal values. Furthermore, the vGRF was conceived as a functional measure of stability and body CoM support which may represent both activity, and body functions and structures domains within the ICF (121).

Neither study integrated patient reported outcome measures, although our participants might have provided valuable information regarding AFO use and compliance in the postoperative rehabilitation, how participation and involvement in life situations were affected, and their expectations regarding continued use of AFOs after the one-year follow-up.

In 3D gait analysis the model with hierarchical order of rotation between body segments imply that variables from adjoining body segments are correlated (76). A change in one segment such as the pelvis most likely affects thigh/hip rotations, and so on, resulting in dependency between curves. Data points from the same curve are also correlated and more so the closer the points are in time. To reduce the risk of type I error that comes with multiple comparisons of correlated and dependent data we aimed to evaluate few, but relevant variables. The use of functional curve analysis in Paper III was a means of reducing the effect of interdependency between several points on the GRF curve, while integrating more information regarding the entire gait curve (164, 172, 179).

Variability and measurement error may conceal or overrate clinically important changes. Intrinsic sources of variation in 3D gait analysis involve natural, biological variability within and between subjects and can be influenced by many factors such as age, height, mass or walking speed (161). Variability is generally higher in children with spastic CP than in typically developing peers, with regards to temporal-spatial, kinematic and kinetic outcome measures within and between days. Furthermore, kinetic variables have been found more repeatable than kinematics (180). Averaging of three trials per condition handled some of the intrinsic variability within participants. The use of one single trial and gait cycle per condition in the functional analysis of vGRF curves may have introduced bias; however, curves were inspected for variability and consistently using the first of three trials. Further concerns involved fitting GRF curves with basis splines (168, 171), which may have introduced unnecessary smoothing of curves on top of already filtered data. To account for the possibility of underestimation, peak vGRF values were also included in the study. Differences in body stature between pre-and postoperative conditions were removed using non-dimensional temporal-spatial variables (158), GRF and kinetics were normalized to bodyweight (35, 36).

In the Plug-in-Gait model (155, 156) extrinsic variation and measurement errors are mainly related to the use of skin-mounted markers, with skin motion artefact (181), and model defined rotation axes that are offset from true anatomical joint axes (76). The model is particularly sensitive to definition of the thigh coordinate system (182), entailing that when thigh markers indicate a direction of the knee joint axis which is offset from the anatomic axis, true knee flexion during the swing phase is projected to the coronal plane as an increase in knee varus or valgus curves (157).The foot segment is modeled as a vector, with joint kinematics and kinetics mainly sagittal plane. After data collection with the current model new models have incorporated optimized joint centre and axes estimations which may reduce extrinsic variability (183).

To minimize known sources of marker placement error, the protocol in our gait lab involved use of the knee varus/valgus curves to identify the presence of crosstalk and thigh rotation offset, and correct the offset during processing (182). Moreover, we mainly utilized sagittal plane kinematic and kinetic outcome variables that are less sensitive to marker placement error and have acceptable variability (159, 161). Forefoot markers were consistently placed proximal on the base of the 2nd metatarsal bone to minimize effect of flexible feet/midfoot break to cause 'false' dorsiflexion during single support. To ensure optimal estimation of ankle dorsiflexion with AFOs and shoes, the heel-to toe drop of the shoe was measured, with heel- and forefoot markers placed accordingly, as described in Baker (157).

Data Analysis

In most statistical analyses there is an assumption that observations are independent. However, this may not hold in situations where repeated measurements are made on the same individual. In the current PhD project, most comparisons were based on repeated measures in a cohort of children with spastic CP, where each child was considered as his or her own control. Even though it is considered a strong design, such data will be correlated, i.e. more similar within each individual and therefore dependent. Statistical methods were selected that accounted for these concerns in the most appropriate ways, using paired T test (Paper I), linear mixed models (Paper II and III) (169, 170), functional mixed effects model (172) and generalised additive models (Paper III) (168). An advantage of mixed model analysis is that correlation between observations can be handled using random effects. Effectively, the output of a simple analysis will be the same as performing paired T-tests. Uneven spacing of the repeated measurements and missing data are accepted which means all valid data can be included for analysis (169, 170, 174). For example, participants who had invalid force plate data in one condition could still have data from the other two conditions included for analysis. Also, normal distribution of the data is not required provided residuals have a normal distribution (173). Mixed model analysis allowed us to compare fixed effect of condition, i.e. clinical intervention, while accounting for random subject effects. In comparison, ANOVA for repeated measures require correction of p-value with increased number of repeated measurements that might give overly conservative levels, disguising a real effect and increasing the risk of type II error(173).

In Paper I we divided the sample into subgroups of children who used ground reaction AFOs and hinged AFOs respectively, and performed the paired samples T test separately in each group. In review, we should have taken into account the repeated measures made on the same variable, adjusted the p-value 0.05/3 and used $p = 0.017$ for the repeated subgroup tests. Since the reported differences were all significant with $p < 0.017$ the conclusions are however valid.

5.2 Results

Postoperative changes and residual gait problems

In all three Papers we found changes that indicated overall improvement in gait quality, kinematics and kinetics walking barefoot at the one-year postoperative follow-up with 3D gait analysis. The GPS improved from an average 17.3° (4.6°) preoperatively to 12.3° (2.8°) postoperatively in children with bilateral CP (Paper I), which is a reduction three times the minimal clinically important difference of 1.6° (166). In children with unilateral CP (Paper II) a reduction of the GPS from 12.6° (3.1°) preoperatively to 10.1° (2.4°) postoperatively meant a clinically important but less substantial improvement. Our findings reflect those by Rutz et al who after multilevel surgery in ambulating children with bilateral CP found that the children with higher GPS and more abnormal preoperative gait patterns improved most, possibly since they have higher potential for improvement (184).

Both groups exceeded the average normal GPS of 5.3° (1.8°) substantially, and single kinematic and kinetic variables deviated from normative ranges postoperatively, suggesting gait problems such as crouch (Paper I) and drop-foot/dynamic equinus (Paper II) were present walking barefoot one year postoperatively. Walking speed and step length was reduced indicating reduced gait capacity. Investigation of the vGRF demonstrated significant changes from a pattern with Ben Lomonding and CoM deceleration deficiency preoperatively (63), which was improved postoperatively (Paper III). However, late stance vGRF below bodyweight indicated remaining deceleration deficiency, insufficient bodyweight support and stance stability walking barefoot one year postoperatively.

Impacts of AFOs

Major impacts walking with AFOs on kinematic, kinetic, and temporal-spatial gait variables at the one-year postoperative 3D gait analysis differed between children with bilateral and unilateral CP. In the bilateral group (Paper I) the GPS improved significantly with AFOs, besides control of excessive stance ankle dorsiflexion, improved knee extension moments, and minimum knee flexion in the children who used ground reaction AFOs. Improvement of the GPS was less pronounced in the unilateral group (Paper II), and the major impacts of AFOs were correction of drop-foot, improved knee extension, ankle dorsiflexion and prepositioning for initial contact. Walking with AFOs caused significantly improved temporal-spatial variables with increased walking speed, step length, and decreased cadence compared to barefoot postoperatively (Paper I & II). Furthermore, increased vGRF magnitudes indicated improved stability in stance, CoM deceleration and ability to support bodyweight with AFOs (Paper III).

In a recent study, Schwartze et al investigated whether walking with AFOs lead to additional improvements of gait after multi-level surgery in 20 children with bilateral CP. They reached similar conclusions with regards to improved walking speed, found a non-significant reduction/improvement of the GPS, and significantly improved Gilette Gait Index (185). However, kinetic variables were not assessed, other kinematic variables were part of the Gilette Gait Index and few were directly comparable with the outcome measures in our study. Stance minimum knee flexion or knee moments were not reported that could have elucidated the presence of crouch postoperatively and correction with AFOs.

Influence of AFO types

Subgroup and fixed effects analyses reflected that the various AFO types were intended for specific gait patterns and affected gait accordingly. In Paper I the children who used ground reaction AFOs experienced significant improvements with regards to reduced excessive maximum ankle dorsiflexion and reduced minimum stance knee flexion, implying that solid- or ground reaction AFOs re-established the plantarflexion –knee extension couple and was indicated in children who walked with residual crouch. This is in line with previous recommendations regarding continued use of solid AFOs after lower limb surgery in children with CP (6). The mean (SD) minimum knee flexion of 13.9° (13°) walking barefoot 1 year postoperatively was reduced to 8.2° (10°) with ground reaction AFOs. Our results, together with reduced maximum ankle dorsiflexion from 15.8° (7.2°) to 5.8° (4.3°), are in accordance with Rogozinski et al who demonstrated that ground reaction AFOs can diminish crouch by restricting excessive stance ankle dorsiflexion. In their study, minimum knee flexion in stance was improved from a mean 29° walking barefoot to 18° with ground reaction AFOs (132). Similarly, Böhm et al found improvement from 36° to 21° minimum knee flexion in good responders to ground reaction AFOs (145). In our cohort there was less severe crouch since contractures had been corrected surgically, which we assumed enhanced the efficacy of AFOs to apply adequate mechanical support during the postoperative period (1). The combined treatment, using ground-reaction AFOs "until stable biomechanical realignment of the lower limbs during gait was achieved" as described by Rodda et al (55) probably collaborated to the improvements one year postoperatively. Effects of ground reaction AFOs have been found in a randomized controlled study of a 12-week therapy program, where the intervention that combined ground reaction AFOs and de-rotational strapping improved stance knee and hip flexion more than conventional treatment with or without straps (147).

Children who used hinged AFOs among the bilateral group experienced few changes, possibly because they had less severe gait problems postoperatively and therefore more moderate impacts of AFOs. Hinged AFOs were prescribed preoperatively and mainly for children with true equinus gait pattern who underwent triceps surae lengthening at a single level. Therefore, the potential for change was different both with regards to AFO type and gait pattern.

In Paper II 23 of 33 (70%) used hinged AFOs. Despite uneven distribution the influence of AFO type was tested as a fixed factor, revealing significantly increased stance maximum ankle dorsiflexion with hinged versus ground reaction AFOs. This is logical since hinged AFOs allowed dorsiflexion and tibial progression over the stationary foot, which was enabled by increased ankle range of motion after surgical correction of ankle equinus. In agreement with previous studies both AFO types corrected dynamic equinus/drop-foot at initial contact (112, 130, 140), and indirectly contributing to improved knee extension at initial contact (112, 134). We found a decrease in ankle power generation during push-off due to restrained ankle plantarflexion in 3rd rocker with AFOs, also confirming the work of others (112, 114, 130, 139).

Late stance vGRF increased most in the children with unilateral CP (Paper III). This was also the group where hinged AFOs were most prevalent (23 of 32). We therefore postulated that increased range for ankle dorsal- and plantarflexion with hinged AFOs was beneficial for CoM deceleration and bodyweight support. Unpublished results indicated increased vGRF magnitudes in both weight acceptance and late stance with hinged AFOs versus solid AFOs when tested across bilateral and unilateral groups and the entire vGRF curve. However, effect of CP type yielded similar results and it is possible that there was a combined effect of AFO and CP type, re discussion about internal validity. Decreased late stance vGRF has been found walking with solid versus hinged AFOs, although the experimental study used a normal adult sample (186). Further multifactorial analyses with larger and more balanced samples and controlled variations in AFO design may provide enhanced guidelines regarding optimal AFO configurations for ground reaction force transfer, CoM and bodyweight support.

Adverse effects of allowing free dorsiflexion but blocking plantarflexion in a hinged AFO may be obstruction of the plantarflexion - knee extension couple, over-lengthening of the soleus muscle and a shift of a tight or spastic gastrocnemius muscle to act as knee flexor instead of plantar flexor (51). The fact that eight children in the bilateral cohort underwent tendo-achilles lengthening and used hinged AFOs at the one-year follow-up was disturbing, but average stance knee and ankle kinematics were within normal ranges postoperatively confirming adequate orthotic prescription.

Immobilization of ankle motion with AFOs prevents the foot rockers and reduces muscle work about the ankle during the gait cycle (116, 140). Previously, variation of rotational stiffness around the ankle in AFOs was found to positively affect energy storage, push-off, and energy cost for patients (137). Optimization with spring-like AFOs improved stance knee and ankle kinematics and kinetics in children with CP (113). In the postoperative period optimized AFOs may enhance compliance and mechanical efficacy of the orthoses. Future studies should therefore not only focus on the effects of conventional AFO types but evaluate the influence of variations in AFO joint rotational stiffness and dynamic AFO designs for defined gait patterns.

Indications for continued use of AFOs at the one-year follow-up

The need and indications for continued use of AFOs depend on whether there are remaining gait problems one year postoperatively, and the efficacy of AFOs to provide the necessary additional corrections. Predictors for clinically important difference in the GPS were evaluated in Paper I where we discovered that children with more severe gait dysfunction preoperatively had a stronger benefit of using AFOs at the one-year follow-up. Possibly, minimum stance knee flexion walking barefoot postoperatively was also a strong predictor of improvement in our study had it been tested as a covariate. Crouch was undoubtedly the strongest indication for continued use of AFOs in this group. The gait pattern was most pronounced in the subgroup that used ground reaction AFOs, which gave more potential for correction. In accordance, Ries & Schwartz found that the more stance ankle dorsiflexion and knee flexion, the larger the impact of ground reaction AFOs to reduce crouch (146).

In unilateral CP (Paper II) 21 children exhibited a type 1 gait pattern with drop-foot or dynamic equinus one year postoperatively. Six children did not change after surgery and remained in type 2 gait pattern with true equinus, one remained in type 4, and two deteriorated and exhibited a crouch pattern on the affected side, possibly due to over lengthening of the triceps surae (46). The main indication for continued use of AFOs was drop-foot postoperatively with insufficient prepositioning for initial contact, although all gait patterns indicated continued use of AFOs (3). Previously, drop-foot or dynamic equinus has been identified as a common manifestation after triceps surae lengthening. The only predictor of normalized ankle prepositioning for initial contact postoperatively was normal selective ankle dorsiflexion control preoperatively (48). Meanwhile, recurrent equinus after triceps surae lengthening is common. Borton and colleagues reported that 38% of children with unilateral CP were in equinus 5-10 years post isolated triceps surae lengthening (50), whereas Joo et al conveyed that 62.5% required repeated triceps surae surgery (98). Neither study recommended prophylactic use of AFOs in the groups that were most at risk. Rather, it was advised that children in the risk groups should be prepared for the possible event of repeated triceps surae lengthening (50, 98).

Special caution has been recommended in patients with young age at surgery who are at risk of residual gait problems that may arise with pubertal growth spurt (49, 50, 53). Prolonged use of orthoses may therefore be indicated particularly in children with young age at surgery where the risk of recurrent equinus is high (27, 50, 98). Likewise, children with bilateral CP have higher incidence of crouch particularly after tendo-achilles lengthening, which also could indicate prolonged use of AFOs (6, 49, 50).

An important purpose of AFOs in ambulating children with CP is to provide stability in stance (43), and AFOs have been promoted to reduce CoM deceleration deficiency (63). We found that late stance vGRF was generally below bodyweight walking barefoot postoperatively, suggesting insufficient stance stability in bilateral and unilateral groups (Paper III). Early studies of ground reaction forces and influence of AFOs in children with CP observed that most children had reduced second peak (FZ_2) of the vertical GRF less than bodyweight when walking barefoot. With optimised AFOs the FZ_2 increased above bodyweight whereas increased stance stability reduced the impact forces in weight acceptance (187). In a larger sample of children with bilateral and unilateral CP our results confirmed increased late stance vGRF force magnitudes with AFOs and stability in stance with AFOs. In contrast, vGRF magnitudes also increased in weight acceptance, a change which had moderate to strong positive correlation with walking speed.

Clinical recommendations concerning continued use of AFOs were noted in the individual patients' journal/chart, based on the 3D gait report and multidisciplinary team consent. They were made independent from the current PhD project and are reported as descriptive variables in Paper I and II. In the study involving bilateral CP (Paper I) 35 % had improvements with AFOs that exceeded the MCID in the GPS of 1.6°(166). Even so, 85% of the children were recommended to continue using orthoses after the postoperative follow-up, possibly due to factors that were not detected by the GPS. It has been suggested that summary measures and gait indices does not contain sufficient detail to evaluate AFO efficacy (131). The GPS is based only on kinematics, while kinetics and temporal-spatial variables are also important to evaluate the impact of AFOs. In the unilateral group (Paper II) 32 of 33 participants were recommended continued use of AFOs. The main indication was remaining drop-foot/dynamic equinus, which was corrected with AFOs, resulting in improved ankle and knee prepositioning. The recommendations corresponded to treatment algorithms for the respective gait pattern (3), and was in this respect reasonable, even though there was no significant improvement with regards to the GPS, and few individual participants experienced an MCID ≥1.6° walking with AFOs versus barefoot postoperatively in this group. An intention to slow or prevent progression of crouch or recurrent equinus by using AFOs until the children reach skeletal maturity was probably a contributing factor and the main reason why some patients prophylactically were recommended continued use, despite moderate gait deficits and kinematic variables within normal ranges postoperatively. An important purpose with AFOs is to protect the foot against destructive load and stabilise the foot lever arm. If there was foot deformity or instability with midfoot break this may have been a clinical indication for recommending continued use of AFOs which could not be measured with the conventional Plug-in-Gait model.

6 Conclusions

We demonstrated the functional impacts of AFOs one year postoperatively in children with bilateral and unilateral CP, assessing 3D kinematic, kinetic and temporal-spatial variables within activity, and body functions and structures domains of the ICF. A new method was explored to compare vGRF curves, examining the mechanical influence of orthoses to support bodyweight and enhance stability in stance. All three papers in this thesis demonstrated additional changes consistent with improvement walking with AFOs at the one-year postoperative follow-up. Children with the most severe gait problems preoperatively, remaining crouch or dynamic equinus/drop-foot postoperatively experienced the strongest benefit of walking with AFOs after the one-year postoperative follow-up.

Impacts of different AFO types were related to postoperative gait patterns and residual gait problems postoperatively. This was most obviuos in the bilateral cohort where residual crouch postoperatively was improved with reduced minimum knee flexion in the group using ground reaction AFOs. In line with earlier studies we found recommendations regarding continued use of AFOs in the majority of the children, verifying that planning for on-going care is an important objective with the analysis.

Paper I: Children with bilateral CP, GMFCS I-III, experienced improvements in the GPS, gait kinematic, kinetic and temporal-spatial variables walking with AFOs compared to barefoot one year after lower limb surgery. Stronger impact of AFOs was found in children who had more severe gait dysfunction preoperatively, and remaining crouch was corrected in children who used ground reaction AFOs.

Paper II: In children with unilateral CP ankle equinus was reduced, but the majority of children had residual drop-foot walking barefoot postoperatively. Besides improved walking speed and step length, correction of drop-foot, dynamic equinus and improved prepositioning for initial contact at the ankle and knee were the main impacts of AFOs and indications for continued use of AFOs after the one-year postoperative follow-up.

Paper III: The vGRF was responsive to evaluate treatment of gait problems with surgery and AFOs in children with CP. Fewer children walked with Ben Lomonding postoperatively, seen as decreased forces in weight acceptance and increased forces in late stance. With AFOs additionally increased vGRF magnitude indicated clinically important improvement in stance stability with reduced CoM deceleration deficiency.

Clinical Implications and Future Perspectives

The one-year postoperative 3D gait analysis is a valuable method to evaluate treatment outcome and impacts of AFOs after lower limb surgery in children with bilateral CP. Practitioners and patients should be prepared that gait problems are not completely resolved at the one-year follow-up, and that rehabilitation may require more time. Prolonged use of AFOs is often necessary, particularly for the children who have severe gait dysfunction preoperatively, who require more surgery and mechanical support to maintain the surgical corrections, but also in children who are at risk of recurrent deformity such as ankle equinus.

To help motivate children and parents and clarify why continued use of AFOs is necessary, recommendations should be well-founded, preferably based on clinically important improvements, i.e. meaningful change from the patient and clinician perspective. Both the MAP of the GPS and the vGRF are two visually effective variables that are feasible to demonstrate results pre- versus post intervention, including the efficacy of AFOs.

Future studies should evaluate the bearing of the surgical procedures on AFO efficacy comparing preoperative AFO condition with the postoperative AFO condition. Stratification by functional level, AFO type, gait pattern and/or type of surgery should be considered.

We have not investigated how many children in the cohorts actually continued using of AFOs, for how long or whether continued use of AFOs gave better prognosis of their gait function. It would be interesting and important to evaluate the prophylactic value of continued AFO use to prevent residual deformity and reduce the risk of recurrence after surgery. According to the research questions there are many ways of conducting such studies. A register study involving the children who were followed in CPRN and CPOP is one possible method; using the reported information on range of movement, AFO use, surgery and motor function. A randomized controlled study starting after the one-year postoperative follow-up could provide higher-level evidence regarding effects of AFOs to prevent recurrent deformity and maintain surgical corrections.

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Papers I-III

II

Original Clinical Article

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Comparison of gait with and without ankle-foot orthoses after lower limb surgery in children with unilateral cerebral palsy

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Abstract

Purpose Children with spastic unilateral cerebral palsy (SUCP) frequently undergo lower limb surgery to improve gait. Postoperatively, ankle-foot orthoses (AFOs) are used to maintain the surgical corrections and provide adequate mechanical support. Our aim was to evaluate changes in gait and impacts of AFOs one-year postoperatively.

Methods In all, 33 children with SUCP, 17 girls and 16 boys, mean age 9.2 years (5 to 16.5) were measured by 3D gait analysis walking barefoot preoperatively and walking barefoot and with AFOs one-year postoperatively. Changes in Gait Profile Scores (GPS), kinematic, kinetic and temporal spatial variables were examined using linear mixed models, with gender, gross motor function and AFO type as fixed effects.

Results The results confirm significant gait improvements in the GPS, kinematics and kinetics walking barefoot one year after surgery. Comparing AFOs with barefoot walking postoperatively, there was additionally reduced ankle plantarflexion by an average of 5.1° and knee flexion by 4.7° at initial contact, enhanced ankle moments during loading response, increased velocity, longer steps and inhibited push-off power generation. Stance and swing phase dorsiflexion increased in children walking with hinged AFOs *versus* children walking with ground reaction AFOs. Changes in the non-affected limbs indicated less compensatory gait postoperatively.

Conclusion Major changes were found between pre- and postoperative barefoot conditions. The main impact of AFOs was correction of residual drop foot and improved prepositioning for initial contact, which could be considered as indications for continued use after the one-year follow-up.

Level of Evidence: Level II - Therapeutic

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Keywords: gait; ankle-foot orthoses; orthopaedic surgery; postoperative; unilateral cerebral palsy

Introduction

Gait deviations are common in children with spastic unilateral cerebral palsy (SUCP). This is mainly due to ankle equinus but involvement at the proximal joints also occurs.[1-3] Early treatment often includes a combination of physiotherapy, serial casting, ankle-foot orthoses (AFOs) and injections of botulinum toxin A to reduce spasticity in the triceps surae muscle and maintain adequate ankle joint range of movement. In cases where fixed deformities impair functional ambulation, orthopaedic surgery may be necessary. In the postoperative rehabilitation period, different types of AFOs are routinely used to provide adequate mechanical support during gait and prevent recurrence of deformities.[2,4-8]

Previous studies using 3D gait analysis (3DGA) have found that surgery at single[7,8] or multiple[5,6] levels improved gait kinematics and kinetics in children with SUCP. Still, residual gait problems, such as drop-foot in the swing-phase are common.[2,8,9] Recurrent equinus has been reported in 38% to 62.5 % of patients with unilateral cerebral palsy (CP) five to ten years after triceps surae lengthening.[10,11] It is, therefore, not surprising that the one-year postoperative evaluation with 3DGA often results in recommendations regarding further treatment, such as prolonged use of orthoses, to prevent recurrent deformities.[8,12]

Several studies have provided valuable documentation regarding effects of orthoses on gait.[13-16] To our knowledge there is no existing study that has evaluated the impact of AFOs after lower limb surgery in children with SUCP. This

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might be important to provide realistic perspectives for the patients, families and caregivers and to establish indications for continued use of AFOs after surgery.

The aim of the present study was to investigate changes in gait function one year after lower limb surgery in children with SUCP. Our objectives were to evaluate if gait function was improved after surgery and whether further changes take place when walking with AFOs compared with barefoot at the one-year postoperative follow-up with 3DGA.

Patients and methods

Participant selection

We included children with SUCP, who underwent preoperative 3DGA and lower limb surgery including triceps surae lengthening to treat ankle equinus, and who used AFOs at the time of postoperative 3DGA. Consecutive sampling during a four-year inclusion period resulted in 43 patients who received written information about the study. Ten patients did not respond or wish to participate which resulted in 33 included patients (17 girls and 16 boys) who gave written informed consent. A total of 22 children were classified as level I and 11 children as level II according to The Gross Motor Function Classification System (GMFCS).[17] The study was approved by the Regional Ethics Committee (REC; 2013/1242).

Data collection

All children were measured with 3DGA in three conditions; preoperatively walking barefoot, postoperatively walking barefoot and postoperatively walking with AFOs and shoes. Data was captured using a Vicon system (Vicon Motion Systems Ltd., Oxford, United Kingdom) with six infrared cameras (Vicon MXF40) and three force plates (AMTI OR6-7, Advanced Mechanical Technology Inc., Watertown, Massachusetts). Two experienced testers (IS or ABH plus one physiotherapist) reached agreement on marker placement, following the Plug-in-Gait model and marker protocol.[18] Participants were walking at self-selected speed across a 12-metre walkway until a minimum of three trials containing valid kinetic and kinematic data was captured. Data processing with Vicon Nexus software included definition of gait events, i.e. initial contact and foot off, which were determined on the force plates and correlated to all gait cycles in the trial. Prior to the walking trials, a standardized physical examination of joint range of movement, muscle strength, tone and selective motor control was performed. In the postoperative conditions, participants were first measured barefoot. After ten minutes rest, measurement commenced with AFOs and with shoes only on the non-affected side. With AFOs the pelvis, thigh and knee markers remained on the skin from the barefoot session. Shank and foot markers were repositioned on AFOs and shoes in optimal agreement with movement and segment axes. Differences in shoe heel height were accounted for by measuring the heel-to-toe drop of the shoe sole using an outside calliper, placing the heel marker accordingly higher than the forefoot marker on the shoes and not assuming that the markers were horizontal during static processing.[19]

According to typical procedure, a multidisciplinary team of child neurologist, certified prosthetist orthotist (CPO) (IS, ABH), physiotherapist and orthopaedic surgeon evaluated the pre- and postoperative 3DGA. This involved assessment of patient's gait curves against normative curves from our reference database of 24 typically developing children (11 girls, 13 boys) with a mean age of 9.8 years (5 to 15). Normal ranges were defined as mean (SD). Gait patterns were categorized according to Winters et al[3] into four types: children with Type 1 pattern walk with dynamic ankle equinus or drop-foot in swing; Type 2 walk with true equinus, with the knee in extension or recurvatum during stance; Type 3 with true equinus and flexed knee during stance; and Type 4 present with a stronger proximal involvement, usually with frontal and transverse plane deviations. Each participant's preoperative gait pattern,[3] physical examination and the treatment algorithms suggested by Rodda and Graham[2] guided the decisions regarding surgery and postoperative follow-up, including the type and function of orthoses. Using the Silverskiolds test, children with passive dorsiflexion to 0° with knee flexed usually underwent gastrocnemius recession and children with passive dorsiflexion less than 0° with knee flexed had tendo-achilles lengthening. Treatment recommendations were specified in the children's gait reports. For descriptive analysis, we reviewed the gait reports to register recommendations regarding continued use of AFOs following postoperative 3DGA, and the distribution of gait patterns pre- and postoperatively.

AFOs

In children with Type 1 or 2 gait patterns,[3] who underwent triceps surae lengthening for equinus, AFOs were constructed to allow ankle dorsiflexion, restrict plantar flexion and lift the foot in swing and categorized as hinged AFOs (HAFOs). HAFOs were made with 2.5-mm to 3-mm polypropylene-butylene and integrated joints (Tamarack, Blaine, Washington), with dorsal leg shell to below the fibular head, a circular total-contact foot part and flexible long sole (past the toes) (Fig. 1a). In children with Type 3 or 4 patterns,[3] who underwent hamstrings lengthening and/or rectus femoris transfer, AFOs should restrict dorsal and plantar flexion and apply an external knee extension moment during stance and were categorized as ground

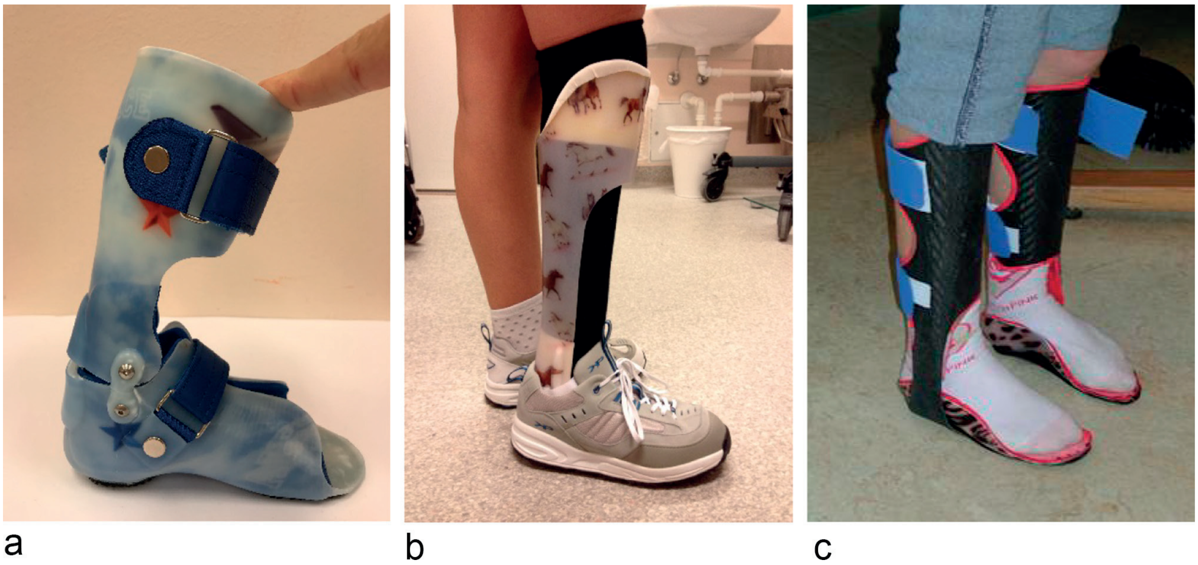

Fig. 1 The different types of ankle-foot orthoses (AFOs) used at the postoperative gait analysis: (**a**) hinged AFO with dorsal shell and foot part connected by integrated Tamarack flexure joints (Tamarack Habilitation Technologies Inc, Blaine, Washington); (**b**) polypropylene ground reaction AFO (GRAFO), with a ventral shell to mid-patella; as shown with shoes and standing alignment; (**c**) carbon composite GRAFOs with ventral shell to below patella.

reaction AFOs (GRAFOs). GRAFOs were fabricated solid, in 5-mm to 6-mm polypropylene, with a ventral shell extending to mid-patella or in carbon composite with a ventral shell to below patella, both with stiff long soles (Figs 1b and 1c).

Casting for postoperative AFOs was made by CPOs after surgical closure. Splints to immobilize ankles in 0° to 5° of dorsiflexion were applied for five weeks. Physiotherapy was started the first day postoperatively and continued throughout the rehabilitation period. Immediately after splint removal, AFOs and shoes were fitted, using a standing alignment with 5° to 10° shank inclination. The children were instructed to use the orthoses all day until the one-year postoperative 3DGA evaluation.

Outcome measures

As a summary measure of gait quality, we calculated the Gait Profile Score (GPS) which is based on nine kinematic Gait Variable Scores (GVS).[20] The GVSs are root mean square differences between patient's sagittal, transverse and frontal plane gait curves and averaged gait curves from our reference database of 24 children with no gait pathology. Smaller GVS and GPS values indicate gait closer to normal. We also analyzed six kinematic and three kinetic variables that were considered relevant to evaluate the outcome after surgery and the impact of AFOs. These included ankle and knee angle at initial ground contact, maximum ankle dorsiflexion during stance and swing phases, stance minimum knee and hip flexion, ankle mean moment during loading response in 0% to 10% of the gait cycle, maximum external dorsiflexion moment and maximum power generation in terminal stance. Temporal-spatial variables were non-dimensional gait velocity, step length and cadence, normalized by body height to account for growth between the pre- and postoperative evaluations.[21]

Statistical analysis

From every participant, data from three trials in each condition were averaged and used in the statistical analysis (SPSS 21 for Windows; IBM Corp., Armonk, New York). Data from both limbs were split and the affected and non-affected side analyzed separately. For kinematic and kinetic data analysis one gait cycle per trial was used whereas temporal-spatial data used all available gait cycles (three to four) within each trial. Distributions of the outcome variables and model residuals were tested using the Kolmogorov-Smirnov test. To account for possible correlation between repeated measurements made on the same individual, changes in each outcome variable were analyzed using linear mixed models.[22] The postoperative barefoot condition was defined as the reference category against which the preoperative barefoot and postoperative AFO conditions were compared, respectively. In the model, participants were defined as random effects, whereas fixed effects included gender, GMFCS level and AFO type (HAFOs *versus* GRAFOs) and fixed effects' interaction with each condition. Variance components were used as covariance structure and model selection was based on significance and Akaike's information criterion. The level of significance was set at $p < 0.05$.

Results

Participants

Individual characteristics, including surgical procedures, AFO types and gait patterns are presented in Table 1. GVS components and GPS are displayed in the movement analysis profile (MAP; Fig. 2). The mean age at time of surgery was 9.2 years (5 to 16.5) and mean time from surgery to postoperative 3DGA was 15.5 months (11 to 27). In all, 23 children underwent tendo-achilles lengthening and ten underwent gastrocnemius recession. Concomitant procedures were performed in ten children with tendo-achilles lengthening and two children with gastrocnemius recession. At the one-year postoperative 3DGA 23 children used HAFOs and ten children used GRAFOs, of which three were made of polypropylene and seven in carbon composite. The postoperative gait reports revealed that the multidisciplinary team recommended continued use of AFOs in 32 children (Table 1).

The most frequent gait pattern preoperatively was Type 2 (n = 22) whereas Type 1 was predominant postoperatively (n = 21). Two children deteriorated to a more severe gait pattern after surgery, nine were unchanged and 22 children had improved and showed a less severe gait pattern type. This included three who were within normal ranges after surgery and not classifiable.

Preoperative barefoot versus postoperative barefoot

The mean GPS on the affected side was significantly reduced from 12.6° (SD 3.1°) preoperatively to 10.1° (SD 2.4°) walking barefoot postoperatively (Table 2). Other significant changes were reduced ankle plantarflexion by 7.2° and knee flexion by 3.7° at initial contact, increased ankle maximum dorsiflexion by 14° in stance and 11° in swing, decreased minimum hip flexion and reduced cadence postoperatively. Significant changes in the kinetic variables included reduced external dorsiflexion moments during loading response and increased stance maximum dorsiflexion moment and ankle power generation (Table 2).

Table 1 General table with participant characteristics, type of surgery, type of ankle-foot orthosis (AFO) and recommendations regarding continued use

ID	Gender	Affected side	Age at surgery (yrs)	GMFCS	Preop pattern	Postop pattern	Surgery	Type of AFO	Recommendation AFO
1	M	Left	12.5	I	Type 4	Type 4	TAL, Psoas	HAFO	1
2	F	Left	6.5	I	Type 2	Type 1	GR	HAFO	1
3	F	Right	8	I	Type 2	Type 1	TAL,TibPT	GRAFO	1
4	F	Right	5.5	I	Type 2	Type 1	TAL	HAFO	1
5	M	Left	9.5	II	Type 2	Type 1	TAL	HAFO	1
6	F	Left	6	I	Type 2	Type 2	TAL	GRAFO	1
7	M	Right	7	I	Type 2	Type 1	TAL	HAFO	1
8	M	Right	16.5	I	Type 2	Type 1	GR	GRAFO	1
9	F	Left	13	I	Type 2	Type 1	TAL	HAFO	1
10	M	Right	15	II	Type 3	Crouch	TAL,TibPT, RFT	HAFO	1
11	F	Right	6.5	II	Type 3	Type 1	TAL, Psoas	HAFO	1
12	M	Right	8.5	I	Type 4	Type 1	TAL, Psoas, Hams	GRAFO	1
13	F	Right	13	II	Type 4	Type 1	GR, Psoas, Hams	GRAFO	1
14	M	Right	8.5	I	Type 2	Type 1	GR, TibAS	GRAFO	1
15	M	Left	7	I	Type 2	Type 1	TAL	HAFO	1
16	F	Right	10	I	Type 2	NC	TAL	HAFO	0
17	F	Right	11.5	II	Type 2	Type 1	TAL, Psoas	HAFO	1
18	F	Left	7	II	Type 2	Type 2	TAL	HAFO	1
19	F	Right	5	I	Type 2	Type 2	GR	HAFO	1
20	F	Left	13.5	I	Type 3	Type 1	TAL, Hams	GRAFO	1
21	M	Left	7	I	Type 2	Type 1	TAL	HAFO	1
22	M	Right	10.5	I	Type 2	NC	TAL	HAFO	1
23	M	Right	5.5	I	Type 3	Type 1	TAL	HAFO	1
24	F	Right	15	II	Type 1	Type 1	GR	HAFO	1
25	M	Left	6.5	II	Type 3	NC	TAL, Hams	GRAFO	1
26	M	Left	8	II	Type 2	Type 2	GR	HAFO	1
27	M	Left	12	II	Type 2	Type 1	GR	HAFO	1
28	F	Right	9	I	Type 1	Type 1	GR	GRAFO	1
29	F	Left	9	I	Type 2	Type 1	TAL	HAFO	1
30	M	Right	6	I	Type 2	Type 2	GR	HAFO	1
31	M	Right	12	II	Type 2	Crouch	TAL, FDO	GRAFO	1
32	F	Left	7.5	I	Type 3	Type 1	TAL, Hams	HAFO	1
33	F	Right	6	I	Type 2	Type 2	TAL	HAFO	1

Recommendation AFO: 0, discontinue; 1, continue

GMFCS, Gross Motor Function Classification System; Preop, preoperative; Postop, postoperative; NC, no classifiable gait deficit; TAL, tendo-achilles lengthening; P, psoas lengthening; GR, gastrocnemius recession; TibPT, tibialis posterior transfer; RFT, rectus femoris transfer; Hams, hamstrings lengthening; TibAS, tibialis anterior shortening; FDO, femoral derotation osteotomy; HAFO, hinged AFO; GRAFO, ground reaction AFO

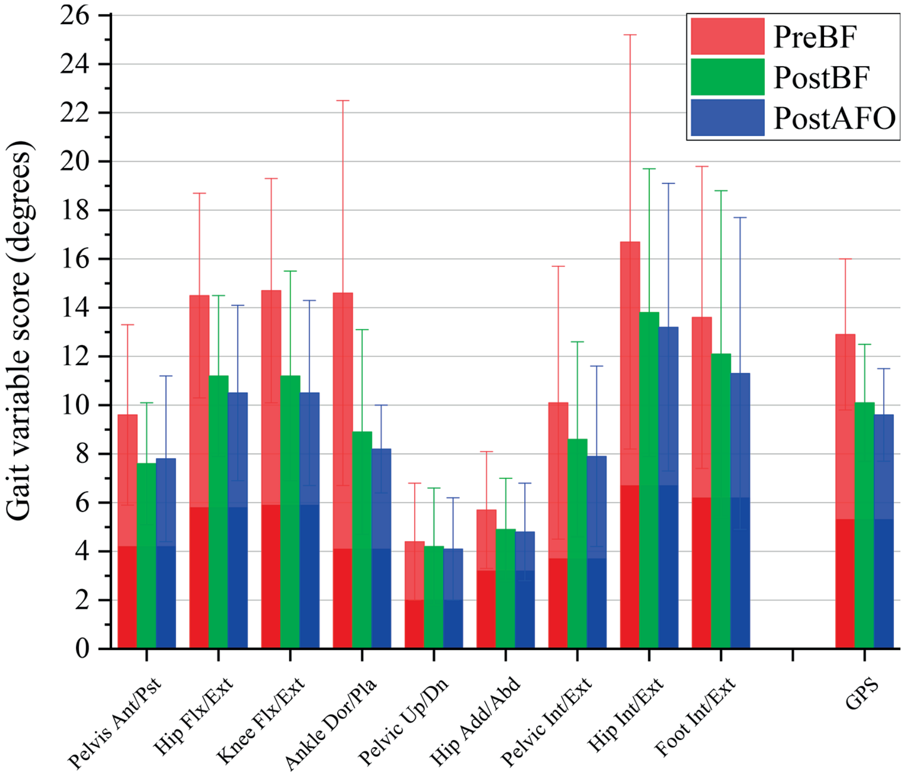

Fig. 2 Movement analysis profile (MAP) with Gait Variable Scores (GVS) and Gait Profile Scores (GPS)[20] in the three measurement conditions: preoperatively walking barefoot (PreBF), postoperatively walking barefoot (PostBF) and postoperatively walking with ankle-foot orthoses (PostAFO). Each column represents the root mean square difference across the gait cycle, averaged and with 1 SD for all participants (n = 33), with mean scores from our normal reference data (n = 24) in the darker base of each column (Ant, anterior; Pst, posterior; Flx, flexion; Ext, extension; Dor, dorsiflexion; Pla, plantarflexion; Dn, down; Add, adduction; Abd, abduction; Int, internal; Ext, external.

Postoperative AFO versus postoperative barefoot

Further reduction of the mean GPS to 9.6° (SD 1.9°) walking with AFOs *versus* barefoot was not significant (Table 2). With AFOs, the main improvements took place at initial contact with a significant reduction of ankle plantarflexion by 5.1° and knee flexion by 4.7°. The moments generated about the ankle joint in 0% to 10% of the gait cycle changed significantly from external dorsiflexion moment walking barefoot, to plantarflexion moments walking with AFOs. Ankle power generation was reduced when children were walking with AFOs. Both gait velocity and step length increased significantly, while cadence was reduced (Table 2).

Non-affected sides

Changes on the non-affected side walking barefoot postoperatively *versus* preoperatively included increased stance maximum dorsiflexion, knee and hip flexion, increased late stance ankle dorsiflexion moment and power generation (Table 3). In the AFO condition and with shoes on the non-affected side, significant changes included reduced knee flexion at initial contact, increased plantarflexion moment in 0% to 10% of the gait cycle, reduced stance ankle maximum dorsiflexion and power generation compared with barefoot postoperatively.

Fixed factors

We found no significant group effect of gender or GMFCS level. The increase in swing phase maximum dorsiflexion in the postoperative AFO *versus* barefoot condition was significant, but only when AFO type was included in the model as a fixed effect (p = 0.015). Interaction effect between AFO type and the postoperative AFO condition indicated that stance ankle maximum dorsiflexion increased by 10.5° (p = 0.010) in children walking with HAFOs *versus* children walking with GRAFOs (see Supplemental Material). Children who used GRAFOs had an estimated 7.6° more knee flexion at initial contact preoperatively (p = 0.027) (Fig. 3).

Discussion

After surgery, improvements were seen for the GPS, key kinematic and kinetic variables on the affected sides. An

Table 2 Changes in gait variables on the affected side

		Affected side			Comparisons (p-value)	
	Reference data	PreBF	PostBF	PostAFO	PreBF *vs* PostBF	PostAFO *vs* PostBF
GPS (°)	5.3 (1.8)	12.6 (3.1)	10.1 (2.4)	9.6 (1.9)	< **0.001**	0.247
Ankle						
Angle at initial contact (°)	-2.2 (3.1)	-18.4 (10)	-11.2 (8)	-6.1 (4.7)	< **0.001**	**0.002**
Maximum dorsiflexion 30% to 60% GC (°)	13.2 (3.9)	-4.8 (12)	9.2 (8.3)	9.9 (7.1)	< **0.001**	0.694
Maximum dorsiflexion in swing (°)	2.9 (3.1)	-16 (10.2)	-5.1 (8.6)	-2.3 (4.5)	< **0.001**	0.098
Mean moment 0% to 10% GC (Nm/kg)	-0.1 (0.08)	0.47 (0.2)	0.3 2 (0.2)	-0.04 (0.2)	**0.001**	< **0.001**
Maximum moment 30% to 60% GC (Nm/kg)	1.2 (0.18)	0.81 (0.2)	1.06 (0.2)	1.13 (0.2)	< **0.001**	0.116
Maximum power 30% to 60% GC (W)	3 (0.9)	1.52 (0.6)	2.16 (0.6)	1.64 (0.7)	< **0.001**	< **0.001**
Knee						
Angle at initial contact (°)	4.9 (4.5)	15.1 (9)	11.4 (7.4)	6.7 (8.7)	**0.022**	**0.004**
Minimum flexion 30% to 60% GC (°)	1.6 (4.4)	4.4 (10.3)	3.5 (8.4)	0.5 (11)	0.613	0.074
Hip						
Minimum flexion 30% to 60% GC (°)	-11.7 (6.4)	-2.9 (7.7)	-5.2 (6.1)	-5.7 (6.6)	**0.038**	0.668
Temporal-spatial						
Non-dimensional velocity (vel/√Hxg)		0.40 (0.04)	0.39 (0.04)	0.45 (0.05)	0.089	
Non-dimensional step length (step/H)		0.33 (0.05)	0.31 (0.05)	0.34 (0.05)	0.075	
Velocity (m/sec)*	1.35 (0.09)	1.19 (0.17)	1.18 (0.17)	1.27 (0.16)		**0.011**
Step length (m)*	0.62 (0.06)	0.53 (0.08)	0.56 (0.07)	0.64 (0.07)		< **0.001**
Cadence (step/min)	133 (8.7)	138 (21)	126 (18)	121 (14)	< **0.001**	0.054

Values are presented as mean (SD)

Reference data, values from our laboratory database of 24 typically developing children

*pre- and postoperative comparisons were performed with non-dimensional values

p-values are from linear mixed model analyses. Bold letters indicate significant difference with $p < 0.05$

PreBF, preoperatively walking barefoot; PostBF, postoperatively walking barefoot; PostAFO, postoperatively walking with ankle-foot orthoses (AFOs); GPS, Gait Profile Score; GC, gait cycle; H, height; g, gravity

Table 3 Changes in gait variables on the non-affected side

		Non-affected side			Comparison (p-value)	
	Reference data	PreBF	PostBF	PostAFO	PreBF *vs* PostBF	PostAFO *vs* PostBF
GPS (°)	5.3 (1.8)	10.1 (1.9)	9.5 (1.9)	9.6 (1.7)	0.064	0.858
Ankle						
Angle at initial contact (°)	-2.2 (3.1)	-2.1 (4.7)	-2.3 (4.7)	-1.9 (5.6)	0.836	0.711
Maximum dorsiflexion 30% to 60% GC (°)	13.2 (3.9)	10.4 (6.4)	13.8 (5.9)	8.7 (6.1)	**0.002**	< **0.001**
Maximum dorsiflexion in swing (°)	2.9 (3.1)	4.1 (4.8)	4.9 (4.7)	3.2 (5.1)	0.400	0.085
Mean moment 0% to 10% GC (Nm/kg)	-0.1 (0.08)	0.053 (0.15)	0.05 (0.15)	-0.06 (0.1)	0.809	< **0.001**
Maximum moment 30% to 60% GC (Nm/kg)	1.2 (0.18)	1.21 (0.3)	1.34 (0.2)	1.4 (0.18)	**0.001**	0.185
Maximum power 30% to 60% GC (W)	3 (0.9)	3.62 (1.14)	4.05 (1)	3.54 (0.8)	**0.011**	**0.003**
Knee						
Angle at initial contact (°)	4.9 (4.5)	7.6 (6.2)	9.8 (6)	6.2 (6.3)	0.068	**0.004**
Minimum flexion 30% to 60% GC (°)	1.6 (4.4)	0.2 (7.1)	4.5 (8.6)	1.9 (7.2)	**0.001**	0.059
Hip						
Minimum flexion 30% to 60% GC (°)	-11.7 (6.4)	-11.8 (5.4)	-9.7 (6.4)	-10.3 (5)	**0.030**	0.543
Temporal-spatial						
Non-dimensional velocity (vel/√Hxg)		0.33 (0.05)	0.31 (0.06)	0.34 (0.06)	0.190	
Non-dimensional step length (stepl/H)		0.39 (0.04)	0.40 (0.04)	0.44 (0.05)	0.860	
Velocity (m/sec)*	1.35 (0.09)	1.19 (0.17)	1.18 (0.17)	1.27 (0.16)		**0.020**
Step length (m)*	0.62 (0.06)	0.52 (0.07)	0.57 (0.07)	0.62 (0.06)		<**0.001**
Cadence (step/min)	133 (8.7)	138 (21)	126 (18)	121 (14)	<**0.001**	0.053

Values are presented as mean (SD)

Reference data, values from our laboratory database of 24 typically developing children

*pre- and postoperative comparisons were performed with non-dimensional values

p-values are from linear mixed model analyses. Bold letters indicate significant difference with $p < 0.05$

PreBF, preoperatively walking barefoot; PostBF, postoperatively walking barefoot; PostAFO, postoperatively walking with ankle-foot orthoses (AFOs); GPS, Gait Profile Score; GC, gait cycle; H, height; g, gravity

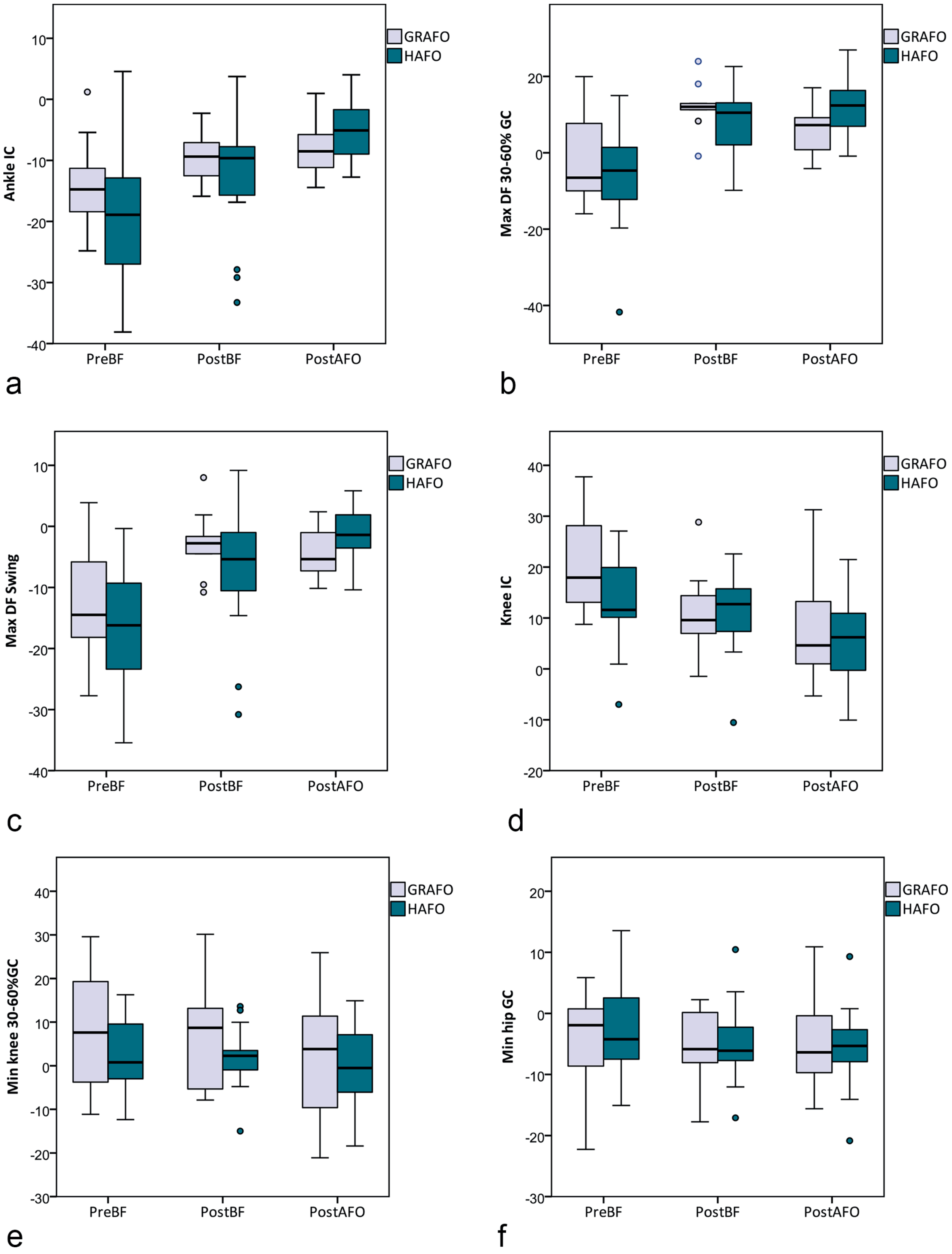

Fig. 3 Box-plots illustrating medians and interquartile ranges for all kinematic variables in the three conditions: preoperatively walking barefoot (PreBF), postoperatively walking barefoot (PostBF), postoperatively walking with ankle-foot orthoses (AFOs) (PostAFO) and clustered by AFO type: (**a**) ankle angle (°) at initial contact (IC), dorsiflexion (DF) positive and plantarflexion negative; (**b**) maximum (max) ankle DF (°) in 30% to 60% of the gait cycle (GC); (**c**) max ankle DF (°) in swing; (**d**) knee angle (°) at IC, knee flexion positive and knee extension negative; (**e**) minimum (min) knee flexion (°) in 30% to 60% GC; (**f**) min hip flexion (°) during GC (HAFO, hinged AFO; GRAFO, ground reaction AFO).

average GPS reduction of 2.5° walking barefoot was more than the previously defined minimal clinically important difference of 1.6°.[23] Still, the average postoperative GPS exceeded the normal range, indicating that the gait problems were not completely corrected. In swing and at initial contact the average ankle plantarflexion implied residual dynamic equinus or drop-foot, which contributed to ground contact with the forefoot and external dorsiflexion moment during loading response. Our results support previous research,[8,9] which stated that while triceps surae lengthening improves dynamic ankle range of movement; swing-phase drop-foot frequently persists, possibly due to inadequate activation of the dorsiflexors. The findings are less consistent with those of Tylkowski et al[7] who reported normalized ankle kinematics in both stance and swing phases after tendo-achilles lengthening in SUCP.

When the children walked with AFOs, the GPS was reduced and closer to normal, but not sufficiently to reach significance. Danino et al[24] questioned whether gait indices such as the GPS are sufficiently sensitive to measure AFO efficiency. In our belief it is an appropriate measure of gait quality, but because it is a summary score calculated across several kinematic components and entire gait cycles, single key variables should also be reported. Similar to previous studies comparing AFOs and barefoot gait in children with unilateral CP, we found improved pre-positioning for initial contact at the ankle[13-15] and knee,[14] and enhanced ankle moments during loading response.[14,15] Our study also confirmed decreased power generation resulting from restricted ankle movement in AFOs.[13-16] However, this decrease in push-off propulsion did not, as previously proposed,[16] have an adverse effect on gait velocity. Velocity increased and gait could be termed more energy-efficient since the children were taking longer steps at a lower cadence walking with AFOs compared with barefoot postoperatively (Table 2). After surgery, there was less knee flexor tightness which resulted in improved terminal swing reach and knee extension at initial contact. Additional knee extension with AFOs *versus* barefoot postoperatively could be explained by less activation of knee flexors secondary to improved ankle prepositioning in the orthoses. Supporting this theory, decreased electromyographic activity in biceps femoris has been found from mid- to terminal swing in children with SUCP walking with AFOs compared with barefoot.[25] Another explanation for additional knee extension is that the distally added weight of shoes and orthoses may increase knee angular momentum.

The most frequent orthoses used in our study were HAFOs, which allow free ankle dorsiflexion and unrestricted tibial progression over the stationary foot during stance. There is concern that this AFO type could overlengthen the soleus muscle instead of treating gastrocnemius tightness.[26,27] Nevertheless, HAFOs are often preferred since they allow more freedom of movement. Also, children with unilateral CP are less at risk compared with bilaterally affected children of developing calcaneal gait secondary to over-lengthening of the triceps surae.[10,28] Our results confirmed increased ankle dorsiflexion in stance and swing phases with HAFOs, supporting their use to maintain functional triceps surae length and to allow range for tibialis anterior activation. Alternatively, preservation of ankle movement during stance and push-off power generation could be optimized using energy-storing carbon fibre springs[14] or joints with dynamic response to plantar- and dorsiflexion.[16]

After surgery, significant changes consistent with improvement were also seen in the non-affected limbs, indicating that changes on the affected side may influence gait bilaterally. Increased ankle dorsiflexion, knee and hip flexion in stance postoperatively suggest that compensatory vaulting, or limb extension, was no longer necessary to ensure opposite foot clearance during swing. However, in the AFO condition, stance ankle dorsiflexion decreased in the non-affected limb. This was surprising, since less compensatory vaulting should have been necessary when AFOs enhanced swing phase clearance on the affected side. One explanation is that the added shoe heel height may leave the ankle on the non-affected side more plantarflexed relative to the floor.

Recommendations to continue with AFOs were in most cases in accordance with the treatment algorithms defined by Rodda and Graham for the various gait types.[2] The main change in pattern after surgery was to Type 1 which requires an AFO to correct drop-foot.[2] However, one child with crouched gait pattern was recommended to continue with HAFO, which is not mechanically appropriate to apply an extension moment at the knee. Also, two children with normalized postoperative gait patterns and no apparent need for orthoses were recommended continued use of AFOs. Individual factors, such as foot deformities, pain or patient preferences which might have indicated use of orthoses, were not described or documented as part of the present study.

Borton et al[10] found that children with SUCP had a prevailing 38% risk of recurrent equinus deformity five to ten years after isolated calf muscle lengthening. Over ten years, Joo et al[11] found that 62.5% of the children with SUCP underwent repeated surgery to treat recurrent equinus. Risk factors for recurrent equinus were young age at surgery (≤ 8 years)[11] and male gender,[10] with higher incidence in unilateral *versus* bilateral CP.[10,11] While neither study assessed use of AFOs after surgery as a factor, both raised doubts concerning the preventive effect of AFOs. Instead, reoperation was recommended in cases where deformity recurred. It appears, however, more viable to prescribe conservative treatment, such as AFOs, in cases where deformity is expected. Our study confirmed the functional efficacy of AFOs one-year postoperatively, particularly in improving swing phase clearance and prepositioning of the foot for initial contact. However, longer-term results are warranted to investigate the role of AFOs in reducing the risk of recurrence after surgery. Previously, maintenance of passive and active ankle range of movement with AFOs has been demonstrated over a one-year study period.[13] Hosl et al[27] found that after on average 16 weeks (SD 4) of AFO use, passive ankle dorsiflexion improved, but gastrocnemius fascicles shortened, and muscle volume decreased. Nevertheless, the adverse changes in muscle morphology were considered as outweighed by functional gains related to increased gait velocity and improved ankle kinematics with AFOs.

There were some limitations to this study. We focused on changes in the sagittal plane and evaluation of transverse and frontal planes was limited to the GVS elements of the GPS. The small number of participants may have influenced the power of the statistical analyses, particularly in analyses of fixed effects and grouped data. In some cases, the time from surgery to postoperative gait analysis was considerably delayed (up to 27 months) and 12 children received concomitant lower limb surgeries, which added heterogeneity to the sample and could have had an impact on the results. Best-practice guidelines recommended a shoes-only instead of barefoot control condition in studies evaluating the effect of AFOs.[29] However, many children experience fatigue during testing and barefoot data was therefore prioritized for comparison with preoperative data. Previously, no clear difference was found in barefoot *versus* shoes-only conditions.[14] Similar results were found by Böhm et al[30] who concluded that barefoot walking is sufficient as control condition when evaluating impacts of AFOs in children with CP. Since preoperative data of participants walking with AFOs was not available, it is difficult to precisely deduce in what way contractures and subsequent surgery influenced AFO efficacy. This is a limitation which should be addressed in future investigations. Further research should also include patient-reported outcomes to evaluate function and satisfaction with the orthoses.

Conclusion

The most clinically significant changes in gait were found between pre- and postoperative barefoot conditions. One year postoperatively, correction of residual drop-foot and improved prepositioning for initial contact at the ankle and knee were the main impacts of AFOs and could be considered indications for continued use.

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COMPLIANCE WITH ETHICAL STANDARDS

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OA LICENCE TEXT

ETHICAL STATEMENT

Ethical approval: The study was approved by the Regional Ethics Committee (REC; 2013/1242). All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Informed consent: Informed consent was obtained from all individual participants included in the study.

ICMJE CONFLICT OF INTEREST STATEMENT

All authors declare that they have no conflict of interest.

AUTHOR CONTRIBUTIONS

IS: Study concept and design, data collection, analysis and interpretation, drafting article and final approval of submitted version.

HS: Interpretation of data, revising article and final approval of submitted version.

ABH: Data collection and interpretation, revising article and final approval of submitted version.

IH: Study concept and design, interpretation of data, revising article and final approval of submitted version.

SUPPLEMENTAL MATERIAL

Supplemental material is available for this paper at https://online.boneandjoint.org.uk/doi/suppl/10.1302/1863-2548.13.180146

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III

Postoperative changes in vertical ground reaction forces, walking barefoot and with ankle-foot orthoses in children with Cerebral Palsy

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Abstract.

Background: Children with cerebral palsy often have problems to support the body centre of mass, seen as increased ratio between excessive vertical ground reaction forces during weight acceptance and decreased forces below bodyweight in late stance. We aimed to examine whether increasing ankle range of motion through surgery and restraining motion with ankle-foot orthoses postoperatively would have impact on the vertical ground reaction force in weight acceptance and late stance.

Methods: Ground reaction forces were recorded from 24 children with bilateral and 32 children with unilateral cerebral palsy, each measured walking barefoot before and after triceps surae lengthening. Postoperatively, the children were also measured walking with ankle-foot orthoses. Changes in vertical ground reaction forces between the three conditions were evaluated with functional curve and descriptive peak analyses; accounting for repeated measures and within-subject correlation.

Findings: After surgery, there were decreased vertical ground reaction forces in weight acceptance and increased forces in late stance. Additional significant changes with ankle-foot orthoses involved increased vertical forces in weight acceptance, and in late stance corresponding to bodyweight (bilateral, from 92% to 98% bodyweight; unilateral, from 94% to 103% bodyweight) postoperatively.

Interpretation: Our findings confirmed that surgery affected vertical ground reaction forces to approach more normative patterns. Additional changes with ankle-foot orthoses indicated further improved ability to support bodyweight and decelerate centre of mass in late stance.

1 Introduction

Reduced motor function in children with cerebral palsy (CP) may affect adequate body centre of mass (CoM) support and stability during stance, which are important prerequisites of functional human gait [1, 2]. CoM support can be expressed by the forces exerted on the ground and the resulting vertical component of the ground reaction force (vGRF). In normal gait, the vGRF has a characteristic M-shape with two peaks, each of approximately equal height, about 120% and 110% of bodyweight, respectively (Fig. 1, blue line).[3-5] The first peak (FZ_1) occurs after loading response with weight acceptance in early stance phase of the gait cycle.[4] The second peak (FZ_2) is associated with push-off in late single stance[4] when the trailing limb decelerates downwards motion of the CoM.[6, 7]

Children with CP often have difficulties to support and decelerate the CoM during late stance. This can be observed as a decrease of the FZ_2 below bodyweight,[4, 8] which often results in rapid weight transfer to the leading limb, enlarged forces (FZ_1) during weight acceptance (Fig. 1, red line) and excessive load on muscles, ligaments and joints. Williams et al named the occurrence of an increased FZ_1 and reduced FZ_2 as 'Ben Lomonding' since the vGRF curve has a similar shape to the Scottish mountain.[8] They categorised 74 ambulating children with spastic CP and found Ben Lomonding in 87%, whereas 66% had difficulty in generating an FZ_2 above bodyweight implying that the child is about to 'collapse', termed CoM deceleration deficiency. It was therefore claimed that clinical interventions to treat gait problems in children with CP should be aimed at improving their ability to support CoM by generating an adequate FZ_2.[8]

In normal gait, the ankle plantarflexor muscles are the main factor to support CoM and increase vGRF magnitudes in late stance.[9] Eccentric work during 2nd rocker controls the GRF producing an external knee extension moment known as the plantarflexion-knee extension couple.[1, 9] Third rocker plantarflexion elongates the lower limb, is the most important determinant to reduce CoM displacement[10] and crucial for efficient step-to step transition during gait.[6] These mechanisms rely on a stable foot lever arm, sufficient range of motion, adequate muscular timing and strength, all of which may be compromised in CP.[1] Weak or overlengthened triceps surae reduce the ankle joint stability and torque, whereas equinus contracture may decrease the area of support and range for plantarflexion during push-off. Massaad et al[11] confirmed that in children with CP there was 1.3 to 1.6 times greater vertical CoM displacement than in typically developing children, indicating a support deficit which was mainly associated with an equinus gait pattern. Surgical intervention which increased ankle dorsiflexion resulted in less abnormal CoM displacement, as calculated from the vGRF.[12] However, in many cases with CP, the plantarflexors may be weakened after surgical release, with reduced ability to stabilise the ankle and generate knee extension moments.[13]

Ankle-foot orthoses (AFOs) are routinely used after lower limb surgery to provide adequate mechanical support during the rehabilitation period and prevent recurrence of deformities.[14-16] Reviewing the prerequisites of normal gait, an important purpose of AFOs in ambulating children with CP is to provide stability in stance.[1] Adequately aligned AFOs have been advocated to reduce CoM deceleration deficiency,[8] and walking with AFOs has previously been associated with increased FZ_2 in spastic CP. [17] Meanwhile, there is concern that restriction of ankle motion in AFOs may inhibit ankle push-off and CoM deceleration in late stance[18-20] Nevertheless, there is limited evidence regarding how clinical interventions such as surgery and orthoses influence the vGRF magnitude and the ability to support the CoM in children with CP.

A conventional method to study changes in GRF data has been to extract discrete scalars, such as minimum or maximum (peak) values.[4, 8, 21] The vGRF has been found to be the most consistent and reproducible kinetic outcome variable [4, 22, 23] and FZ_2 the least variable peak value,[4] which was not affected by stature or changes in gait speed in typically developing children.[3] However, for pathological gait vGRF curves may be more complex, causing difficulties in defining peak values.[21]

An alternative approach is to examine information from entire curves. Frequency domain analysis throughout stance has been shown to reduce the variability of vertical and horizontal GRF components in unimpaired[5, 23] and CP[23] gait. Transforming gait curves to functions of time has previously demonstrated benefits over more traditional methods to assess kinematic gait curves.[24] More recently, Zhang et al[25] proposed a functional mixed-effects analysis to study kinematic gait curves in children with CP walking with and without AFOs. Such approaches would enable comparisons of vGRF curves with greater reliability,[4] before and after intervention, over the entire and parts of the stance phase, and corresponding to the periods where FZ_1 and FZ_2 occur.

In the present study we aimed to evaluate the impact of surgical triceps surae lengthening to treat ankle equinus and postoperative AFOs in children with CP, using the vGRF as outcome measure. Our objective was to explore the use of functional curve analysis, studying changes in vGRF curve shape and magnitude while accounting for within-subject correlation in repeated measures, between pre- and postoperative, barefoot and AFO walking. Our hypothesis was that increasing the ankle range of motion through triceps surae lengthening and controlling motion with AFOs would cause significant changes on the vGRF indicating improved ability to support bodyweight.

2 Methods

2.1 Participants

The study was based on three repeated measures in a cohort of children with preoperative baseline data, who were initially part of two other studies[15, 16] to compare walking with AFOs versus barefoot after lower limb surgery. The study was approved by the Regional Committee for Medical Research Ethics –South East Norway (REC; 2013/1242). We included children who underwent triceps surae lengthening to treat ankle equinus, used AFOs postoperatively and were ambulatory in level I-II of the gross motor function classification system (GMFCS).[26] Fifty-six patients; 32 (16 girls and 16 boys) with unilateral, and 24 (8 girls and 16 boys) with bilateral spastic CP, were eligible for inclusion and gave written informed consent. Their main demographic information is presented in Table 1.

Equinus surgery included gastrocnemius recession (n=24) or tendo-achilles lengthening (n=32), as recommended in the children's preoperative three-dimensional gait analyses. AFOs and shoes were fitted after removal of postoperative splints, and children instructed to use the orthoses all day until the postoperative gait assessment about one year after surgery. The types of AFOs used in the current study included 33 children with hinged AFOs and 23 with solid AFOs. The hinged AFOs allowed dorsiflexion and restricted plantarflexion to assist foot clearance in swing. Solid AFOs restricted ankle motion entirely and were mainly employed in cases of weak triceps surae and crouch gait patterns.[27]

2.2 Data collection

Kinematic, kinetic and temporal-spatial data was collected using a Vicon system (Vicon Motion systems Ltd, Oxford, UK) with six infrared cameras (MXF40), Plug-in-Gait model and marker protocol.[28, 29] With AFOs, joint width measures were adjusted and markers were positioned on the devices, aligned with segment and motion axes. Ground reaction forces were collected at 1000 Hz using three strain-gauge force plates (AMTI OR6-7, Advanced Mechanical Technology Inc., Watertown, MA, USA) embedded level with the floor in a 12-meter walkway. The children walked with a self-selected speed until at least three trials with clean left and right foot strikes on separate force plates were obtained. Gait events, i.e. initial contact and foot off were determined on the force plates and the vGRF curves inspected for consistency in Vicon Nexus software. Force plate data was noise-reduced using a 4th order zero-lag low-pass Butterworth filter with a cut-off frequency of 5 Hz.

Gait data was collected under three different conditions: 1) Preoperatively walking barefoot (PreBF), 2) Postoperatively walking barefoot (PostBF), and 3) Postoperatively walking with AFOs (PostAFO). Postoperatively, participants were first measured walking barefoot and subsequently with AFOs.

2.3 Data analysis

To retain independence, data from one limb per participant was used in the statistical analyses. This implied the affected side in children with unilateral CP; the most affected, or left side when no side difference was found, in children with bilateral CP.

2.3.1 vGRF curve analysis

For functional analysis of the vGRF curve a single representative trial containing valid force plate data was selected from each participant and condition and exported to ASCII format, In MatLab (version R2018a, Mathworks, Natick, MA, USA) vGRF data was normalized to bodyweight (N/kg) and time-normalized from 0-100% of stance phase.

To analyze the outcome as a smooth function of time, normalized vGRF data was fitted using generalised additive models as described in Wood et al,[30-32] R statistical programming language and *mgcv* library with cubic spline basis set.[33] Repeated measures of subjects were accounted for by inclusion of subject-specific random smooth effects whose associated smoothing parameters were assumed to be uniform across subjects. Statistical significance of shape and intercept was assessed with F-tests, to examine how similar two curves were to one another relative to estimates of background variability. The hypotheses tested refer to similarity of the smooth curves in the interval being modelled. For the current study, the fitted vGRF curves were examined across the 0-100% time interval (T) of stance phase, denoted T(Stance); in the interval of the first peak in 15-35% of stance, denoted T(FZ_1); and the interval of the second peak in 65-85% of stance, denoted T(FZ_2).

To evaluate changes on the vGRF following triceps surae surgery we tested PostBF relative to PreBF. To evaluate changes walking with AFOs versus barefoot postoperatively we tested

PostAFO relative to PostBF. Comparisons between these condition pairs were tested with the model:

vGRF = s(time | population average) + s(time | individual effect) + s(time | condition), where s( ) denotes smooth function(s) of time for the indicated strata using a cubic spline basis, and time denotes the interval (T) being examined. This model was chosen based on superiority of model fitness using the Akaike and Bayesian information criteria. To assure identifiability of each term and generalizability of the model, we limited the flexibility of the individual and condition smooth term effects. The smoothing parameter of the model was estimated via generalized cross validation.

Besides testing effects of condition, covariates comprised topographical CP type (unilateral versus bilateral), which was tested in each condition stratum. Within the PreBF, PostBF, and PostAFO conditions, we therefore fit the model:

vGRF = s(time | population average) + s(time | individual effect) + s(time | CP type), where notation is used as previously.

2.3.2 Descriptive variables

Descriptive variables were averaged across three trials in each condition. To quantify changes in FZ_1 and FZ_2 magnitudes, we used the highest force values in 15-35% and 65-85% of stance, respectively, and normalized to bodyweight (N/kg). To describe change in dynamic ankle range of motion between conditions we calculated the kinematic maximum ankle dorsiflexion in late stance. Walking speed was normalised to non-dimensional quantities to account for changes in body height pre-to-postoperatively using the formula Non-dimensional speed($m\ s^{-1}$)= speed/√h g, where h is the body height (m) and g is the acceleration due to gravity (9.81 $m\ s^{-2}$).[34] Kolmogorov-Smirnov tests confirmed normally distributed residuals whereby changes in descriptive variables between conditions were investigated using linear mixed models[35] (SPSS 21 for Windows, IBM corp. Armonk, NY, USA). PostBF was the reference category against which PreBF and PostAFO conditions were compared, respectively. Individual deviations from the population average trend were tested using subject-specific random effects. Since some vGRF peak values have been found sensitive to walking speed,[3] we also evaluated the correlation of FZ_1 and FZ_2 with non-dimensional speed.[34]

The level of significance for all hypothesis tests was set at $P < .05$.

3 Results

3.1 vGRF curve analysis

CP type (bilateral versus unilateral) yielded highly significant differences in the vGRF curve across T(Stance); in PreBF ($P<.001$), PostBF ($P<.001$) and PostAFO ($P<.001$) conditions. We therefore performed comparisons between conditions separately in bilateral and unilateral CP groups. Results of the functional curve analysis are illustrated in Figures 2 and 3.

3.1.1 Impact of surgery; PostBF versus PreBF

In the bilateral CP group significant changes were found across the entire period of T(stance) ($P<.001$) with reduced forces in T(FZ_1) ($P<.001$) and increased forces in T(FZ_2) ($P<.001$) postoperatively (Fig. 2a). Similar significant changes were also found in the unilateral CP group across T(stance) ($P<.001$), T(FZ_1) ($P<.001$) and T(FZ_2) ($P=.013$) (Fig. 2b).

3.1.2 Impact of walking with AFOs; PostAFO versus PostBF

In both the bilateral and unilateral CP groups differences between conditions were significant across T(stance) ($P<.001$) and with curves indicating higher forces in T(FZ_1) ($P<.001$) walking with AFOs compared to barefoot postoperatively (Fig. 3a-b). In T(FZ_2) differences were also highly significant for both groups ($P<.001$). However, with AFOs increased vGRF magnitudes were more distinct in the unilateral group (Fig. 3b).

All tests of curve shape and intercept (magnitude) between conditions were significant. Significant differences were found even when curves from different conditions seemed highly similar, as seen in Figure 2b and T(FZ_2) interval. Although the difference in shape was less distinct ($P=.01$), narrow confidence interval indicated high statistical precision of the effect estimate. Generally, all confidence intervals for the T(FZ_2) were narrower than those for the T(FZ_1) suggesting higher precision and less variation in the change occurring in late compared to early stance.

3.2 Descriptive results

Results from descriptive analyses are presented in Table 2. Linear mixed model analyses and graphs illustrating the mean (1SD) vGRF across stance phase of the uni- and bilateral groups in PreBF, PostBF and PostAFO conditions may be found in Supplements.

In the bilateral CP group peak FZ_1 decreased from an average 124 to 104% bodyweight in PostBF versus PreBF ($P<.001$) (Table 2 and Supplements). The mean FZ_2 was below bodyweight with no significant difference ($P=.339$). In children with unilateral CP there was no significant difference in FZ_1 between PostBF and PreBF conditions (P=.072), whereas FZ_2 increased from 88 to 94% bodyweight postoperatively (P=.007).

Comparing PostAFO with PostBF in the bilateral group, FZ_1 increased from an average 104 to 115% bodyweight (P=.007) and FZ_2 increased from 92 to 98% bodyweight (P=.001). In the unilateral group FZ_1 increased from 113 to 125% bodyweight (P=.005) and FZ_2 from 94 to 103% bodyweight (P<.001). (Table 2 and Supplements).

In both bilateral and unilateral groups there was ankle equinus preoperatively, seen as negative maximum ankle dorsiflexion values in PreBF, and enhanced ankle range of motion in PostBF (P<.001) (Table 2). Maximum ankle dorsiflexion was reduced in PostAFO versus PostBF for the bilateral (P=.016) whereas no difference was found in the unilateral group. Walking speed decreased in PostBF versus PreBF (P=.009, bilateral; P=.088 unilateral) and increased in PostAFO versus PostBF (P=.047, bilateral; P=.013 unilateral). Across groups we found a moderate-to-strong positive correlation between non-dimensional speed and FZ_1 (PreBF r=.45, P<.001), PostBF r=.68 P<.001, PostAFO r=.67 P<.001), whereas the correlation between speed and FZ_2 was weak and insignificant.

4 Discussion

After surgery, vGRF decreased in weight acceptance and increased in late stance. Additional changes with AFOs versus barefoot postoperatively involved increased vGRF in weight acceptance and in late stance.

Preoperative graphs (Fig.2) and descriptive values indicated an enlarged ratio between excessive vGRF during weight acceptance and decreased forces below bodyweight in late stance. The pattern is consistent with deceleration deficiency and Ben Lomonding which has been described as a typical gait pattern in children with CP.[8] Although the average FZ_1 did not exceed normative ranges[3, 5] standard deviations revealed variability and a higher frequency of excessive forces in weight acceptance. However, similar variability has been found in normal gait.[3] After triceps surae lengthening we found decreased force magnitudes during weight acceptance and increased forces in late stance. Since the vGRF curve reached more normative patterns postoperatively, we may infer that enhanced ankle dorsiflexion and plantarflexion range improved the children's ability to decelerate CoM in late stance. Similar assumptions were made by Massaad et al[12] where surgical treatment of equinus gait in a limited sample of seven children with spastic CP was related to a decrease in vertical CoM displacement. Nevertheless, we found that the average late stance vGRF was less than bodyweight, suggesting remaining CoM deceleration deficiency walking barefoot postoperatively. Reasons could be overlengthened and/or weak triceps surae postoperatively, especially in the bilateral cases as indicated by their range of maximum ankle dorsiflexion.

Walking with AFOs, results from curve and peak analyses confirmed higher forces in both weight acceptance and late stance periods compared to barefoot postoperatively. An explanation for enlarged vGRF in weight acceptance may be increased walking speed with AFOs. The FZ_1 increased with increasing speed, while there was no positive correlation between speed and FZ_2. In agreement, a longitudinal study of typically developing children found that FZ_1 amplified with increased speed, whereas FZ_2 showed consistency and little variability.[3] Similarly, the magnitude of vertical weight acceptance forces, termed 'collision', and step-to step transition has been found to increase with speed in dynamic walking models.[6]

Using the definition of deceleration deficiency, a clinically important improvement would imply an increase of late stance vGRF ≥ bodyweight. Descriptive analysis confirmed increased vGRF equivalent to bodyweight, supporting our hypothesis that control of ankle motion with AFOs improves CoM deceleration and support in late stance. However, late stance forces improved most in the children with unilateral CP where maximum ankle dorsiflexion with AFOs resembled the barefoot condition. In this group hinged AFOs were predominant (n=23) which allowed a greater range of motion. Hence, dynamic AFOs may be beneficial to increase late stance vGRF, provided triceps surae strength and plantarflexion-knee extension coupling is adequate.[14] Kitaoka et al found that ankle immobilisation with solid AFOs was associated with reduced late stance vGRF. With hinged AFOs there was enhanced midfoot stabilisation, but vertical force components were not affected.[36] However, their results refer to normal adult gait and are not entirely pertinent to CP. Stabilisation of flexible feet with AFOs may have enhanced efficient force transfer in our participants, however we did not include evaluation of subtalar foot motion that may confirm this theory. Previously, lever arm dysfunction that caused impairment of the midtarsal locking mechanism has been associated with a decreased second peak of the vGRF.[37] Further studies are warranted to clarify how differences in AFO mechanical design, ankle and foot stabilisation affect vGRF magnitudes during gait in persons with neuromuscular diagnoses.

Testing the CP type as a covariate, the functional curve analysis revealed highly significant differences between bilateral and unilateral groups in each of the tested conditions. Besides dysfunction in both lower limbs and diminished gait speed, all participants with bilateral CP used AFOs on both sides which may have contributed to the differences, and supporting the decision to analyse groups separately.

It is difficult to explain why Ben Lomonding occurs in children with unilateral CP. If excessive weight acceptance forces in the leading limb results from late stance deceleration deficiency in the opposite, trailing limb[8] insufficient late stance stability is suggested in the non-affected limbs. However, ankle equinus in the affected limb with inadequate prepositioning for initial contact and reduced contact area with the ground most likely contributed to the pattern. In addition, compensatory strategies such as vaulting may have caused sub-optimal CoM support in non-affected limbs. Previously, White et al found asymmetric vGRF patterns between more and less affected limbs in children with CP, although their study did not differentiate between topographical CP types.[23] In future studies consecutive force plate recording from affected and non-affected limbs may help explain vGRF patterns in children with unilateral CP.

Examining the entire vGRF using functional curve analyses revealed significant differences between all compared conditions, and the graphs demonstrated where differences were most pronounced. Differences in intercept, i.e. magnitude, and shape of the vGRF were determined within the specified intervals. The differences found by curve analysis were at large identified by analysis of peak values and both methods handled within-subject dependencies in the data caused by repeated measurements. Still, peak value analysis did not pick up significant differences in the FZ_2 area for the bilateral and FZ_1 for the unilateral group in PostBF versus PreBF comparisons. In early works by Jacobs et al[21] functional representations of vGRF curves were found to be particularly useful to study pathological gait where patterns are less consistent than in unimpaired gait. Several investigators promoted analyses that examine oscillations of ground reaction forces throughout stance[5, 23] since larger areas of the curves provide a more representative variable, and coefficients of variations are reduced. Furthermore, functional curve analyses have advantages over alternatives such as multiple pointwise tests along the curves, which require corrections of *P* values (Bonferroni etc) to more conservative levels.[24, 25, 35, 38] Limitations exist in testing effects of relevant continuous covariates such as speed when the dependent variable is a function. [24] In comparison, the linear mixed model analysis with FZ_1 and FZ_2 as continuous dependent variables enables evaluation of both categorical and continuous covariates.[35]

In our analyses we claimed differences in one single component of the vGRF, and in pre-specified time periods that were thought to be of clinical importance. Choices regarding timing and lengths of sub-intervals were based on previous work[39] and visual inspection of the vGRF graphs, to achieve an adequate curve representation in areas where the highest magnitudes occurred. Even so, it would be advantageous to statistically test significant differences along the course of the entire curve during stance phase. Røislien et al[24] suggested the influence of covariates be expressed as estimated mean effects with 95% confidence interval. The area in the gait cycle where the confidence interval did not include zero corresponded to a *P* value below 0.05 and statistically significant effect.[24] Statistical parametric mapping is an alternative method where GRF components may be analysed as one multi-component vector changing through time and space, and covariates may be tested across a defined time domain, using *P* values to explain significant differences.[38] Both approaches could be adequate in future studies.

Limitations included heterogeneity in the cohort, especially with regards to surgery where 22 children underwent isolated triceps surae lengthening and 34 received concurrent surgical procedures as part of single-event multilevel surgery. Increasing the sample size and controlling for type of surgery, including triceps surae surgery (ex. gastrocnemius recession versus tendo-achilles lengthening), would be relevant. The unbalanced number of hinged versus solid AFOs was a limitation that made it difficult to test and quantify the impact of AFO type in the subgroups. Use of a shoes-only control condition could differentiate possible effects of shoes from AFOs. Other variables to explain CoM support and stability in stance such as ankle and knee moments and power, point of GRF application and pressure distribution on the foot were not within our focus but may be relevant for further research.

5 Conclusions

Our results indicate that the vGRF is responsive to evaluate treatment of gait problems with surgery and AFOs in children with CP. Decreased forces in weight acceptance and increased forces in late stance imply that fewer children walked with Ben Lomonding postoperatively. Walking with AFOs versus barefoot postoperatively, vGRF magnitude in late stance increased equivalent to bodyweight, which could be considered a clinically important improvement indicating reduced deceleration deficiency and more adequate CoM support.

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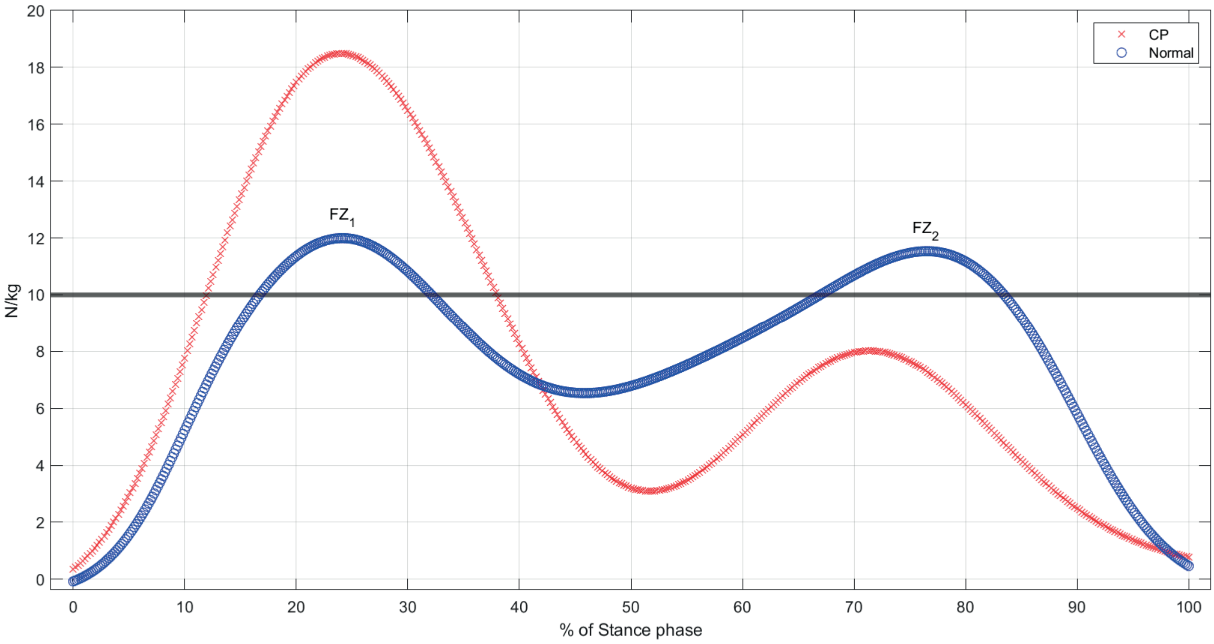
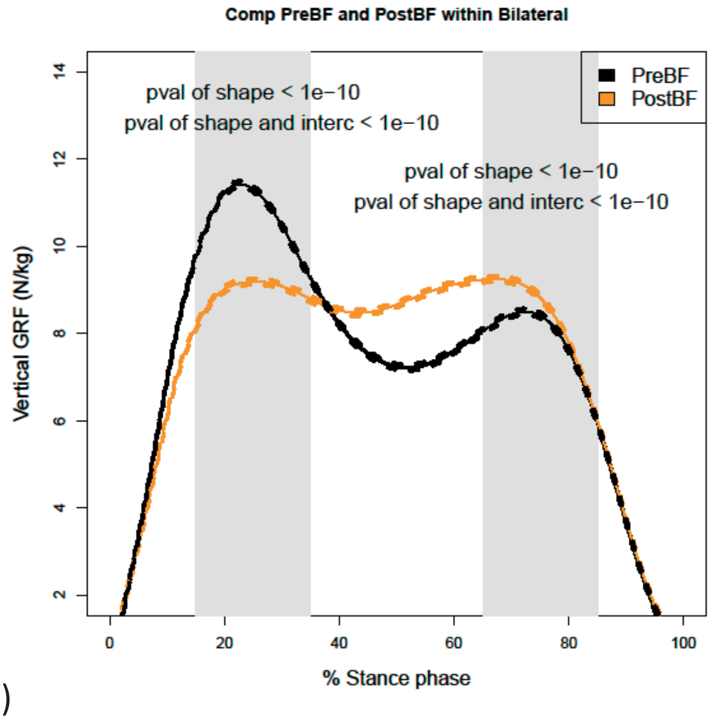
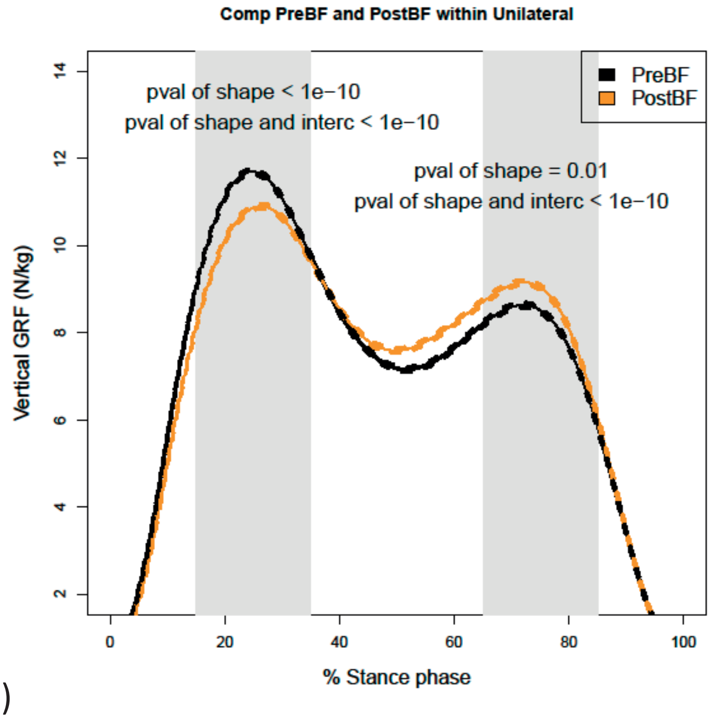


Figure 1. Example of differences in vertical ground reaction force patterns in a child with unimpaired gait (blue) and a child with bilateral spastic cerebral palsy (red). Forces are normalised to bodyweight (N/kg) and time-normalized from 0-100% of stance phase**.** The horizontal line at 10 N/kg illustrates 100% bodyweight.



a)



b)

Figure 2. Functional curve analysis comparing Preoperative barefoot (PreBF; black) and Postoperative barefoot (PostBF; orange) conditions in 0-100% stance for a) bilateral group, and b) unilateral group. The difference in the curves shown is an average (solid) condition effect with confidence interval (dashed) over the population being modeled and controlling for individual variation. Grey bars indicate P values for tests of curve shape and intercept (magnitude) in subintervals 15-35% stance; T(FZ_1), and 65-85% stance; T(FZ_2).

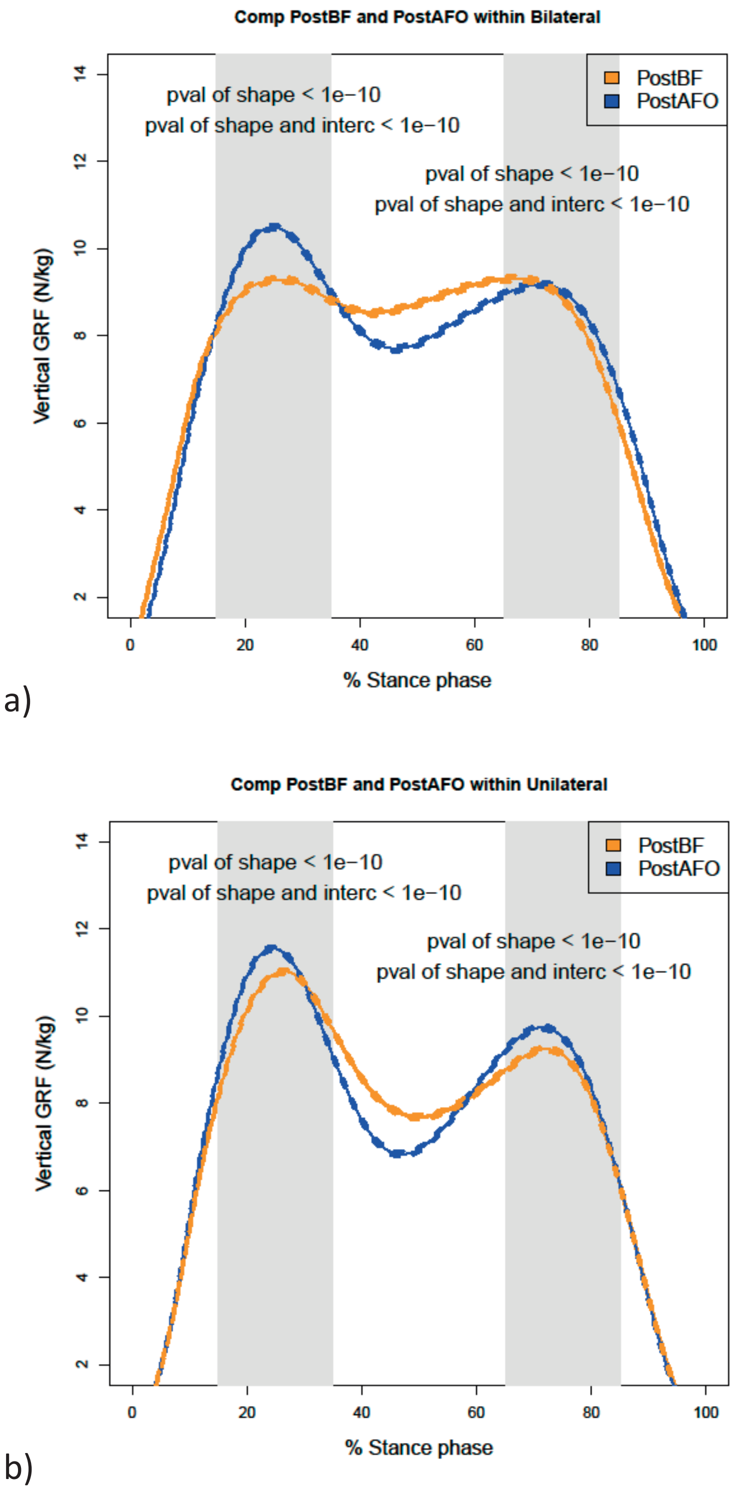


Figure 3. Functional curve analysis comparing Postoperative AFO (PostAFO; blue) and Postoperative barefoot (PostBF; orange) conditions in 0-100% stance for a) bilateral group, and b) unilateral group. The difference in the curves shown is an average (solid) condition effect with confidence interval (dashed) over the population being modeled and controlling for individual variation. Grey bars indicate P values for tests of curve shape and intercept (magnitude) in subintervals 15-35% stance; T(FZ_1), and 65-85% stance; T(FZ_2).

Table 2. Results from analyses of descriptive variables

	Bilateral n=24			**Comparisons *p* value**	
	PreBF	PostBF	PostAFO	PostBF vs PreBF	PostAFO vs PostBF
FZ_1 (N/kg)	12.38 (2.5)	10.42 (1.3)	11.49 (1.7)	**<0.001**	**0.007**
FZ_2 (N/kg)	8.99 (.8)	9.16 (.7)	9.79 (.6)	0.339	**0.001**
Max ankle DF stance phase (°)	-5.54 (11.6)	13.5 (6.3)	7.8 (4.6)	**<0.001**	**0.016**
ND speed (speed/√Hxg)	0.28 (.07)	0.25 (.05)	0.28 (0.6)	**0.009**	**0.047**
	Unilateral n=32				
	PreBF	PostBF	PostAFO	PostBF vs PreBF	PostAFO vs PostBF
FZ_1 (N/kg)	11.82 (1.6)	11.29 (1.6)	12.46 (1.9)	0.072	**0.001**
FZ_2 (N/kg)	8.87 (1.1)	9.45 (.8)	10.31 (.8)	**0.007**	**<0.001**
Max ankle DF stance phase (°)	-4.2 (10.5)	9.8 (8.5)	9.9 (6.9)	**<0.001**	0.956
ND speed (speed/√Hxg)	0.33 (.05)	0.31 (.05)	0.34 (.05)	0.088	**0.013**

Values are presented as mean ± standard deviation (SD). P values are from linear mixed model analyses. Bold letters indicate significant differences with $P < 0.05$
PreBF, preoperatively walking barefoot; PostBF, postoperatively walking barefoot; PostAFO, postoperatively walking with ankle-foot orthoses; DF, dorsiflexion; FZ_1 denotes 1st peak within 15-35% of stance; FZ_2 denotes 2nd peak within 65-85% of stance; DF, dorsiflexion; ND, non-dimensional, N, Newton; H, body height; g, gravity (9.81m/s^2)

Table 1. Demographics of participants with bilateral and unilateral cerebral palsy

	n(F)	Age, Pre (years)	Age Post (years)	Mass Pre (kg)	Mass Post (kg)	Height Pre (cm)	Height Post (cm)	HAFO /SAFO
Bilateral	24 (8)	10.3 (2.6)	12.3 (2.7)	34.1 (9.7)	42.5 (10.4)	138.8 (16.4)	149.6 (13.5)	10/14
Unilateral	32 (16)	8.5 (3.1)	10.5 (3.2)	31.5 (15.9)	38.5 (17.6)	132.6 (20.4)	143.1 (17.5)	23/9

Values are presented as mean ± standard deviation (SD)
N, number of participants; F, female; Pre, preoperative gait analysis; Post, postoperative gait analysis; HAFO, hinged ankle-foot orthoses; SAFO, solid ankle-foot orthoses

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