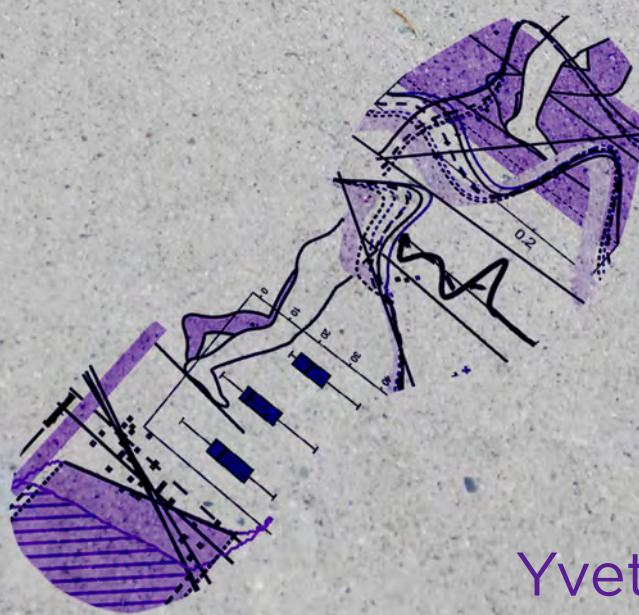




# Maximizing the efficacy of ankle foot orthoses in children with cerebral palsy



Yvette L. Kerkum



# Maximizing the efficacy of ankle foot orthoses in children with cerebral palsy

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als de weg ernaar toe  
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# **Maximizing the efficacy of ankle foot orthoses in children with cerebral palsy**

ACADEMISCH PROEFSCHRIFT

ter verkrijging van de graad Doctor aan  
de Vrije Universiteit Amsterdam,  
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door

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geboren te Lelystad

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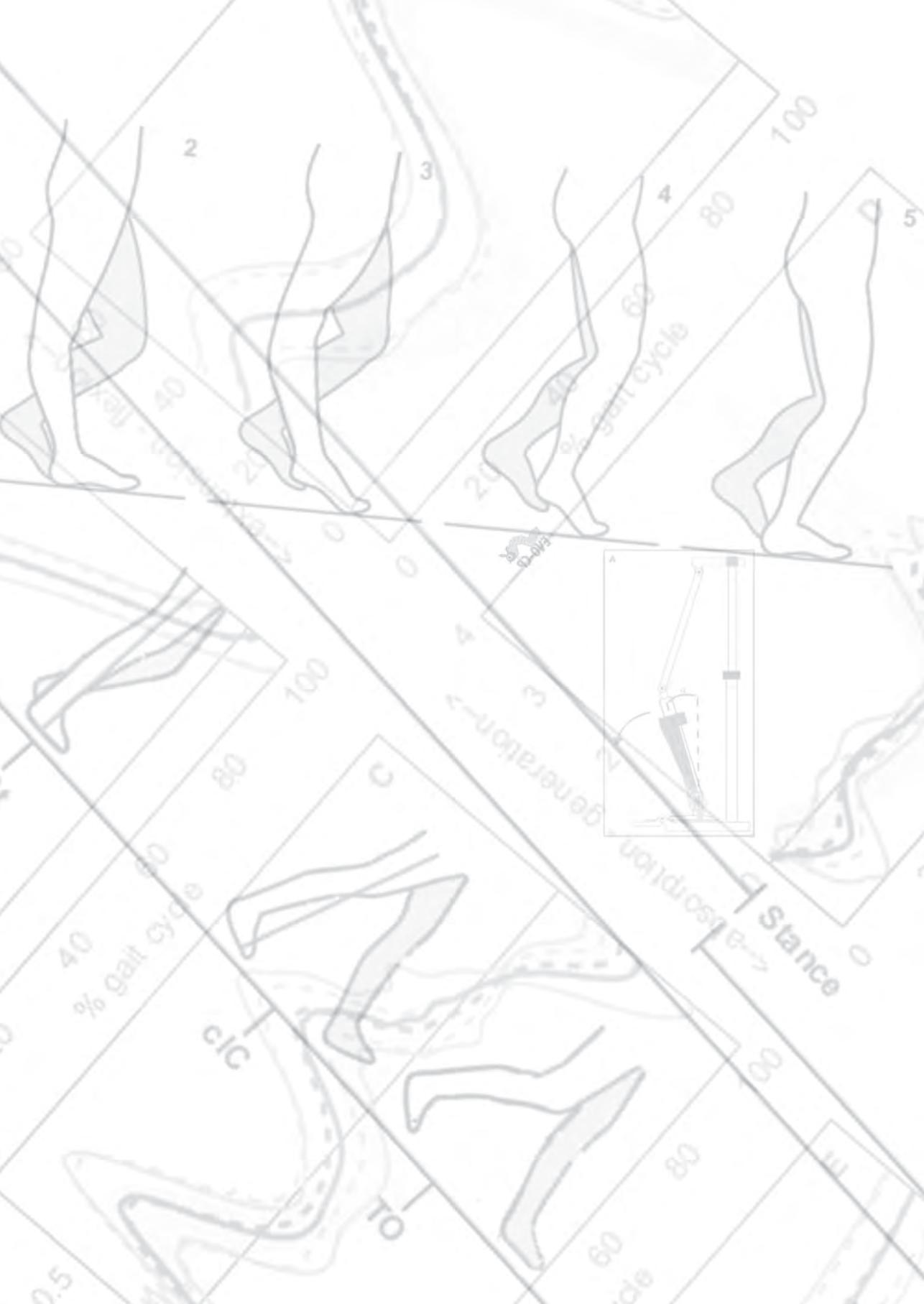
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# CONTENT

<b>CHAPTER I</b>	11
General Introduction	
<b>CHAPTER II</b>	29
Optimising ankle foot orthoses for children with cerebral palsy walking with excessive knee flexion to improve their mobility and participation; protocol of the AFO-CP study	
<b>CHAPTER III</b>	49
The shank-to-vertical angle as a parameter to evaluate tuning of ankle foot orthoses	
<b>CHAPTER IV</b>	67
Defining the mechanical properties of a spring-hinged ankle foot orthosis to assess its potential use in children with spastic cerebral palsy	
<b>CHAPTER V</b>	79
The effects of varying ankle foot orthosis stiffness on gait biomechanics and walking energy cost in children with spastic cerebral palsy who walk with excessive knee flexion	
<b>CHAPTER VI</b>	109
An individual approach to optimizing ankle foot orthosis stiffness to improve mobility in children with spastic cerebral palsy walking with excessive knee flexion	
<b>CHAPTER VII</b>	129
Acclimatization of the gait pattern to wearing an ankle foot orthosis in children with spastic cerebral palsy	
<b>CHAPTER VIII</b>	147
General Discussion	
Summary	171
Samenvatting	177
About the author	183
Dankwoord	189



# General introduction

Walking is one of the most important activities in daily life, as it enables participation in daily life activities. Although walking seems an easy task for most people, it is a delicate combination of movements of the different parts of the body. When a central neurological disorder such as cerebral palsy (CP) affects motor control, and consequently walking ability, the complexity of walking becomes apparent. In pediatric rehabilitation medicine, an important treatment goal is to acquire, maintain or improve walking ability of children with CP. To this end, one of the most common interventions in these patients is the use of an ankle foot orthosis (AFO). Evidence for the efficacy of these orthoses to improve gait in children with CP is however considered ambiguous, as both positive and negative effects have been reported. This thesis will focus on how efficacy of AFOs in CP might be improved.

## CEREBRAL PALSY

With a prevalence of 2-3 per 1000 live births, CP is the most common cause of children's disability in Western Europe<sup>[1-3]</sup>. It is described as "a group of permanent disorders of the development of movement and posture, causing activity limitations, that are attributed to a non-progressive disturbance that occurred in the developing fetal or infant brain"<sup>[1,4]</sup>. Several etiological factors could underlie CP and are typical for a particular time of onset: prenatal (e.g. intoxication), perinatal (e.g. infarction), and postnatal (e.g. infection)<sup>[3,5]</sup>. There are several risk factors for CP, such as prematurity, infection in the mother and/or child, and intrapartum asphyxia<sup>[6]</sup>.

The term CP covers a broad variety of clinical presentations, which can be categorized into groups or classes. First, CP can be described in terms of motor disorders, with spastic, ataxic, and dyskinetic sub-types<sup>[5]</sup>. A mix of types may also occur, in which the dominant type defines the motor disorder classification<sup>[4,7]</sup>. The type that will be discussed in this thesis is the spastic CP, which is the most common type as it accounts for approximately 80 percent of the patients with CP<sup>[3]</sup>. Spastic CP is defined as a posture- and movement-dependent muscle tone regulation impairment<sup>[5]</sup>, which can be divided into impaired muscle control and impaired biomechanical muscle properties<sup>[5,8]</sup>. Impaired muscle control includes both deficit symptoms (e.g. muscle weakness and loss of selective motor control), and excess symptoms, such as spasticity<sup>[9]</sup> and muscle co-contractions<sup>[5]</sup>. Impaired biomechanical muscle properties include increased muscle stiffness and abnormal muscle length<sup>[10,11]</sup>. Consequently, children with spastic CP have a risk to develop secondary impairments, such as joint contractures and bony deformities<sup>[12]</sup>. As a second categorization, spastic CP can be classified based on disease distribution<sup>[5]</sup>.

Children with unilateral CP are affected on one side, while bilateral CP involves both sides of the body<sup>[5,13]</sup>. Third, the level of impaired overall motor functioning in children with CP can be categorized with the Gross Motor Function Classification System<sup>[14]</sup> (GMFCS). The GMFCS describes five levels of motor functioning, where lower levels indicate better motor function. Levels IV and V describe the group of children who are not able to walk independently. This thesis is focused on children with GMFCS levels I-III, i.e. referring to children who are able to walk independently with or without restrictions (level I and II respectively), or with a walking device (level III). Although the majority of children with CP (approximately 70%) are able to walk with or without assistive devices, motor impairments often lead to walking limitations<sup>[5]</sup>. In pediatric rehabilitation medicine, an important treatment goal is to acquire, maintain or improve walking ability of children with CP.

## GAIT

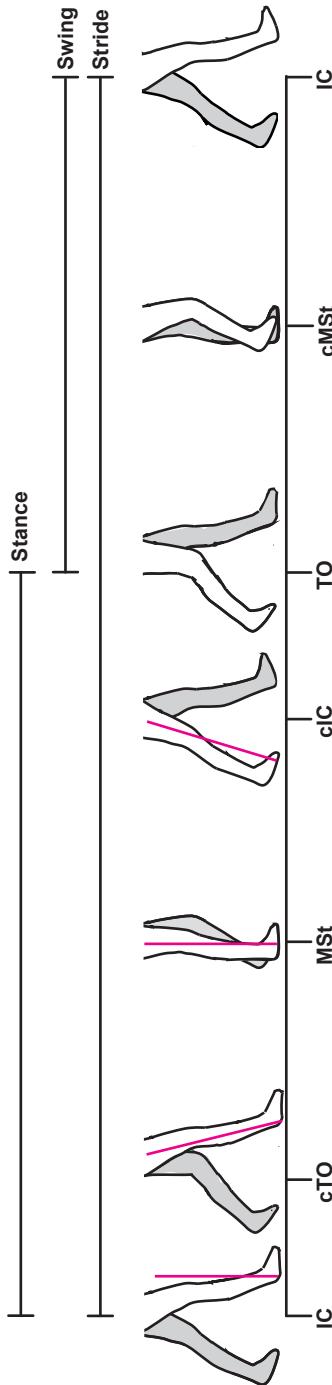
### The gait cycle

Gait is described by different phases of one leg (i.e. the leading leg) within a gait cycle. One gait cycle, or stride, starts with initial contact, which refers to the first contact of the foot with the ground and ends with the same leg hitting the ground again. Each stride is divided into a stance phase and a swing phase, where the stance phase accounts for approximately 60% of the gait cycle. These two main phases can be sub-divided into separate events, such as midstance (see Figure 1.1). Push-off describes the leg's transition from the stance phase into the swing phase.

### Normal gait

Each phase of the gait cycle is characterized by a specific position and orientation of the body's segments and joints. In relation to these orientations, the ground reaction force, i.e. the force exerted by the ground on the body, acts on the joints. A close alignment of the ground reaction force to the joint rotation centers results in low net moments, and accordingly, in low muscle forces to maintain posture and balance. The course of the ground reaction force and its alignment to the joint rotation centers during normal gait are shown in Figure 1.1.





**Figure 1.1.** The gait cycle, divided into the stance phase (i.e. initial contact to toe-off) and the swing phase (toe-off to initial contact). The phase between initial contact and contralateral toe-off is referred to as the loading response. Following loading response, the leading leg progresses towards midstance, which is defined as the moment of the contralateral leg passing the stance leg. The contralateral leg swings in front of the leading leg, until contralateral initial contact occurs. Within a small period of double support, the weight is shifted from the trailing leg to the contralateral leg, also referred to as preswing or push off. This is followed by the moment of toe-off, which concludes the leg's transition from the stance phase into the swing phase. In swing, the leg passes the contralateral leg at contralateral midstance to continue to initial contact to start a new gait cycle. The magenta line indicates the course of the ground reaction force, and its alignment with respect to the joints during normal walking. The center of pressure starts at the heel and slowly progresses towards the hallux as the stance phase continues.

Abbreviations: IC, initial contact; cTO, contralateral toe-off; MSt, midstance; cIC, contralateral initial contact; TO, toe-off; cMSt, contralateral midstance.

Adequate functioning of the ankle and foot is essential for normal walking, and can be described in terms of rockers<sup>[15]</sup>. The first rocker, also called heel rocker, represents the time span from the first heel contact (i.e. initial contact) to full contact of the foot with the surface. This involves lowering of the foot to the surface, which is controlled by eccentric contraction of the ankle dorsiflexor muscles. During the second rocker (ankle rocker), the tibia progresses over the foot, which remains flat on the ground. This is due to an eccentric contraction of the ankle plantar flexor muscles and allows continued forward movement of the body. The third rocker (forefoot rocker) represents the phase from contralateral initial contact to toe-off. At this stage, a large burst of power is generated about the ankle, which is due to a fast shortening (concentric contraction) of the plantar flexor muscles resulting in plantar flexion movement.

In addition to sufficient rocker functions, knee joint function in the sagittal plane is also considered an important feature during gait. The knee acts like a shock absorber as it flexes during early stance, while maximal extension is reached just before contralateral initial contact occurs. Using the force of the plantar flexion during the third rocker (push-off), and psoas muscle activation, the knee and hip will flex to reach sufficient clearance of the foot during the swing phase.

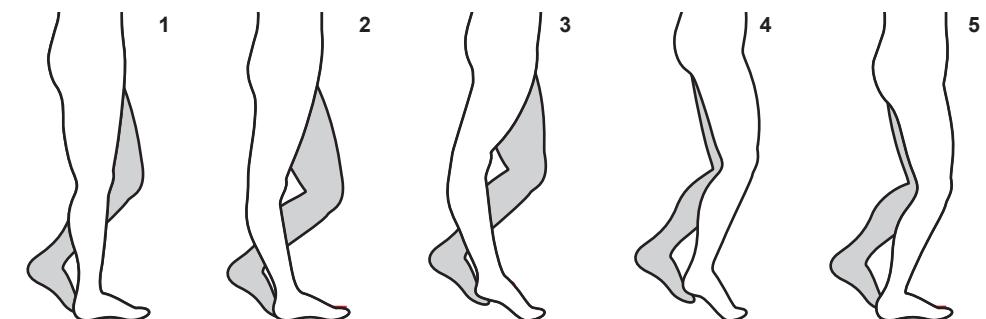
Normal gait is a complex yet highly energy efficient process<sup>[16]</sup>. Several biomechanical adjustments are used by the body to minimize the energy consumption [ $J \cdot kg \cdot min^{-1}$ ] during walking, such as a minimal excursion of the center of mass, and transfer of energy between segments by bi-articular muscles<sup>[16]</sup>. The energy consumed in relation to the covered distance is also dependent on a person's walking speed, expressed by the energy cost of walking [ $J \cdot kg \cdot m^{-1}$ ] (also referred to as gait efficiency) and calculated by dividing the energy consumption by walking speed. Healthy individuals select a comfortable walking speed at which gait efficiency is maximal<sup>[17]</sup>. In healthy children, gait efficiency improves with age until adulthood<sup>[17,18]</sup>.



### Gait in cerebral palsy

Due to symptoms of impaired motor control, muscle weakness, abnormal joint position, decreased joint range of motion, and a decreased muscle length, gait is often hampered in CP<sup>[16]</sup>. The clinical representation of gait in CP is very heterogeneous, and therefore several efforts have been made to categorize these into gait patterns<sup>[19]</sup>. In the Netherlands, the classification of Becher<sup>[5,20]</sup> is generally used for gait in spastic CP. This classification describes five gait types, mainly based on the deviations of the knee and ankle joint angles at midstance (see Figure 1.2). Gait type 1 describes a gait pattern characterized by a normal stance phase, but insufficient foot lift during the swing phase. Gait types 2 and 3 show hyperextension of the knee, respectively with or without full foot contact. Gait types 4 and 5 describe a pattern with excessive knee flexion, either with ankle plantar flexion (type 4) or excessive dorsiflexion (type 5). This thesis only discusses children who walk in gait types 4 or 5.

The ankle and foot rocker functions are impaired in most CP walking types. Weakness of the dorsiflexor muscles, for example, commonly causes mid- or forefoot contact during the first rocker. Consequently, the tibia is positioned excessively inclined already in the beginning of stance, allowing no forward tibia progression during the second rocker, and an excessively flexed knee joint at this stage. This effect is frequently enhanced by weakness of the plantar flexor muscles. The abnormal second rocker function leads to the posterior alignment of the ground reaction force with respect to the knee joint rotation center, and accordingly, in an increased knee moment. The push-off power is also often impaired as a result of abnormal ankle joint moments in late stance. The rapid plantar flexion movement that is needed for an effective push-off is impeded by the persistent



**Figure 1.2.** The gait types according to the classification of Becher. This thesis focusses on the gait that are characterized by excessive knee flexion, either without foot contact (type 4) or with full foot contact (type 5) at midstance.

knee flexion and posterior alignment of the ground reaction force with respect to the knee joint rotation center in late stance. Abnormal timing of plantar flexion movement (i.e. early or late heel rise)<sup>[16]</sup>, caused by short calf muscles, plantar flexor weakness or impaired muscle activation<sup>[21]</sup>, can further deteriorate the third rocker function. A reduced push-off prevents a rapid movement of knee and hip into flexion, <sup>[16]</sup>resulting in inadequate clearance, and insufficient knee extension at the end of the swing phase. The reduced knee extension during this phase leads to mid- or forefoot contact at initial contact, consequently reducing step length. As muscle shortening in spastic CP occurs according to the movement pattern, it seems apparent that gait deviations are related to an impaired muscle length<sup>[20]</sup>. In turn, this may reduce the (passive) range of motion of the adjacent joints, i.e limited muscle stretch, which has been shown to be an important cause in the development of muscle contractures<sup>[12]</sup>. Muscle contractures negatively affect the gait deviations<sup>[16]</sup>, leading to a vicious circle in which gait further deteriorates over time.

The underlying impairments often limit walking ability in CP, as gait deviations are associated with increased energy consumption. This especially applies to children walking with excessive knee flexion during stance, as these children are liable to show deterioration of walking in (pre-)puberty<sup>[22,23]</sup>, and their gait patterns are particularly energy consuming<sup>[17,24]</sup>. In fact, it has been shown that walking energy consumption in CP can be two to three times higher compared to typically developing children<sup>[17,24-26]</sup>. To minimize the increase in energy consumption, patients often lower their walking speed to maintain walking over longer distances<sup>[25,26]</sup>. As such, an increased walking energy cost is commonly observed, which reflects poor gait efficiency<sup>[25,26]</sup>. Although the nature of the association between underlying impairments, gait deviations and the increased energy consumption during walking in CP is not yet unraveled, abnormal knee and ankle kinematics and kinetics during gait are considered key features. First, the increased internal knee extension moment require high muscle forces to maintain posture<sup>[27]</sup>, which could be related to a higher energy consumption during walking<sup>[17,28]</sup>. Second, reduced ankle range of motion and ankle push-off power generation have been shown associated with a lower walking speed, which subsequently increases walking energy cost<sup>[29-31]</sup>. To compensate for the reduced ankle push-off, power is often generated in the hip joint around toe off <sup>[16,32,33]</sup>. This has been shown to be mechanically less efficient<sup>[29,34]</sup>, and is thus likely to further increase the walking energy cost.

## IMPROVING GAIT IN CEREBRAL PALSY

### Treatment with AFOs

An ankle foot orthosis (AFO) is a commonly applied rehabilitation intervention in children with CP to maintain muscle length (i.e. passive range of motion), as well as to maintain or improve stability, standing, and/or walking ability<sup>[35]</sup>. An AFO is a medical device that imposes a mechanical constraint to the ankle and foot, aiming to compensate for a loss of function (i.e. resisting forces that act upon the body), or to counteract an excess of function (i.e. resisting forces from within the body). As such, an AFO can directly affect movements of the ankle and foot, and, dependent on its design, it can also stabilize the knee and hip joints<sup>[36]</sup>. AFOs are available in various different designs, e.g. hinged or non-hinged, and with a ventral shell or dorsal shell. In addition, AFOs can be made of different materials like carbon fiber or polypropylene. Consequently, each AFO holds specific mechanical properties, such as stiffness around the ankle. When prescribing an AFO, its design and mechanical properties should counteract the patient's underlying impairments, and be designed such that it counteracts the specific gait deviations as much as possible, aiming to effectively improve gait.

### AFO efficacy in cerebral palsy

The efficacy of AFOs on gait can be described in terms of its mechanical effects (i.e. gait biomechanics), and/or in terms of the patient's gain in walking ability (e.g. gait efficiency)<sup>[37]</sup>. In general, the effects of AFOs on gait have been widely investigated, with studies mainly evaluating the efficacy of AFOs on gait biomechanics. These studies generally report improvements in terms of spatio-temporal parameters<sup>[38-45]</sup> and joint kinematics<sup>[38,40-44,46]</sup> and kinetics<sup>[40,41,43,46]</sup>. Although improvement of gait biomechanics has been shown to be closely coupled to improvement of gait efficiency<sup>[17]</sup>, also in the context of orthotic interventions<sup>[40,46,47]</sup>, evidence for the efficacy of AFOs on gait efficiency remains inconclusive<sup>[48]</sup>.

These ambiguous results within AFO research have been acknowledged in some reviews<sup>[48-50]</sup>, where various gaps in current literature have been addressed. First, the AFO's efficacy is expected to be largely dependent on the match between the AFO's mechanical properties and patient's specific underlying impairments. Sufficient description of participants, especially in terms of motor impairments and gait patterns is however mostly lacking. Also the AFOs used in studies are described with global reference to design and materials, and rarely described in terms of mechanical properties<sup>[48-50]</sup>.

The absence of such information hampers to unravel the nature of the optimal match between the patient and the AFO, and to define the causes of the AFO's (in)efficacy. Moreover, a variety of AFOs in relation to the underlying impairments and gait deviations of participants is introduced by differences in the prescription process, which is currently largely dependent on clinical experience<sup>[48,49]</sup> as clear prescription guidelines are scarce<sup>[36,51]</sup>. The insufficient information and variety between studies prevents a fair comparison of results, and reduces the potential to perform meta-analyses to provide more substantial evidence to improve prescription guidelines<sup>[50]</sup>. Evidence for AFO efficacy in CP could be improved by good quality research using strong designs that can control for confounding factors<sup>[48,50]</sup>, providing unambiguous characteristics of the participants' walking biomechanics and the mechanical properties of the applied AFOs.

Another problem within AFO research concerns the lack of consistency in outcomes to report AFO efficacy. To evaluate of the effects AFOs on gait, Brehm et al.<sup>[52]</sup> suggested a core set of outcomes, covering measures on all domains of the International Classification of Functioning, Disability and Health (IFC) framework<sup>[53]</sup>. This is the common framework for the assessment of functioning and treatment planning of patients in rehabilitation medicine<sup>[53]</sup>. The ICF framework uses the domains of 'body functions and structures' 'activities' and 'participation' to describe the impact of a disease or disability on human functioning, which can also be affected by 'personal factors', and 'environmental factors'. The domain 'body functions and structures' describes a person's functioning on the level of the physiological functions of body systems and the body's anatomical structures. The 'activity' domain describes human functioning in terms of daily-life activities, and can be sub-divided into the capacity and performance qualifiers. The first refers to what a person can do in, for example, a laboratory setting, while the latter describes what a person actually does in day-to-day life. The 'participation' domain refers to a person's participation in daily life situations, such as sports, and social events.

From the patient's perspective, it is most relevant to assess the effect of an AFO on outcome measures that express the gain for the patient; for example, a measure that quantifies walking capacity such as walking speed or walking energy cost. Additionally, an assessment of daily walking activity (i.e. walking performance) may give insight in the patient's functioning in daily life. The effects of an AFO on gait biomechanics, assessed with a 3D gait analysis, can also be evaluated, representing the biomechanical functioning of an AFO (i.e. at level of body functions and structures)<sup>[37]</sup>. It has been suggested that outcome measures in studies on AFO efficacy should cover both the activity level and the level of body functions and structures<sup>[37]</sup>. This could reveal mutual relations between

outcome measures, giving insight the underlying working mechanism of AFOs, which may contribute to proving the efficacy of a treatment algorithm, i.e. how to prescribe a well-matched AFO<sup>[37]</sup>.

## **Optimizing AFO treatment**

Several factors that could improve AFO treatment in order to prescribe a well-matched AFO have been addressed in the literature, some of which will be discussed in this thesis.

### ***Adjusting the AFO alignment***

Firstly, literature suggests that an appropriate orientation of the shank (i.e. close to normal) during midstance is a main determinant for efficient walking. In normal walking, the shank is 10 to 12 degrees inclined with respect to the vertical at midstance, therewith positioning the knee joint in the middle of the base of support. It is assumed that this facilitates balance and appropriate ground reaction force alignment with respect to the knee and hip joints, and therefore contributes to conservation of energy<sup>[54]</sup>. Healthy individuals have the ability to adapt the orientation of the body segments to achieve adequate alignment independent from the footwear they are wearing as long as it allows ankle range of motion.

When wearing an AFO, for most AFOs the ankle is fixed into a pre-defined angle determined by the AFO's neutral angle (i.e. the angle of the AFO when no force is applied). As ankle range of motion is restricted, the orientation of the shank at midstance is now defined by the combination of the AFO's neutral angle, and the shoe's heel-sole differential, i.e. the difference in height between the heel and the forefoot. Adjusting the heel-sole differential of the AFO-footwear combination is therefore expected to influence the orientation of the shank during walking. Subsequently, adjusting the AFO alignment could impact on the efficacy of the AFO, which is considered to be affected by the (mal-)alignment of the ground reaction force to the lower limb joint rotation centers<sup>[36,55,56]</sup>. Although recently more interest has been shown in the AFO alignment in CP, evidence for the effects are lacking, and evaluating the alignment is currently not completely incorporated into clinical practice<sup>[55]</sup>.

To successfully implement a proper evaluation of the AFO's alignment into the prescription process in CP, a parameter to quantify the AFO alignment seems required. The Shank-to-Vertical-Angle (SVA) has been proposed as a relatively simple control

parameter to evaluate the effects of adaptations to the heel-sole differential<sup>[54]</sup>. Although effects of such adjustments, as quantified by the SVA, have already been reported in some studies<sup>[55]</sup>, the response of the SVA to manipulations to the AFO-footwear combination and its relation to joint kinematics and kinetics have not been investigated so far.

### **AFO stiffness**

Secondly, children walking with excessive knee flexion in stance are typically prescribed with a rigid ventral shell AFO that is manufactured with a rigid footplate<sup>[57]</sup>. This type of AFO aims to reduce knee flexion by shifting the ground reaction force anterior to the knee joint rotation center, to create an external knee extensor moment in stance, which is done through a force by muscular contraction of the ankle plantar flexors in normal gait. Literature shows that rigid AFOs can effectively reduce knee flexion during stance<sup>[43]</sup>, which may contribute to walking energy cost improvement<sup>[46]</sup>. However, the rigid properties also impede walking by impairing the rocker functions. This especially accounts for the third rocker, as the rigid AFO obstructs plantar flexion, therewith reducing push-off power<sup>[42,58-61]</sup>. As ankle range of motion and push-off power are considered key features for efficient gait<sup>[29,31]</sup>, this could negatively impact on the walking energy cost.

The AFO's impeding effect on third rocker function could be reduced by using spring-like AFOs. Research has shown that spring-like AFOs (e.g. carbon fiber AFOs) can improve the gait pattern, while less constraining voluntary push-off<sup>[62,63]</sup>, which might be beneficial in terms of the walking energy cost<sup>[29,31]</sup>. Model studies<sup>[64]</sup>, as well as studies in healthy adults<sup>[65]</sup> and adult patient populations<sup>[66,67]</sup>, already showed that both joint kinematics and kinetics, as well as the walking efficiency can be influenced by applying spring-like AFOs with different degrees of stiffness. As a result, gait efficiency could be maximized by choosing the appropriate AFO stiffness for each individual patient<sup>[66]</sup>. This individual approach of selecting an appropriate AFO stiffness may also apply to children with CP who walk with excessive knee flexion. It is thought that an optimal AFO stiffness, which is the stiffness that results in maximized gait efficiency, could be found. This optimum is expected to reflect the trade-off between a sufficient reduction of the knee flexion and minimal obstruction of third rocker function, although this hypothesis has not yet been investigated.

### **Acclimatizing to a new AFO**

Thirdly, AFO efficacy is often assessed by its effect on gait biomechanics, as the mechanical constraint of the AFO is primary expected to alter the gait pattern in terms of joint kinematics and kinetics, and eventually the overall gait performance. When applying an AFO, it could be expected that children with CP may need time to improve their gait pattern to the AFO by adjusting their muscle activation pattern (i.e. motor learning) to the new ankle mechanics<sup>[68]</sup>. When evaluating effects of an AFO on gait, acclimatization time for the gait pattern to adapt to the mechanical constraints as induced by a new AFO is therefore generally recommended to comply with the learning effect<sup>[50]</sup>. Accordingly, most testing protocols in research studies allow for acclimatization, although the permitted time varies between six weeks and less than one day<sup>[50]</sup>. While inadequate acclimatization time has been reported as a potential confounding factor for the efficacy of AFOs in some studies<sup>[39,69]</sup>, it is unknown whether such an acclimatization period is actually required to reliably assess gait biomechanics of a newly prescribed AFO.

In summary, AFOs are widely used to improve gait in children with CP. Scientific evidence for the effectiveness of AFOs is however scarce and inconclusive. Insight in underlying working mechanisms of AFOs is also lacking. Considering the variation in reported effects of AFOs on gait in CP, an individual approach to optimize AFO prescription seems essential to maximize treatment efficacy.

## **AIM**

This thesis aims to evaluate factors that can guide an individual optimization of AFO prescription in order to maximize AFO efficacy in children with CP who walk with excessive knee flexion in stance.

## **OUTLINE**

The chapters of this thesis are primarily based on results of the AFO-CP trial, which was initiated to get insight in underlying working mechanisms of AFOs and to provide evidence for AFO effectiveness in children with CP walking with excessive knee flexion in stance. The study specifically aimed to individually optimize AFO stiffness to maximize the gait efficiency in these children. The protocol of the AFO-CP study is described in **Chapter II**.

In **Chapter III**, an instrumented treadmill was used to investigate the Shank-to-Vertical Angle as a parameter to evaluate the tuning process within AFO prescription. This study was performed in healthy adults and describes the effects of adjusting heel height and footplate stiffness of an AFO-footwear combination (i.e. AFO and shoes) on the Shank-to-Vertical Angle and lower limb joint angles and moments.

In **Chapter IV**, the BRUCE instrument was used to measure the mechanical properties of an adjustable spring-hinged AFO to assess its potential use in children with spastic CP. The spring-hinged AFO was set into different stiffness configurations, of which the mechanical characteristics were measured and discussed in relation to AFO function in children with CP walking with excessive knee flexion.

The effects of a rigid AFO and two spring-like AFO configurations of the spring-hinged AFO on gait were investigated in **Chapter V**. The effects of the AFOs were compared to walking with shoes-only. In addition to relevant biomechanical parameters and walking energy cost, also AFO contributions to ankle work were calculated.

In **Chapter VI**, the results of the stiffness variations on gait were used to select the optimal AFO stiffness for each patient. This selection was based on the maximal knee extension in stance and walking energy cost while walking with the different AFOs. Children wore their optimal AFO for three months, after which effect of the stiffness-based optimization on daily life activity was assessed.

**Chapter VII** investigates the effects of an acclimatization period to wearing a newly prescribed AFO on gait biomechanics in children with CP. Although an acclimatization time for the gait pattern to adapt to the new AFO is generally applied, the actual need for such acclimatization to reliably assess the effects of an AFO on gait biomechanics is not known. This chapter evaluates the effects of an AFO directly after tuning, and four weeks later in a subset of relevant biomechanical gait parameters.

In **Chapter VIII** the overall aim of this thesis is discussed and directions for future research are given.



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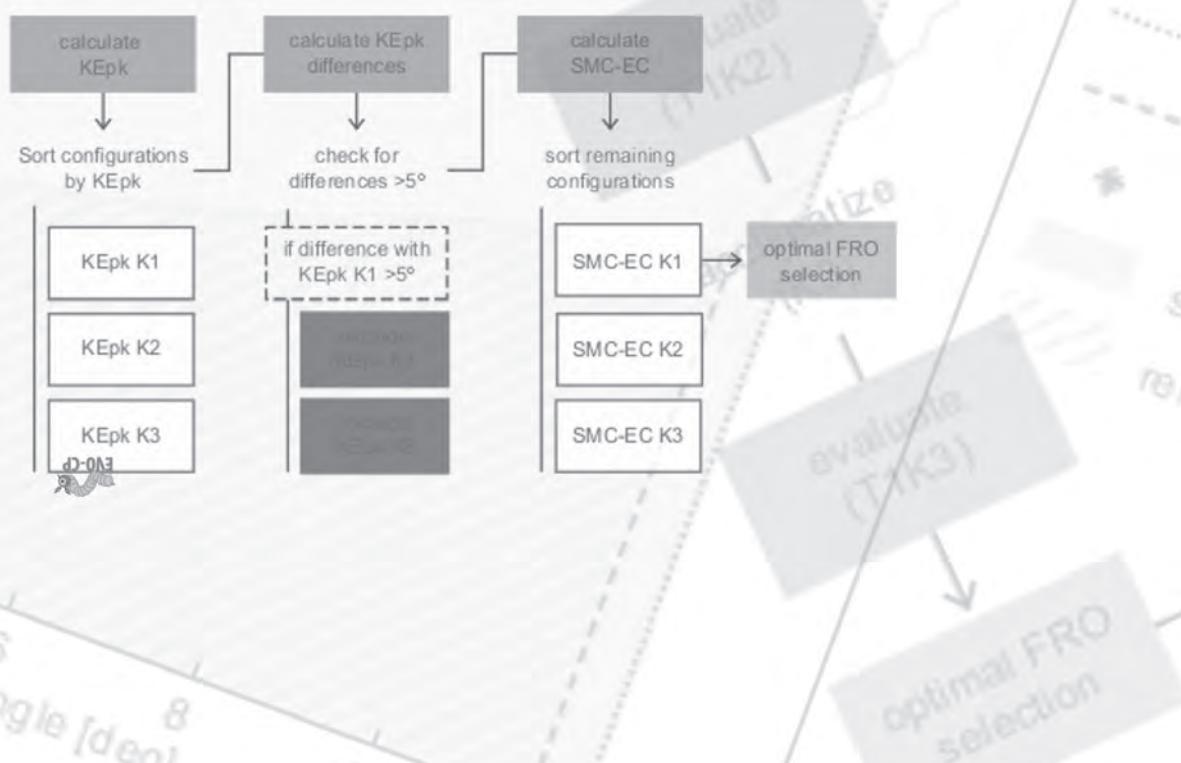
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## CHAPTER II

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# Optimising ankle foot orthoses for children with cerebral palsy walking with excessive knee flexion to improve their mobility and participation; protocol of the AFO-CP study

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## ABSTRACT

Ankle foot orthoses with a ventral shell, also known as floor reaction orthoses (FROs), are often used to reduce gait-related problems in children with spastic cerebral palsy (CP), walking with excessive knee flexion. However, current evidence for the effectiveness (e.g. in terms of walking energy cost) of FROs is both limited and inconclusive. Much of this ambiguity may be due to a mismatch between the FRO ankle stiffness and the patient's gait deviations. The primary aim of this study is to evaluate the effect of FROs optimised for ankle stiffness on the walking energy cost in children with spastic CP, compared to walking with shoes alone. In addition, effects on various secondary outcome measures will be evaluated in order to identify possible working mechanisms and potential predictors of FRO treatment success. A pre-post experimental study design will include 32 children with spastic CP, walking with excessive knee flexion in midstance, recruited from our university hospital and affiliated rehabilitation centres. All participants will receive a newly designed FRO, allowing ankle stiffness to be varied into three configurations by means of a hinge. Gait biomechanics will be assessed for each FRO configuration. The FRO that results in the greatest reduction in knee flexion during the single stance phase will be selected as the subject's optimal FRO. Subsequently, the effects of wearing this optimal FRO will be evaluated after 12-20 weeks. The primary study parameter will be walking energy cost, with the most important secondary outcomes being intensity of participation, daily activity, walking speed and gait biomechanics. The AFO-CP trial will be the first experimental study to evaluate the effect of individually optimised FROs on mobility and participation. The evaluation will include outcome measures at all levels of the International Classification of Functioning, Disability and Health, providing a unique set of data with which to assess relationships between outcome measures. This will provide insights into working mechanisms of FROs and will help to identify predictors of treatment success, both of which will contribute to improving FRO treatment in spastic CP in term.

## INTRODUCTION

With an incidence of 2-3 per 1000 living births, cerebral palsy (CP) is the most frequent cause of motor disorders in childhood in Western countries<sup>[1]</sup>. Spastic motor disorders are most common in children with CP, with symptoms of spasticity, muscle weakness and decreased selective motor control<sup>[2]</sup>, often causing limitations in mobility<sup>[3]</sup>, which may lead to a restricted participation in everyday life<sup>[4]</sup>.

Although more than half of all children with bilateral spastic CP walk independently with or without an assistive device<sup>[5]</sup>, most experience mobility-related problems such as reduced walking speed and/or an increased walking energy cost<sup>[6-12]</sup>. These problems are often caused by gait deviations<sup>[13-16]</sup>, which can be corrected by prescribing ankle foot orthoses (AFOs). An AFO imposes a mechanical constraint on the ankle, either to compensate for loss of function<sup>[17-19]</sup> or to counteract an excess of function<sup>[20,21]</sup>. An AFO therefore acts by applying control to the ankle and foot and, dependent on its design, it can indirectly stabilise the knee and hip joints<sup>[22]</sup>. As such, AFOs aim to improve, i.e. normalise joint kinetics, joint kinematics and spatio-temporal parameters<sup>[17,23-26]</sup>. Improvements in joint kinetics and kinematics have been shown to be closely coupled to an improved walking energy cost, which leads to benefits in walking ability; an effect also noted in the context of orthotic interventions<sup>[23,25-27]</sup>. This applies especially to children who walk with excessive knee flexion in midstance, since this walking pattern is particularly energy consuming<sup>[9,10]</sup> and these children are liable to show deterioration in walking ability in (pre-) puberty<sup>[28,29]</sup>.

A variety of AFO types are available, depending on the specific gait deviations of the child. For children who walk with excessive knee flexion, orthoses with a ventral shell, also known as floor reaction orthoses (FROs), are commonly prescribed<sup>[20]</sup>. Although FROs are widely used in spastic CP, evidence supporting their effectiveness is so far lacking. The decision-making process leading to FRO prescription is still based on expert opinion and experience (i.e. a trial-and-error approach), resulting in differences in treatment paradigms with respect to both the indication and the mechanical construction of FROs<sup>[30,31]</sup>. This is reflected in current literature, as studies have shown that wearing an FRO can be effective in decreasing walking energy cost, but may also have no effect<sup>[32]</sup> or even be adverse in some children in terms of walking energy cost or walking speed<sup>[26,32]</sup>.

This variation in FRO effectiveness might be partly explained by the match of the mechanical properties of the orthosis to a patient's specific gait deviations. Research in adults with neurological disorders has shown that walking energy cost with a typical spring-like AFO could be optimised by choosing the correct AFO ankle stiffness<sup>[33]</sup>,

suggesting that there may be an optimal match between a patient's characteristics and the mechanical properties of an AFO. A similar principal might also apply to FROs.

A conventional FRO is a rigid type of AFO, and includes a ventral shell and a rigid foot-plate. The biomechanical mechanism of an FRO is to create a knee extensor moment during midstance and terminal stance, by shifting of the ground reaction force forward<sup>[21]</sup>. Although an FRO might be effective in this respect, ankle push-off power is obstructed by an impeded plantar flexion in terminal stance and preswing. To enhance push-off power, a more spring-like FRO could potentially be beneficial, since it could store energy at the beginning of the stance phase that is released and returned in preswing. Achieving a sufficiently high stiffness to counteract knee flexion while including the potential benefit of spring-like properties in terms of walking energy cost may result in an optimal FRO stiffness based on the least compromise between these two goals.

Designing and evaluating the efficacy of such an optimal FRO requires an evaluation of the effects of different degrees of FRO ankle stiffness on various aspects of gait, i.e. function, mobility and participation. This implies a need for a set of outcome measures that covers all domains of the International Classification of Functioning, Disability and Health (ICF)<sup>[34]</sup>. Evaluating the effects of an intervention on more than one of the ICF domains will provide insights into mutual relations, thereby aiming to identify possible working mechanisms<sup>[35]</sup>, which will contribute to improved FRO treatment.

FRO treatment could be further improved by identifying those children who could benefit from FROs<sup>[30]</sup>. Rogozinski et al.<sup>[21]</sup> explored clinical examination parameters that might explain the efficacy of FROs in CP children walking with excessive knee flexion. They found a strong, negative correlation between knee and hip flexion contractures and peak knee extension, achieved during walking with an FRO. Other studies have shown that child characteristics and environmental factors predict the response to rehabilitation interventions such as Botulinum toxin A injections<sup>[36-38]</sup> and surgery<sup>[39-41]</sup>. Specific patient characteristics might also be relevant predictive factors for FRO efficacy.

In summary, evidence supporting the efficacy of FROs in children with spastic CP walking with excessive knee flexion remains inconclusive. Understanding of both the underlying working mechanisms and the factors predictive of treatment success is still lacking. Therefore, this project has two main goals:

1. To study the effect of an FRO optimised for ankle stiffness on walking energy cost in children with SCP walking with excessive knee flexion, compared to walking with shoes alone.

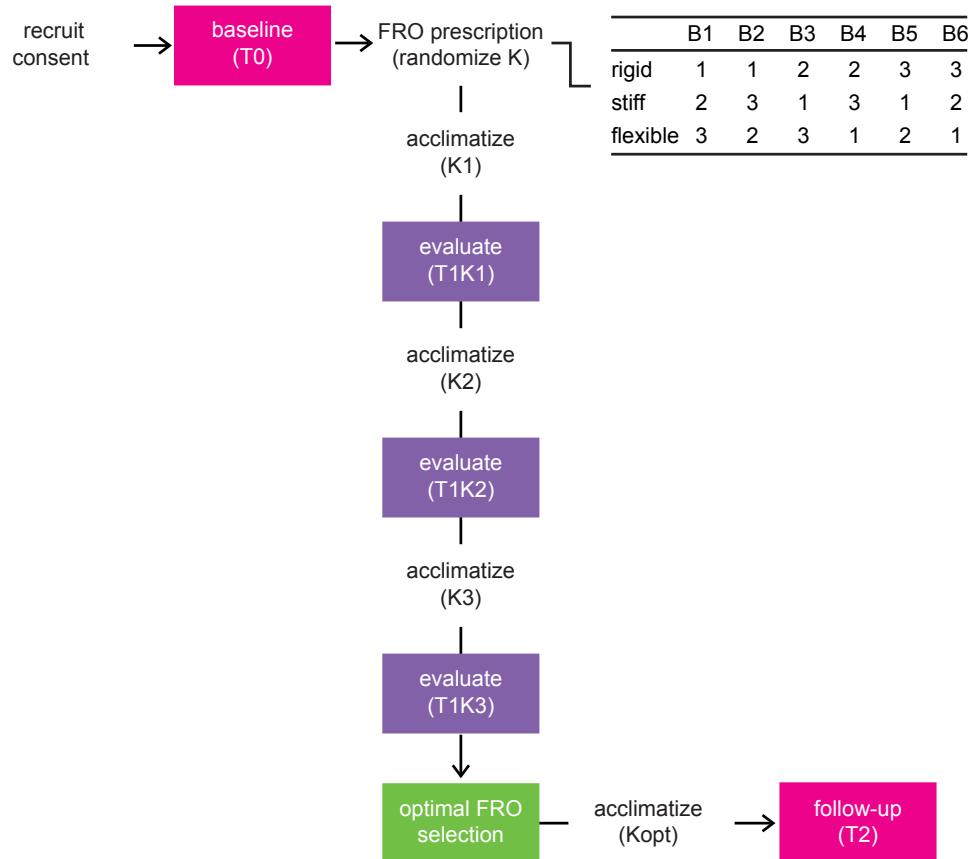
2. To identify the possible working mechanisms of an FRO, and the predictors for success of FRO treatment in children with spastic CP, walking with excessive knee flexion.

## METHODS

### Design

A pre-post experimental study consisting of two repeated measurements, i.e. at baseline, T<sub>0</sub>, walking with shoes only (control) and at 12-20 weeks follow-up, T<sub>2</sub>, walking with an optimised FRO (case) will be performed to evaluate FRO efficacy in children with spastic CP (see Figure 2.1). The study protocol has been approved by the Medical Ethics Committee of the VU University medical center in Amsterdam.

Following completion of study enrolment, baseline measurements (T<sub>0</sub>) will be performed barefoot, with shoes only and with the subject's current orthoses (if applicable). Stiffness (K) of the new FRO will be varied into three configurations: rigid, stiff, and flexible. A balanced block randomisation will be applied for six possible sequences of stiffness configurations, to ensure that the same number of patients is allocated to each sequence. Every configuration will be worn for an accommodation period of four to eight weeks, after which FRO efficacy will be evaluated (T<sub>1K1</sub>, T<sub>1K2</sub> and T<sub>1K3</sub>). An analysis of the evaluation of all FRO configurations will allow the selection of the stiffness with the maximal benefit for a particular subject, referred to as the subject's optimal FRO (the selection procedure is explained further below). Following this selection, the optimal FRO will be worn for twelve to twenty weeks, after which the follow-up measurements (T<sub>2</sub>) will be taken.



**Figure 2.1.** Schematic representation of the study design. Following baseline measurements (T<sub>0</sub>), the subjects will be prescribed an interventional FRO. The stiffness of this FRO will be varied (rigid, stiff and flexible) and the order of FRO stiffness will be block randomised. Accommodation time for each stiffness will last 4-8 weeks, after which effects will be evaluated (T<sub>1K1</sub>, T<sub>1K2</sub>, and T<sub>1K3</sub>). Following these evaluations, an optimal FRO for the subject will be selected. Follow-up measurements (T<sub>2</sub>) will be carried out at 12-20 weeks.

Abbreviations: B, block; FRO, floor reaction orthosis; K, AFO stiffness. K<sub>1</sub>, K<sub>2</sub>, and K<sub>3</sub> represent either rigid, stiff or flexible stiffness configurations.

## Participants

### **Inclusion and exclusion criteria**

Our aim is to include 32 children with spastic CP (Gross Motor Function Classification System<sup>[42]</sup> (GMFCS) levels I, II and III, provided that the child is able to perform a 3D-gait analysis without walking aids) who are candidates for a (new) FRO. Children will be recruited from the outpatient clinic of the VU University medical center, Amsterdam and affiliated rehabilitation centres.

**Table 2.1.** Inclusion and exclusion criteria.

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#### **inclusion criteria**

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Spastic CP;

6-14 years;

A gait pattern characterised by excessive knee flexion (jump gait, apparent equinus or crouch gait)<sup>[56]</sup>;

GMFCS I, II, or III (provided that the patient is able to walk independently for at least 15 meters)

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#### **exclusion criteria**

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Any orthopaedic surgery or other surgical interventions that might influence mobility in the past 6 months;

Botulinum toxin A injections in the past 3 months, Intrathecal Baclofen therapy in the past 6 months, or SDR in the past year;

Impairments that could contraindicate fitness testing;

Plantar flexion contractures or knee contractures  $>10^\circ$  or hip endorotation  $>20^\circ$  in midstance;

Other medical conditions influencing mobility;

Severe behavioural problems.

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Abbreviations: GMFCS, Gross Motor Function Classification System<sup>[42]</sup>; SDR, Selective Dorsal Rhizotomy.

Study information will be provided to potential participants in the form of a patient information letter and a brochure. Patients and parents willing to participate will be contacted by the primary investigator, who will verify inclusion and exclusion criteria (see Table 2.1). When a patient meets the inclusion criteria, oral and written informed consent will be obtained from both parents, and from children aged 12 years and older, in accordance with the declaration of Helsinki.

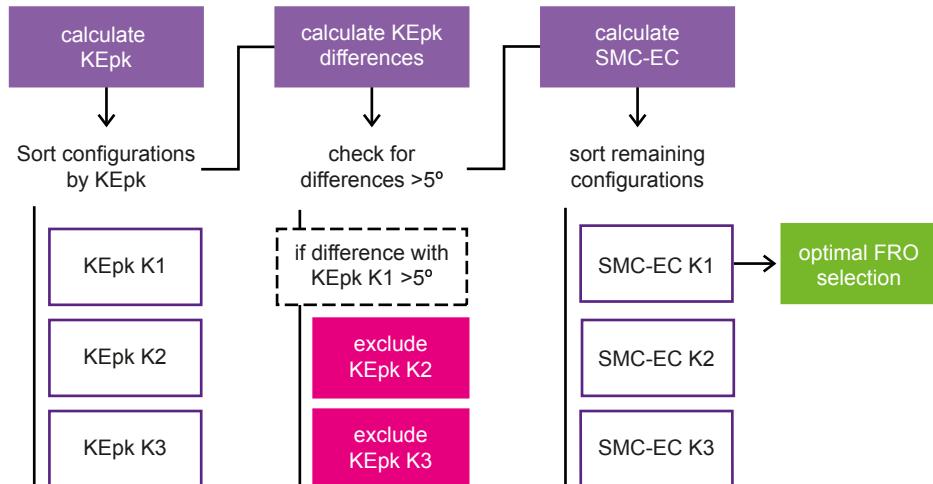
### **Sample size**

The sample size will be based on a power analysis of the expected changes (i.e. T<sub>0</sub> versus T<sub>2</sub>) in the primary outcome, walking energy cost (EC) [ $J \cdot kg^{-1} \cdot m^{-1}$ ]. According to literature, walking energy cost in children with CP may be 30-50% higher than in healthy children<sup>[10-12]</sup>. Spastic CP children with GMFCS levels I, II and III show a mean net EC of 5.02 ( $\pm 1.70$ )  $J \cdot kg^{-1} \cdot m^{-1}$ <sup>[26]</sup>. A reduction of 25% in this value ( $\approx 1.26 J \cdot kg^{-1} \cdot m^{-1}$ ) is considered to be a clinically significant change<sup>[25,26]</sup>. Assuming a power of 80% and a significance level of 0.05, detecting a clinically significant change will require a sample size of 29 children<sup>[43]</sup>. Allowing for a dropout of approximately 10%, a sample size of 32 will be sufficient.

### **Investigational AFO**

Investigational FROs will be composed of prepreg carbon, manufactured using the Mälmo-technique (Otto Bock HealthCare GmbH, Duderstadt, Germany). For fair evaluation of efficacy, the investigational FRO will be fabricated with a rigid footplate. To further ensure a fair comparison, tuning of the FRO-footwear combination following the Owen method will be carried out for each configuration<sup>[44]</sup>.

Investigational FROs will be fabricated with an integrated Neuro Swing<sup>®</sup> system hinge (Fior & Gentz, Lüneberg, Germany), which is available in different sizes. The size of the hinge is dependent on the body weight and length of the patient. For this study, it is expected that only the 14mm and 16mm hinges will be used. The hinge holds an anterior and posterior shaft, and comes with a package of five springs, each with a different degree of stiffness. Ankle stiffness can be adjusted within the same orthosis, using different spring forces towards plantar and dorsal flexion. In this study, the hinge will be prepared in three configurations: rigid, stiff and flexible. The rigid configuration (i.e.  $\pm 4.3 Nm \cdot deg^{-1}$ ) will entirely prevent dorsal or plantar flexion. For the stiff and flexible configurations, the spring force for dorsal flexion will be varied using the strongest spring (i.e.  $\pm 1.2 Nm \cdot deg^{-1}$  [14mm] and  $\pm 2.4 Nm \cdot deg^{-1}$  [16mm]) and the second strongest spring (i.e.  $\pm 0.5 Nm \cdot deg^{-1}$



**Figure 2.2.** Flowchart of the optimal FRO stiffness selection procedure. After sorting the different stiffness configurations based on peak knee extension angle in single support (KEpk), absolute differences in peak knee extension angle will be calculated. KEpk of K2 and/or KEpk of K3 will be excluded when more than five degrees. Otherwise, the remaining configurations will be sorted by net non-dimensional walking energy cost (this can be either two or three remaining configurations). The stiffness that results in the lowest walking energy cost will be selected as the subject's optimal FRO.

Abbreviations: KEpk, peak knee extension angle during single support; SMC-EC, net non-dimensional walking energy cost relative to speed matched control cost; SS, single support. K<sub>1</sub>, K<sub>2</sub> and K<sub>3</sub> represent either rigid, stiff, or flexible FRO stiffness configurations.

[14mm] and  $\pm 1.0 \text{ Nm}\cdot\text{deg}^{-1}$  [16mm]), respectively. The spring force towards plantar flexion will be very compliant (i.e.  $\pm 0.01 \text{ Nm}\cdot\text{deg}^{-1}$  [14mm] and  $\pm 0.04 \text{ Nm}\cdot\text{deg}^{-1}$  [16mm]) for both configurations.

### Accommodation procedures

The accommodation period for all three FRO configurations will include a gradual increase in the length of time the FRO is worn each day, in order to minimise the risk of adverse events. Patients will be contacted one week after setting each new FRO configuration, to check for adverse events such as pain, discomfort, or pressure sores. If the patient has no complaints, the accommodation period will continue until the next visit (four to eight weeks later). When adverse events are reported, the investigator will identify the causes and make an appropriate decision according to protocol. The accommodation period will not start until all complaints are resolved.

### Optimal AFO selection procedure

Following a standard procedure, evaluation of FRO efficacy of the three configurations (T1K1, T1K2 and T1K3) will lead to selection of the subject's optimal FRO configuration (see Figure 2.2). Since clinical assessment of FRO effectiveness in children walking with excessive knee flexion is mainly based on knee kinematics in stance, the minimum amount of knee flexion (i.e. peak knee extension) in the single support phase will be the main discriminating parameter. The configuration that results in smallest peak knee flexion will be selected as the subject's optimal FRO. Differences of less than 5° will be considered equal, since this angle lies within the variability of 3D-gait analysis<sup>[45]</sup>. Should minimum knee flexion in single support be unable to discriminate between the remaining configurations, walking energy cost (expressed as net non-dimensional energy cost relative to speed-matched control cost (SMC-EC)<sup>[46,47]</sup> will be decisive. In this situation, the FRO that results in the lowest SMC-EC will be selected as the subject's optimal FRO.

### Outcome measures

Outcome measures for this study are categorised in accordance with the ICF<sup>[34]</sup> and cover the components 'body functions and structures' and 'activities and participation', as well as personal and environmental factors<sup>[34]</sup>. An overview of all outcome measures is presented in Table 2.2.

### Primary outcome

Our primary outcome measure is walking energy cost, which will be measured during a 6-minute walking test on an indoor oval track. Subjects will be asked to walk at a self-preferred comfortable speed, during which oxygen uptake and carbon dioxide production will be measured using the accurate and reliable Metamax 3B portable gas analysis system (Cortex Biophysik, Leipzig, Germany). Calculations will be based on measurements during a steady state of walking, defined as a period of at least one minute in which fluctuations in walking speed, oxygen uptake and carbon dioxide production show the least change<sup>[47]</sup>.

Mean steady-state breath-by-breath oxygen uptake values and respiratory exchange ratios will be computed. Using these values, gross and net energy consumption will be calculated and normalised according to the net non-dimensional scheme of Schwartz et al.<sup>[47]</sup>. The primary outcome measures will be expressed as net EC and as SMC-EC. Furthermore, non-dimensional walking speed (N\_speed) (a secondary outcome measure) will be calculated.

EC measurements in children with CP are sufficiently sensitive, as shown by Brehm et al.<sup>[48]</sup>. The net non-dimensional normalisation scheme of Schwartz et al.<sup>[47]</sup> is suggested to be the preferred method for reporting oxygen consumption data for subjects who have not reached their full stature, since it is largely independent of mass, height and age.

### **Secondary outcome**

Secondary outcome measures include daily activity, gait biomechanics, walking speed (N\_speed) and diversity, intensity and enjoyment of participation (assessed with the Children's Assessment of Participation and Enjoyment [CAPE]). Two of these outcome measures (daily activity and gait biomechanics) are further explained below.

#### **Daily activity**

Daily activity will be measured for one week with a StepWatch3<sup>TM</sup> Activity Monitor 3.0 (SAM) (Cyma Corporation Seattle, WA, USA), which is an ankle worn accelerometer that measures the average amount of steps per minute over a broad spectrum of cadences. The SAM will be attached to the ankle of the dominant leg. Subjects will be instructed not to remove the SAM at any time, except when taking a bath or shower or when swimming. For adequate interpretation of the data, subjects will be asked to keep a diary of their activity program during each day of the week.

Daily activity will be determined as 1) average total steps per day, 2) percentage of time children were active, 3) percentage of time children were inactive, 4) ratio of medium to low activity levels and 5) percentage of time children show high activity levels. A calibrated SAM has been shown to be an accurate tool for recording daily steps in children with CP<sup>[49,50]</sup>.

**Table 2.2.** Overview of tests performed at different measurement moments.

		To	T1K <sup>a</sup>	T2
<b>primary study parameters</b>				
activities and participation	ECWT	x	x	x
<b>secondary study parameters</b>				
body functions and structures	3D-gait analysis	x	x	x
activities and participation	SAM <sup>c</sup>	x	x	x
	CAPE <sup>c</sup>		x	
<b>effect modifiers</b>				
body functions and structures	physical fitness test	x		
environmental and personal factors	physical examination <sup>b</sup>	x		
	gait pattern	x		
	intake questionnaire	x		
	BSS	x		
	FMS	x	x	x
	FAQ	x	x	x
	GMFCS	x		
<b>other outcomes</b>				
	GAS	x		x
	FRO properties	x	x	x
	motivation diary	x	x	x
	satisfaction	x	x	x

<sup>a</sup>T1 will be repeated for each FRO configuration (i.e. rigid, stiff and flexible).

<sup>b</sup>The physical examination includes passive Range of Motion, selective motor control and gross motor function tests.

<sup>c</sup>SAM and CAPE data will be assessed in the week prior to the ticked measurement moment.

Abbreviations: BSS, Bronnen van Steun en Spanning; CAPE, Children's Assessment of Participation and Enjoyment; ECWT, Energy Cost of Walking Test; FAQ, Functional Assessment Questionnaire; FMS, Functional Mobility Scale; FRO, floor reaction orthosis; GAS, Goal Attainment Scaling; GMFCS, Gross Motor Function Classification System; SAM, StepWatch<sup>TM</sup> Activity Monitor.

### Gait biomechanics

Joint kinematics will be assessed in the laboratory, using a three-dimensional motion analysis system (OptoTrak, Northern Digital, Waterloo, Canada), while the subject walks on a 10m walkway at a self-preferred comfortable speed. Marker clusters will be attached to the feet, shanks, thighs, pelvis and trunk. To determine anatomical coordinate systems, anatomical landmarks will be palpated according to Cappozzo et al.<sup>[51]</sup>. Joint kinetics will be calculated by assessment of the ground reaction force, using an integrated force plate (OR6-5-1000, AMTI, Watertown, USA).

At baseline, all subjects will be measured walking bare foot and with shoes-only. An additional condition (old AFO-footwear combination) will be included for children who have (suitable) old orthoses. Follow-up recordings will be made while walking with the new FRO-footwear combination. Six trials, with the subject stepping on the force plate, will be completed for each condition (i.e. three trials for each leg). Data on joint kinematics, and kinetics around the hip, knee and ankle will be averaged. Spatio-temporal parameters, such as step length [m], step width [m] and cadence [ $\text{steps}\cdot\text{min}^{-1}$ ] will also be calculated.

### Effect modifiers

As potential effect modifiers, the following outcome measures will be assessed: demographic variables, disease characteristics, personal and family characteristics, level of functional mobility and physical fitness (explained below).

### Physical fitness

Physical fitness will be measured by means of an aerobic and anaerobic exercise test on a bicycle ergometer. The aerobic test will be performed according to the protocol described by Balemans et al.<sup>[52]</sup> and aerobic fitness will be defined as oxygen uptake over the 30 seconds with the highest sustained load ( $\text{VO}_{2\text{ peak}}$ ) [ $\text{ml}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$ ]. Anaerobic power will be determined using the 20 seconds Wingate Anaerobic cycling Test (20s-WAnT), a sprint test against a constant breaking torque<sup>[53]</sup>. Anaerobic fitness will be defined by the mean anaerobic power over 20 seconds ( $\text{P}_{20\text{ mean}}$ ) [ $\text{W}\cdot\text{kg}^{-1}$ ] and by the highest power output within the 20 seconds, the peak anaerobic power ( $\text{P}_{20\text{ peak}}$ ) [ $\text{W}\cdot\text{kg}^{-1}$ ]. Measurement procedures, equipment and protocols for both tests will be as described by Balemans et al.<sup>[52]</sup>.

### **Other outcomes**

Other study outcome measures will include 1) the patient's personal treatment goals, measured with Goal Attainment Scaling (GAS), 2) treatment adherence, assessed with a motivation diary and with the @monitor<sup>[54]</sup>, 3) satisfaction with the FRO, as perceived by the patient and parents and 4) FRO stiffness, measured with BRUCE, which is a recently developed device for measuring mechanical AFO properties<sup>[55]</sup>.

### **Statistical analysis**

#### **Subject population**

Demographic variables and disease characteristics will be summarised using descriptive statistics. Furthermore, the means, medians, standard deviations and 95% confidence interval (CI) of primary and secondary outcome measures will be presented for all visits. In addition, correlations between parameters will be examined using correlation coefficients and graphical techniques.

#### **Evaluation of FRO efficacy**

Evaluation of the efficacy of a subject's optimal FRO will be based on analyses of pre/post-intervention differences in primary and secondary outcome measures. The pre-intervention (control) condition will be for shoes-only. Mean data for these measurements (assessed at T1K2) will be compared to follow-up measurements (T2), using paired sample t-tests.

To identify working mechanisms, multivariate linear regression analyses will be applied to investigate which of the changes in gait biomechanics are associated with changes in walking energy cost (model 1) and daily activity (model 2). First, a univariate regression analysis (ANOVA) will be performed to determine which factors are significantly associated with changes in the biomechanics of gait ( $p \leq 0.1$ ), followed by the analysis of significant factors ( $p \leq 0.05$ ) in a multivariate regression analysis model.

#### **Identifying prognostic factors**

Multivariate regression analysis will also be applied to investigate to what extent child characteristics and FRO stiffness represent determinants for success of FRO treatment, defined as decreased walking energy cost (model 1), improvement in daily activity (model 2) and positive GAS scores (model 3). Initially, a univariate regression analysis

(ANOVA) will be performed to determine which factors are significantly associated with FRO treatment outcomes ( $p \leq 0.1$ ). Significant factors ( $p \leq 0.05$ ) will then be included in a multivariate regression model. Model analysis will include factors such as level of physical fitness, baseline disease characteristics, gait pattern, level of functional mobility, environmental factors and FRO characteristics.

## DISCUSSION

This study will evaluate the effects of varying degrees of FRO ankle stiffness on different aspects of gait. Based on earlier studies, an optimal match is expected between specific patient characteristics and FRO stiffness. Assuming that there is an optimal FRO stiffness for each subject, this study might lead directly to an optimised FRO treatment for these patients. In addition, the study will evaluate FRO efficacy, using outcome measures that are relevant in the patient's daily life (i.e. walking energy cost and daily activity), thereby emphasising clinical relevance.

Because the stiffness of an FRO should be based on the specific gait deviations of the child, the inclusion criteria of this study will be specifically defined. This will result in a relatively homogeneous study population, enabling a fair comparison of subjects. On the other hand, these strict criteria may make it difficult to generalise results to the wider treatment and prescription of FROs, also because the design of the investigational FRO design differs from conventional FROs. Nonetheless, it is expected that the results of the study will allow an optimal FRO treatment to be defined in this specific patient group.

This study will be the first to investigate broadly the efficacy of an individually optimised FRO, including evaluation of effects on multiple ICF levels. This will result in a unique data set with which to assess mutual relations between outcome measures. We anticipate that this analysis will aid in identifying both the underlying working mechanisms of FRO and the factors important to treatment success. In conclusion, the data generated by this study may provide not only novel insights, but may also contribute to improved FRO treatment in spastic CP in the (near) term.

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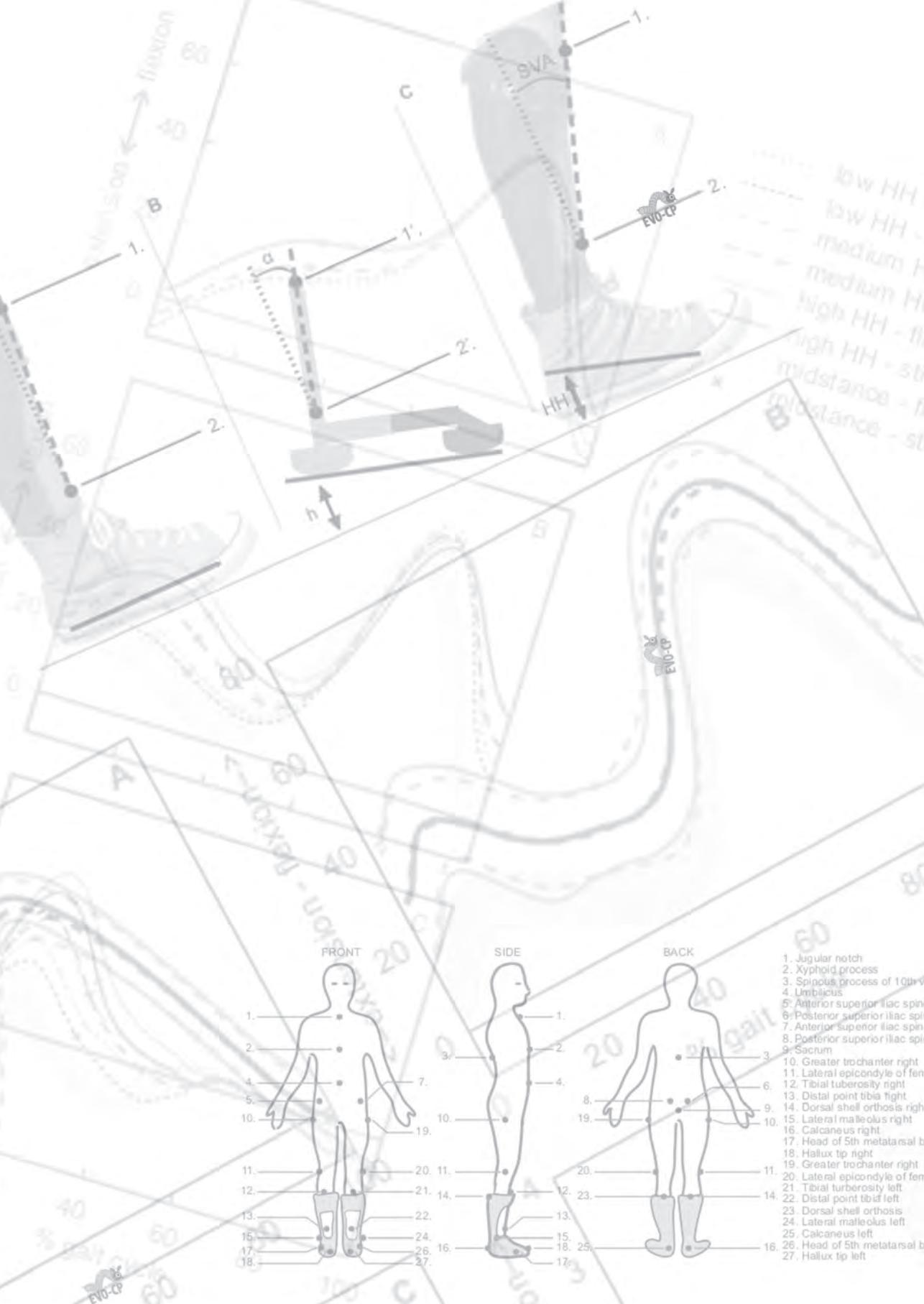
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# The shank-to-vertical angle as a parameter to evaluate tuning of ankle foot orthoses

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## ABSTRACT

The effectiveness of an ankle foot orthosis-footwear combination (AFO-FC) may be partly dependent on the alignment of the ground reaction force with respect to lower limb joint rotation centers, reflected by joint angles and moments. Adjusting (i.e. tuning) the AFO-FC's properties could affect this alignment, which may be guided by monitoring the shank-to-vertical angle (SVA). This study aimed to investigate whether the SVA during walking responds to variations in heel height and footplate stiffness, and if this would reflect changes in joint angles and net moments in healthy adults. Ten subjects walked on an instrumented treadmill and performed six trials while walking with bilateral rigid AFOs. The AFO-FC heel height was increased, aiming to impose a SVA of 5°, 11° and 20°, and combined with a flexible or stiff footplate. For each trial, the SVA, joint flexion-extension angles and net joint moments of the right leg at midstance were averaged over 25 gait cycles. The SVA significantly increased with increasing heel height ( $p<0.001$ ), resulting in an increase in knee flexion angle and internal knee extensor moment ( $p<0.001$ ). The stiff footplate reduced the effect of heel height on the internal knee extensor moment ( $p=0.030$ ), while the internal ankle plantar flexion moment increased ( $p=0.035$ ). Effects of heel height and footplate stiffness on the hip joint were limited. Our results support the potential to use the SVA as a parameter to evaluate AFO-FC tuning, as it is responsive to changes in heel height and reflects concomitant changes in the lower limb angles and moments.

## INTRODUCTION

Ankle foot orthoses (AFOs) are frequently applied in patients with neurological disorders, aiming to normalize joint kinematics and joint kinetics during walking<sup>[1-4]</sup>. Although it has been shown that AFOs can significantly improve sagittal joint kinematics and kinetics<sup>[2,3,5-7]</sup>, inadequate alignment of ground reaction force (i.e. distant from the joint rotation centers) during walking negatively impacts the effectiveness<sup>[4,8,9]</sup>.

Tuning of the AFO optimizes the alignment of the ground reaction force with respect to the joint rotation centers, enhancing normalization of the joint kinematics and kinetics<sup>[8-12]</sup>. Such tuning can be described as the process in which the properties of an AFO-footwear combination (AFO-FC) are manipulated. Commonly used adjustments comprise changing the footplate stiffness to affect the point of application of the ground reaction force, and altering the heel-sole differential (i.e. the difference in height between the heel and forefoot of the shoe), which affects shank orientation<sup>[8]</sup>. The combined effect of the AFO-FC's ankle angle and heel-sole differential can be described in terms of the shank-to-vertical angle (SVA). The SVA, i.e. tibia inclination, is the angle between the anterior surface of the tibia and the vertical in the global sagittal plane<sup>[8,13]</sup>. It is clinically often measured using sagittal video recordings<sup>[13]</sup>. The SVA is considered inclined, when the shank is tilted forward, or reclined, when it is tilted backward with respect to the vertical. Owen<sup>[13]</sup> suggested that an appropriate shank orientation at midstance aligns the ground reaction force to the joint rotation centers, which contributes to stability, facilitates adequate switching from flexion to extension moments at the knee and hip, and lowers vertical center of mass excursion. Accordingly, the SVA at midstance may be an important and relatively simple parameter to evaluate the effects of adjustments to the AFO-FC during its tuning process<sup>[8,13]</sup>, also because information on the ground reaction force and calculations of joint moments are not always available in clinical practice.

Several studies in patients with neurological disorders report the SVA, and describe a normalization of gait parameters following changes of the heel-sole differential<sup>[11,12,14,15]</sup>. However, in all available studies, the SVA was measured while the patient was in a static position, whereas there is no evidence showing that the SVA in this position represents the SVA at midstance<sup>[8]</sup>. Evidence on the effects of changing the footplate stiffness on the SVA, as well as on joint kinematics and kinetics is also lacking. Yet, in clinical practice, such manipulations of footplate stiffness, in addition to changing the heel-sole differential are commonly applied. Since tuning of these AFO-FC properties is generally guided by monitoring the SVA at midstance, insight is needed in how the SVA responds to changes

of the heel-sole differential and footplate stiffness, in order to assess its potential as a parameter to evaluate the effects of such manipulations.

To this end, we evaluated in healthy young adults i) whether the SVA at midstance can be influenced during walking with an AFO-FC by applying commonly used manipulations within the process of tuning (i.e. changing AFO-FC heel-sole differential and footplate stiffness), and ii) how changes in the SVA, as a result of the manipulations, are reflected in ankle, knee and hip flexion-extension angles and net internal joint moments at midstance. We hypothesized that the SVA would be responsive to changes in AFO-FC heel height and that this would be reflected by increased knee and hip flexion angles and net internal joint extension moments at midstance. As a stiff footplate mainly aims to shift the ground reaction force forward without affecting joint flexion-extension angles, we expected no response of the SVA to changes in footplate stiffness, while it was expected to affect internal net joint moments.

## METHODS

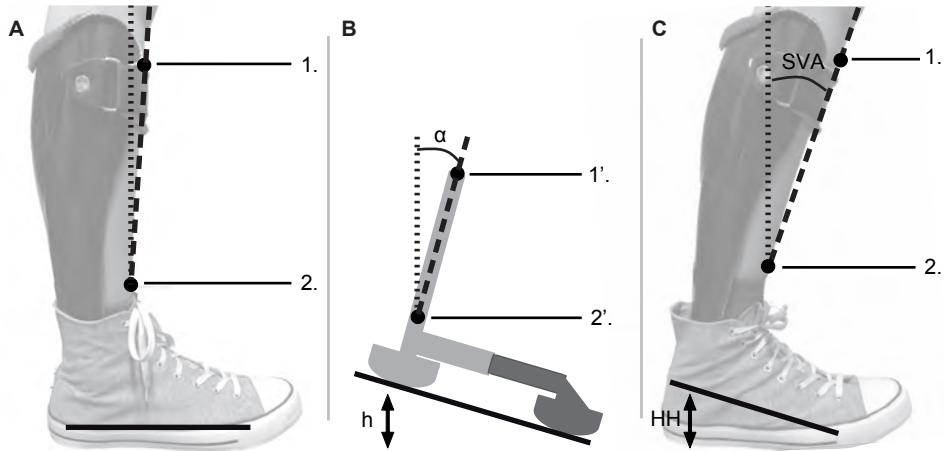
### Participants

Ten healthy young adults (3 male; mean (SD) age: 24 (3) years; mean (SD) body mass index: 22.8 (2.2)) participated. All subjects provided written informed consent in accordance with the procedures of the Institutional Review Board of the VU University.

### Materials

For this study, two pairs of prepeg carbon AFOs were manufactured (European shoe size: 39 and 43) (see Figure 3.1A). Each participant chose the best fitting pair. The stiffness of the AFOs at the ankle and metatarsal joints was measured using BRUCE<sup>[16]</sup>, which is an instrument to define AFO mechanical properties. The AFOs were rigid at the ankle ( $7.9 \text{ Nm}\cdot\text{deg}^{-1}$ ), aiming to immobilize the ankle joint at  $0^\circ$ .

According to Owen<sup>[13]</sup>, important kinematic characteristics at midstance (e.g. thigh inclination) can only be preserved with an SVA ranging from  $7^\circ$ - $15^\circ$ , while an SVA of  $10^\circ$ - $12^\circ$  is suggested to be optimal. In the current study, AFO-FC heel-sole differential was varied using three heel heights by applying insole wedges, aiming to impose an SVA of  $5^\circ$ ,  $11^\circ$  and  $20^\circ$  in static position. As such, the effects of SVA manipulations near the presumed optimum and outside the suggested optimal range were investigated. The height of the wedges was pre-defined for both AFO-FCs, using a dedicated instrument to measure heel height and heel-sole differential of an AFO-FC when doffed (Vertical Inclinometer



**Figure 3.1.** A) Picture of the AFO-FC of the right leg without insole wedges. B) Schematic representation of the VICTOR<sup>[17]</sup>, with virtual markers 12' and 13' as analogue reference points of the anatomical markers at the tibial tuberosity (Figure 2, #12) and tibia (Figure 2, #13). VICTOR was used to determine the height of the insole wedges to impose a SVA of 5°, 11° and 20° during the walking trials. Wedges were added to increase the height (h) of the heel probe until the inclination angle (α) reflected the pre-defined angles of 5°, 11° and 20°. C) Picture of the AFO-FC including insole wedges (pre-defined using VICTOR (h)), resulting in the heel height (HH). The SVA during walking was calculated as the angle between the line at the anterior surface of the tibia (dashed) (i.e. the line connecting the marker at the tibial tuberosity (#12) and tibia (#13)) and the vertical (dotted) in the global sagittal plane. The SVA was expected to represent α.

Dotted, the vertical as used for SVA calculation; dashed, line at the anterior surface of the tibia, representing the long axis of the shank in the global sagittal plane; solid, estimated position of the footplate in the shoe.

on a Rail (VICTOR)<sup>[17]</sup>) (see Figure 3.1B). Using VICTOR, low (size 39: 0.6 cm; size 43: 1.3 cm), medium (size 39: 2.8 cm; size 43: 2.8 cm) and high (size 39: 4.9 cm; size 43: 5.3 cm) heel heights were specified (see Figure 3.1). These heel heights were combined with two different degrees of footplate stiffness, which could be changed by adding a stiff inlay footplate ( $0.89 \text{ Nm}\cdot\text{deg}^{-1}$ ) to the AFO's flexible footplate ( $0.06 \text{ Nm}\cdot\text{deg}^{-1}$ ). The provided shoes (i.e. flexible sneakers) were large enough to allow for the insole wedges.

### Measurements

Subjects walked on the GRAIL system (Motek Medical BV, Amsterdam, the Netherlands), consisting of a split-belt instrumented treadmill (ForceLink<sup>®</sup>, Culemborg, the Netherlands) and a passive marker motion capture system (Vicon, Oxford, UK), collecting marker trajectories. Ground reaction forces were captured from force sensors mounted underneath both treadmill belts, and synchronized with kinematic data at 120 Hz.

Reflective markers were placed at anatomical landmarks according to the Human Body Model<sup>[18,19]</sup> (see Figure 3.2). The SVA was calculated as it is defined in clinical practice<sup>[13]</sup>, i.e. using the line over the anterior surface of the tibia, representing the long axis of the shank, and calculated as the angle between this line and the vertical in the global sagittal plane (see Figure 3.1). In order to do so, additional markers were added to the Human Body Model (see Figure 3.2): at the tibial tuberosity (#12 and #21) and at a distal point on the tibia (#13 and #22, i.e. at 75% of the lower leg, measured from the tibial tuberosity (#12 and #21) to the floor and vertically in line with the marker at tibial tuberosity in the frontal plane). Other additional markers were placed at the dorsal shell of each AFO (#14 and #23), which were horizontally aligned with the tibial tuberosity marker (#12 and #21) in the sagittal plane and vertically aligned to the calcaneus marker (#16 and #25) in the sagittal plane. These markers were used to determine movements of the shank in the AFO, therewith evaluating the immobilization of the ankle. This was done for interpretation of the results, as inadequate immobilization is expected to affect joint flexion-extension angles and moments. The Human Body Model foot markers (#16-18 and #25-27) and the markers at the lateral malleoli (#15 and #24) were positioned on the shoe. None of the markers were replaced between different trials.

### Procedure

After being provided with the AFO-FC, the subject accommodated to walking on a treadmill until he/she felt comfortable. Subsequently, the subject's comfortable walking speed was determined following a standardized protocol. Following this protocol, the participant started walking at an initial speed of  $0.8 \text{ m}\cdot\text{s}^{-1}$ . Treadmill speed was then gradually increased with  $0.1 \text{ m}\cdot\text{s}^{-1}$  until the participant indicated the speed as comfortable. From thereon, speed was further increased until comfortable speed  $+0.3 \text{ m}\cdot\text{s}^{-1}$  and gradually decreased until the participant indicated the speed as comfortable again. The mean of both self-selected speeds represented the subject's comfortable

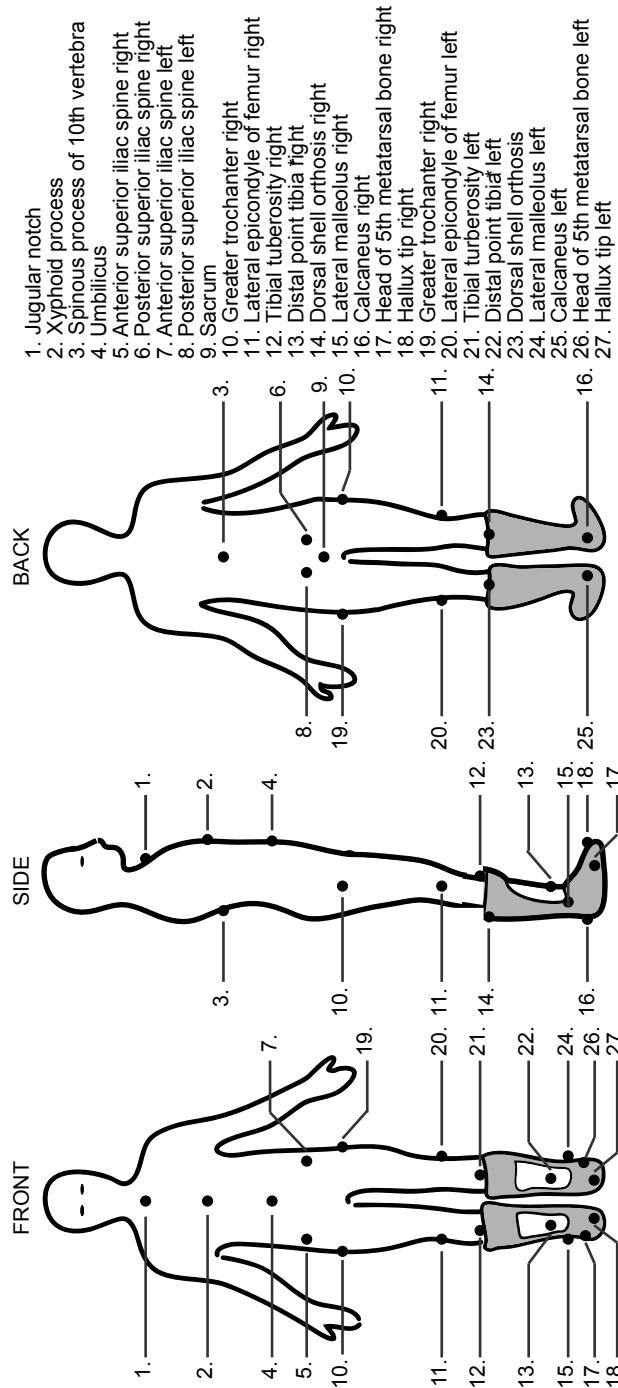


Figure 3.2. Marker model according Human Body Model, with six additional markers (i.e. marker numbers 12, 13, 14, 21, 22, and 23).

\*The markers referring to the distal point of the tibia were positioned at 75% of the lower leg, measured from the tibial tuberosity to the floor and in line with the tibial tuberosity marker in the frontal plane

walking speed. Thereafter, subjects performed six walking trials of 2 minutes at this comfortable speed. For each trial, AFO-FC heel height was set into low, medium or high, and combined with either the stiff or flexible footplate. The sequence of these six combinations was randomly applied.

### ***Data processing***

Joint flexion-extension angles and net internal moments were calculated using the Human Body Model and D-flow software<sup>[18,19]</sup>. Joint flexion-extension angles were calculated using the orientation of the distal segment with respect to the orientation of proximal segment and expressed in the sagittal plane of the proximal segment. The SVA, calculated in the global sagittal plane, was defined as the angle between the anterior surface of the tibia and the vertical<sup>[13]</sup> (see Figure 3.1). Another line was created using the position of the marker at the dorsal shell of the AFO (#14 and #23) and the lateral malleolus marker (#15 and #24) in the global sagittal plane. The angle between the two lines represented changes of the position of the shank with respect to the AFO (i.e. Shank-to-AFO angle). Assuming that this angle would be unchanged with a fully immobilized ankle joint, smaller angles would indicate movement of the shank towards the AFO's dorsal shell. Calculations of the SVA and Shank-to-AFO angle were done using Matlab 2011 (The Mathworks, USA).

Marker and force plate data were low pass filtered at 6 Hz using the Human Body Model<sup>[19]</sup>. To select only strides with foot placement on a single belt, a stride was excluded if i) the force of that stride deviated more than 100% from the mean force of all strides, or ii) the length of the stride deviated more than 20 samples from the median length of all strides. For further processing, only correctly recorded strides were selected, based on two criteria i) single-belt foot placement and ii) sufficient marker data (i.e. no occlusion) to calculate the considered parameters. Subsequently, remaining strides were normalized to 100% gait cycle and the SVA, lower limb joint flexion-extension angles and net internal moments, and Shank-to-AFO angles of the right leg were determined at midstance, defined as the moment that the malleolus marker of the contralateral leg (#24) passed the malleolus marker of the ipsilateral leg (#15). The parameters were limited to midstance, as the SVA is clinically used to evaluate the effects of tuning at this stage of the gait cycle<sup>[13]</sup>. The parameters were averaged over 25 steps, which were selected starting from the end of the trial.



## Statistics

The effects of the different AFO-FC conditions on the SVA, joint flexion-extension angles and net moments, and Shank-to-AFO angle were analyzed for statistical significance using a two-way repeated measures analysis of variance (ANOVA) with two within-subject factors (i.e. heel height (three levels) and footplate stiffness (two levels)), using Bonferroni post-hoc adjustments ( $\alpha=5\%$ ). Statistical analyses were done using IBM SPSS Statistics, version 20 (SPSS Inc, Chicago, USA).

## RESULTS

### SVA

The SVA at midstance significantly increased with increasing heel height (see Figure 3.3A). The SVA during walking (mean (SD) walking speed:  $0.96 (0.07) \text{ m}\cdot\text{s}^{-1}$ ) was larger in all heel height conditions compared to the imposed SVA of  $5^\circ$ ,  $11^\circ$  and  $20^\circ$  in static position (see Table 3.1). Footplate stiffness had no effect on SVA, and also no interaction effect of heel height and footplate stiffness on SVA was found ( $F=0.71$ ,  $p=0.505$ ) (see Figure 3.3A).

### Knee joint

The effects of the AFO-FC manipulations were most prominent at the knee joint, with the knee flexion angle and internal knee extensor moment at midstance significantly increasing with increasing heel height (see Table 3.1). The stiff footplate tended to decrease the knee flexion angle and internal knee extensor moment at midstance, although this was only significant for the internal knee extensor moment. An interaction effect of heel height and footplate stiffness on the knee flexion angle ( $F=3.54$ ,  $p=0.050$ ) and internal knee extensor moment ( $F=4.06$ ,  $p=0.035$ ) was found, indicating an inhibiting effect of the stiff footplate for the low and high heel height conditions, but not for the medium heel height condition (see Table 3.1; Figure 3.3B-C).

**Table 3.1.** Main effects of heel height and foot plate stiffness manipulations on mean (SE) SVA, joint flexion-extension angles and internal net moments at midstance (n=10).

SVA	angle [deg]	heel height						footplate stiffness			
		low	med	high	F	p	post hoc	flex	stiff	F	p
ankle	angle [deg]	14.3 (0.81)	17.4 (0.79)	24.3 (1.52)	71.7	<0.001	m <sup>a</sup> ;  h <sup>a</sup> ; m-h <sup>a</sup>	18.6 (1.07)	18.7 (0.92)	0.07	0.805
ankle	angle [deg]	10.5 (1.75)	13.4 (2.31)	16.7 (3.12)	4.86	0.020		14.3 (2.40)	12.7 (2.04)	2.75	0.131
moment	moment [Nm·kg <sup>-1</sup> ]	0.69 (0.07)	0.74 (0.10)	0.91 (0.07)	3.81	0.042		0.70 (0.07)	0.86 (0.08)	6.11	0.035
knee	angle [deg]	17.2 (0.99)	24.5 (1.45)	32.7 (2.05)	49.8	<0.001	m <sup>a</sup> ;  h <sup>a</sup> ; m-h <sup>b</sup>	25.8 (1.58)	23.8 (1.14)	3.74	0.085
knee	moment [Nm·kg <sup>-1</sup> ]	0.20 (0.05)	0.50 (0.08)	0.73 (0.08)	53.2	<0.001	m <sup>b</sup> ;  h <sup>a</sup> ; m-h <sup>b</sup>	0.53 (0.07)	0.43 (0.07)	6.63	0.030
hip	angle [deg]	13.1 (2.57)	16.4 (2.73)	19.1 (2.59)	29.9	<0.001	m <sup>b</sup> ;  h <sup>a</sup> ; m-h <sup>b</sup>	16.6 (2.60)	15.8 (2.61)	2.17	0.175
moment	moment [Nm·kg <sup>-1</sup> ]	-0.01 (0.05)	-0.02 (0.04)	0.02 (0.04)	2.27	0.132		-0.01 (0.05)	0.00 (0.04)	0.00	0.935

Positive values represent joint flexion angles, and plantar flexion, knee extension and hip flexion moment.

l, m and h represent low, medium and high heel height respectively.

<sup>a</sup>p<0.001  
<sup>b</sup>p<0.05

### Ankle and hip joint

Increasing heel height resulted in a significant increase in ankle dorsal flexion angle, hip flexion angle and internal plantar flexion moment at midstance. The internal ankle plantar flexion moment further increased as a result of the stiff footplate, whereas ankle angle, hip angle, and internal hip moment were not affected by footplate stiffness (see Table 3.1). No interaction effects of heel height and footplate stiffness were found for ankle angle ( $F=1.66$ ,  $p=0.218$ ), hip angle ( $F=0.24$ ,  $p=0.790$ ), ankle moment ( $F=0.32$ ,  $p=0.732$ ), and hip moment ( $F=0.05$ ,  $p=0.953$ ).

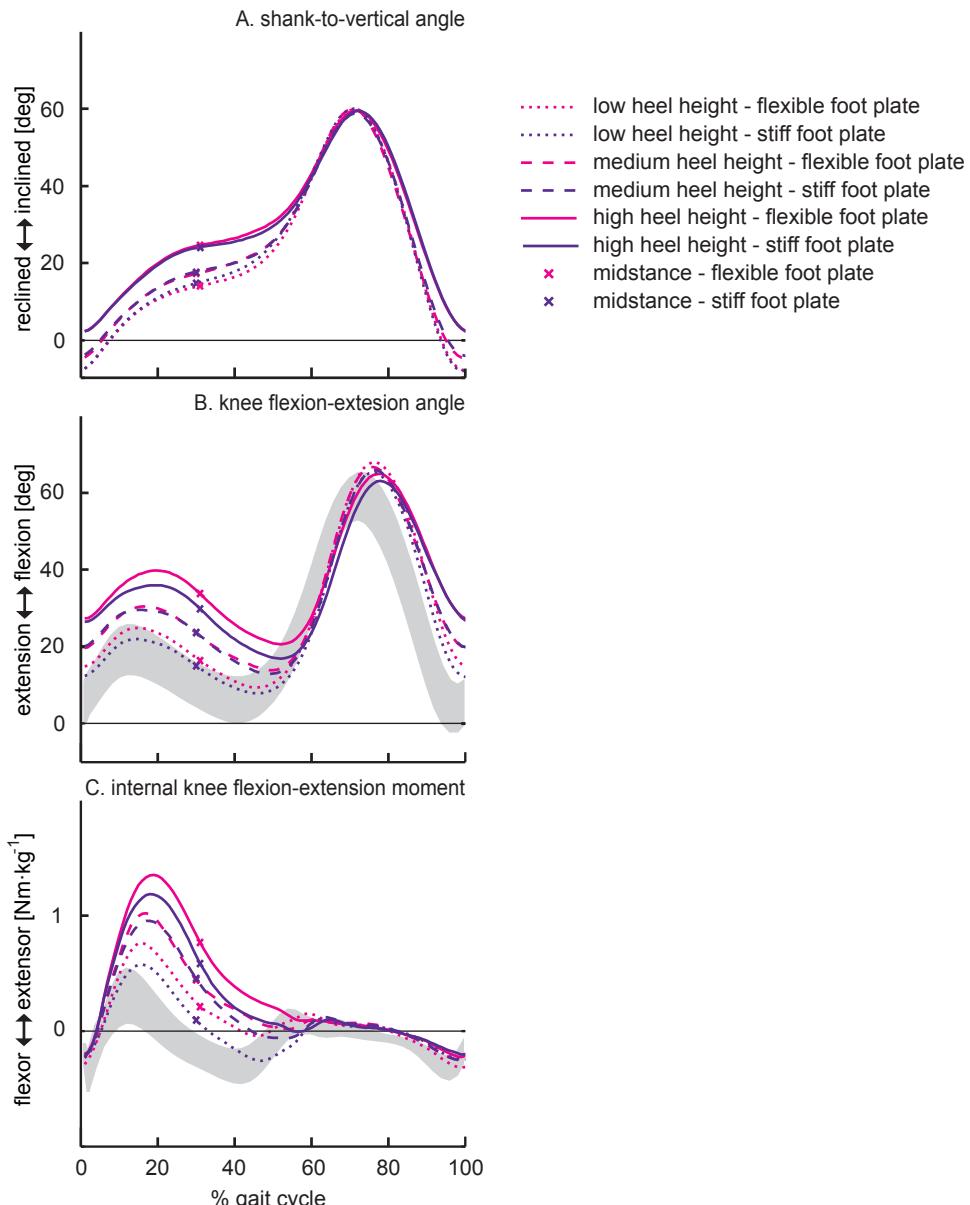
### Shank-to-AFO angle

Mean (SE) Shank-to-AFO angle at midstance significantly decreased with increasing heel height ( $F=46.9$ ,  $p<0.001$ ), with a mean (SE) angle of  $18.3^\circ$  ( $1.23$ ) for the low,  $15.3^\circ$  ( $0.92$ ) for the medium, and  $13.0^\circ$  ( $0.64$ ) for the high heel height condition. Mean (SE) Shank-to-AFO angle was  $15.4^\circ$  ( $0.91$ ) while walking with the flexible footplate, and  $15.6^\circ$  ( $0.92$ ) with the stiff footplate ( $F=0.945$ ,  $p=0.356$ ). No interaction effect of heel height and footplate stiffness was found ( $F=1.14$ ,  $p=0.341$ ).

## DISCUSSION

The present study demonstrates that the SVA is responsive to changes in the AFO-FC heel height, which resulted in an increase in lower limb joint flexion angles and net internal extension moments. In line with our hypothesis, the stiff footplate did not affect the SVA, although it did alter the net internal ankle and knee joint moments. The stiff footplate also affected the knee flexion angle, which is in contrast with our hypothesis.

A recent study of Jagadamma and colleagues<sup>[12]</sup> showed the effects of tuning rigid AFOs on joint kinematics and kinetics in children with spastic cerebral palsy. In that study, tuning was based on inclining the SVA, starting from  $12^\circ$ , until the ground reaction force alignment during stance was closest to normal. They found that increasing the SVA resulted in an increased knee angle and a non-significant increase in peak hip flexion in stance. Another study of Jagadamma et al<sup>[11]</sup> also showed that when the SVA was increased from  $5.6^\circ$  to  $10.8^\circ$  after tuning, the peak knee flexion angle in stance increased. This is comparable to our study, as the SVA increased with increasing heel height, resulting in an increase in knee and hip flexion angles. Our results also show an increase in ankle dorsiflexion angle with increasing heel height, while the rigid AFOs aimed to



**Figure 3.3.** Mean ( $n=10$ ) shank-to-vertical angle, knee flexion-extension angle, and internal knee flexion-extension moment for different conditions, normalized to 100% gait cycle. Shaded area indicates normal walking.

immobilize the ankle in zero degrees. We presume that this was the effect of an offset between the foot markers and the position of the bony landmarks. More specifically, the foot markers (placed on the shoe) were not replaced between the trials, while the insole wedges lifted the foot inside the shoe.

Also comparable to our results, the tuned AFO-FC in the Jadamma study<sup>[12]</sup> resulted in an increased internal peak knee extensor and ankle plantar flexion moment, whereas the peak hip moment remained unchanged. While we did expect to see these changes at the ankle and knee joints, the unchanged internal hip moment between conditions was in contrast to our hypothesis. In our study, subjects may have positioned the thigh such that the ground reaction force was aligned close to the hip joint at midstance, independent from shank kinematics, therewith showing similar internal hip joint moments between heel height conditions at this stage. Although AFO-FC heel height manipulations and the resulting changes in joint angles and moments found in Jagadamma's studies<sup>[11,12]</sup> were smaller compared to our study, the nature of their effects was similar. Hence, our study confirms the responsiveness of the SVA to changes in AFO-FC heel height, though providing a more systematic change of heel height and, additionally, analyzing the effect of adjusting footplate stiffness.

Since literature on the effect of footplate stiffness on joint angles and moments is lacking, our results might best be compared to a study on the effect of different AFO footplate lengths<sup>[20]</sup>. Similar to the non-significant decrease in knee flexion angle as a result from the stiff footplate in our study, Fatone and colleagues<sup>[20]</sup> found a non-significant decrease in knee flexion angle while walking with the full-length footplate. The increase in the internal ankle plantar flexor moment as a result of the stiff footplate is also in agreement with that study<sup>[20]</sup>, and may be explained by the ground reaction force shifting forward early in stance. On the contrary, Fatone's study<sup>[20]</sup> showed a non-significant increase in the internal peak knee extensor moment in early stance while walking with a full-length footplate, compared to the three-quarter footplate. Yet, as they found that subtle changes in sagittal AFO-FC alignment had relatively less effect on the knee moments during stance compared to changes in the length of the footplate, Fatone et al.<sup>[20]</sup> suggested that adjustments in footplate length should be used to control the knee joint moments during stance. The interaction effect of heel height and footplate stiffness on the internal knee extensor moment found in our study, emphasizes the importance of considering footplate characteristics within AFO-FC tuning. In this context, tuning using footplate stiffness characteristics should however preferably be done using the ground reaction force, as the stiff footplate showed no effect on SVA.

A limitation of the study is the calculation of the SVA, which was expressed in the global sagittal plane. Although the SVA was calculated according to methods used in other research and in clinical (2D) settings, it may have introduced a small underestimation of the SVA. Another limitation is poor fitting of the AFOs to some subjects, enabling compensation to the AFO-FC manipulations. This is supported by the results on the Shank-to-AFO angle, which decreased with increasing heel height, indicating that the lower leg was pushed more into the dorsal shell of the AFO when heel height increased. Moreover, the AFO may have been lifted inside the shoe, therewith affecting joint flexion-extension angles and moments.

Our results indicate that the SVA is responsive to AFO-FC heel height manipulations in young healthy adults walking with bilateral rigid AFOs. An increase in SVA was accompanied by increased joint flexion angles and internal net extension moments, especially at the knee joint. Whereas the SVA was not responsive to changes in footplate stiffness, the stiff footplate increased the internal ankle plantar flexion moment, and an interaction effect of heel height and footplate stiffness showed an opposite effect of the stiff footplate on the internal knee extensor moment in the low and high heel height conditions. These findings emphasize the consideration of footplate characteristics in the tuning process. In conclusion, the SVA may serve as a parameter to evaluate AFO-FC tuning, which has to be elaborated on in the clinical target population.

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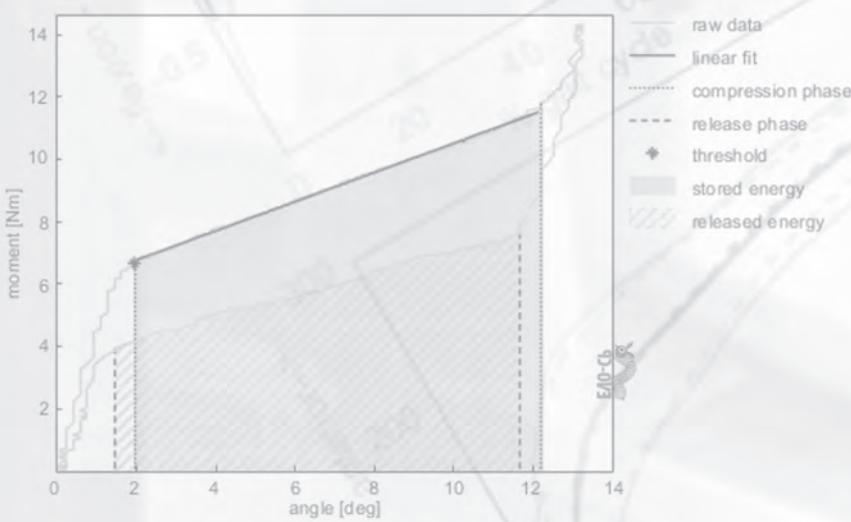
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# Defining the mechanical properties of a spring-hinged ankle foot orthosis to assess its potential use in children with spastic cerebral palsy

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## ABSTRACT

A rigid ventral shell ankle foot orthosis (AFO) may improve gait in children with spastic cerebral palsy (CP) whose gait is characterized by excessive knee flexion in stance. However, these AFOs can also impede ankle range of motion (RoM) and thereby inhibit push-off power. A more spring-like AFO can enhance push-off and may potentially reduce walking energy cost. The recent development of an adjustable spring-hinged AFO now allows adjustment of AFO stiffness, enabling tuning towards optimal gait performance. This study aims to quantify the mechanical properties of this spring-hinged AFO for each of its springs and settings. Using an AFO stiffness tester, two AFO hinges and their accompanying springs were measured. The springs showed a stiffness range of 0.01 to 1.82 Nm·deg<sup>-1</sup>. The moment-threshold increased with increasing stiffness (1.13 to 12.1 Nm), while RoM decreased (4.91 to 16.5 degrees). Energy was returned by all springs (11.5 to 116.3 J). These results suggest that the two stiffest available springs should improve joint kinematics and enhance push-off in children with spastic CP walking with excessive knee flexion.

## INTRODUCTION

Gait in children with spastic cerebral palsy (CP) is often hampered by excessive knee flexion during the stance phase of gait, which may lead to walking limitations in terms of increased walking energy cost and/or a decreased speed<sup>[1,2]</sup>. To counteract excessive knee flexion and improve gait, children with spastic CP are commonly provided with a rigid ventral shell ankle foot orthosis (AFO)<sup>[3]</sup>. This AFO consists of an anterior support to the tibia and a rigid footplate that aims to create a knee extension moment during single limb support by shifting the ground reaction force forward<sup>[4]</sup>.

Although a rigid ventral shell AFO may be effective in counteracting knee flexion, its high stiffness has the disadvantage of limiting the ankle Range of Motion (RoM), thereby inhibiting push-off power<sup>[5,6]</sup> and reducing the possibility to store and release energy. Dependent on the AFO stiffness, it has been shown that a more spring-like AFO can store energy during single stance, which can then be returned in preswing<sup>[7]</sup>. A study in adults with plantar flexor weakness showed that this storage and release of energy is beneficial in terms of reducing walking energy cost<sup>[8]</sup> and that this benefit can be optimised by choosing the correct AFO stiffness<sup>[9]</sup>. As a similar optimisation may also be possible for children with spastic CP, with a decreased walking energy cost potentially yielding improved walking ability<sup>[10,11]</sup>, the effects of different degrees of AFO stiffness on gait performance should be investigated in these children.

Our on-going AFO-CP trial<sup>[12]</sup> includes a spring-hinged AFO with adjustable mechanical properties, that is used to evaluate the effects of different degrees of AFO stiffness on gait performance in children with spastic CP. This evaluation requires that the mechanical characteristics of the AFO are known. However, as no studies are available in the literature, the aim of the present study was to quantify the mechanical properties of the spring-hinged AFO for each of its springs and settings. We hypothesize that the AFO's stiffness range should be sufficient to counteract excessive knee flexion, and the energy returned by the springs should augment push-off power.

## METHODS

### Equipment

In this study, we tested the mechanical properties of the AFO Neuro Swing<sup>®</sup> hinge, developed by Fior & Gentz (Germany). This hinge allows several mechanical properties to be varied within the same orthosis by applying different compression springs. The hinge includes anterior and posterior shafts into which springs can be inserted, and five pre-compressed springs are available per hinge. A cap, consisting of two screws, fixes the spring on each side of the hinge. The offset (i.e. the angle of the hinge in unloaded condition) can be set with the outer screw, while the inner screw preloads the spring, thereby increasing the force threshold and limiting the RoM towards the end of the spring's elastic range.

Two test AFOs were manufactured for this study, each with a different hinge size (14 mm and 16 mm). The AFO's ventral shell was composed of carbon composite and an aluminium footplate was attached to the hinge by an aluminium bar.

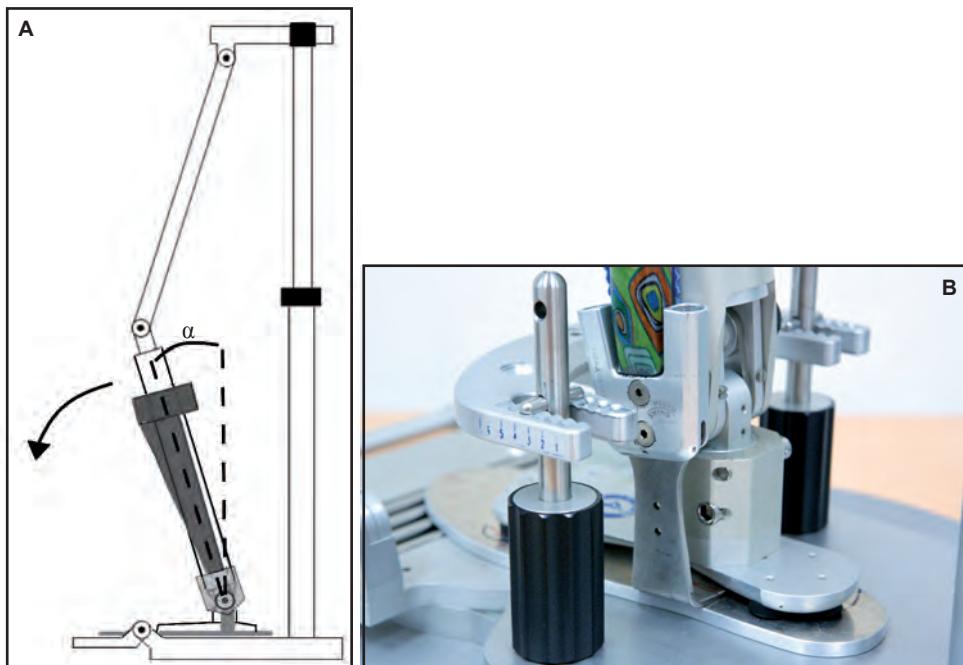
### Measurement protocol

The mechanical properties of both hinges were measured with a recently developed stiffness-testing device, named BRUCE (see Figure 4.1A), which has been shown to provide reliable measurements (ICC = 1.00) of AFO properties<sup>[13]</sup>. The anterior shell of the AFO was attached to BRUCE by a Velcro strap. The hinge's rotation axis was aligned manually with the measuring "ankle" axis of BRUCE (see Figure 4.1B). To avoid misalignment, the AFO was repositioned in BRUCE several times and hysteresis was measured for each position. The position that resulted in the least hysteresis was chosen as the most optimal axes alignment. The offset was set to 0°, while the inner screws of the hinge did not limit the RoM of the springs, enabling evaluation of the springs' maximal capacity.

After fixation and alignment of the test AFO into BRUCE, the dummy leg was manually pushed towards dorsiflexion (see Figure 4.1A) and released towards plantar flexion slowly (i.e. mean angular velocity of 14 deg·s<sup>-1</sup>), three times for each spring. In so doing, the spring was fully compressed and released, while the exerted net moment and ankle angle were continuously measured. Three additional measurements were carried out while the inserted spring (i.e. the 5<sup>th</sup> spring in the 16 mm hinge) was fully pre-compressed, i.e. acting like a rigid configuration with no RoM towards dorsiflexion.

### Data processing and analysis

The data were analysed using custom-made software, based on Matlab 2011 (The Mathworks, USA). The angle-moment relationship was plotted, after which the elastic range of both the compression phase and release phase was chosen manually by the processor. The following mechanical properties were calculated and averaged over the three measurements: i) the springs' elastic range, measured as the hinge's range of motion during the compression and release phase and averaged over both phases, referred to as the RoM [deg], ii) the exerted moment at the start of the RoM, referred to as the threshold [Nm], iii) the stiffness [ $\text{Nm}\cdot\text{deg}^{-1}$ ], which was derived from the slope of the linear fit on the angle-moment relation of the compression phase, iv) the amount of energy [J] that was stored during compression, v) the amount of returned energy [J] while releasing the spring, and vi) the efficiency, expressed as  $E_{\text{RLS}}$  as a percentage of  $E_{\text{STOR}}$  [% $E_{\text{STOR}}$ ]. Only the stiffness was calculated for the rigid configuration, as no other properties are applicable for this setting.

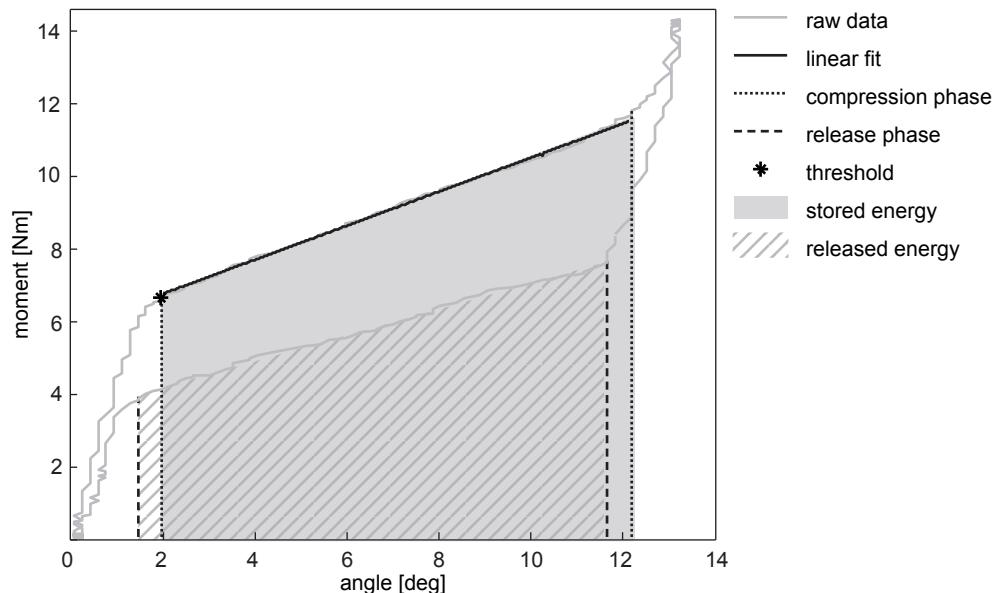


**Figure 4.1.** A) Line drawing of the measurement setup. Sagittal view of the test-AFO placed into BRUCE, with  $\alpha$  indicating the angle of the hinge and the arrow indicating the direction of applied force. B) Detailed picture of the test-AFO placed into BRUCE (sagittal-posterior view), showing the alignment of the rotation axis of the hinge and measuring “ankle” axis of BRUCE.

## RESULTS

The rigid configuration showed a stiffness of  $8.07 \pm 0.63 \text{ Nm}\cdot\text{deg}^{-1}$ . The stiffness of the springs in the 14 mm hinge ranged from  $0.01 \text{ Nm}\cdot\text{deg}^{-1}$  to  $1.16 \text{ Nm}\cdot\text{deg}^{-1}$ , with comparable stiffness values for the 3<sup>rd</sup> and 4<sup>th</sup> spring. For the 16 mm hinge, stiffness ranged from  $0.03 \text{ Nm}\cdot\text{deg}^{-1}$  to  $1.82 \text{ Nm}\cdot\text{deg}^{-1}$ , showing a more gradual increase in stiffness within springs. The threshold increased with increasing stiffness, although it was almost the same for the 3<sup>rd</sup> and 4<sup>th</sup> spring in the 16mm (see Table 4.1).

The hinges' 4<sup>th</sup> spring showed the largest energy return (see Table 4.1 and Figure 4.2). Efficiency was comparable for all springs in the 14 mm hinge, but increased with increasing stiffness in the 16 mm hinge. The springs' RoM decreased with increasing stiffness in both hinges (see Table 4.1).



**Figure 4.2.** Typical result of a recording of the test AFO using the second stiffest spring of the 14mm hinge.

**Table 4.1.** Mechanical properties of the five springs fitting into the 14mm and 16mm hinges (mean (SD) of three repetitions).

spring	stiffness [Nm·deg <sup>-1</sup> ]	threshold [Nm]	RoM [deg]	E <sub>STOR</sub> [J]	efficiency [%E <sub>STOR</sub> ]	E <sub>RLS</sub> [J]
<b>14mm</b>						
1	0.01 (0.00)	1.13 (0.06)	13.8 (0.3)	18.1 (0.2)	63.3 (0.7)	11.5 (0.2)
2	0.25 (0.00)	3.14 (0.15)	13.9 (0.3)	69.9 (1.3)	64.1 (1.6)	44.8 (0.8)
3	0.51 (0.00)	5.44 (0.16)	9.52 (0.37)	75.8 (2.3)	60.8 (0.9)	46.1 (1.5)
4	0.47 (0.00)	6.56 (0.23)	10.3 (0.1)	95.4 (0.9)	65.4 (0.7)	62.3 (0.2)
5	1.16 (0.03)	9.90 (0.13)	4.91 (0.08)	62.9 (2.3)	63.7 (0.4)	40.1 (1.7)
<b>16mm</b>						
1	0.03 (0.01)	1.86 (0.71)	15.7 (0.3)	36.7 (0.8)	53.2 (1.6)	19.5 (0.2)
2	0.23 (0.00)	3.94 (0.09)	16.5 (0.2)	99.0 (1.5)	66.2 (2.4)	65.5 (1.4)
3	0.54 (0.00)	6.82 (0.08)	11.1 (0.1)	110.5 (1.6)	71.3 (2.0)	78.8 (2.6)
4	0.99 (0.01)	6.54 (0.18)	12.4 (0.3)	156.5 (3.5)	74.3 (1.6)	116.3 (1.7)
5	1.82 (0.02)	12.1 (0.6)	7.21 (0.26)	136.5 (7.6)	79.0 (3.7)	107.6 (1.2)

Abbreviations: RoM, range of motion; E<sub>RLS</sub>, released energy; E<sub>STOR</sub>, stored energy.

## DISCUSSION

The aim of this study was to quantify the mechanical properties of a spring-hinged test AFO for each of its springs and settings. Our measurements showed that spring stiffness ranged between 0.01 to 1.82 Nm·deg<sup>-1</sup>, which was considerably lower than the stiffness of the rigid configuration. Additionally and in contrast to the rigid configuration, the spring-hinged AFO allowed energy storage of 18.1 J to 156.5 J that returned as 11.5 J to 116.3 J. This energy return may be beneficial in terms of reducing walking energy cost. On the other hand, the lower stiffness might counteract excessive knee flexion less effectively compared to the rigid configuration.

Current literature on AFOs in children with spastic CP rarely includes clear and unambiguous mechanical descriptions of the orthosis<sup>[14]</sup>. However, Bregman and colleagues<sup>[15]</sup> tested the mechanical properties of AFOs in adults with plantar flexor weakness and reported stiffness ranging from 0.5 to 5.4 Nm·deg<sup>-1</sup>. Although the springs used in our study showed much lower stiffness values, they also exhibited a threshold force before the spring entered its elastic range. This threshold may prevent ankle dorsal flexion at low ankle moment values (up to approximately 0.5 Nm·kg<sup>-1</sup>, depending on the child's weight), hence supporting knee extension at the beginning of the stance phase. As the ankle moment increases in midstance, it will compress the spring through its elastic range until the dorsiflexion stop is hit. This stop will prevent excessive ankle dorsal flexion in late stance, thereby contributing to a normalization of knee kinematics<sup>[3]</sup>, which has been shown to reduce walking energy cost in children with spastic CP<sup>[10]</sup>. As the RoM of the stiffest spring is the most limited, this is expected to be the most effective in normalizing ankle and knee kinematics, although at the expense of potential energy return.

The area beneath the curve, derived from the relation between RoM and the exerted net moment, represents the stored energy within the spring. If RoMs were similar for all springs, the most energy would be stored by the stiffest spring. However, the restricted RoM of the stiffest spring of either hinge, required for normalization of joint kinematics, also limited its energy storage. Therefore, the maximal potential energy return was determined for the second stiffest spring, which was 62.3 J for the 14 mm and 116.3 J for 16 mm hinge. As these values are comparable to those of Bregman and colleagues, in which an AFO energy return of approximately 70 J resulted in the greatest walking energy cost reduction, our measured values suggest potential for reduced walking energy cost in children with spastic CP<sup>[9]</sup>.

One limitation of our study was that the alignment of the hinges' rotation axis and BRUCE's measuring axis was done by eye. This may have resulted in a slight misalignment, leading to dry friction between the AFO and the device and resulting in hysteresis that is not attributable to the hinge<sup>[13]</sup>. However, this potential effect was compensated for by repositioning the AFO in BRUCE several times and measuring hysteresis for each position. Secondly, in the recordings we did not allow for the different angular velocities to which the hinge will be subjected during gait. However, other studies measuring AFOs at different speeds do not show a substantial influence of angular velocity<sup>[13]</sup>.

In conclusion, our evaluation of the mechanical properties of the spring-hinged AFO indicates that the two stiffest available springs should be adequate for use in children with spastic CP walking with excessive knee flexion. While the energy return of the second stiffest spring may best make the most contribution to enhanced push-off power, the stiffest spring is expected to normalize joint kinematics most effectively. The spring-hinged AFO should now be evaluated in clinical practice for potential to contribute to improved gait performance in children with spastic CP.

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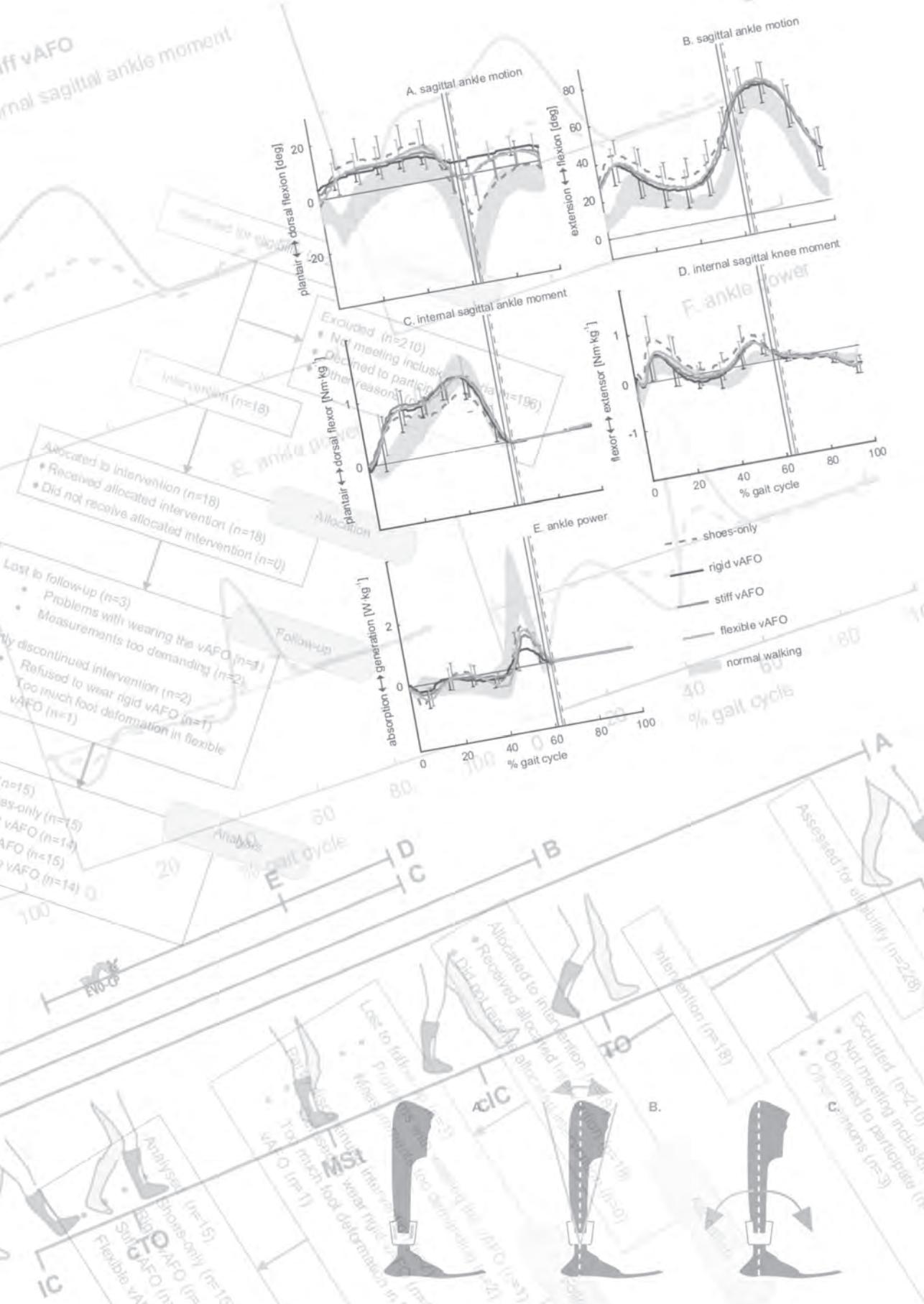
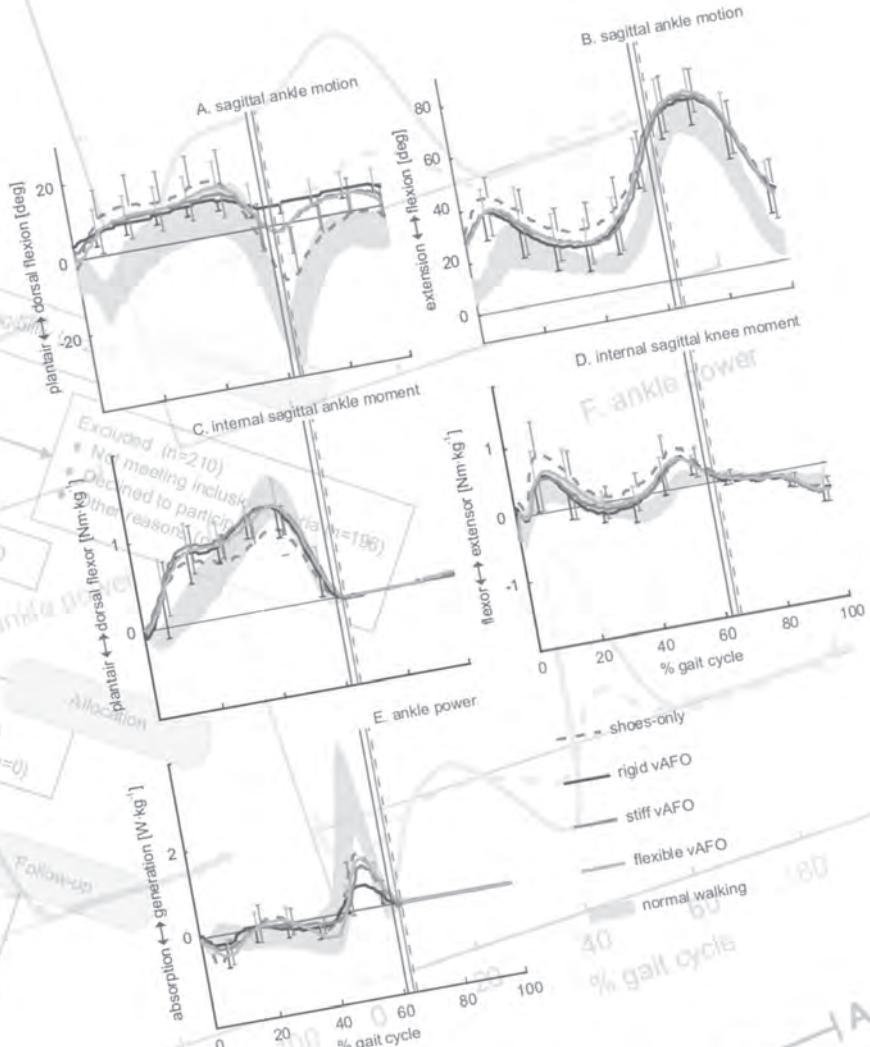
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# The effects of varying ankle foot orthosis stiffness on gait in children with spastic cerebral palsy who walk with excessive knee flexion

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## ABSTRACT

Rigid ankle foot orthoses (AFOs) are commonly prescribed to counteract excessive knee flexion during the stance phase of gait in children with cerebral palsy (CP). While rigid AFOs may normalize knee kinematics and kinetics effectively, it has the disadvantage of impeding push-off power. A spring-like AFO may enhance push-off power, which may come at the cost of reducing the knee flexion less effectively. Optimizing this trade-off between enhancing push-off power and normalizing knee flexion in stance is expected to maximize gait efficiency. This study investigated the effects of varying AFO stiffness on gait biomechanics and efficiency in children with CP who walk with excessive knee flexion in stance. Fifteen children with spastic CP (11 boys, mean (SD) 10 (2) years) were prescribed with a ventral shell spring-hinged AFO. The hinge was set into a rigid, or spring-like setting, using both a stiff and flexible performance. At baseline (i.e. shoes-only) and for each AFO, a 3D-gait analysis and 6-minute walk test with breath-gas analysis were performed at comfortable speed. Lower limb joint kinematics and kinetics were calculated. From the 6-minute walk test, walking speed and the net energy cost were determined. A generalized estimation equation ( $p<0.05$ ) was used to analyze the effects of different conditions. Compared to shoes-only, all AFOs improved the knee angle and net moment similarly. Ankle power generation and work were preserved only by the spring-like AFOs. All AFOs decreased the net energy cost compared to shoes-only, but no differences were found between AFOs, showing that the effects of spring-like AFOs to promote push-off power did not lead to greater reductions in walking energy cost. These findings suggest that, in this specific group of children with spastic CP, the AFO stiffness that maximizes gait efficiency is primarily determined by its effect on knee kinematics and kinetics rather than by its effect on push-off power.

## INTRODUCTION

Gait in children with Cerebral Palsy (CP) is often characterized by abnormal gait biomechanics, such as excessive knee flexion during stance. Associated with such gait deviations, an elevated walking energy cost is often observed<sup>[1-3]</sup>, which may contribute to activity limitations<sup>[4,5]</sup>. To treat these gait-related problems in CP, ankle foot orthoses (AFOs) are commonly prescribed.

When prescribing an AFO, the specific gait deviations and functional deficits of the patient should be clearly identified, such that these can be optimally addressed by the design and mechanical properties of the AFO<sup>[6]</sup>. A rigid ventral shell AFO is typically used for children who walk with excessive knee flexion in stance<sup>[6]</sup>; a gait pattern that is particularly energy consuming<sup>[3,7,8]</sup>. Mechanically, a ventral shell AFO aims to shift the ground reaction force more anterior relative to the knee, which reduces the external flexion moment. This is expected to reduce knee flexion and decrease the elevated internal knee extensor moment during stance<sup>[6]</sup>. Accordingly, this may reduce walking energy cost<sup>[9,10]</sup>.

Although a ventral shell AFO may be effective in reducing knee flexion and subsequent walking energy cost, its high stiffness has the disadvantage of impeding ankle range of motion. Ankle range of motion during gait has been shown to be a key kinematic factor in gait efficiency<sup>[11,12]</sup>. In fact, a reduced ankle range of motion during gait, especially towards plantar flexion, limits push-off power about the ankle, which almost always leads to an increased walking energy cost<sup>[11,13]</sup>. Besides, a common strategy to compensate for reduced push-off power is to deliver work around the hip<sup>[14-17]</sup>, which may also increase walking energy cost<sup>[14,18]</sup>.

The metabolic penalty of limiting the ankle push-off power may be reduced by applying spring-like AFOs. These AFOs allow dorsiflexion in the beginning of stance phase, thereby storing energy within the AFO. This energy can be returned in pre-swing, which may support push-off power, therewith enhancing gait efficiency in terms of walking energy cost<sup>[19,20]</sup>. Considering the key role of ankle range of motion during gait, an AFO that would additionally allow plantar flexion in late stance might support push-off power and gait efficiency even further<sup>[21,22]</sup>.

The efficacy of spring-like AFOs to improve gait is however partly dependent on their stiffness. This has been shown in simulation models<sup>[19]</sup>, as well as in studies in healthy adults<sup>[21]</sup> and in adult patient populations<sup>[23-28]</sup>, where results indicated that changing the AFO stiffness significantly affected knee and ankle kinematics and kinetics, as well as walking energy cost. Results also indicated that the reduction in walking energy cost

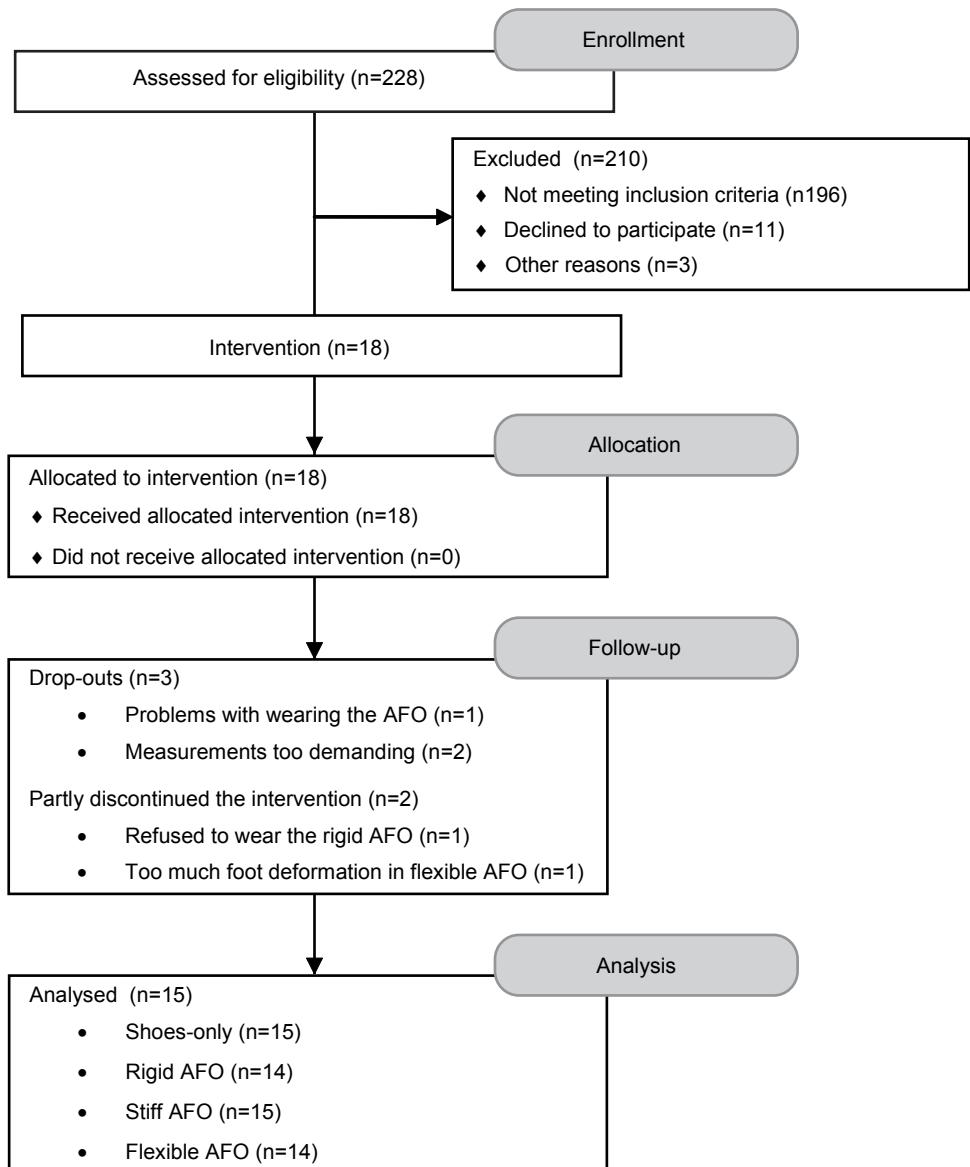
could be improved by choosing the appropriate AFO stiffness<sup>[27]</sup>. Such stiffness-based maximization of gait efficiency may also apply to children with CP<sup>[29]</sup>, which is relevant considering that AFOs are not always effective in terms of reducing walking energy cost in these children<sup>[9,10,30]</sup>, while this is an important goal of AFO prescription<sup>[31]</sup>. However, the effects of different degrees of AFO stiffness on gait biomechanics and walking energy cost have not previously been reported in this patient group.

The aim of this study was to investigate the effects of varying AFO stiffness on lower limb joint kinematics and kinetics and walking energy cost in children with spastic CP whose gait pattern is characterized by excessive knee flexion in stance. Stiffer AFOs were expected to normalize knee flexion most effectively, though at the expense of obstructing ankle range of motion and push-off power. Contrarily, the less stiff AFOs were expected to enhance push-off power, but to be less effective in counteracting knee flexion. We hypothesized that the optimal AFO stiffness (i.e. at which walking energy cost would be lowest), would be defined by a trade-off between improving knee kinematics and kinetics, and enhancing ankle push-off power.

## METHODS

### Participants

Data used in the study were collected in the context of the AFO-CP trial<sup>[32]</sup>. Participants in the AFO-CP trial were recruited from the rehabilitation department of a university hospital in the Netherlands and its affiliated rehabilitation centers (see Figure 5.1). Children diagnosed with spastic CP and aged between 6 and 14 years old were included. Other inclusion criteria were a Gross Motor Function Classification System (GMFCS)<sup>[33]</sup> level I, II or III, and a barefoot gait pattern that was characterized by excessive knee flexion in (mid)stance (i.e. more than 10° in midstance). Children with ankle plantar flexion contractures, knee flexion contractures and/or hip flexion contractures of more than 10° were excluded. Institutional review board approvals were obtained prior to the start of the study, and all participants (above 12 years old) and their parents provided written informed consent. All measurements were performed in accordance to the Declaration of Helsinki.

**Figure 5.1.** Trial flow diagram.

Abbreviations: AFO, ankle foot orthosis.

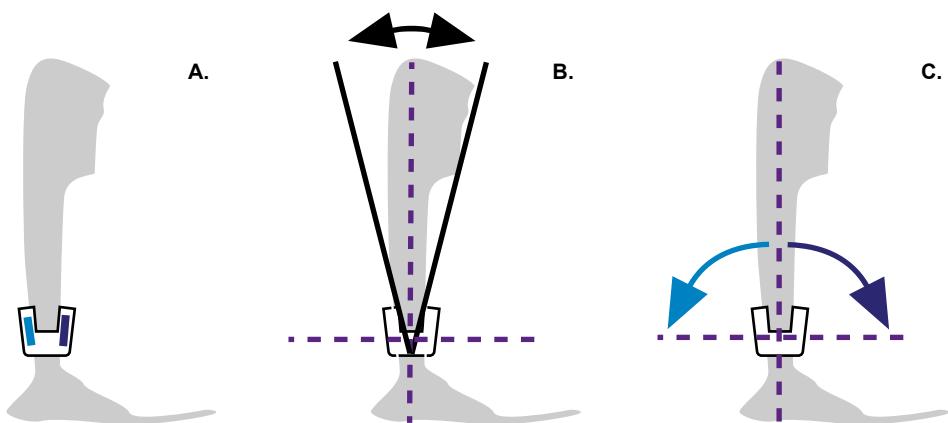
### Materials

Participants were prescribed with a new ventral shell AFO, which was designed with a ventral shell and rigid footplate. The AFOs were made of pre-preg carbon fiber, and manufactured with an integrated ankle hinge (Neuro Swing®, Fior & Gentz, Lüneburg, Germany). This hinge allows mechanical characteristics (e.g. stiffness) to be varied within the same orthosis, as it holds a shaft for dorsal and plantar flexion in which pre-compressed springs with different mechanical properties can be inserted<sup>[29]</sup> (see Figure 5.2). The hinge is available in different sizes, each accompanied with a spring package covering a range of stiffness degrees. The size of the hinge (14mm or 16mm) was individually determined following a standard prescription protocol (Fior & Gentz, Lüneburg, Germany), which is based on weight and height of the child. For this study, the hinge was set into three stiffness configurations: i) rigid, ii) stiff and iii) flexible. For the rigid configuration, the hinge's spring-like properties were eliminated, aiming to act like a conventional rigid ventral shell AFO with limited range of motion. For the stiff and flexible configurations, stiffness towards dorsiflexion was varied by applying the stiffest available spring and one less stiff spring in the hinge's ventral shaft. These springs were expected to sufficiently improve gait in children with spastic CP who walk with excessive knee flexion<sup>[29]</sup>. The most compliant spring available was used towards plantar flexion for both configurations.

The AFOs were worn in combination with the children's own shoes (i.e. shoes with flat, flexible soles), referred to as the AFO-footwear combination. For shoes-only measurements, children were instructed to wear shoes that they normally used when walking without AFOs.

### Procedure

At the start of each measurement session (i.e. AFO stiffness evaluation), measures of bare foot height [m] and weight [kg] were determined using an electronic scale (DGI 250D, KERN DE v. 3.3 10/2004, Kern & Sohn GmbH, Balingen, Germany). Leg length (from the trochanter head to the lateral malleolus) was measured while the participant was standing upright, with knees extended as much as possible. Stiffness of the new AFO was randomly (i.e. block-randomized) set into one of the three configurations. After setting the hinge, the AFO-footwear combination was tuned following a common clinical protocol<sup>[34]</sup>.



**Figure 5.2.** Picture of the spring-hinged ventral shell ankle foot orthosis, including possible adjustments using the hinge. The hinge allows: A, the stiffness to be varied towards dorsal flexion and plantar flexion; B, adjustment of the alignment of the ventral shell with respect to the foot; C, the range of motion to be varied, although this is also dependent of the spring inserted (stiffer springs allow less range of motion).

Figures adapted from Fior & Gentz.

Each AFO stiffness configuration was worn for an acclimatization period of four weeks, after which efficacy of that AFO was evaluated. This evaluation consisted of a 3D gait analysis to measure gait biomechanics and a 6-minute walk test to measure walking energy cost. Additionally, the mechanical properties of the AFO<sup>[35]</sup> and various shoe parameters were assessed<sup>[36]</sup>. Next, the hinge was set into the second stiffness configuration and the procedure was repeated until all three stiffness configurations were evaluated. For the shoes-only (i.e. baseline) condition, the same set of measurements was performed, and these were conducted during the assessment of the second AFO stiffness configuration.

### **Measurements**

#### ***Gait biomechanics***

Gait analyses were performed in our gait laboratory. Participants were instructed to walk up and down a 10m-walkway with integrated force plate (OR6-5-1000, AMTI, Watertown, USA) at a comfortable walking speed. Kinematic data were collected using an optoelectronic motion capture system (OptoTrak 3020, Northern Digital, Waterloo, Canada). Technical clusters of three markers were rigidly attached to the trunk, pelvis, thighs, shanks (including the AFOs' ventral shell) and feet, and anatomically calibrated by probing 32 bony landmarks<sup>[37]</sup>. The bony markers of the foot (i.e. calcaneus and metatarsal joints I and IV) were probed on the shoe, and horizontally aligned in the sagittal plane of the foot while the foot was flat on the ground. The foot segments included the AFO's foot part and shoe, where no movement between these components was assumed. Segment movements were tracked (sample frequency: 100 Hz) and synchronized with force plate data (sample frequency: 1000 Hz). Data collection trials were repeated until three strides with correct foot placement (i.e. within the borders of the force plate) of the most affected leg were recorded.

#### ***Walking energy cost***

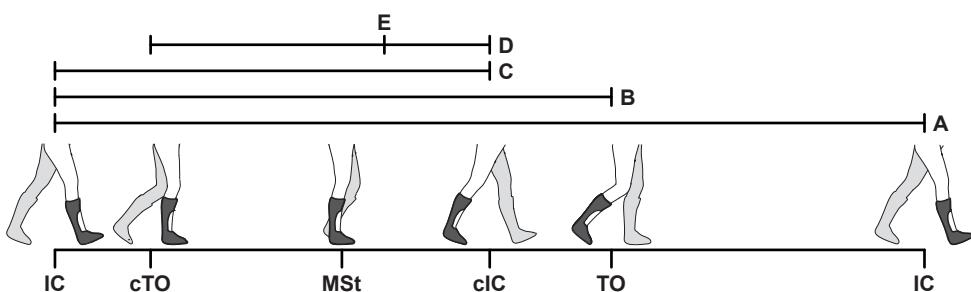
A portable breath gas-analysis system (Metamax 3B, Cortex Biophysik, Leipzig, Germany) was used to record breath-by-breath oxygen uptake (VO<sub>2</sub>) and carbon dioxide production (VCO<sub>2</sub>) values. Each measurement started with a rest test. Participants were seated, while the equipment was put on and the facemask was fitted. Then participants sat down quietly watching a movie for six minutes. They were instructed not to talk or laugh during the measurement. After completion of the rest test, participants performed

a 6-minute walk test at comfortable walking speed on a 40m indoor oval track, which has been shown to be a sufficiently sensitive and reliable protocol for energy cost measurements in children with CP<sup>[1,38]</sup>.

### AFO properties and shoe parameters

The AFO's mechanical properties were measured using the Bi-articular Reciprocal Universal Compliance Estimator (BRUCE)<sup>[35]</sup>, which is an instrument to measure AFO mechanical properties. Each AFO was placed into the BRUCE, such that the rotation axis of the hinge was aligned with the "ankle" axis of the BRUCE. The ventral shaft of the AFO was fixated to the BRUCE by a Velcro strap. The AFO was then manually pushed towards dorsiflexion and plantar flexion, while the exerted moment and ankle angle were continuously recorded. For each movement direction, measurements were repeated three times<sup>[29]</sup>.

Shoe parameters (i.e. the height of the shoe sole and the heel-sole differential angle) were obtained with the Vertical Inclinometer on a Rail (VICTOR)<sup>[36]</sup>, a dedicated instrument to define these parameters.



**Figure 5.3.** Representation of relevant phases of the gait cycle. Phases of the gait cycle were defined as i) stance: initial contact to toe-off; ii) step: initial contact to contralateral initial contact; iii) single support (SS): contralateral toe-off to contralateral initial contact. Definitions of specific gait events and mean timing [%gait cycle]: i) contralateral toe-off (cTO) [11%]; ii) midstance (MSt): the moment that the malleolus marker of the contralateral leg passed the malleolus marker of the ipsilateral leg [33%]; iii) contralateral initial contact (cIC) [50%]; iv) toe-off (TO) [64%]; v) timing of minimal knee flexion angle during single support (peak knee extension angle) (TKEpk): [38%].

Abbreviations: cTO, contralateral toe-off; cIC, contralateral initial contact; IC, initial contact; TKEpk, timing of peak knee extension angle; MSt, midstance; SS, single support; TO, toe-off.

### Data processing

#### Gait biomechanics

Optoelectronic marker data and force plate data of the three recorded trials were analyzed using custom-made software (BodyMech, [www.bodymech.nl](http://www.bodymech.nl)) based on MATLAB version R2011a (The Mathworks, Natick, USA). For each trial, initial contact and toe-off in the gait cycle of the ipsilateral leg were determined using force-plate data, while foot angular velocity was used to determine the gait events of the contralateral leg<sup>[39]</sup>. Based on these gait events, relevant phases of the gait cycle were determined (see Figure 5.3). Furthermore, walking speed [ $\text{m}\cdot\text{s}^{-1}$ ] was determined and averaged over three trials.

3D lower limb joint flexion-extension angles [deg] were calculated from the anatomically calibrated cluster marker data, according to ISB anatomical frames<sup>[37,40]</sup>. As bony landmarks of the foot were probed on the shoe, a coordinate frame of the shoe (including the AFO and foot) was calculated. Shoe parameters (i.e. height of the shoe sole and the heel-sole differential angle) obtained using VICTOR<sup>[36]</sup>, were used to correct the measured ankle angle for the offset between probing positions and the actual position of the bony landmarks, and for orientation of the foot in the shoe, which is dependent on the heel sole differential. Lower limb net joint flexion-extension moments [ $\text{Nm}\cdot\text{kg}^{-1}$ ] were calculated with force plate data using inverse dynamics<sup>[41]</sup>, expressed with respect to the proximal segment frame<sup>[42]</sup>, and normalized to body weight. Also lower limb joint powers [ $\text{W}\cdot\text{kg}^{-1}$ ] and work [ $\text{J}\cdot\text{kg}^{-1}$ ] (i.e. integral of net ankle power) were calculated.

From the mean joint angles, moments and powers as a function of the gait cycle, we determined specific relevant parameters, primarily at the knee and ankle joints. At the knee joint, these included the knee flexion-extension angle and moment at midstance, peak knee extension angle during single support (KEpk) and the knee moment at timing of KEpk. Ankle joint parameters included range of motion (RoM) during the stride and peak power generation during push-off, where push-off was defined as the period in late stance and pre-swing in which the net ankle power was positive. Positive and negative work over the gait cycle, as well as the net work (i.e. positive + negative) during push-off were determined for all lower limb joints.

### Walking energy cost

Breath-by-breath  $\text{VO}_2$  and  $\text{VCO}_2$  values in minute three to six of both the rest and the walk test were used to calculate the mean steady-state energy consumption values (ECSrest and ECSwalk) [ $\text{J}\cdot\text{kg}^{-1}\cdot\text{m}^{-1}$ ]<sup>[43]</sup>. The mean walking speed [ $\text{m}\cdot\text{min}^{-1}$ ] was measured over the same time frame of the walk test. From these assessments, the net energy cost (EC) [ $\text{J}\cdot\text{kg}^{-1}\cdot\text{m}^{-1}$ ] was calculated as  $(\text{ECSwalk} - \text{ECSrest}) / \text{walking speed}$ . To control for the influence of different body dimensions of children, net EC values were normalized according to the scheme by Schwartz et al.<sup>[44]</sup> to calculate the net non-dimensional energy cost, which was expressed as a percentage of speed-matched control cost (SMC-EC)<sup>[45]</sup>.

### AFO mechanical properties

Angle-moment relation curves resulting from the BRUCE assessments were analyzed using custom-made software based on Matlab version R2011a (The Mathworks, Natick, USA). First, the AFO's neutral angle was determined, which is the angle of the AFO when no force is exerted. Subsequently, we determined stiffness [ $\text{Nm}\cdot\text{deg}^{-1}$ ], range of motion [deg] (i.e. the spring's elastic range), and the threshold [Nm] (i.e. the exerted moment at the start of the spring's elastic range)<sup>[29]</sup>. For the rigid configuration, only the stiffness was determined, as other variables were not applicable.

To calculate the AFO's contributions to the ankle work, first the contributions to the net ankle moment were determined. For the rigid AFO, stiffness was multiplied by the AFO's deflection angle (i.e. the migration of the AFO from its neutral angle) for each point in time. To align these deflection angles during gait to the angles as measured using BRUCE, it was assumed that the AFO had negligible displacement during sway<sup>[20]</sup>. Considering the low stiffness towards plantar flexion of the stiff and flexible AFO, and thus the possibility of plantar flexion movement, this assumption could not be preserved. As such, the alignment was done using the angle of the AFO-footwear combination when exceeding the spring's threshold during gait. Using the moments exerted by the AFO and the ankle's angular velocity for each point in time, contributions to net ankle power and work [ $\text{J}\cdot\text{kg}^{-1}$ ] over the gait cycle and during push-off could be calculated.

**Table 5.1.** Mean (SD) baseline participant characteristics (n=15).

age	[yrs]	10 (2)
weight <sup>a</sup>	[kg]	37.2 (9.0)
height <sup>a</sup>	[cm]	141 (9.0)
sex	[boy/girl]	11/4
GMFCS	[I/II/III]	2/11/2
selective motor control <sup>b</sup>	[good/moderate/poor]	11/3/1
AFO use	[unilateral/bilateral]	1/14

<sup>a</sup>Weight and height were assessed at the start of each measurement moment, but presented here as average values at baseline (i.e. first measurement occasion).

<sup>b</sup>Selective motor control of both legs was assessed using the modified Trost test, which measures the ability to dorsiflex the ankle and extend the knee in an isolated movement<sup>[52]</sup>. Ankle dorsiflexion and knee extension of each leg were scored as 0 (no selective, only synergistic movement), 1 (diminished selective movement) or 2 (full selective movement) and summed to a total score of 0 to 8. These total scores were categorized into poor (total score of 0 to 2), moderate (total score of 3 to 5) or good (total score of 6 to 8) selective motor control<sup>[53]</sup>.

Abbreviations: AFO, ankle foot orthosis; GMFCS, Gross Motor Function Classification System.

## Statistics

Statistical analyses were done with SPSS version 20 (SPSS Inc, Chicago, USA), using an alpha level of 0.05 for all tests of significance. Descriptive statistics (means and standard deviations (SD)) were used to summarize socio-demographic characteristics, disease characteristics, gait-related outcomes, and AFO mechanical properties. Differences in gait-related outcomes between conditions were analyzed with generalized estimating equation analyses, with conditions (i.e. shoes-only, rigid AFO, stiff AFO and flexible AFO) as within-subject factor. Exchangeable correlation structures were assumed. Walking speed, as measured during the gait analyses, was added to the model as covariate<sup>[12]</sup>.

**Table 5.2.** Baseline passive range of motion and spasticity values of relevant joints and muscles of the most affected leg (n=15).

angle of interest <sup>a</sup>		muscles	RoM Median [min max]	spasticity scale <sup>b</sup> [0/1/2/3]
hip extension	+ = extension		10 [0 20]	n/a
knee extension	+ = extension		0 [-10 0]	n/a
popliteal angle		Hamstrings	55 [45 70]	[10/1/3/0]
ankle dorsiflexion (flexed knee)	+ = dorsal flexion	Soleus	10 [0 25]	[13/1/0/1]
ankle dorsiflexion (extended knee)	+ = dorsal flexion	Gastrocnemius	0 [-10 10]	[13/1/0/1]

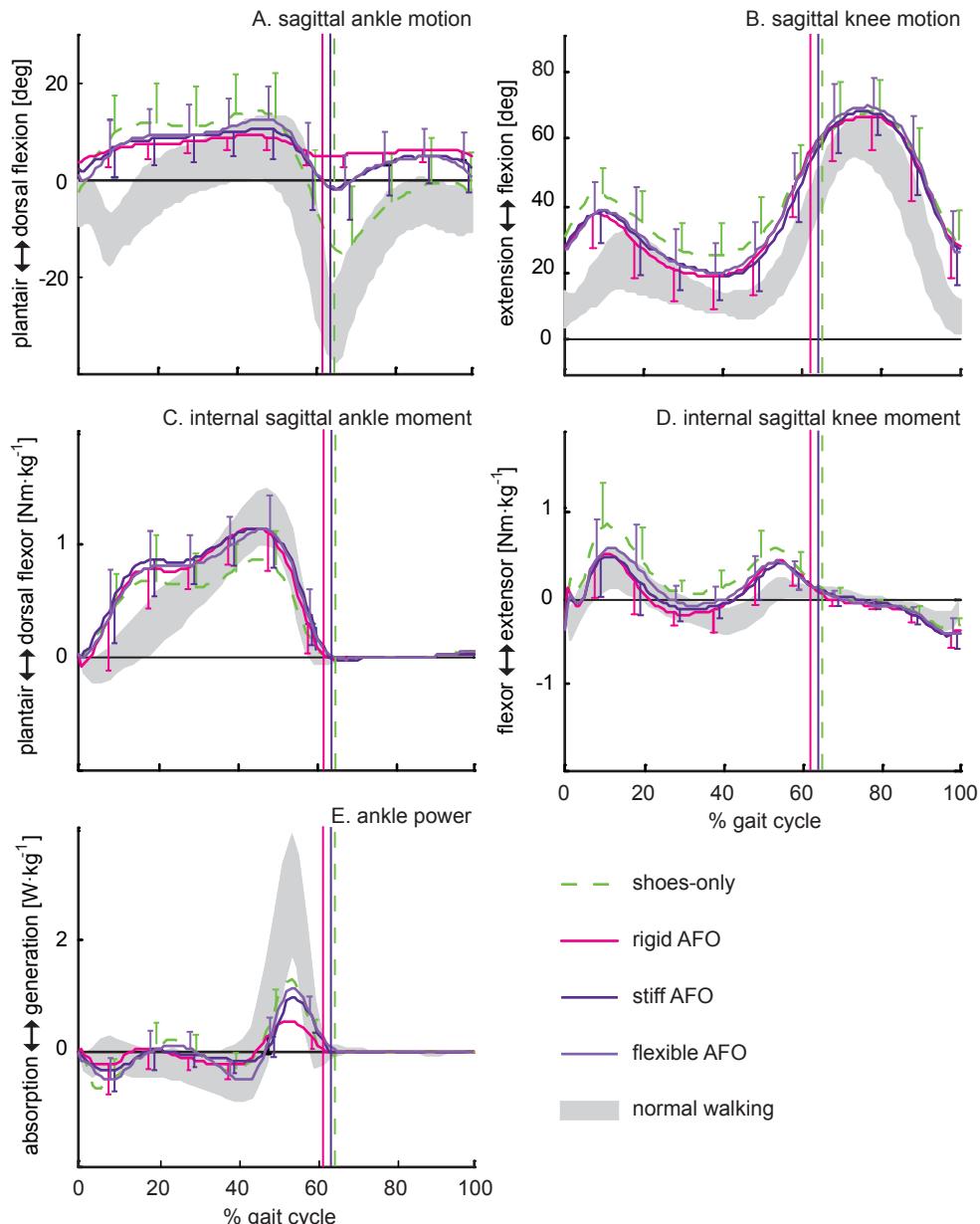
<sup>a</sup>Hip extension was measured with the patient in prone position. All other measurements were performed with the patient in supine position. Comprehensive descriptions of positions and movements are described elsewhere<sup>[54,55]</sup>. The popliteal angle was missing in one patient.

<sup>b</sup>Spasticity was tested according to the Spasticity Test protocol<sup>[55]</sup>, using a 4-point spasticity scale: 0, normal or increased muscle resistance over the whole range of motion; 1, increase in muscle resistance somewhere in the range of motion; 2, catch and release; 3, catch blocking further movement<sup>[54,55]</sup>.

Abbreviations: RoM, range of motion; min, minimum; max, maximum; n/a, not applicable.

## RESULTS

Fifteen children with spastic CP (11 boys, 4 girls) were included in the study. Social-demographic and disease characteristics of these children are presented in Table 5.1. Data from the physical examination are presented in Table 5.2. In 13 children, the effects of all three AFO configurations were evaluated. In one child, only the flexible and stiff AFOs were evaluated because this child refused to wear the rigid AFO. Another child could not acclimatize to the flexible AFO, because of too much foot deformation within the AFO leading to pressure marks, and therefore only the stiff and rigid AFOs were evaluated.



**Figure 5.4.** Mean (n=15) of the most relevant gait parameters as a function of the gait cycle. Vertical lines indicate timing of toe-off (with similar timing for stiff and flexible AFOs).

Abbreviations: AFO, ankle foot orthosis.

## Gait biomechanics

During the gait analyses, the mean (SD) walking speed while walking with shoes-only was  $1.09 (0.21) \text{ m}\cdot\text{s}^{-1}$ . Speed was significantly lower while walking with AFOs, i.e.  $1.07 (0.24)$ ,  $1.00 (0.21)$ , and  $1.05 (0.17) \text{ m}\cdot\text{s}^{-1}$  for the rigid, stiff and flexible configuration respectively (Wald  $\chi^2=10.3$ ,  $p=0.016$ ).

Differences in knee joint angles and between walking with shoes-only and walking with the AFO were comparable for all AFOs. All AFOs decreased the knee flexion angle at contralateral toe-off, midstance, and at timing of KEpk. Also the internal knee flexion-extension moment at midstance and at timing of KEpk were significantly improved by all AFOs (see Table 5.3 and Figure 5.4C-D).

At the ankle joint, we found that ankle RoM was significantly reduced by all AFOs compared to walking with shoes-only, though ankle RoM was significantly less reduced by the stiff and flexible AFO. Peak ankle power generation was reduced by the rigid AFO, while it was preserved by the stiff and flexible AFO (see Table 5.3 and Figure 5.4A,C,D). Ankle work was reduced most by the rigid AFO. The AFOs' contribution to the ankle work over the gait cycle was smaller for the rigid AFO, compared to the spring-like AFOs, while no significant differences were found in the AFOs' contributions to ankle work during push-off between AFOs (see Table 5.4 and Figure 5.5).

## Walking energy cost

Walking speed during the 6-minute walk test was comparable between all conditions. Compared to walking with shoes-only, the net EC was significantly reduced with 9.8%, 11.5%, and 8.2% by the rigid, stiff and flexible AFO respectively. No significant differences were found between AFOs (see Table 5.5). On average, the overall (i.e. all AFOs) reduction in net EC was  $0.67 \text{ J}\cdot\text{kg}^{-1}\cdot\text{m}^{-1}$  (11%), with large individual differences. While some participants showed an improvement, i.e. reduction, in net EC with at least one of the AFOs, others showed no response or even an increase of their energy cost while walking with the AFO (see Figure 5.6). When comparing the SMC-EC, only a significant reduction was found for the rigid and stiff AFO compared to walking shoes-only (see Table 5.5).

**Table 5.3.** Results of generalized estimating equation analyses for relevant gait parameters (mean (SD)), with positive values representing flexion angles, internal extension moments, and power generation.

			condition				statistics			
			shoes (n=15)	rigid (n=14)	stiff (n=15)	flexible (n=14)				
hip	angle	cIC	[deg]	19.9 (12.7)	17.3 (9.7)	16.8 (12.2)	16.4 (14.7)	4.41	0.220	b-f♦
moment	cIC	[Nm·kg <sup>-1</sup> ]		-0.62 (0.33)	-0.62 (0.27)	-0.65 (0.43)	-0.62 (0.34)	1.02	0.795	
knee	angle	cTO	[deg]	41.7 (9.4)	36.3 (11.1)	37.6 (10.3)	38.2 (10.8)	9.39	0.025	b-r♦; b-s♦
angle	MSt	[deg]		34.8 (13.4)	31.8 (8.6)	30.5 (11.0)	29.7 (14.6)	5.37	0.147	b-f♦
angle	SS	[deg]		22.7 (8.7)	16.7 (10.0)	18.1 (8.6)	18.4 (9.3)	31.7	<0.001	b-r§; b-s§;
angle	(KEpk)	[deg]		0.08 (0.15)	-0.15 (0.17)	-0.12 (0.15)	-0.07 (0.16)		b-f♦	
moment	MSt	[Nm·kg <sup>-1</sup> ]		0.02 (0.18)	-0.21 (0.23)	-0.13 (0.18)	-0.09 (0.18)	38.1	<0.001	b-r§; b-s§;
moment	TKEpk	[Nm·kg <sup>-1</sup> ]						24.6	<0.001	b-r§; b-s♦;
ankle	angle	IC	[deg]	-2.6 (7.6)	3.7 (2.2)	2.3 (5.9)	1.0 (6.1)	14.1	0.003	b-r§; b-s♦;
angle	MSt	[deg]		11.4 (8.4)	7.9 (2.6)	9.1 (5.1)	9.4 (6.1)	3.24	0.356	b-f♦
RoM	Stride	[deg]		35.4 (8.1)	7.0 (2.4)	15.4 (4.3)	19.5 (3.9)	267	<0.001	b-r§; b-s§;
										b-f§; r-s§;

moment (PFpk)	stance	[Nm·kg <sup>-1</sup> ]	0.95 (0.21)	1.21 (0.18)	1.19 (0.19)	25.9	<0.001	b-r §; b-s §; b-f §		
power (PGpk)	PO	[W·kg <sup>-1</sup> ]	1.49 (0.71)	0.73 (0.30)	1.21 (0.43)	1.43 (0.53)	91.0	<0.001	b-r §; r-s §; r-f §	
timing PGpk	GC	[%GC]	55 (2)	54 (3)	56 (2)	56 (3)	3.93	0.269		
CoP	excursion <sup>a</sup>	step	[mm]	126 (35)	189 (38)	174 (43)	181 (27)	56.6	<0.001	b-r §; b-s §; b-f §
excursion <sup>b</sup>	MSt	[%Step]		82 (9.6)	89 (6)	90 (6)	88 (5)	15.3	0.002	b-r §; b-s §; b-f §

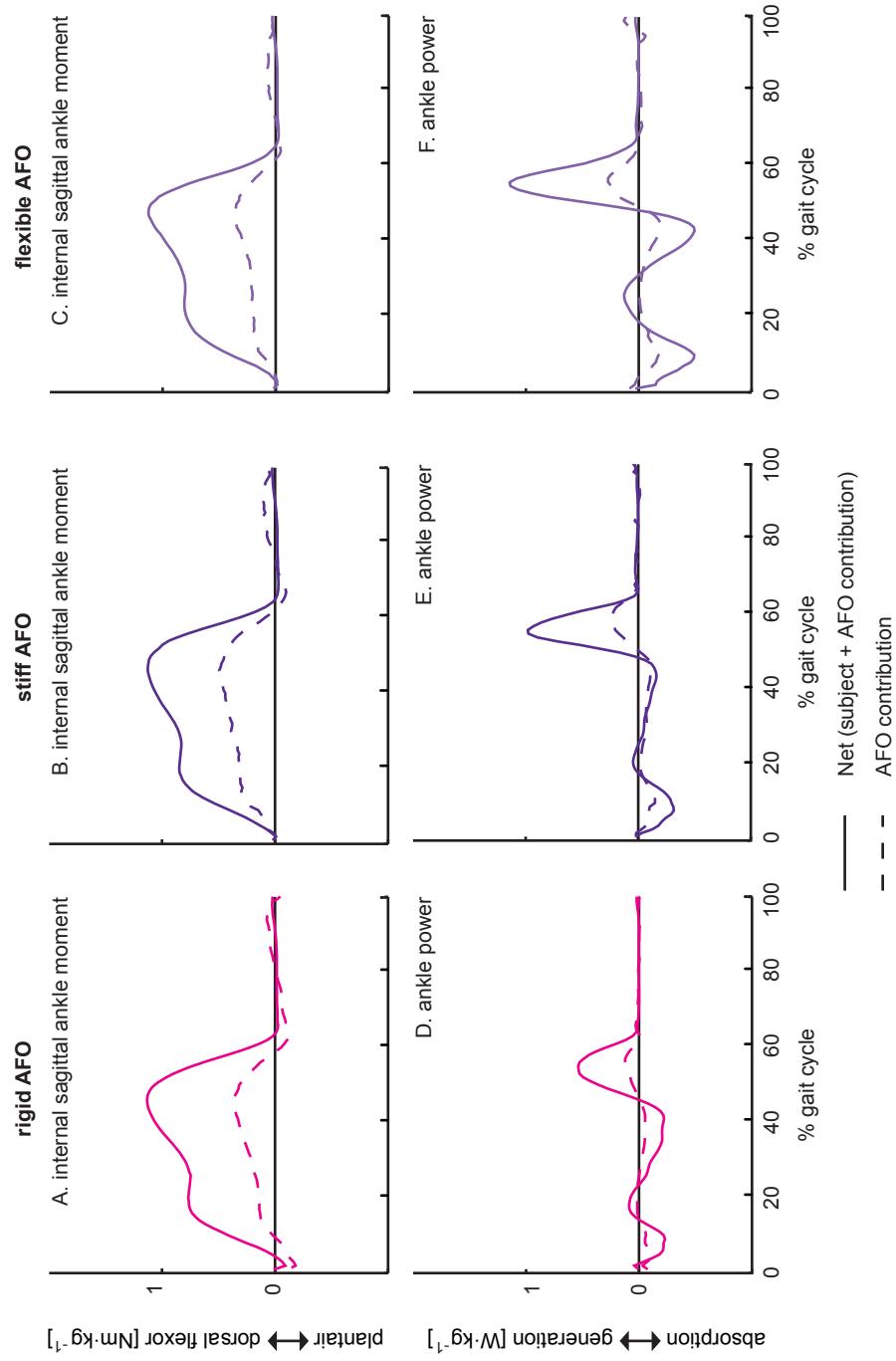
♦ = p&lt;0.05

§ = p&lt;0.001

<sup>a</sup>Center of Pressure (CoP) excursion during the step was determined by continuously calculating the CoP position with respect to the position of the calcaneus at initial contact.

<sup>b</sup>The relative position of the CoP at midstance was calculated as CoPMSt / CoPStep \* 100%.

Abbreviations: KEpk, peak knee extension angle during single support, PFpk, peak internal plantar flexion moment during stance; PGpk, peak power generation during push-off; CoP, centre of pressure; cIC, contralateral initial contact; cTO, contralateral toe-off; MSt, midstance; SS, single support; TKEpk, timing of peak knee extension angle during single support; IC, initial contact, GC, gait cycle; PO, push-off; b, baseline (shoes-only); r, rigid AFO; s, stiff AFO; f, flexible AFO.



**Figure 5.5.** Mean ( $n=15$ ) net internal ankle moment and ankle power for walking with different degrees of AFO stiffness, with mean AFO contributions as a function of the gait cycle. The area underneath the power curves (panel D-F) represents the net ankle work and AFO work.

Abbreviations: AFO, ankle foot orthosis.

### AFO mechanical properties

The rigid AFO was much stiffer (mean (SD) 3.8 (0,7) Nm·deg<sup>-1</sup>) towards dorsiflexion than the stiff (mean (SD) 1.6 (0.4) Nm·deg<sup>-1</sup>) and flexible AFO (mean (SD) 0.7 (0,2) Nm·deg<sup>-1</sup>). AFO properties of the stiff compared to the flexible AFO were different towards dorsal flexion, as the stiff AFO showed a smaller RoM and higher threshold. The mechanical properties towards plantar flexion were comparable between these two spring-like AFOs (see Table 5.6).

## DISCUSSION

This study in children with spastic CP showed that, compared to walking shoes-only, rigid AFOs and spring-like AFOs comparably reduced the knee flexion angle and internal knee flexor moment during stance. Favorable effects on ankle RoM and power generation were found for the spring-like AFOs, but not for the rigid AFOs. Results further showed that all AFOs improved gait efficiency compared to walking with shoes-only, but no significant differences were found between AFOs.

This is the first clinical study investigating the effects of rigid versus spring-like AFOs on gait in children with CP. Earlier, we evaluated the potential value of the spring-like AFOs that were used in the current study. Results of that study suggested that the threshold of the springs within the AFO could reduce knee flexion by preventing dorsiflexion in the beginning of the stance phase until approximately 0.5 Nm·kg<sup>-1</sup>. In the current study however, it appeared that the flexible and stiff AFO were only able to prevent dorsiflexion until a net ankle moment of respectively 0.3 Nm·kg<sup>-1</sup> and 0.4 Nm·kg<sup>-1</sup>. This was reflected in the ankle angle, which gradually increased towards dorsiflexion in the two spring-like AFOs, but approached the maximal dorsiflexion angle already in early-stance (see Figure 5.4A). However, for optimal performance, the spring should however be compressed into its elastics range at midstance. Accordingly, the AFOs did not improve knee angles into normal values, although knee flexion in early stance was reduced by all AFOs (see Table 5.3 and Figure 5.4B). Nonetheless, ankle and knee flexion-extension angles from midstance onwards were improved by all AFOs compared to shoes-only. These improvements are comparable to the study of Rogozinski et al.<sup>[46]</sup>, who investigated the efficacy of a similar type of AFO (i.e. ventral shell) in children with CP.

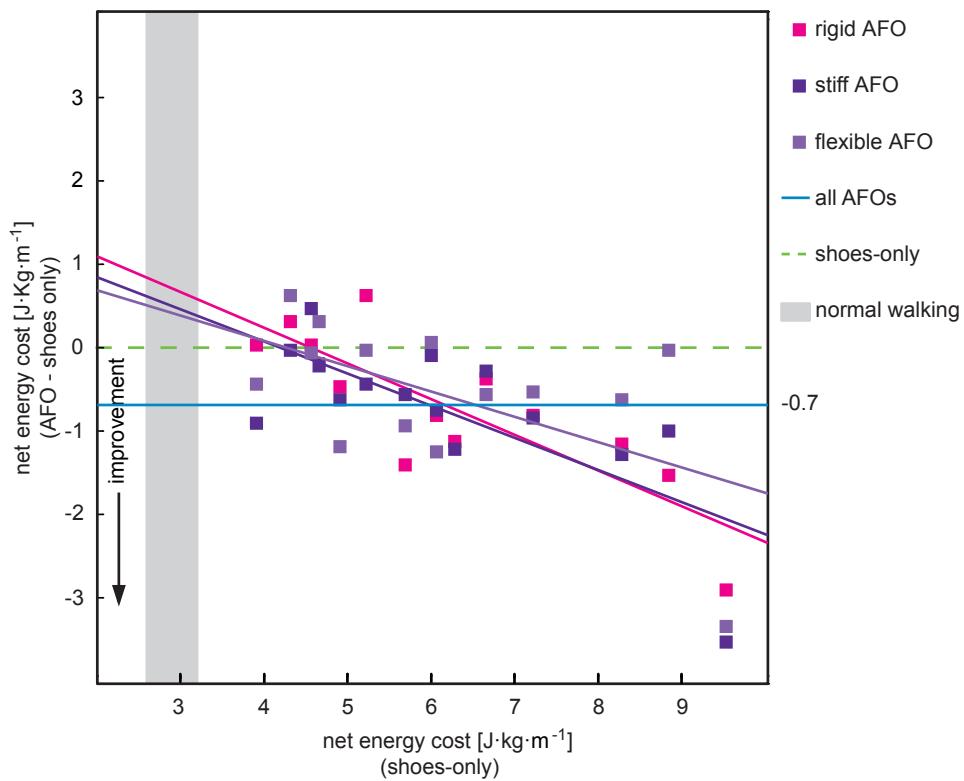
Table 5.4. Results of generalized estimating equation analyses for mean (SD) hip, knee, ankle and AFO work.

			condition			statistics	
			shoes (n=15)	rigid (n=14)	stiff (n=15)	flexible (n=14)	Wald $\chi^2$
hip	work pos	GC	[J·kg <sup>-1</sup> ] (0.11)	0.47 (0.17)	0.51 (0.37)	0.46 (0.10)	8.72 0.033
	work neg	GC	[J·kg <sup>-1</sup> ] (0.05)	-0.10 (0.04)	-0.08 (0.18)	-0.12 (0.05)	5.13 0.162
	work net	PO	[J·kg <sup>-1</sup> ] (0.04)	0.04 (0.05)	0.06 (0.07)	0.06 (0.04)	3.21 0.360
knee	work pos	GC	[J·kg <sup>-1</sup> ] (0.09)	0.17 (0.07)	0.11 (0.07)	0.12 (0.07)	10.3 0.016
	work neg	GC	[J·kg <sup>-1</sup> ] (0.11)	-0.46 (0.17)	-0.49 (0.27)	-0.54 (0.10)	2.66 0.446
	work net	PO	[J·kg <sup>-1</sup> ] (0.08)	-0.19 (0.17)	-0.21 (0.12)	-0.23 (0.08)	3.55 0.315
ankle	work pos	GC	[J·kg <sup>-1</sup> ] (0.09)	0.19 (0.03)	0.08 (0.06)	0.14 (0.08)	41.1 <0.001
	work neg	GC	[J·kg <sup>-1</sup> ] (0.07)	-0.15 (0.02)	-0.07 (0.05)	-0.09 (0.05)	37.2 <0.001
	work net	PO	[J·kg <sup>-1</sup> ] (0.06)	0.13 (0.03)	0.06 (0.04)	0.11 (0.04)	71.3 <0.001
							r-f§; r-ss§; r-f§; b-ss♦; b-r♦; b-ss♦; r-f§; s-f♦; b-r§; r-ss§; r-f§; b-ss♦; r-f§

AFO	work pos	GC	[J·kg <sup>-1</sup> ]	n/a	0.03 (0.02)	0.06 (0.05)	0.05 (0.03)	6.90	0.032	r-s♦; r-f♦
work neg	GC	[J·kg <sup>-1</sup> ]	n/a	-0.03 (0.02)	-0.07 (0.06)	-0.06 (0.04)	11.5	0.003	r-s♦; r-f♦	
work net	PO	[J·kg <sup>-1</sup> ]	n/a	0.01 (0.02)	0.03 (0.05)	0.03 (0.03)	3.26	0.196		

♦ =  $p < 0.05$ § =  $p < 0.001$ 

Abbreviations: AFO, ankle foot orthosis; pos, positive; neg, negative; GC, gait cycle; PO, push-off; b, baseline (shoes-only); r, rigid AFO; s, stiff AFO; f, flexible AFO.



**Figure 5.6.** Overview of individual net energy cost responses. The x-axis represents baseline (i.e. shoes-only) net energy cost values and the y-axis indicates the change in net energy cost as a result of walking with each AFO. Vertically aligned dots thus represent the same participant.

Abbreviations: AFO, *ankle foot orthosis*

Table 5.5. Results of generalized estimating equation analyses for mean (SD) walking speed and energy cost.

		condition			statistics	
		shoes	rigid (n=14)	stiff (n=15)	flexible (n=14)	Wald $\chi^2$
speed	[m·min <sup>-1</sup> ]	58.6 (11.3)	57.8 (8.0)	57.5 (8.4)	58.8 (7.4)	1.53
net EC	[J·kg <sup>-1</sup> ·m <sup>-1</sup> ]	6.1 (1.7)	5.5 (1.1)	5.4 (1.2)	5.6 (1.5)	11.8 0.008 b-r♦; b-s♦; b-f♦
SMC-EC	[%]	269 (73)	245 (51)	242 (51)	251 (62)	7.85 0.049 b-r♦; b-s♦

♦ =  $p < 0.05$ 

Abbreviations: EC, net energy cost; SMC-EC, net non-dimensional energy cost relative to speed-matched control cost; b, baseline (shoes-only); r, rigid AFO; s, stiff AFO; f, flexible AFO.

**Table 5.6.** Mean (SD) mechanical properties of the ankle foot orthoses

			rigid (n=14)	stiff (n=15)	flexible (n=14)
stiffness	dorsal	[Nm·deg <sup>-1</sup> ]	3.8 (0.7)	1.6 (0.4)	0.7 (0.2)
	plantar	[Nm·deg <sup>-1</sup> ]	4.6 (1.3)	0.12 (0.17)	0.11 (0.13)
range of motion	dorsal	[deg]	n/a	6.6 (1.1)	11.8 (1.0)
	plantar	[deg]	n/a	14.3 (1.8)	13.7 (2.5)
threshold	dorsal	[Nm]	n/a	16.5 (5.3)	9.8 (3.2)
	plantar	[Nm]	n/a	-2.2 (2.0)	-2.2 (1.9)

Abbreviations: n/a, not applicable

The comparable reductions of the knee flexion angles during stance between AFOs were in contrast to our hypothesis. This might be explained by the hinge's limited RoM, as the spring-like AFOs can be expected to act rigidly when hitting the hinge's dorsal stop<sup>[29]</sup>. In addition, the ankle RoM measured during walking with the rigid AFO was still 7° (mainly movement in stance). This can be considered as slack, counteracting its extending effect on the knee angle. The rigid AFO was however most effective in reducing the internal knee extensor moment in late stance (at TKEpk), though only significantly compared to the flexible AFO. With forward CoP excursion being similar between AFOs, this difference in knee moment might be explained by changes either in magnitude or direction of the ground reaction force in the sagittal plane (i.e. distance to the knee rotation center), possibly caused by altered trunk positions during walking<sup>[47,48]</sup>. Nonetheless, also the flexible and stiff AFO normalized the internal knee extensor moment over the whole stance phase compared to walking with shoes-only (see Figure 5.4D).

Differences between AFOs were observed in ankle kinematics and kinetics. The ankle power generation was reduced by the rigid AFO, while this was preserved by the spring-like AFOs compared to walking with shoes-only (see Table 5.3 and Figure 5.4E). Nonetheless, peak ankle power generation was only half of reference values of typically developing children (see Figure 5.4E). The potential beneficial effect of spring-like AFOs on push-off function is in accordance with studies comparing different AFO designs in

children with hemiplegia<sup>[49,50]</sup>, showing that spring-like AFOs allow for the storage and return of energy without constraining remaining voluntary push-off. Similar to ankle power, a greater reduction in ankle work was found while walking with the rigid AFO compared to the stiff and flexible AFO. This is in accordance with studies in adult patient populations<sup>[20,51]</sup>. The AFO's energy return (i.e. AFO work) was also smallest for the rigid AFO, which is related to the limited range of motion. Contrary to our expectations, the energy return of the stiff and flexible AFO was comparable (see Table 5.4; see Figure 5.5). The stiffness properties between these two spring-like AFOs might not have been sufficiently different to reveal differences in AFO work during walking in this study.

Compared to walking with shoes-only, the overall mean net EC was significantly lower when walking with AFOs (-11%). This reduction in energy cost, was due to a decrease in energy consumption, as walking speed was not significantly changed by the AFOs. This is similar to a study of Buckon et al.<sup>[10]</sup>, who evaluated the effects of different AFO designs made of polypropylene in children with spastic diplegia. Brehm et al.<sup>[9]</sup> reported a decrease in net EC of only 6% in children with CP while walking with posterior leaf spring or rigid polypropylene AFOs compared to barefoot walking. Sub-analyses in that study revealed that the mean reduction in net EC was much larger when comparing responders to non-responders. Although our study sample was too small to perform such sub-analyses, our results also indicate varying responses between subjects. Differences in the patients' underlying impairments, such as spasticity, could explain the variety in gait biomechanics and walking energy cost. As our patient population had low levels of spasticity, possibly other factors might explain the variety in results. Unfortunately, our sample size was too small to analyze such underlying mechanisms. Nonetheless, our results suggest that most beneficial effects on net EC are seen in the children with highest baseline energy cost levels (see Figure 5.6).

The lack of significant differences in net EC between the three AFOs (see Table 5.5) indicate that the potential benefit of preserving push-off power by the spring-like AFOs may not necessarily enhance walking energy cost. Although we did not relate changes in biomechanical parameters to changes in net EC as a result of varying AFO stiffness, Brehm et al.<sup>[9]</sup> found that changes in knee flexion during stance were significantly related to changes in net EC, while changes in push-off power were not. These results may suggest that, in children with CP who walk with excessive knee flexion, the normalization of knee kinematics and kinetics are dominant with regard to gait efficiency improvement. Alternatively, our study sample may have been too small to show differences in gait efficiency between AFO stiffness levels. Additionally, the stiffness properties between

AFOs may not have been sufficiently distinct. Furthermore, the pre-defined stiffness levels were not matched to specific patient-related characteristics and underlying impairments. The nature of the optimal match between AFO stiffness and patient characteristics has not yet been unraveled, making it difficult to define the optimal stiffness level in relation to each individual patient. Additional studies, evaluating the effects of AFO stiffness levels outside the currently investigated range and in a larger group of children with CP, are needed to further study the relation between changes in gait biomechanics and changes in net EC. This may provide clues to improve and optimize AFO treatment aimed at enhancing gait performance in these children.

Accurate measurements of ankle kinematics in a shod condition are a challenge. In our study, we used VICTOR<sup>[36]</sup> to minimize the effects of probing on the shoe instead of the foot. The calculations of ankle kinematics and AFO contributions were however based on the assumption that no movement occurred between AFOs, shoes, and feet. Possibly, small movements may have occurred, interfering with the results, which can be considered a limitation. Secondly, our study population was homogeneous regarding levels of spasticity, passive range of motion, selective motor control, and gait pattern (i.e. excessive knee flexion), which limits the generalizability of results to other subgroups within CP. However, such a gait-based selection of patients is essential to adequately evaluate the effects of AFO mechanical properties on gait. The small sample size is a third limitation of the study, which could explain that some differences were not statistically significant. Despite these limitations, this is the first study providing accurate descriptions of AFO stiffness and its effects on gait in CP, some of which were evident and clinically important.

In conclusion, despite the homogeneity within our study sample of children with spastic CP, various responses to different degrees of AFO stiffness were seen. Overall, both rigid AFOs and spring-like AFOs reduced the knee flexion angle and internal knee flexion moment comparably in the stance phase of gait, while favorable effects on ankle power generation were only found for the spring-like AFOs. These favorable effects of spring-like AFOs on push-off power did however not lead to greater reductions of walking energy cost. These findings might suggest that, in children with CP who walk with excessive knee flexion in stance, the optimal AFO stiffness that maximizes gait efficiency is primarily defined by its effect on knee kinematics and kinetics during stance and less by its effect on ankle push-off power.

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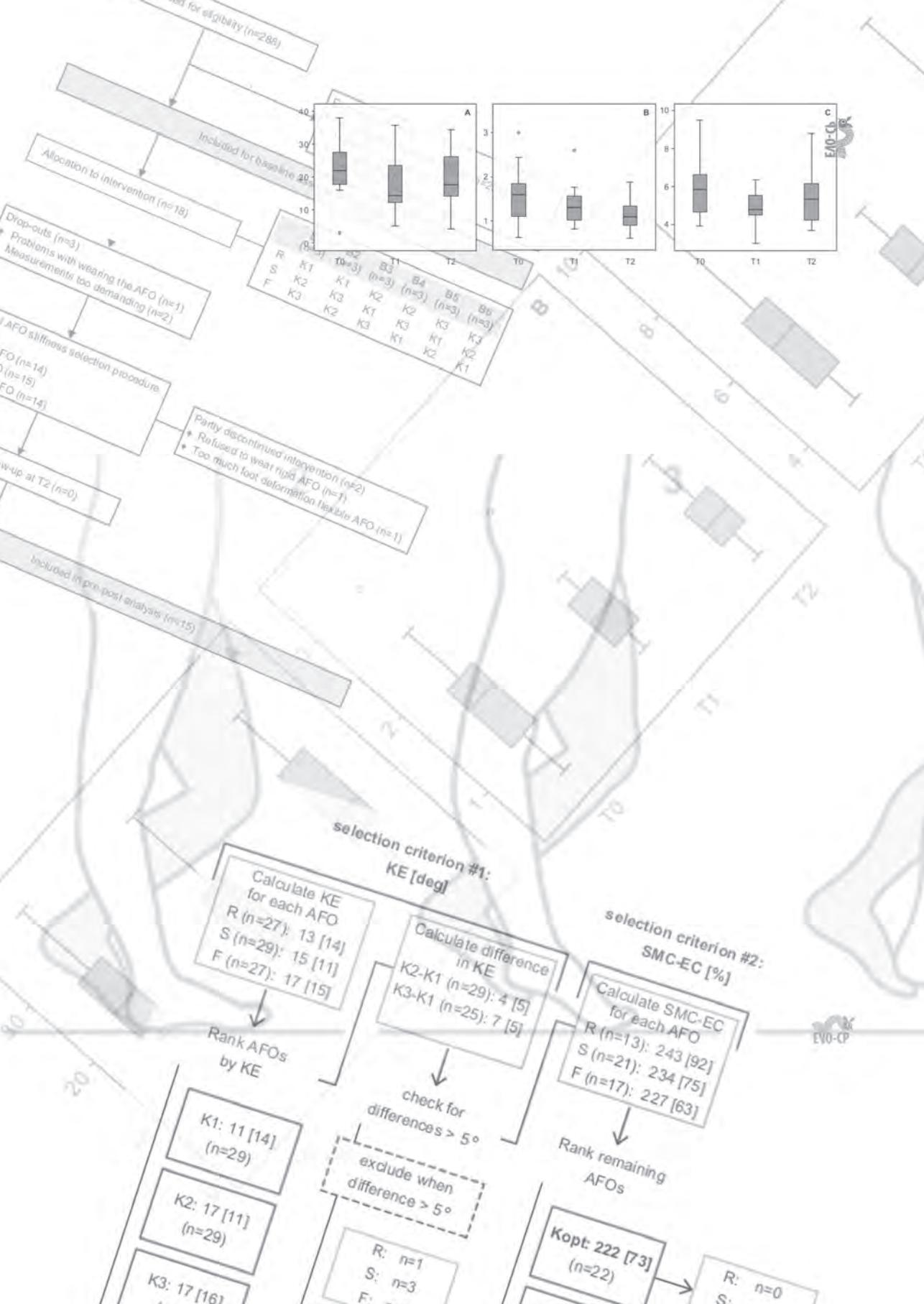
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# An individual approach for optimizing ankle foot orthoses to improve mobility in children with spastic cerebral palsy walking with excessive knee flexion

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## ABSTRACT

Ankle foot orthoses (AFOs) are commonly prescribed to promote gait in children with cerebral palsy (CP). The AFO prescription process is however largely dependent on clinical experience, resulting in confusing results regarding treatment efficacy. To maximize efficacy, the AFO's mechanical properties should be tuned to the patient's underlying impairments. This study aimed to investigate whether the efficacy of a ventral shell AFO to reduce knee flexion and walking energy cost could be improved by individually optimizing AFO stiffness in children with CP walking with excessive knee flexion. Secondarily, the effect of the optimized AFO on daily walking activity was investigated. Fifteen children with spastic CP were prescribed with a hinged AFO with adjustable stiffness. Effects of a rigid, stiff, and flexible setting on knee angle and the net energy cost (EC) [ $J \cdot kg^{-1} \cdot m^{-1}$ ] were assessed to individually select the optimal stiffness. After three months, net EC, daily walking activity [strides·min<sup>-1</sup>] and knee angle [deg] while walking with the optimized AFO were compared to walking with shoes-only. A near significant 9% ( $p=0.077$ ) decrease in net EC ( $-0.5 J \cdot kg^{-1} \cdot m^{-1}$ ) was found for walking with the optimized AFO compared to shoes-only. Daily activity remained unchanged. Knee flexion in stance was reduced by  $2.4^\circ$  ( $p=0.006$ ). These results show that children with CP who walk with excessive knee flexion show a small, but significant reduction of knee flexion in stance as a result of wearing individually optimized AFOs. Data suggest that this also improves gait efficiency for which an individual approach to AFO prescription is emphasized.

## INTRODUCTION

Children with cerebral palsy (CP) have a wide variety of motor impairments (e.g. spasticity and muscle weakness), often resulting in gait deviations, such as excessive knee flexion in stance. The gait pattern of children who walk with excessive knee flexion is prone to deteriorate, as it is associated with the development of knee flexion contractures<sup>[1]</sup> and elevated walking energy cost levels, reflecting poor gait efficiency<sup>[2]</sup>. Interventions in these children therefore primarily aim to reduce knee flexion to prevent deterioration, which could improve gait efficiency<sup>[3]</sup> and walking activity in daily life.

A rigid ventral shell ankle foot orthosis (AFO) is a commonly applied intervention in children with CP walking with excessive knee flexion to reduce knee flexion<sup>[4]</sup> in stance and improve gait efficiency<sup>[3,5]</sup>. Despite the frequent use of AFOs in CP, the prescription process of these orthoses is currently largely dependent on clinical experience, and prescription guidelines are scarce<sup>[6]</sup>. Considering the diversity in underlying impairments within CP, the varying effects of AFOs on gait efficiency as reported in the literature<sup>[3,5]</sup> might be partly explained by an inadequate match between the patient's impairments and the AFO's mechanical properties, including its ankle stiffness<sup>[7,8]</sup>.

To maximize treatment outcome, an AFO is designed to improve the most important deviation in gait biomechanics, while adverse effects on other gait features should be minimized. A rigid ventral shell AFO for example, primarily aims to counteract excessive knee flexion during stance, which has been associated with gait efficiency improvements<sup>[3]</sup>. The AFO's properties however also obstruct ankle range of motion, therewith impeding ankle push-off power and negatively impacting gait efficiency<sup>[9,10]</sup>. Applying a more compliant, spring-like AFO may enhance push-off power and subsequent gait efficiency<sup>[7,11]</sup>, while ideally still counteracting excessive knee flexion. The optimal AFO stiffness that will maximally enhance gait efficiency may rely on a trade-off between counteracting knee flexion during stance, and preserving remaining push-off power<sup>[12]</sup>.

As the aforementioned trade-off is expected to be primarily dependent on the patient's specific underlying impairments, an individual optimization of AFO stiffness seems essential to maximize treatment outcome<sup>[8]</sup>. Such an optimization requires an extensive evaluation of the effects of AFOs on multiple gait-related outcomes<sup>[13]</sup>. Brehm et al.<sup>[14]</sup> suggested a core set of outcome measures for studies on lower limb orthoses, covering all levels of the International Classification of Functioning, Disability and Health (ICF) framework. Such a core set is also useful for the process of AFO stiffness optimization, and includes outcomes quantifying the AFO's effect on gait biomechanics, gait efficiency and daily walking activity<sup>[14,15]</sup>. In this context, we tested the hypothesis

that the efficacy of AFOs to reduce knee flexion and improve gait efficiency in children with CP who walk with knee flexion in stance can be improved by individually tuning the mechanical properties of the AFO, i.e. optimizing the AFO stiffness to the underlying impairments of the patient. Secondarily, we investigated whether this stiffness-optimized AFO would also improve daily walking activity in these children.

## **METHODS**

### **Study design**

We performed a pre-post experimental study (AFO-CP study<sup>[13]</sup>; Dutch National Trial Register no. NTR3418), consisting of two repeated measurements; at baseline (T<sub>0</sub>), walking with shoes only, and at 12–20 weeks follow-up (T<sub>2</sub>), walking with the optimized AFO. Additional measurements were performed to provide data for the optimal AFO stiffness selection (T<sub>1</sub>).

Institutional review board approvals were obtained prior to the start of the study and all measurements were performed in accordance to the Declaration of Helsinki. Parents of all participants and participants above 12 years old provided written informed consent.

### **Participants**

Children diagnosed with spastic CP who were indicated for a new AFO were recruited from the rehabilitation department of a university medical center, and two affiliated rehabilitation centers. Children could be included in the study when they were 6–14 years old, classified with a Gross Motor Function Classification System<sup>[16]</sup> level I, II or III, and presented a barefoot gait pattern that was characterized by excessive knee flexion in stance (i.e. >10° knee flexion at midstance). Children were excluded if they had hip and/or knee flexion contractures of >10°, as these have been shown to impede the effect of AFOs<sup>[4]</sup>.

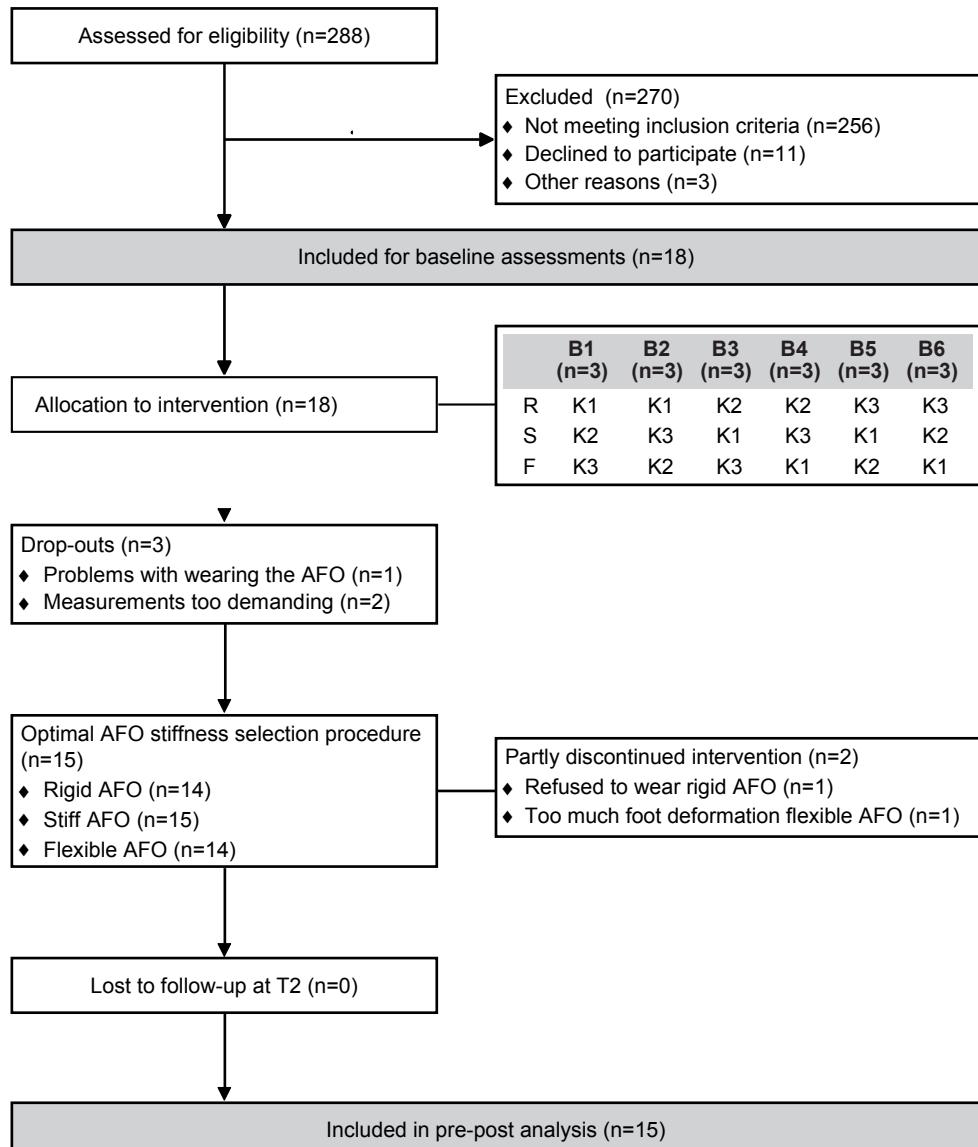


Figure 6.1. Study flowchart. The allocation of the different degrees of stiffness (i.e. rigid (K1), stiff (K2), and flexible (K3)) was block-randomized: B1 to B6 represent the different blocks.

Abbreviations: AFO, ankle foot orthosis; R, rigid AFO stiffness; S, stiff AFO stiffness; F, flexible AFO stiffness.

### Intervention

For shoes-only measurements, participants wore their own shoes. Children were prescribed with a ventral shell AFO with a full-length stiff footplate, which were worn in sneakers with flat flexible soles. The AFOs were made out of pre-preg carbon fibers and manufactured with an integrated hinge (Neuro Swing®, Fior&Gentz, Germany). This hinge holds shafts towards ankle plantar flexion and dorsiflexion in which springs with various mechanical properties can be inserted<sup>[12]</sup>. For each participant, the hinge was randomly set into three stiffness configurations (i.e. rigid, stiff and flexible), and the effects of each configuration on gait were evaluated (see Appendix A for the detailed protocol). After this, the optimal AFO stiffness was selected (T1) according to a predefined decision scheme (see Appendix A), which was based on ranking the AFO's effect on knee extension (KE) and, in addition, on gait efficiency (i.e. walking energy cost). Outcome was assessed after three months of wearing the optimized AFO.

### Outcomes

The primary outcome in our study was walking energy cost. Secondary outcomes included daily walking activity, knee angle and ankle power. Additionally, compliance to the optimized AFO was measured. Extensive descriptions of these outcomes are described elsewhere<sup>[13]</sup>.

Walking energy cost was assessed with a 6-minute rest test, followed by a 6-minute walk test at comfortable speed on a 40-meter indoor oval track. During the rest tests and the walk test, breath-by-breath oxygen uptake ( $VO_2$ ) and carbon dioxide production ( $VCO_2$ ) values were recorded using a portable gas analysis system (Metamax 3B, Cortex Biophysik, Germany). Participants were instructed not to talk or laugh during the assessments.

Daily walking activity was assessed using the ankle-worn biaxial StepWatch™ Activity Monitor 3.0 (SAM) (Orthocare Innovations, USA), which registers accelerations of one leg in the frontal-sagittal plane. Children were asked to wear the SAM for seven consecutive days (five weekdays, two weekend days) during waking hours<sup>[17]</sup>.

Gait biomechanics were assessed by 3D-gait analysis that was performed in a gait laboratory. Participants were instructed to walk on a 10m-walkway with integrated force platform (OR6-5-1000, AMTI, USA) at comfortable speed. Technical marker clusters of three markers were rigidly attached to the body segments and anatomically calibrated by probing bony landmarks<sup>[18]</sup>. Segment movements were tracked using an optoelectronic

motion capture system (Optotrak 3020, Northern Digital, Canada) and synchronized with the force plate data. Measurements were repeated until three successful steps of both legs were recorded (i.e. within the borders of the force plate).

Time of wearing the optimized AFO [hours·day<sup>-1</sup>] was measured during the seven days that the SAM was worn, using a temperature-based monitor (the @monitor, Academic Medical Center, The Netherlands), which was mounted in the shell of the AFO. This device has been shown to reliably assess the use of footwear and assistive devices<sup>[19]</sup>.

### Data processing

To calculate walking energy cost, breath-by-breath  $\text{VO}_2$  and  $\text{VCO}_2$  values from minute three to six of the rest and the walk test were used to determine the mean steady-state energy consumption values (ECSrest and ECSwalk). The mean walking speed [ $\text{m} \cdot \text{min}^{-1}$ ] was measured over the same time frame of the walk test. Accordingly, the net energy cost (EC) [ $\text{J} \cdot \text{kg}^{-1} \cdot \text{m}^{-1}$ ] was calculated as  $(\text{ECSwalk} - \text{ECSrest}) / \text{walking speed}$ . Net EC values were normalized to calculate the net non-dimensional energy cost<sup>[20]</sup>, and the net non-dimensional energy cost as a percentage of speed-matched control cost (SMC-EC) [%].

Regarding the SAM, data were excluded from the analysis if i)  $>3$  hours of data were missing within the time interval of being awake, and ii) a day had less than eight hours of registration time. A minimum of three correctly recorded days was required to calculate the average daily stride rate. Daily stride rate was sub-divided into stride rate levels, according to existing thresholds<sup>[21]</sup>: 0 strides·min<sup>-1</sup> (SR0), 1 to 15 strides·min<sup>-1</sup> (SR1-15), 16 to 30 strides·min<sup>-1</sup> (SR16-30), 31 to 60 strides·min<sup>-1</sup> (SR31-60), and  $>60$  strides·min<sup>-1</sup> (SR>60).

For gait analysis, optoelectronic marker data and force plate data of three trials of the most affected leg were analyzed using custom-made software (BodyMech, [www.bodymech.nl](http://www.bodymech.nl)). Initial contact and toe-off of the trailing leg were determined using foot angular velocity, and single support of the leading leg was defined. Joint and segment kinematics were calculated according to ISB anatomical frames<sup>[18]</sup>. The peak knee extension angle (KE) [deg] was defined as the minimal knee flexion angle during single limb support. The shank-to-vertical angle (SVA) [deg], defined as the angle of the shank's anterior surface with respect to the vertical in the sagittal plane, at midstance was also calculated<sup>[22]</sup>. Using force plate data and inverse dynamics, peak ankle power generation (AP) [ $\text{W} \cdot \text{kg}^{-1}$ ] was calculated. All data were processed using MATLAB version R2011a (The Mathworks, USA).

## Statistical analyses

The sample size for this study was based on a power analysis of the expected changes in the net EC (i.e. shoes-only versus stiffness-optimized AFO), assuming a power of 80% and a significance level of 0.05. A sample size of 32 children was planned<sup>[13]</sup>.

Descriptive statistics (mean and standard deviation (SD) or median and interquartile range (IQR)) were used to summarize the participants' demographic and disease characteristics, as well as all outcome measures. Effects of the optimized AFO (T<sub>2</sub>) on net EC, walking speed, daily walking activity, and biomechanical gait parameters were compared to walking with shoes-only (T<sub>0</sub>) using Wilcoxon Signed Rank Tests. One-tailed tests were performed for the net EC and KE, as the AFO was optimized based on these parameters and a one-sided (i.e. decrease) effect was therefore hypothesized. Analyses were done with SPSS Statistics 20. An alpha level of 0.05 was used for all tests of significance.

**Table 6.1** Participant's demographic and disease characteristics at baseline (n=15).

age	[yrs]	10 (2)
weight	[kg]	37.2 (9.0)
height	[cm]	141 (9.0)
sex	[boy/girl]	11/4
GMFCS	[I/II/III]	2/11/2
(most) affected side	[right/left]	11/4
selective motor control <sup>a</sup>	[good/moderate/poor]	11/3/1

<sup>a</sup>Selective motor control of both legs was assessed using the modified Trost test, which measures the ability to dorsiflex the ankle and extend the knee in an isolated movement. Ankle dorsiflexion and knee extension of each leg were scored as 0 (no selective, only synergistic movement), 1 (diminished selective movement) or 2 (full selective movement) and summed to a total score of 0 to 8. These total scores were categorized into poor (total score of 0 to 2), moderate (total score of 3 to 5) or good (total score of 6 to 8) selective motor control<sup>[31]</sup>.

Abbreviations: GMFCS, Gross Motor Function Classification System;

## RESULTS

### Participant flow and recruitment

210 out of 228 children that were screened for eligibility to participate in the study did not meet the inclusion criteria. The majority was excluded based on age and gait pattern. 32 children were invited to participate, of which 18 children were enrolled in the study. Two participants dropped out as the measurements were too demanding, and one participant dropped out because he had too many problems with the fitting of the AFOs. Accordingly, data of 15 children (29 limbs) were included in the analyses (see Figure 6.1). Demographic and disease characteristics of these children are shown in Table 6.1. The participants' characteristics as assessed during the physical exam are shown in Table 6.2.

**Table 6.2** Results of the physical exam as assessed at baseline (n=15).

angle of interest <sup>a</sup>	muscles	RoM Median [min max]	spasticity scale <sup>b</sup> [0/1/2/3]
hip extension	[+ = extension]	10 [0 20]	n/a
knee extension	[+ = extension]	0 [-10 0]	n/a
popliteal angle	Hamstrings	55 [45 70]	[10/1/3/0]
ankle dorsiflexion (flexed knee)	Soleus	10 [0 25]	[13/1/0/1]
ankle dorsiflexion (extended knee)	Gastrocnemius	0 [-10 10]	[13/1/0/1]

<sup>a</sup>Hip extension was measured with the patient in prone position. All other measurements were performed with the patient in supine position. Comprehensive descriptions of positions and movements are described elsewhere<sup>[32]</sup>. The popliteal angle was missing in one patient.

<sup>b</sup>Spasticity was tested according to the Spasticity Test protocol<sup>[32]</sup>, using a 4-points spasticity scale: 0, normal or increased muscle resistance over the whole range of motion; 1, increase in muscle resistance somewhere in the range of motion; 2, catch and release; 3, catch blocking further movement<sup>[32]</sup>.

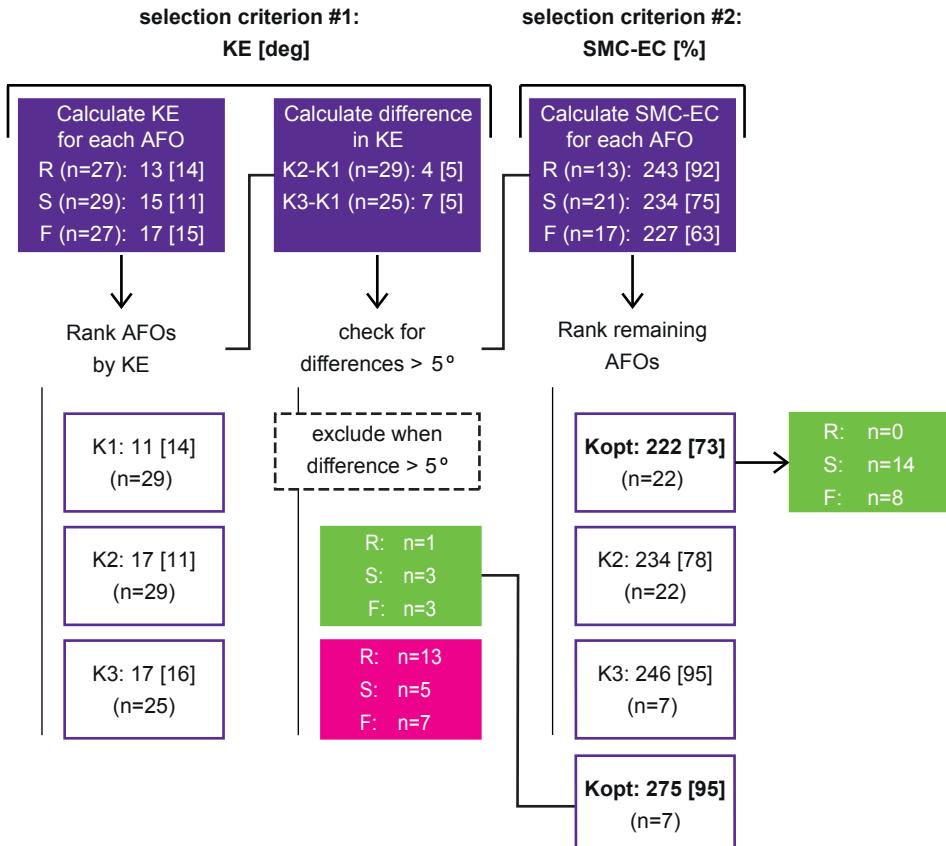
Abbreviations: RoM, range of motion; n/a, not applicable.

### Optimal stiffness selection

Ranking of the AFO based on its effect on KE resulted in an immediate decision for 7 out of 29 legs, of which one was prescribed with the rigid, three with the stiff, and three with the flexible AFO as most optimal. Based on SMC-EC, the stiff (n=14) and flexible (n=8) AFO were prescribed as most optimal for the remaining legs (see Figure 6.2). At the moment of selecting the optimal stiffness (T1), the optimized AFO reduced the net EC in all participants, resulting in a median [IQR] net EC of 4.8 [1.5] J·kg·m<sup>-1</sup>, accounting for a 20% decrease compared to baseline (see Table 6.3 and Figure 6.3).

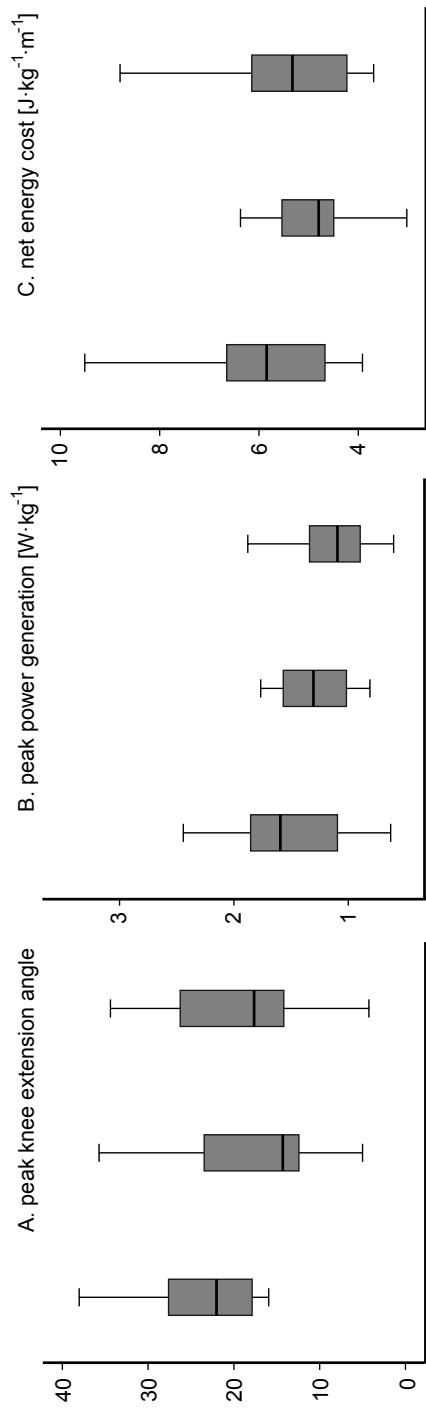
### Effects of the optimized AFO

Walking energy cost data of one participant was excluded from analysis, because equipment failed during the measurement. At follow-up, 11 out of 14 children showed a decrease in net EC compared to baseline, resulting in a median reduction of 9% ( $p=0.077$ ) for walking with the optimized AFO compared to shoes-only. The optimized AFO did not affect walking speed ( $p=1.000$ ). SAM data of 11 participants were sufficient for analysis. Daily stride rate was not affected by the optimized AFO compared to baseline on all stride rate frequencies ( $p>=0.148$ ). The optimized AFO significantly reduced the SVA by 5.2° ( $p=0.002$ ), and the KE by 2.4° ( $p=0.006$ ) compared to walking shoes-only. The peak ankle power generation was not significantly reduced ( $p=0.064$ ) (see Table 6.3). The optimized AFO was worn for median [IQR] of 8.9 [5.0] hours·day<sup>-1</sup>.



**Figure 6.2.** Optimal AFO stiffness selection decision scheme. On the first selection criterion (i.e. peak knee extension angle during single support (KE)), the first ranked AFOs (K1) resulted in a median [IQR] KE of 11 [14], while this was 17 [11] and 17 [16] for the second (K2) and third (K3) ranked AFO stiffness respectively. AFOs were excluded from further analysis when KE of K1 was  $>5^\circ$  smaller compared to K2 and/or K3. Accordingly, the first criterion was decisive for 7 legs (middle panel, green square), which represented a median [IQR] walking energy cost (SMC-EC) of 275 [95] percent. Based on KE, the rigid AFO was excluded for selection in 13 legs, the stiff AFO in 5 legs, and the flexible AFO in 7 legs (middle panel, pink square). SMC-EC was decisive for the remaining legs, resulting in a median [IQR] optimal SMC-EC of 222 [73] percent. In total, the assigned optimal AFO stiffness was rigid for one leg, stiff for 17 legs, and flexible for 11 legs. Eight participants were prescribed with bilateral stiff AFOs as optimal, four with bilateral flexible AFOs, two with a stiff and a flexible AFO, and one with a rigid and a flexible AFO.

Abbreviations: AFO, ankle foot orthosis; KE, peak knee extension during single limb support; SMC-EC, net non-dimensional walking energy cost, calculated as a percentage of speed-matched control cost. Kopt, optimal AFO stiffness; K1, K2 and K3 represent first, second and third ranked AFO stiffness respectively; R, rigid AFO stiffness; S, stiff AFO stiffness; F, flexible AFO stiffness.



**Figure 6.3.** Boxplots of the peak knee extension angle during single support (n=15), peak power generation (n=15), and the net energy cost (n=14) at T0 (i.e. shoes-only), T1 (i.e. AFO with optimal stiffness at moment of optimal stiffness selection), and T2 (i.e. stiffness-optimized AFO at three months follow-up).

**Table 6.3.** Wilcoxon signed rank test for median [IQR] gait efficiency (n=14), daily walking activity (n=11), and gait biomechanics of the most affected leg (n=15), at baseline (To; shoes-only), and follow-up (T2; optimized ankle foot orthosis).

		To	T1	T2	T2-To <sup>b</sup>	Z	p
<i>gait efficiency</i>							
speed	[m·min <sup>-1</sup> ]	61.7 [14.2]	58.2 [8.1]	61.6 [29.0]	-0.1 (<1%)	-0.03	1.000
net EC <sup>a</sup>	[J·kg·m <sup>-1</sup> ]	5.8 [2.1]	4.8 [1.5]	5.3 [2.1]	-0.5 (-9%)	-1.48	0.077
<i>daily walking activity</i>							
total	[strides/day]	3986 [2579]	n/a	2513 [3715]	-1473	-0.09	0.966
SR1-15	[strides/day]	984 [1082]	n/a	976 [567]	-16	-0.45	0.700
SR16-30	[strides/day]	946 [924]	n/a	963 [1245]	2	-0.71	0.520
SR31-60	[strides/day]	1692 [1955]	n/a	836 [1949]	-855	-0.36	0.765
SR>60	[strides/day]	0 [90]	n/a	18 [99]	18	-1.54	0.148
<i>gait biomechanics</i>							
KE <sup>a</sup>	[deg]	22.0 [11.7]	14.3 [15.2]	19.6 [17.2]	-2.4	-2.44	0.006
SVA	[deg]	25.4 [7.8]	20.2 [8.4]	20.2 [9.2]	-5.2	-2.90	0.002
AP	[W·kg <sup>-1</sup> ]	1.6 [0.9]	1.3 [0.7]	1.2 [0.5]	-0.4	-1.87	0.064

<sup>a</sup>Tested one-tailed.

<sup>b</sup>Difference in percentage between T2 and To was calculated as:  $(T2-To)/((To+T2)/2)*100\%$

Abbreviations: KE, peak knee extension angle at single support; KM, internal knee moment at timing of KE; AP, peak ankle power generation; net EC, net energy cost; SMC-EC, net non-dimensional energy cost expressed as a percentage of speed-matched control cost; SRo, stride frequency of o strides per minute; SR1-15, stride frequency of 1 to 15 stride per minute; SR16-30, stride frequency of 16 to 30 strides per minute; SR31-60, stride frequency of 31 to 60 strides per minute; SR>60, stride frequency of more than 60 strides per minute; n/a, not applicable.

## DISCUSSION

This study aimed to individually optimize AFO stiffness in children with CP walking with excessive knee flexion in order to improve treatment outcome in terms of gait efficiency, knee angle and secondarily, daily walking activity. Our results show that the individually optimized AFO improved gait efficiency by 9% compared to walking with shoes-only, although this difference was not statistically significant. Daily walking activity remained unchanged, while the AFO significantly reduced knee flexion by 2.4°.

While no previous studies have reported the effects of stiffness-optimized AFOs on gait in CP, effects of various AFO designs have been previously compared<sup>[5,23,24]</sup>, some of which also used walking energy cost assessments to select the most beneficial AFO<sup>[5,25]</sup>. In our study, we found that the knee extension angle in stance (i.e. first selection criterion) was decisive in only 7 out 29 legs, indicating that both rigid and spring-like AFOs affected knee angle comparably in most children. This is in line with other literature<sup>[24]</sup>, showing similar improvements in stance phase knee kinematics by solid and spring-like AFOs. Unexpectedly, the rigid AFO performed worse on knee extension compared to the spring-like AFOs in 13 legs. We observed that some children avoided knee extension, and thus stretching of the calf muscles, by walking on the tip of the rigid AFOs' footplate, which could explain the persistent knee flexion in this condition. Since only rigid AFOs are stiff enough to allow such a walking pattern, it may be suggested that spring-hinged AFOs are more suited to improve knee extension, and subsequently prevent development of muscle contractures and improve gait efficiency. This idea is supported by the fact that the spring-like AFOs were selected as optimal for the majority of participants based on net EC reductions, confirming the beneficial effect of spring-like properties on gait efficiency, which has also been shown in adult populations<sup>[8,11]</sup>.

At the moment of optimal stiffness selection, the AFOs resulted in a 20% decrease in net EC compared to walking shoes-only, indicating a relatively large improvement compared to literature<sup>[3,5]</sup>. At follow-up, we found a 9% decrease in the net EC compared to shoes-only. Several factors might explain this smaller decrease (i.e. less profitable) in net EC at follow-up. First, the mechanical properties of the AFO may have changed over time, therewith less effectively reducing knee flexion and enhancing push-off power. Also the participant's development (e.g. growth) could have interfered with the AFOs' effect on net EC. On the other hand, considering that the majority of AFOs were optimized based on energy cost reductions, the decrease in net EC at the moment of optimal stiffness selection may have been overestimated. In these cases, the selection

was based on absolute differences in energy cost, regardless of the magnitude of the differences between AFOs. Walking energy cost measurements are however subject to large variability, resulting in a large smallest detectable difference. Hence, unjustified assignments of an optimal stiffness could have occurred. Nonetheless, 11 out of 14 subjects showed a decrease in their net EC while walking with the optimized AFO at follow-up. Five subjects showed a decrease of >10% indicating that individually optimizing AFO stiffness can result in clinically meaningful changes. Also the diversity in assigned optimal stiffness levels emphasizes an individual approach to optimizing treatment in CP in order to maximize the gain for the patient. The lack of statistical significance and the absence of a larger effect on net EC is most likely related to the small sample size, as the study was underpowered<sup>[13]</sup>. Although homogeneity in the study population was required to enable the stiffness optimization as performed in our study, the very specific inclusion criteria restricted the enrollment of children into the study. This is a serious limitation of this study, making it difficult to draw firm conclusions on the potential benefit of optimizing AFO stiffness on gait efficiency in children with CP.

Literature suggests that the AFO's efficacy is dependent on the SVA, which is a parameter to quantify the alignment of the ground reaction force with respect to the joints when wearing an AFO<sup>[22,26]</sup>. The SVA of 20° as found in our study was much larger compared to findings in other literature<sup>[26]</sup>, suggesting inadequate alignment. However, literature showed that the SVA might be less important in children with CP with a knee flexion gait pattern, as in our study, as the AFO's performance is not affected by changes in SVA in these children<sup>[27]</sup>. Nevertheless, the optimized AFO significantly improved the SVA compared to walking shoes-only, which was accompanied by a small but significant improvement in knee extension, which was comparable to other literature<sup>[3,5,23]</sup>.

In our study, the AFOs were used for a median [IQR] of 8.8 [5.0] hours·day<sup>-1</sup>, which is comparable to Wren et al.<sup>[28]</sup>. Despite this relatively intensive use, daily walking activity did not increase with the optimized AFO. Wren et al.<sup>[28]</sup> evaluated the effects two AFO designs (adjustable dynamic, and dynamic) on gait biomechanics and daily walking in children with CP. They found a favorable effect of the adjustable AFO on push-off power, like in our study, while this was not reflected in an improvement of daily walking activity. The authors suggested that their findings could be related to an inadequate accommodation. In our study, baseline daily activity was measured while wearing their old AFO, which was also spring-hinged in some participants, possibly causing an insufficient contrast between the baseline and follow-up walking conditions. Although an association between the level of physical activity and walking energy cost has been

found in children with CP<sup>[29]</sup>, it is unclear whether energy cost improvements can actually lead to increased activity levels. Besides, improving daily activity is challenging, because it involves a behavioral change<sup>[30]</sup>.

In conclusion, our study in children with CP who walk with excessive knee flexion shows that individually optimizing AFO stiffness significantly improves the gait pattern by a reduced knee flexion in stance. In addition, data suggest that gait efficiency can also be improved, although we cannot draw firm conclusions on the improvement in gait efficiency given the limited sample size. Nonetheless, the variety in the assignment of an optimal stiffness emphasizes an individual approach to AFO prescription in CP to maximize its effects on the gait pattern and gait efficiency.

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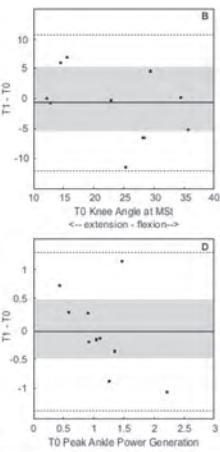
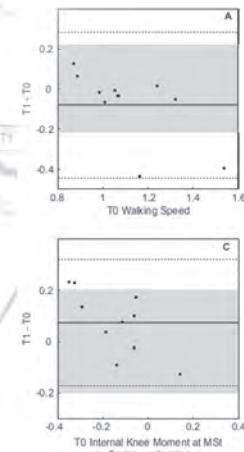
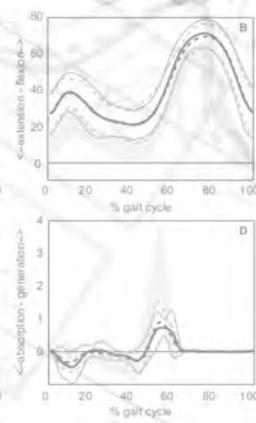
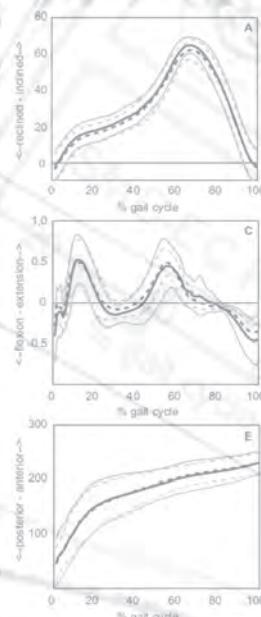
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## **APPENDIX A. PROTOCOL FOR DEFINING THE OPTIMAL STIFFNESS**

Following new ankle foot orthosis (AFO) prescription, the AFO's hinge was randomly (i.e. block randomized) set into one of the three configurations, which varied in stiffness towards dorsiflexion: rigid [mean (SD) 3.8 (0.7) Nm·deg<sup>-1</sup>], stiff [mean (SD) 1.6 (0.4) Nm·deg<sup>-1</sup>], and flexible [mean (SD) 0.7 (0.2) Nm·deg<sup>-1</sup>]. The stiffness was measured using the Biarticular Reciprocal Universal Compliance Estimator (BRUCE) device, according to a standard protocol<sup>[12]</sup>. The AFO-footwear combination was tuned according to a clinical protocol, based on ground reaction force alignment at midstance and terminal stance. Participants were instructed to gradually increase time of wearing to avoid pressure sores. After acclimatizing to the AFO for 4 to 6 weeks, effects of the AFO stiffness on gait were evaluated by means of a walking energy cost test and a 3D-gait analysis. When this evaluation was completed, the hinge was set in the next stiffness and the procedure was repeated. From each stiffness evaluation, the peak knee extension angle during single support was derived from the gait analysis, where all available steps within the recorded trials of both legs were analyzed, with a minimum of three steps per participant. From the walking energy cost test, the net non-dimensional energy cost was determined, which was expressed as a percentage of speed-matched control cost (SMC-EC).

Following the three stiffness evaluation assessments, the optimal AFO stiffness was individually determined for each participant according to a decision scheme, which was based on two decision criteria<sup>[13]</sup>. First, AFOs were ranked based on peak knee extension angle during single support (KE), with lower peak values indicating better performance. AFO performance was considered equal when the difference in KE was smaller than 5 degrees (i.e. smallest detectable difference of sagittal knee kinematics). If one AFO resulted in a KE that was more than 5 degrees lower compared to the two other AFOs, that AFO configuration was immediately chosen as optimal AFO stiffness. When two or three AFOs showed equal performance on KE, the effect on the walking energy cost was decisive. The AFO stiffness resulting in the lowest SMC-EC was chosen as optimal AFO stiffness. This decision-making process was performed for each leg separately. As such, different AFO stiffness configurations could be assigned to both legs within the same participant.



• individual data points    SDC    mean difference     $\pm 1.96$  S.D. of mean difference

## CHAPTER VII

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# Acclimatization of the gait pattern to wearing an ankle foot orthosis in children with spastic cerebral palsy

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## ABSTRACT

Ankle foot orthoses (AFOs) can be prescribed to improve gait in children with cerebral palsy (CP). Before evaluating the effects of AFOs on gait, a period to adapt or acclimatize is usually applied. It is however unknown whether an acclimatization period is actually needed to reliably evaluate the effect of a new orthosis on gait. This study aimed to investigate whether specific gait parameters in children with CP would change within an acclimatization period after being provided with new AFOs. Ten children with CP, walking with excessive knee flexion in midstance (8 boys; mean (SD) 10.2 (1.9) years; Gross Motor Function Classification System level I-II) were provided with ventral shell AFOs. The orthoses were worn in combination with the child's own shoes and tuned, based on ground reaction force alignment with respect to the lower limb joints. Directly after tuning (T<sub>0</sub>) and four weeks later (T<sub>1</sub>), 3D-gait analysis was performed using an optoelectronic motion capture system and a force plate. From this assessment, ten spatiotemporal, kinematic and kinetic gait parameters were derived for the most affected leg. Differences in parameters between T<sub>0</sub> and T<sub>1</sub> were analyzed using paired t-tests or Wilcoxon signed rank tests ( $p<0.05$ ). Over the course of four weeks, no significant differences ( $p\geq0.080$ ) were observed for any investigated parameter. These results imply that the biomechanical effect of ventral shell AFOs on gait in independent walking children with CP is immediately apparent, i.e. there is no further change after acclimatization.

## INTRODUCTION

Gait in children with spastic cerebral palsy (CP) is often affected by symptoms of spasticity and lower extremity muscle weakness, which limit the patient's ability to walk. To improve gait and reduce walking limitations, children with CP can be provided with ankle foot orthoses (AFOs)<sup>[1]</sup>. Studies evaluating the effects of AFOs on gait in these children generally report improvement in terms of spatiotemporal parameters<sup>[2-9]</sup>, joint kinematics<sup>[2-5,7-11]</sup> and kinetics<sup>[3,4,7,8,10]</sup>, and walking ability<sup>[3,3,10]</sup>.

When evaluating the effects of AFOs on gait, acclimatization to the new orthosis, i.e. ensuring that the gait pattern is completely adapted to the altered ankle function as induced by the prescribed AFO, is recommended to represent daily use<sup>[12]</sup>. Yet, although most testing protocols in previous studies permitted acclimatization time, varying from less than one day to more than six weeks<sup>[12]</sup>, it is currently unknown whether acclimatization time is actually needed to adapt to the provided AFO. The need for such an acclimatization period particularly depends on the patient's response to the mechanical constraint applied by the AFO. On the one hand, it may be hypothesized that the effect of the AFO on gait is solely a biomechanical response to which children with CP are able to adapt immediately, implying that acclimatization time would not be needed. On the other hand, children with CP may need time to adjust their gait pattern to the new AFO by improving their muscle activation pattern in terms of muscle timing, which would require a period of learning (i.e. motor learning)<sup>[13]</sup>, and thus acclimatization time to account for the learning effect.

Previously, the effects of AFOs on the gait pattern and on muscle timing in CP have been evaluated in three studies<sup>[7,14,15]</sup>. Radtka et al.<sup>[7,14]</sup> measured the effects of two types of AFOs in a small group of mildly involved children with spastic CP, showing that after acclimatizing for four weeks, both AFOs altered joint kinematics and kinetics, while no accompanying changes in lower extremity muscle timing were found compared to barefoot walking. Likewise, Rethlefsen et al.<sup>[15]</sup> found no difference in timing of calf muscle activity between walking with shoes and walking with AFOs, whereas changes in ankle kinematics and kinetics during gait were observed. While these findings seem to indicate that compared to barefoot walking, AFOs can change the gait pattern without affecting muscle timing, none of the studies specifically aimed to compare the gait pattern before and after acclimatization.

Accordingly, it is currently unknown whether an acclimatization period is needed for a reliable evaluation of the biomechanical effects of AFOs on gait in children with CP, while it is recognized that knowledge about the effects of acclimatization time would

improve the quality of AFO research<sup>[12]</sup>. This study aimed to investigate whether relevant biomechanical gait parameters in children with CP would change within an acclimatization period after being provided with a new AFO. Because previously mentioned studies showed no changes in muscle timing after being provided with a new AFO<sup>[7,14,15]</sup>, we hypothesized that these parameters would not change within the acclimatization period.

## METHODS

The data used in this study originates from AFO-CP trial<sup>[16]</sup>, which is aimed at optimizing AFO treatment in children with CP. For the present study, data were used from participants who were provided with ventral shell AFOs and evaluated in the gait laboratory directly after delivery of the AFOs and four weeks later.

### Participants

Participants in the AFO-CP trial were recruited from the outpatient clinic of a university hospital in the Netherlands and two of its affiliated rehabilitation centers. The main inclusion criteria were a confirmed diagnosis of CP and a barefoot gait pattern that was characterized by excessive knee flexion in midstance. Other inclusion criteria have been described elsewhere<sup>[16]</sup>. Institutional review board approvals were obtained prior to study initiation, and all participants (above 12 years old) and their parents provided written informed consent.

### Materials

Participants in the AFO-CP trial were provided with floor reaction AFOs, which were designed with a ventral shell and a rigid footplate. AFOs were manufactured using prepeg carbon, with an integrated ankle hinge (NeuroSwing®, Fior & Gentz, Lüneburg, Germany), which allows mechanical properties to be adjusted within the same orthosis by applying springs with different degrees of stiffness<sup>[17]</sup>. The AFO was worn in combination with the child's own shoes, referred to as the AFO-footwear combination (AFO-FC). Participants were instructed not to change shoes in between measurements.

### Procedures

The AFO-FC was tuned based on a clinical protocol. Following this protocol, the participant was asked to walk up and down the walkway, while a high-speed video

camera recorded in the sagittal plane (sample frequency 50 Hz; Basler Pilot piA640-210 gc GigE, Basler, Ahrensburg Germany). Ground Reaction Force (GRF) data was collected with a force plate (sample frequency 1000 Hz; OR6-5-1000, AMTI, Watertown, USA) and synchronized with the video recordings. Data collection continued until a successful force plate strike for each foot was recorded. GRF alignment with respect to the knee and hip joints in midstance and terminal stance was immediately assessed using CMAX software (ProCare, Groningen, The Netherlands). Heel wedges were added and/or the neutral angle of the hinge was changed until maximal knee extension was achieved. Immediately after tuning (T0) and four weeks later (T1), a three dimensional (3D) gait analysis was performed. Also the stiffness of the AFO worn on the most affected leg was determined at T0 and T1.

## Measurements

Selective motor control of both legs was assessed using the modified Trost test, which measures the ability to dorsiflex the ankle and extend the knee in an isolated movement<sup>[18]</sup>. Ankle dorsiflexion and knee extension of each leg (i.e. four single joint movements) were scored as 0 (no selective, only synergistic movement), 1 (diminished selective movement) or 2 (full selective movement) and summed to a total score of 0 to 8. Subsequently, total scores were categorized into poor (total score of 0 to 2), moderate (total score of 3 to 5) or good (total score of 6 to 8) selective motor control<sup>[19]</sup>.

Gait analyses were performed in our gait laboratory. Participants were instructed to walk up and down the 10m-walkway at a self-selected comfortable walking speed. During the measurements, kinematic and kinetic data were collected using an optoelectronic motion capture system (OptoTrak 3020, Northern Digital, Waterloo, Canada) combined with a forceplate (OR6-5-1000, AMTI, Watertown, USA). Technical clusters of three markers were rigidly attached to the body segments and anatomically calibrated by probing 30 bony landmarks<sup>[20]</sup>. Trunk, pelvis, thighs, shanks and feet movements were tracked at a sample frequency of 100 Hz, while force plate data were collected at a sample frequency of 1000 Hz. For each measurement (i.e. T0 and T1), data of three successful trials of the most affected leg (i.e. foot placement within the borders of the force plate) were collected.

The stiffness of the AFO was measured using BRUCE, which is an instrument that has been found to be reliable in measuring AFO properties<sup>[21]</sup>.

### Data processing

Optoelectronic marker data and force plate data of the three trials were analyzed using custom-made software (BodyMech, [www.bodymech.nl](http://www.bodymech.nl)) based on MATLAB version R2011a (The Mathworks, Natick, USA). Initial contact and toe-off in the gait cycle of the ipsilateral leg were determined using force-plate data. Foot angular velocity was used to determine gait events of the contralateral leg<sup>[22]</sup>. Accordingly, relevant phases of the gait cycle could be determined: i) the step of the most affected leg, defined as ipsilateral initial contact to contralateral initial contact, ii) single support, defined as contralateral toe-off to contralateral initial contact and iii) midstance, defined as the moment that the malleolus marker of the contralateral leg passed the malleolus marker of the ipsilateral leg. The following spatiotemporal gait parameters were calculated and averaged over three trials: walking speed [ $\text{m}\cdot\text{s}^{-1}$ ], step length [m], single support time [s], and cadence [ $\text{steps}\cdot\text{min}^{-1}$ ].

3D joint angles of the ankle, knee, and hip joints were calculated from the optoelectronic marker data, using ISB anatomical frames<sup>[20,23]</sup>. Combined with force plate data, the joint moments were calculated using inverse dynamics<sup>[24]</sup>, expressed with respect to the proximal segment frame<sup>[25]</sup>, and normalized to body weight (measured barefoot). Also joint power was calculated. This resulted in the mean (over the 3 trials) joint kinematics and kinetics as a function of the gait cycle. Kinematic parameters that were considered relevant in the context of AFO evaluation included the minimal knee flexion angle in the single support phase [deg], the knee angle at midstance [deg] and the shank-to-vertical angle (SVA) at midstance<sup>[26]</sup> [deg]. Calculation of the SVA was based on two markers: one at the tuberositas tibiae and one at the tibia (i.e. at approximately 75% at the distal side of the shank and in line with the tuberositas tibiae in the frontal plane). The angle of the line connecting these two markers relative to the vertical in the sagittal plane represented the SVA<sup>[26]</sup>. The considered kinetic parameters included the net internal knee moment at midstance [ $\text{Nm}\cdot\text{kg}^{-1}$ ] and peak ankle power [ $\text{W}\cdot\text{kg}^{-1}$ ]. From the GRF, the forward Center of Pressure (CoP) excursion [mm] during the step of the most-affected leg was determined by continuously calculating the CoP position with respect to the position of the calcaneus marker at initial contact.

## Statistics

Descriptive statistics were used to summarize socio-demographic characteristics; disease characteristics; AFO stiffness; and the considered gait parameters. Depending on the distribution, data were described as the mean with the standard deviation (SD) (i.e. single support time, step length, SVA, knee flexion angle at midstance, minimal knee flexion angle in single support, internal knee moment and CoP excursion) or the median with the interquartile range (IQR) (i.e. walking speed, cadence and ankle power). Differences in AFO stiffness and gait parameters between T0 and T1 were analyzed with paired t-tests or Wilcoxon signed ranks tests, with a significance level set at  $p < 0.05$ . Bland-Altman plots were constructed to evaluate the individual changes with respect to the mean change. Furthermore, individual changes were related to the smallest detectable change (SDC), which was taken from studies in young healthy adults<sup>[27]</sup> (i.e. kinematic and kinetic gait parameters) and healthy children<sup>[28]</sup> (i.e. walking speed). Statistical analyses were done using SPSS version 20 (SPSS Inc, Chicago, USA).

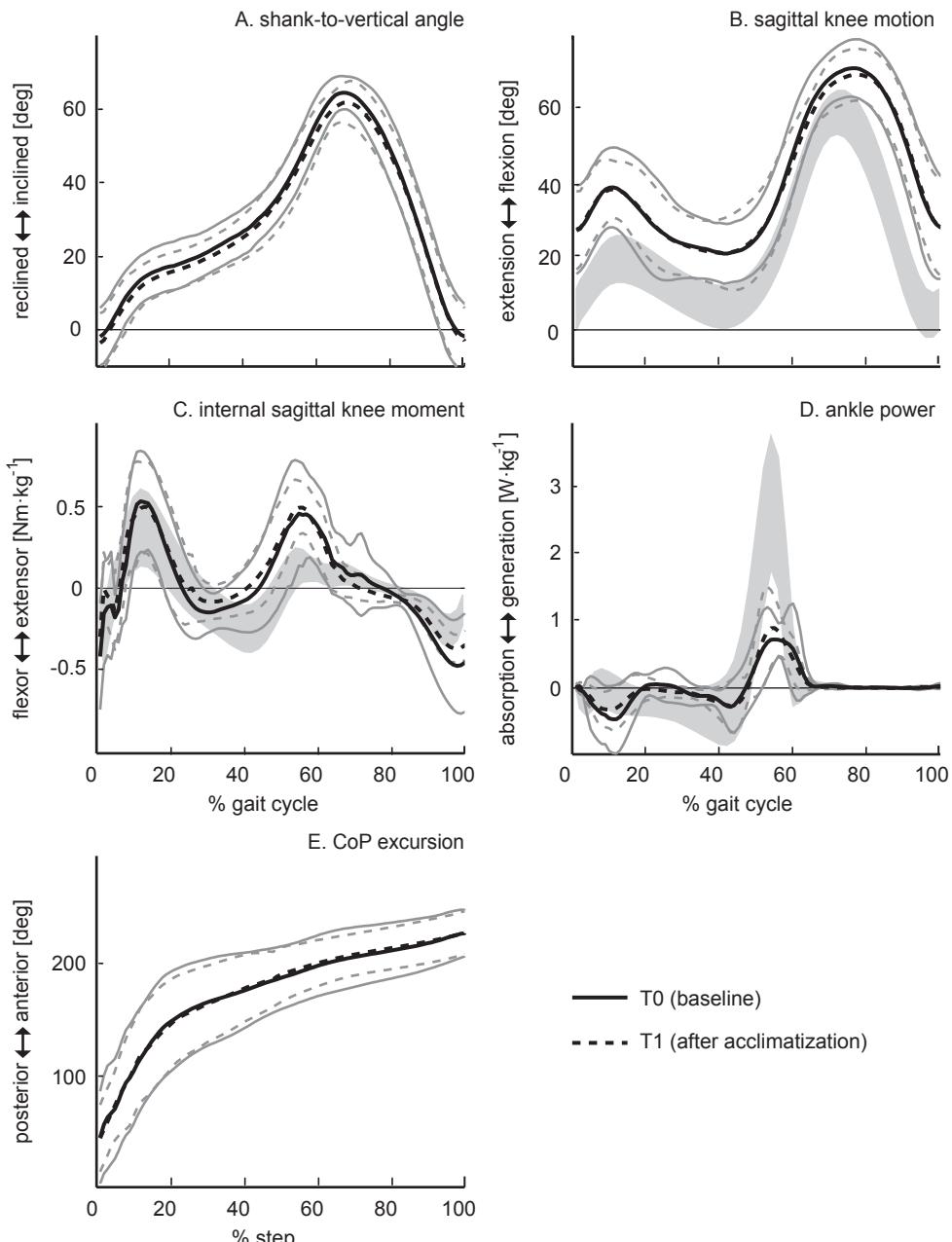
## RESULTS

For the present study, data of 10 children with spastic CP was available. Disease characteristics such as Gross Motor Function Classification System (GMFCS) level<sup>[29]</sup> and selective motor control, as well as social-demographic characteristics of these children are presented in Table 7.1.

**Table 7.1.** Mean (SD) participant characteristics

sex	[boys / girls]	8 / 2
age	[years]	10.2 (1.9)
weight	[kg]	38.6 (8.1)
height	[m]	1.41 (0.09)
limb distribution	[unilateral / bilateral]	1 / 9
GMFCS level	[I / II]	1 / 9
selective motor control	[poor / moderate / good]	1 / 3 / 6

Abbreviations: GMFCS, Gross Motor Function Classification System<sup>[29]</sup>



**Figure 7.1.** Mean (n=10) of relevant gait parameters at T0 (baseline) and T1 (after four weeks acclimatization time). Solid gray, upper and lower boundary of one standard deviation (T0); dashed gray, upper and lower boundary of one standard deviation (T1); shaded, normal walking.

Abbreviations: CoP = Center of Pressure.

Table 7.2. Mean (SD) spatiotemporal, kinematic and kinetic gait parameters at baseline and after acclimatization (n=10).

		baseline (T0)	acclimatization (T1)	$\Delta (T1-T0)$	p
<b>spatiotemporal parameters</b>					
speed <sup>a</sup>	[m·s <sup>-1</sup> ]	1.06 [0.30]	1.01 [0.22]	n/a	0.241
single support time	[s]	0.41 (0.05)	0.44 (0.06)	0.03 (0.06)	0.141
step length	[m]	0.52 (0.08)	0.50 (0.09)	-0.02 (0.03)	0.080
cadence <sup>a</sup>	[steps·min <sup>-1</sup> ]	108 [16]	108 [18]	n/a	0.333
<b>kinematic parameters</b>					
SVA at MSt	[deg]	21.7 (6.39)	20.3 (5.26)	-1.34 (4.73)	0.392
knee flexion angle at MSt	[deg]	23.1 (8.92)	22.5 (8.41)	-0.62 (5.78)	0.741
minimal knee flexion angle in SS	[deg]	18.3 (9.31)	18.8 (9.54)	0.50 (6.84)	0.823
<b>kinetic parameters</b>					
internal knee extensor moment at MSt	[Nm·kg <sup>-1</sup> ]	-0.14 (0.15)	-0.07 (0.11)	0.08 (0.13)	0.092
peak ankle power <sup>a</sup>	[W·kg <sup>-1</sup> ]	1.07 [0.56]	0.95 [0.35]	n/a	0.878
CoP <sub>exc</sub>	[mm]	185 (45)	187 (30)	-1.70 (42.3)	0.902

<sup>a</sup>Data presented as median [Interquartile Range] and analyzed non-parametrically.

Abbreviations: CoP<sub>exc</sub>, forward centre of pressure excursion; MSt, midstance; SS, single support phase; SVA, shank-to vertical angle; n/a, not applicable.

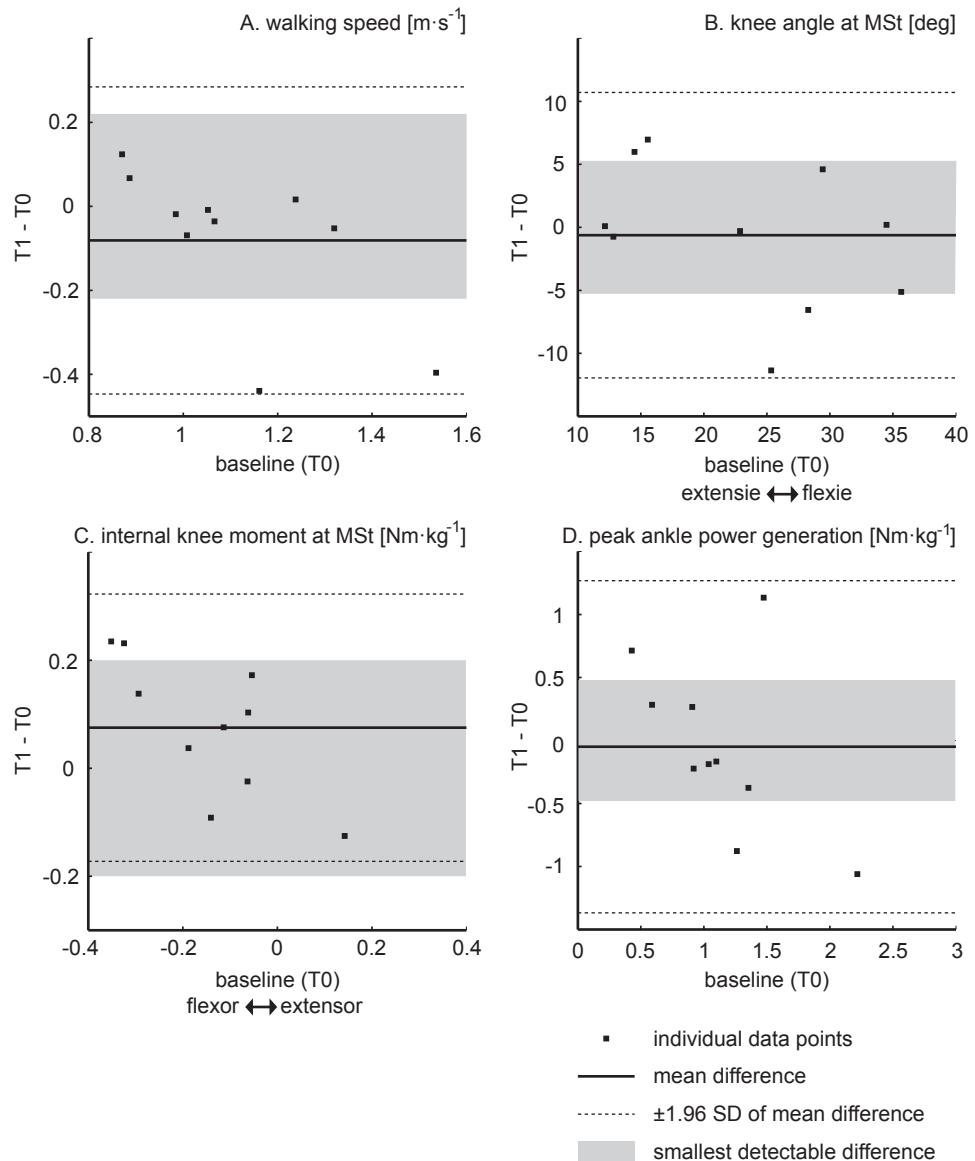
### AFO stiffness

Stiffness of one AFO could not be obtained at baseline due to technical problems. The median [IQR] stiffness of the remaining 9 AFOs at baseline was 0.88 [4.5] Nm·deg<sup>-1</sup> and did not significantly change ( $p=0.594$ ) after wearing the AFO for four weeks (0.97 [4.0] Nm·deg<sup>-1</sup>).

### Gait parameters

No significant changes were found over time in spatiotemporal parameters. The mean walking speed decreased with 5% (see Table 7.2), which was mainly caused by two individuals, who showed a relatively large decrease in their walking speed (see Figure 7.2A). The other eight participants showed only small changes in walking speed, without exceeding the SDC of 0.22 m·s<sup>-1</sup><sup>[28]</sup>.

Joint kinematics and kinetics and forward CoP excursion at baseline and after acclimatization showed equal curves over the gait cycle (see Figure 7.1), with no significant differences found over the course of four weeks (see Table 7.2). Bland-Altman plots showed no systematic changes in the knee angle at midstance, internal knee moment at midstance and peak ankle power (see Figure 7.2B-D). Yet, the change in knee angle exceeded the SDC of 5.3°<sup>[27]</sup> in half of the participants (see Figure 7.2B), while for the peak ankle power and the internal knee moment four and two participants exceeded the SDC of 0.48 W·kg<sup>-1</sup> (see Figure 7.2D), and 0.20 Nm·kg<sup>-1</sup> (see Figure 7.2C), respectively<sup>[27]</sup>. Mean forward CoP excursion during the step did not change over the course of four weeks (see Figure 7.1E).



vii

**Figure 7.2.** Figures representing individual changes in relevant gait parameters after acclimatization time (y-axis) with respect to the initial value at baseline (x-axis).

Abbreviations: MSt, midstance.

## DISCUSSION

This study in children with spastic CP showed that the gait pattern in terms of spatiotemporal parameters and joint kinematics and kinetics did not significantly change after acclimatizing for four weeks to a newly prescribed floor reaction AFO. This is in accordance with our hypothesis, and may suggest that changes in gait that occur after applying AFOs in these children are a direct biomechanical response to the imposed mechanical constraints.

To our knowledge, this is the first study evaluating acclimatization of the gait pattern to a newly prescribed AFO in CP. Accordingly, our results could not be directly compared to previous studies. A comparison of our results to the effects of AFOs versus to barefoot walking may however give some insight in the magnitude of the changes found after acclimatization relative to effects of AFOs compared to walking without AFOs, and thus the possible influence of acclimatization time on gait. As barefoot walking data were not assessed in this study, our results could be best compared to a study of Rogozinski et al.<sup>[8]</sup>, who investigated the effect of floor reaction AFOs on gait in children with CP, although in their study acclimatization time was not specified. In the Rogozinski study<sup>[8]</sup>, an increase in the mean walking speed of  $0.11 \text{ m}\cdot\text{s}^{-1}$  was found when walking with the AFO, compared to barefoot walking. This is a larger change than the mean increase of  $0.05 \text{ m}\cdot\text{s}^{-1}$  after acclimatization found in our study. Furthermore, Rogozinski et al.<sup>[8]</sup> found a decrease of  $11^\circ$  in the minimal knee flexion angle in stance, as well as a decrease of  $0.3 \text{ Nm}\cdot\text{kg}^{-1}$  in the sagittal knee moment, while we found marginal changes of  $0.5^\circ$  and  $0.08 \text{ Nm}\cdot\text{kg}^{-1}$  respectively. Although this comparison of joint kinematic and kinetic parameters may have been affected by the lower walking speed of participants in the Rogozinski study<sup>[8]</sup>, it does seem to indicate that an AFO changes gait to a greater extent immediately after application (i.e. compared to barefoot walking), than it does after acclimatization time (i.e. compared to the immediate effects). This suggests that the greater part of the biomechanical effects of an floor reaction AFO on gait in children with CP are immediately apparent and that acclimatization time may not affect gait outcome significantly in these children.

In our study, the largest mean change was found in the internal knee moment, showing a decrease of  $0.08 \text{ Nm}\cdot\text{kg}^{-1}$  after acclimatization. This change in knee moment may either be related to a change in the position of the CoP or to a change in the magnitude and/or direction of the GRF. Since the position of the CoP was similar at baseline and after acclimatization as shown by the equal CoP excursion curves (see Figure 7.1E), the change in internal knee moment is most likely related to a change in GRF characteristics. More

specifically, the decrease in internal knee moment may have been caused by a decrease of the GRF (i.e. magnitude), and/or the ground reaction vector pointing more posteriorly in the sagittal plane (i.e. direction), thus being more closely aligned to the knee rotation center, thereby decreasing the internal knee moment. The mean decrease in internal knee moment, although not reaching statistical significance, was a 50% change, which may be considered clinically relevant. However, the largest part of this mean change was caused by two participants, who showed a decrease in the internal knee moment which exceeded the SDC values as reported in healthy subjects<sup>[27]</sup> (see Figure 7.2C). Also for other gait parameters, inter-individual changes were seen, both in the magnitude as well as the nature of changes in gait parameters, as shown by the large standard deviations of the mean differences (see Table 7.2 and Figure 7.2). Together with the small sample size, this variability might, at least in part, account for the lack of statistical significance for some of the investigated parameters.

Although the fact that our study may have been underpowered must be considered, we presume that the observed individual changes as found in our study are mostly related to the variability of gait in spastic CP<sup>[30]</sup> and not to adaptation of the gait pattern within the acclimatization period. Following this hypothesis, it may be suggested that changes in gait are an immediate biomechanical response to the mechanical constraints, indicating that the muscle-timing pattern of these children is not affected by a new AFO. On the other hand, children may have adapted their muscle-timing pattern immediately to the imposed mechanical constraints. This could have been possible, as the disordered muscle activation during gait in children with spastic CP is related to the loss of selective motor control<sup>[13]</sup>, that was classified as moderate to good for the children participating in the current study. Participants may thus have been able to adapt their muscle-timing pattern immediately after applying the new AFO, therewith directly changing their gait pattern. Unfortunately, this cannot be confirmed with data on muscle timing, since these were not measured in the present study. Future studies should include such measures of muscle timing to further validate our hypothesis. Moreover, since only a selection of gait parameters was evaluated in our study, inclusion of other gait parameters should also be considered as to indefinitely determine whether or not acclimatization time is needed in the evaluation in AFOs in children with CP. Finally it must be noted that the main treatment goal of an orthosis is not always improvement of the gait pattern, but may also be aimed at prevention of joint contractures and/or increasing range of motion<sup>[1]</sup>. Although children may not need acclimatization time for the gait pattern, children with CP will need time to achieve these goals.

A limitation of this study is the small sample size of 10 children. Yet, our study population was quite homogeneous regarding GMFCS level<sup>[29]</sup>, selective motor control and barefoot walking pattern. On the other hand, the homogeneity within the study population, together with the specific type of AFO (i.e. a hinged floor reaction AFO), which was prescribed for all participants, limits generalizability of the results to all children with spastic CP.

In conclusion, the results of our study indicate that, in independently walking children with spastic CP (i.e. GMFCS level I and II), an acclimatization period of four weeks after being provided with a new floor reaction AFO does not significantly affect the gait pattern in terms of spatiotemporal parameters, knee flexion angle, internal knee moment and ankle power. This finding suggests that, in research or in clinical practice, inclusion of an acclimatization period to reliably evaluate the biomechanical effects of an AFO on gait may not be needed in this patient group. Research in a larger group of children with spastic CP and encompassing more outcome measures is needed to confirm this conclusion.

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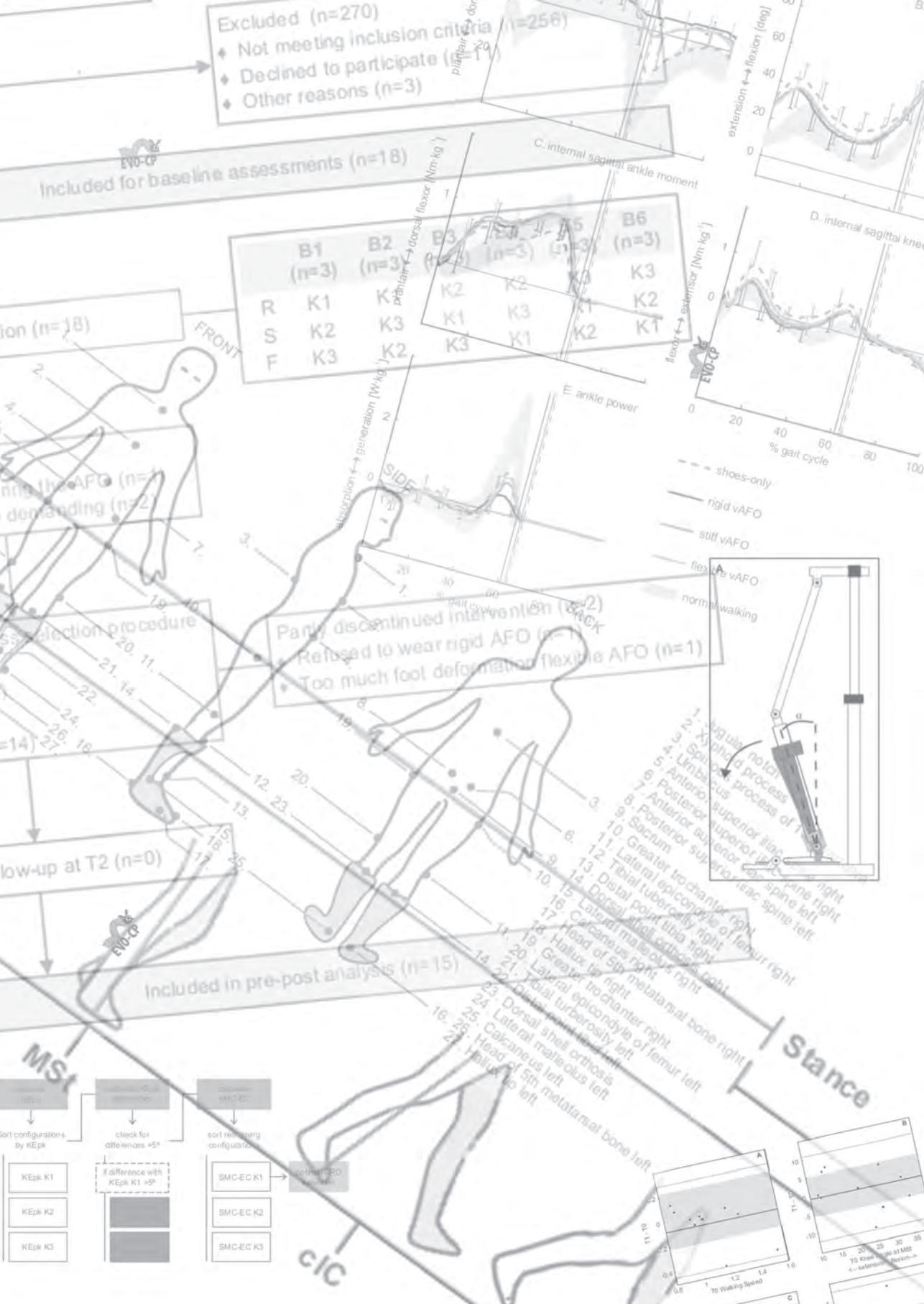
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# General discussion

In children with cerebral palsy (CP), ankle foot orthoses (AFOs) are commonly prescribed to improve gait. Ideally, an AFO should adequately normalize gait biomechanics and prevent deterioration of functions, while simultaneously maximizing the functional gain for the patient, such as improving gait efficiency or walking activity in daily life. To achieve optimal efficacy of the AFO on these two levels, the mechanical properties should match the patient's underlying impairments<sup>[1,2]</sup>. However, there is paucity in treatment algorithms on how to prescribe a well-matched AFO in children with CP. This lack of knowledge is reflected in the literature, as there is ambiguous evidence for the effects AFOs on gait, especially with respect to the functional gain for the patient, which indicates that treatment can be optimized. The general aim of this thesis was to evaluate factors that guide optimization of AFO treatment in children with CP, in order to maximize the functional benefits that can be obtained from the AFO for the individual patient. To this end, the AFO-CP trial was initiated, which aimed to evaluate the effects of different degrees of AFO stiffness on gait biomechanics and gait efficiency in children with CP who walk with excessive knee flexion during stance, in order to individually select the optimal AFO stiffness. In addition, the effects of aligning the AFO-footwear combination (AFO-FC), and applying acclimatization time to wearing an AFO-FC were investigated. In this final chapter the main findings of the presented studies are critically discussed and clinical implications and ideas for future research are provided.

## MAIN FINDINGS

### AFO alignment

In chapter III, the effect of aligning the AFO-FC on joint kinematics and kinetics and the shank-to-vertical angle (SVA) in healthy adults was evaluated. The results showed that the SVA reflected changes in joint kinematics and kinetics, controlled by changes in the AFO-FC heel height, and we concluded that the SVA could serve as a control parameter to quantify the alignment of the ground reaction force over the joint rotation centers<sup>[3-5]</sup>.

While our study did not aim to identify an optimal SVA during walking, an SVA 10-12° at midstance has been suggested as optimal<sup>[5]</sup>. Studies on alignment of the AFO-FC have however been mostly performed in children who walk with hyperextension of the knee during stance<sup>[4,6]</sup>. In chapter VII, we measured the SVA in a group of children with CP walking with excessive knee flexion during stance. In that study, a new AFO was applied, after which the heel height and AFO's neutral angle was altered to optimize the alignment. This was defined as the AFO's setting in which the ground reaction force

alignment in stance was closest to normal, which was evaluated by the representation of the ground reaction force in 2D video recordings. After this tuning process, a mean (SD) SVA of 21.1° (6.4) was found, which is remarkably higher than the proposed optimum of 10-12°. As this SVA value was accompanied by a normalization of the internal knee flexion moment ( $0.14 \text{ Nm}\cdot\text{kg}^{-1}$ ), results indicate that the proposed optimum in literature may not apply to children with CP walking with excessive knee flexion in stance. This is supported by findings of Butler et al<sup>[7]</sup>, who found that changing the heel-sole differential is ineffective in children with moderate to severe knee flexion (i.e.  $>20^\circ$ ) during barefoot walking. Moreover, healthy subjects (chapter III) showed an SVA of 17.4° (0.8) while walking with the medium heel height AFO-FC, which is comparable to the aforementioned SVA in children with CP. This SVA was however accompanied by an internal knee extensor moment of  $0.50 \text{ Nm}\cdot\text{kg}^{-1}$ , which is far from normal (i.e. internal knee flexion moment of  $\pm 0.2 \text{ Nm}\cdot\text{kg}^{-1}$ )<sup>[8]</sup>. This finding indicates that AFOs deteriorate the gait pattern of healthy individuals, which is also reported in other studies<sup>[9-13]</sup>, and it is therefore relevant to assess the potential of the SVA within the target population. Furthermore, these findings illustrate the difficulty of interpreting the SVA in relation to joint kinetics. Factors such as footplate stiffness (as shown in chapter III), footplate length<sup>[14]</sup>, and posture of the upper-body<sup>[15]</sup> interfere with the alignment of the ground reaction force with respect to the lower limb joint rotation centers. The SVA alone might therefore not be sufficient to control the alignment of an AFO-FC. The roll-over shape has been proposed as a potential parameter to quantify alignment of prostheses and orthoses<sup>[16]</sup>, also in the context of children<sup>[17]</sup>, although research on the roll-over shape in children with CP has not been performed so far.

Research in healthy adults shows that the SVA could serve as a control parameter to evaluate the effects of heel height adaptations to an AFO-FC. This should be confirmed in the target population, for example in gait patterns that are characterized by excessive knee flexion in stance. As footplate stiffness of the AFO was found to interfere with the efficacy of AFO-FC heel height adaptations on joint kinematics and kinetics, while not reflected by the SVA, other parameters should also be considered to quantify the AFO's alignment.

### AFO stiffness

#### **Effects of modulating AFO stiffness**

Another factor that is known to interfere with AFO efficacy is the AFO's stiffness. In chapter IV, we used the Bi-articular Reciprocal Universal Compliance Estimator (BRUCE)<sup>[18]</sup> to quantify the mechanical properties of a stiffness adjustable spring-hinged AFO to assess its potential use in children with CP. From these assessments we concluded that the two stiffest available springs of the spring-hinged AFO should be adequate for use in children with CP who walk with excessive knee flexion in stance. We expected that these springs could reduce the elevated walking energy cost in this target population, as the springs held sufficient stiffness to counteract the knee flexion, while the energy return could enhance push-off power. We hypothesized that knee flexion would decrease with increasing stiffness, while push-off power would be more enhanced by the more flexible (spring-like) AFOs. As both mechanisms are associated with walking energy cost reduction, the cost stiffness that would result in the largest energy reduction was expected to rely on a trade-off between counteracting knee flexion and enhancing push-off power.

In chapter V, we evaluated the effects of varying the stiffness of a spring-hinged ventral shell AFO on gait in a group of children with CP walking with excessive knee flexion in stance. To this purpose, the spring-hinged AFO was set into a rigid hinge setting (i.e. no spring-like properties) and two spring-like hinge settings (i.e. stiff and flexible), which were randomly applied to the participants. For each hinge setting, the effects on gait biomechanics (i.e. knee extension angle and push-off power) and walking energy cost were assessed. In contrast to our hypotheses, no significant differences in knee extension angle during stance were found between the three AFOs (i.e. rigid, stiff and flexible). All AFOs also showed comparable improvements in the internal knee flexion moment, although the rigid AFO reduced the internal knee flexion moment more effectively compared to the flexible AFO. Considering the reducing effect of a stiff footplate on the knee flexion angle and internal moment<sup>[14]</sup> as described in chapter III, it is expected that the AFOs' effects on the knee joint angle and moment in all hinge settings was primarily defined by the stiff footplate, and less by the AFOs' ankle stiffness.

Although variations in AFO stiffness did not significantly affect knee joint kinematics and kinetics, differences between AFOs were seen at the level of the ankle joint. The spring-hinged AFO was able to control ankle range of motion according to its settings, showing a decreased ankle range of motion with increasing stiffness. Moreover, our

hypothesis that a spring-like AFO could preserve remaining push-off power<sup>[6,19]</sup> (chapter IV) was confirmed by the results in chapter V, showing that both the stiff and flexible spring-like AFOs preserved ankle power generation compared to shoes-only walking, while the rigid AFOs reduced the ankle power generation. Previously, adequate ankle power generation (third rocker function) has been associated with a rapid plantar flexion movement<sup>[20]</sup>. Although we did not investigate any relations between gait parameters, our results suggest an inverse association between ankle range of motion and push-off power generation. The possibility of sufficient ankle plantar flexion within the spring-like AFOs could therefore be a key feature with regard to preserving push-off power, especially in children without severe calf muscle weakness who are able to actively perform plantar flexion movement<sup>[21]</sup>.

The spring-hinged AFOs were also expected to enhance push-off power by (partly) taking over ankle work during gait through the storage and release of energy by the AFO<sup>[22,23]</sup>. In general, the amount of stored energy by the AFO is dependent on an interaction between the AFO's ankle range of motion and ankle stiffness<sup>[22]</sup>. In our study, the dorsiflexion stop and relatively low stiffness values of the spring-hinged AFO limited the amount of energy that could be stored by the AFO. The hysteresis that was present in all hinge settings further decreased the amount of released energy that was expected to enhance push-off power during walking. These low levels of energy return were reflected by the small contribution of the AFO to ankle work during walking in all hinge settings (chapter V). Nonetheless, over the whole gait cycle, the rigid AFO contributed less to ankle work compared to the spring-like AFOs, emphasizing favorable effects of the spring-like AFOs on ankle biomechanics, which we suggested would also be beneficial in terms of reducing walking energy cost<sup>[21,23-26]</sup>.

Regarding walking energy cost, the rigid AFO as well as the two spring-like AFOs significantly reduced the walking energy cost compared to walking with shoes-only, with an overall reduction of 11%, which is comparable to earlier findings<sup>[27,28]</sup>. In contrast to our expectations and to earlier findings<sup>[25,29]</sup>, we did however not find any significant differences in walking energy cost between AFO stiffness conditions. That is, the favorable effects of the spring-like AFOs on ankle range of motion and push-off power were not unambiguously reflected in a reduction of walking energy cost. Considering that all AFOs comparably improved the knee extension angle during stance and based on findings of Brehm et al.<sup>[28]</sup>, showing that normalization of knee extension after treatment with AFO's in children with CP was associated with a reduced walking energy cost, it is therefore suggested that energy cost reductions as a result of wearing the AFO were

mostly defined by the AFOs' effect on knee biomechanics, and less by their effect on ankle biomechanics (chapter V). On the other hand, the difference in stiffness between AFOs may not have been discriminating enough to detect changes in the walking energy cost. Furthermore, as the number of included patients did not reach the goals that were set, our small study sample may also explain the lack of significant differences in walking energy cost between AFOs.

### ***Effects of a stiffness-optimized AFO***

The results of the gait analyses and walking energy cost assessments reported in chapter V were used to individually optimize AFO stiffness in our study population (chapter VI). This optimization process was initially based on the AFOs performance on the knee extension angle during stance. When no difference in effect on knee angle was found between AFOs, the optimal AFO stiffness selection was based on the reduction in walking energy cost, i.e. the stiffness showing the largest reduction was chosen as optimal AFO stiffness. In chapter VI, the effects of the optimized AFO on walking energy cost, and gait biomechanics were investigated.

In line with the findings in chapter V, performance on peak knee extension angle was not a discriminating factor to select the optimal AFO stiffness in most participants. When individually comparing the peak knee extension angle between AFOs (chapter VI), the rigid AFO only resulted in the best (i.e. at least 5° more extension) knee angle in 1 of 29 evaluated legs. Even more striking, the knee angle was least improved (i.e. >5° more flexion) by the rigid AFO in 13 of 29 evaluated legs. Previously, Rogozinski et al.<sup>[30]</sup> aimed to identify clinical parameters that were related to efficacy of a rigid AFO on gait biomechanics, and found that hip and knee contractures negatively impact on efficacy. As we excluded children with knee flexion contractures of more than 10°, this could not have affected our results. Clinical observations (2D video recordings) however showed that some children tried to walk on the tip of the toes (i.e. footplate), therewith avoiding stretching of the calf muscles (i.e. knee extension), which could (partly) explain the fact that the rigid AFOs did not improve knee kinematics in some children. The rigid AFO might also have induced a flexed knee gait pattern to compensate for a reduced balance control, caused by the rigid AFO<sup>[13]</sup>. In contrast to our findings of chapter V, the results of chapter VI suggest that spring-like AFOs may be more effective in achieving knee extension compared to rigid AFOs in some children with CP who walk with excessive knee flexion in stance. The optimized AFO however reduced the peak knee extension

angle in single support by  $2.4^\circ$ , which is a much smaller reduction compared to Rogozinski et al.<sup>[30]</sup> who found a mean reduction of  $11^\circ$ . Nonetheless, based on our findings, the rigid AFO is expected to have been even less effective in our study population, supporting the hypothesis that the efficacy of an AFO is (partly) defined by the patient's characteristics.

As the knee angle was not a discriminating parameter in most children, the majority was assigned with the AFO that resulted in the lowest walking energy cost level. Within this selection process, only spring-like AFOs (either stiff or flexible) were selected as optimal stiffness. This finding supports that functional benefits from spring-like AFOs can be obtained for a subgroup of children. The optimized AFO improved the walking energy cost at 3 months follow-up in 11 participants, although not significantly, with an overall walking energy cost reduction of 9% compared to walking shoes-only. In line with earlier findings<sup>[25,29,31]</sup>, our results suggest that optimizing AFO stiffness could lead to a clinically relevant improvement in the walking energy cost at the individual level, especially when the patient's main rehabilitation treatment goal is to reduce fatigue related to an increased walking energy cost.

Although the optimized AFO led to small and/or statistically non-significant changes at follow-up, larger effects were found at the moment of selecting the optimal AFO stiffness. At that stage, the optimized AFO reduced the knee flexion angle by  $8^\circ$ , which led to a knee flexion angle at stance that was close to normal (i.e.  $14.3^\circ$ ) and comparable to findings of Rogozinski et al.<sup>[30]</sup>. This is a clinically relevant reduction in terms of improving the gait pattern, and preventing muscle contractures. As a mean overall reduction in knee flexion angle of approximately  $4^\circ$  by all AFOs was found in chapter V, our findings in chapter VI indicate that individually optimizing AFO stiffness could improve AFO treatment efficacy in terms of knee angle during walking, and accordingly, preventing muscle contractures. Furthermore, results showed that the individually optimized AFO initially reduced walking energy cost by 20%, indicating an overall clinically relevant reduction in the walking energy cost. The reduced AFO's efficacy might be explained by a deterioration of the AFO's mechanical function over time. As it is expected that the AFO's efficacy is dependent on the match between the AFO's mechanical properties and patient characteristics, growth and development of the child may also have a significant impact on the efficacy of an optimized AFO at follow-up. In other words, the optimization of AFO stiffness may be time specific and should therefore be reconsidered when the patient's characteristics change over time.

While rigid AFOs are most commonly used in children with CP who walk with excessive knee flexion, our results show that a rigid AFO design is not necessarily required to improve knee extension in these children. In fact, spring-like AFOs are equally effective in achieving knee extension, and can even be more effective in a subgroup of children with CP who walk with excessive knee flexion during stance. Spring-like AFOs also preserve remaining ankle push-off power, as opposed to rigid AFOs, which is likely to be related to the ankle range of motion, which is less obstructed by hinged spring-like AFOs. Although our results did not unambiguously show that the favorable effects of spring-like AFOs on ankle push-off power lead to larger energy cost reductions compared to rigid AFOs, they suggest that individually optimizing AFO stiffness could be beneficial in terms of reducing walking energy cost a subgroup of children with CP. The variety in the assignment of an optimal stiffness emphasizes an individual approach to AFO prescription in children CP who walk with excessive knee flexion in order to maximize treatment efficacy in these children.

### Acclimatizing to an AFO

In chapter VII, we evaluated whether treatment efficacy might also be affected by incorporating acclimatization time for the gait pattern to adapt to the mechanical constraints of a newly applied AFO. As comparable effects of the AFOs on specific gait biomechanics before and after acclimatization were found, we concluded that acclimatization time is not required to reliably assess the AFO's effects on gait biomechanics (chapter VII). This finding could be explained by different mechanisms, which are related to the nature of the response to the newly prescribed AFO, i.e. whether muscle activation is affected by the applied AFO. As studies on the effects of AFOs on muscle timing and activation show mixed results<sup>[32-34]</sup>, it was unclear whether the muscle activation pattern is affected by AFOs in children with CP.

We hypothesized that an acclimatization time would be required when children needed time to adjust their muscle activation (i.e. motor learning) to the mechanical constraints as induced by the AFO. As no changes in biomechanical parameters were seen after acclimatization in our study, we suggested that children show a biomechanical response to newly applied AFO (i.e. no motor learning). This may suggest that muscle activation is not affected by the applied AFO. On the other hand, the response to the

AFO may also be related to muscle force and the level of selective motor control of the children. A mechanical constraint induced by the AFO may only lead to improvement of gait when the patient has the ability (e.g. sufficient motor control) to adapt his/her posture and movements accordingly. For example, a rigid AFO applies a force at the tibia to reduce knee flexion, but a sufficient amount of muscle force of the hip extensors is needed to get the patient in an upright position. Likewise, a spring-like AFO may preserve push-off power, but this can only result in a larger step length, and accordingly an increased walking speed, when the patient has the control to flex the hip and extend the knee joint simultaneously at the end of the swing phase. Although no specific inclusion criteria regarding selective motor control or muscle force were defined for study participation, the majority of subjects in our study had moderate to good selective motor control and sufficient muscle force. Children might therefore have been able to immediately adapt their gait pattern (and muscle activation) to the mechanical constraints of the AFO. As such, children with lower levels of motor control might need time to adapt their gait pattern to the mechanical constraints of the AFO. To confirm the exact nature of the response to a newly applied AFO, research on AFOs should include assessments of muscle activation.

Acclimatization time to adjust to the constraints as induced by an AFO is not required when assessing the effects of a new AFO on gait biomechanics in children with CP who walk with excessive knee flexion in stance.

## METHODOLOGICAL CONSIDERATIONS

### Study population

So far, many studies on AFO efficacy in CP included patients presenting with a variety of gait patterns without tuning of the AFO's design or materials to specific gait deviations<sup>[1,35]</sup>. However, to enable a fair evaluation of the effects of AFOs at a group level, it is important to select a homogeneous group of children with CP, for which the same type of orthosis is prescribed. The in- and exclusion criteria of the AFO-CP trial were formulated such that a homogeneous study population could be created, which included children diagnosed with spastic CP with a gait type characterized by excessive knee flexion. Furthermore, participants were mostly classified as GMFCS II, and showed a relatively high baseline (i.e. shoes-only) walking speed of  $0.98 \text{ m}\cdot\text{s}^{-1}$ , indicating that participants were moderately affected. Although this homogeneity is considered a major strength of the study, it limits the generalizability of the results to CP in general. In other words, our results might not be applicable to children with other types of CP or children who are presented with other gait deviations.

Despite the homogeneity of our study population, various individual responses to the AFO on gait were found (chapter V, VI and VII), reflected by relatively large standard deviations around the means. Moreover, results of chapter V showed that one or more of the AFOs did not reduce the walking energy cost in some children. While the AFO-CP study initially aimed to identify factors that could predict AFO efficacy, as has been found for other interventions<sup>[36-41]</sup>, our sample size was too small to perform such an evaluation. Nonetheless, the results of chapter V indicate that children who show higher baseline walking energy cost values show the largest energy cost improvements as a result of walking with AFOs. This is likely related to the fact that energy cost levels closer to normal can be less reduced compared to higher levels. Nonetheless, specific patient characteristics, such as spasticity, selective motor control and physical fitness, possibly underlying the higher baseline walking energy cost levels, could also be related to the AFO's efficacy in children with CP.

### Mechanics of an adjustable AFO

The spring-hinged AFO as used in the AFO-CP study allowed the AFO's stiffness to be varied without the process of making a completely new orthosis. This was considered an important advantage, as various possible confounding variables (e.g. differences in footplate length or stiffness, neutral angle) could be controlled, and problems with

fitting the AFO were kept to a minimum. However, the specific mechanical properties of the spring-hinged AFO, as described in chapter IV, implicate a different mechanism during walking compared to other AFO designs that hold spring-like properties (e.g. dorsal leaf spring AFOs without a hinge at the ankle).

Bregman et al.<sup>[18,23]</sup> quantified the stiffness of carbon fiber dorsal leaf spring AFOs, showing that the relation between ankle angle displacement and exerted net moment is linear. In contrast, the springs of the spring-hinged AFO hold a threshold, indicating that a certain force is needed to get the spring in its elastic (linear) range. After exceeding the threshold, the properties of the AFO change according to the stiffness of the spring (see Figure 4.2). As the AFO's exerted net moment during gait is dependent on the stiffness and the deflection angle of the ankle, higher net moments can be achieved with stiffer springs within a certain range of motion. Considering the fairly low stiffness and limited range of motion of the spring-hinged AFO, a threshold was required to achieve sufficient moments from midstance onwards without exceeding normal ankle range of motion. In addition to the non-linear behavior within one movement direction, the properties towards dorsiflexion and plantar flexion can be independently adjusted within the spring-hinged AFO, while carbon fiber dorsal leaf spring AFOs showed a similar stiffness towards both these movement directions. The mechanical functioning of the spring-hinged AFO towards both movement directions has the advantage that it may enhance first and third rocker function, without compromising the AFO's effect on the second rocker. As such, hampering effects of AFOs on ankle parameters can be minimized using adjustable AFOs, e.g. the spring-hinged AFO. Furthermore, adjustable (stiffness) characteristics within an AFO allow individual tuning to the gait pattern, which is promising considering the significance of tuning according to the patient's specific impairments.

The thresholds and stiffness values as described in chapter IV appeared to be too low for optimal performance. While we hypothesized that the threshold of the spring-like AFOs could prevent excessive ankle dorsiflexion in the beginning of the stance phase up to  $0.5 \text{ Nm}\cdot\text{kg}^{-1}$ , the AFOs only prevented dorsiflexion until  $0.3\text{--}0.4 \text{ Nm}\cdot\text{kg}^{-1}$ . Moreover, the flexible and stiff AFOs could reach maximum net moments of 18 to 27 Nm, while the mean weight of the participant indicates a maximum ankle plantar flexion moment of approximately 44 Nm. This was reflected by the gait biomechanics, showing that the children used the complete range of motion that was allowed by the settings of the hinge, indicating that the second rocker was primarily controlled by the AFO's dorsiflexion stop, and less by its stiffness. Accordingly, it is expected that much higher stiffness values are

needed to counteract excessive dorsiflexion in these children when range of motion is not controlled by a dorsiflexion stop. Assuming that a maximum dorsiflexion angle of 15° during stance is accompanied by a net ankle moment of 1.2 Nm·kg<sup>-1</sup>, a child of 35 kg would need an AFO holding a stiffness of 2.8 Nm·deg<sup>-1</sup> (assuming linear behavior). Lower stiffness values could also be considered in combination with a sufficient threshold.

### Design and measurements

#### Study design

The design of the AFO-CP trial (chapter II) was based on a baseline measurement and a post-intervention measurement where participants served as their own control (i.e. a single-subject design). The use of such a design has been advised as it may overcome the difficulty of the heterogeneity of CP and could control for confounding factors<sup>[1,35,42]</sup>. In addition, heterogeneity in our study population was minimized by applying very strict in- and exclusion criteria. This was necessary to prescribe a similar AFO design and apply a general stiffness optimization process to all participants. Although the homogeneity in our study population can be considered as a major strength, our strict inclusion criteria led to a small sample size that was only half of our estimated sample size (chapter II). Most children were excluded based on age and gait pattern.

The study design included an extensive set of outcome measures, of which some were repeated multiple times. As such, participation to the AFO-CP trial was physically demanding for all participants and their parents. The protocol may therefore have introduced a selection bias, as only very motivated AFO users might have been willing to participate in the study. This is supported by the fact that only two participants dropped out of the study because the measurements were too demanding.

Including walking with shoes-only as baseline walking condition is another strength of our study design. In doing so, we avoided that effects of the footwear were attributed to the AFO<sup>[1]</sup>. The shoes that participants wore at baseline were however different from those used while walking with AFOs, as these are mostly a few sizes larger to fit the AFOs. Nonetheless, effects of these differences are considered marginal.

### ***Walking energy cost measurements as optimization criterion***

The energy cost of walking is dependent on a person's age, weight and height<sup>[43-45]</sup>, making it difficult to interpret and compare walking energy cost outcomes between or within children<sup>[46]</sup>. To reduce confounding effects, we used the normalization scheme of Schwartz et al.<sup>[46]</sup> for the selection of the optimal AFO stiffness. This normalization scheme results in a non-dimensional speed outcome and a net non-dimensional energy cost outcome, where the energy cost was expressed as a percentage of speed-matched control cost<sup>[47]</sup>. This is considered to be the most appropriate outcome parameter to compare the walking energy cost of different walking conditions among children. Nonetheless, walking energy cost measurements are subject to a relatively large day-to-day variability, especially in children with CP<sup>[48,49]</sup>. Subsequently, the variations in walking energy cost responses that were measured as a result of modulating AFO stiffness in chapter V did not always exceed the smallest detectable change of walking energy cost assessments<sup>[49]</sup>. Variability in, for example, resting energy consumption values may have introduced variations that were incorrectly attributed to the AFO. As the selection of the most optimal AFO stiffness was based on energy cost measurements in the majority of participants, regardless of the magnitude of variety in intra-individual energy cost responses, the optimal AFO stiffness could thus have been incorrectly selected in some participants. Nonetheless, the individually optimized AFO (chapter VI) did lead to a decrease in walking energy cost in 11 out of 14 participants, indicating improvement in this outcome in a much larger part of the study population compared to that found in other studies<sup>[27,28,50,51]</sup>. In contrast to current clinical practice, in which gait biomechanics primarily define AFO prescription, our results emphasize the use of walking energy cost assessments to optimize AFO prescription, although multiple assessments within patients should be considered to account for the variability<sup>[52]</sup>.

### ***3D gait analysis measurements in shod conditions***

Measuring joint kinematics with 3D gait analysis in a barefoot condition has been found to be reliable in healthy subjects<sup>[53]</sup> and patients with CP<sup>[54,55]</sup>. However, accurately measuring foot and ankle kinematics in a shod condition is a challenge, and becomes even more difficult when combining the shoes with AFOs<sup>[56]</sup>. The inaccuracy of the measurements became apparent when interpreting the ankle angle while wearing the rigid AFO (chapter III and V). While the rigid AFO aimed to fix the ankle in a neutral position, a mean ankle range of motion of 7° was measured (chapter V), which has also

been found by other studies measuring ankle kinematics while wearing solid or rigid AFOs<sup>[30,50,57]</sup>. This ankle movement could be attributed to deformation of the AFO<sup>[57]</sup>, but movements of different components of the AFO-FC and foot may also explain the apparent range of motion. In this context some limitations related to 3D gait analyses for assessing ankle kinematics in shod and AFO conditions should be considered.

First, the biomechanical model used in our gait analyses does not allow separate measurement of movements of the foot, AFO, and footwear. This limitation has been acknowledged in the literature before<sup>[56]</sup>. Nonetheless, we assumed that the effects of movements of different components of the AFO-FC on ankle kinematics were mostly negligible in our studies. The custom-made AFOs fitted closely to the patient's foot, and the foot part of the AFO aimed to correct foot deformations, making it plausible that movements of the foot within the AFO were minimized. To check for movements of the AFO relative to footwear, we compared the trajectory of the virtual calcaneus bone marker (i.e. probed on the shoe) expressed in the coordinate system of the foot, based on the (technical) foot cluster to the trajectory of the same marker calculated from the coordinate system of the shank (including the AFO). As these trajectories largely overlapped, movements between the shoe and the AFO were assumed negligible.

$$\begin{pmatrix} \cos(\alpha) & -\sin(\alpha) & 0 & 0 \\ \sin(\alpha) & \cos(\alpha) & 0 & h \\ 0 & 0 & 1 & 0 \\ 0 & 0 & 0 & 1 \end{pmatrix} \quad \text{Equation 8.1}$$

As a second limitation, the marker cluster of the foot was attached to the shoe, while the bony landmarks of the foot were probed on the shoe, assuming the foot, the AFO's foot and the shoe to be one rigid segment. The coordinate system of the foot is conventionally constructed by a horizontal alignment of the bony landmarks on the calcaneus and the metatarsal joints I and V in the sagittal plane. The AFO-footwear combination however prevents the exact localization of the anatomical structures of the foot, but can be approximated using projections of the anatomical structures on the shoe. Obviously this introduces some random errors. However, the anatomical coordinate system of the foot in a shod condition also shows an offset that is strongly determined by the shoe's heel-sole differential and the height of the shoe sole (chapter III). To correct for these offsets in defining the coordinate system of the shod foot, we developed the Vertical Inclinometer on a Rail (VICTOR). This device can be used to

measure the heel sole-differential and heel height of the AFO-FC (see Figure 3.1). The coordination system of the foot can be defined by applying a transformation matrix of the shoe (see Equation 8.1), including the inclination angle caused by the heel-sole differential ( $\alpha$ ) and heel height ( $h$ ), to the coordinate system of the shoe (i.e. shod foot). Although the use of VICTOR will increase the accuracy of calculating ankle flexion-extension angles during gait, it relies on assuming that effects of the AFO-footwear combination can be described in the sagittal plane.

## FUTURE RESEARCH DIRECTIONS

- The study in chapter III showed that the SVA could serve as a control parameter to evaluate heel height adjustments applied to an AFO-FC in healthy adults. Future research should focus on the association between the SVA and joint kinematics and kinetics in pathological gait to assess the potential of the SVA within the target population. Furthermore, the potential of other parameters to quantify the alignment of the AFO-FC, e.g. the roll-over shape, should be evaluated in gait of children with CP.
- Our results suggest that the improvement of the internal knee joint moment as a result of wearing an AFO are primarily defined by the length and stiffness of the footplate, and less by the AFO's stiffness around the ankle joint (chapter III and chapter V). This should be confirmed in studies aiming to investigate the effects of different footplate characteristics on gait in children with CP.
- Although we could not show significant differences in walking energy cost between rigid AFOs and spring-like AFOs on a group level (chapter V), specific changes in gait biomechanics are expected to underlie changes in walking energy cost<sup>[28]</sup>. Unfortunately, our sample size was insufficient to directly investigate such associations. Future research should aim to unravel the relation between changes in gait biomechanics and the change in walking energy cost after treatment with an AFO in children with CP to get more insight in underlying working mechanisms of AFOs, i.e. insight in how an AFO should improve gait biomechanics in order to maximize the functional gain for the patient. Such research should include a large sample of children with CP, presenting with a wide variation in biomechanical gait characteristics.

- While our results illustrate that current techniques within the field of orthotics do not yet completely meet the requirements for optimal AFO performance (chapter IV and chapter V), adjustable AFO's enable better tuning of the AFO's mechanical properties to the patient's (gait) impairments and are therefore promising to improve AFO efficacy in term. Exact tuning of the threshold and stiffness according to body weight could improve the AFO's effects on the ankle joint kinematics and kinetics, although this should be confirmed by research including a larger variety in AFO stiffness degrees, specifically stiffness degrees between our stiff ( $\pm 1.6 \text{ Nm}\cdot\text{deg}^{-1}$ ) and rigid AFO ( $3.8 \text{ Nm}\cdot\text{deg}^{-1}$ ).
- Our results showed that the ankle range of motion in combination with the spring's energy return could improve knee flexion, while the hampering effects on ankle push-off power were minimized. Future research should aim to unravel the association between ankle range of motion, push-off power, AFO contributions, and the gait efficiency to provide further directions for technical developments as to improve the mechanical functioning of AFOs to maximize gait efficiency. To this purpose, the effects of different AFO settings or designs (e.g. using carbon-fiber AFOs, damping hinges without spring-like properties, and variations in ankle range of motion) on ankle parameters, in relation to changes in walking energy cost should be investigated.
- Within our AFO optimization process and its evaluation, we mainly focused on lower limb biomechanics, and its relation to the walking energy cost. Although the primary aim of an AFO is to improve lower limb biomechanics during walking, movements of other segments (e.g. the trunk and arms) may however interfere with the AFO's efficacy on walking energy cost. For example, a rigid AFO may improve knee extension, while negatively impacting on balance control<sup>[13,58]</sup> for which a subject has to compensate, therewith increasing walking energy cost. In other words, an AFO that is optimized based on lower limb biomechanics may not be optimal on all gait-related features. Moreover, the AFOs in our study (chapter VI) were specifically optimized for walking, while effectiveness for other relevant daily life activities, e.g. running, standing and walking the stairs, should also be considered. Future research should therefore focus on the AFOs effects on the body as a whole and on effectiveness of AFOs on other activities than walking.
- Our results showed that specific AFO properties could be beneficial in terms of gait biomechanics and/or the energy cost of walking in subgroups of children with CP. It is expected that specific patient characteristics underlie the (in)efficacy of AFOs in some children. Future research in a larger sample of children with CP is needed to identify factors

that can predict the efficacy of an AFO. Furthermore, our study only included children with spastic CP walking with excessive knee flexion, therewith limiting generalizability to CP in general. Future studies should focus on children with CP presenting other gait patterns, to improve AFO treatment efficacy for the CP population as a whole.

## CLINICAL IMPLICATIONS

- Our results indicate that spring-like AFOs improve ankle biomechanics more effectively compared to rigid AFOs in children with CP who walk with excessive knee flexion, while they comparably improve knee biomechanics in children with CP who walk with excessive knee flexion in stance. Clinicians should therefore consider to prescribe spring-like AFOs in this specific group of children with CP. When prescribing a spring-like AFO for this population, it is essential to use a stiff footplate to ensure adequate improvements of the knee angle and moment during stance.
- The various responses to the different degrees of AFO stiffness emphasize an individual approach to AFO prescription in children with CP, which should be guided by proper evaluations of the AFO's effects on gait. An extensive evaluation on multiple aspects of gait, such as performed in the AFO-CP trial, will mostly not be feasible in clinical practice. Especially since such evaluations should be repeated for new AFO prescriptions as personal and disease characteristics may have changed over time (e.g. growth, other applied interventions). However, it is emphasized to evaluate the AFOs' efficacy on a selection of outcome parameters that are significant in the context of the clinical indication of the prescription. When an AFO is primarily prescribed to reduce the energy cost of walking for example, an evaluation of different degrees of AFO stiffness on this outcome measure may maximize the treatment efficacy. It is not feasible to evaluate multiple AFO stiffness levels in each patient in clinical practice. Therefore, the efficacy of AFO prescriptions should be continuously evaluated on multiple outcome measures in clinical practice. A structured, long-term evaluation may enable to identify key features that could guide the prescription process, which could improve the efficacy of AFO prescription in CP.
- Our results showed that an extensive acclimatization period to a newly prescribed AFO is not required to reliably assess its effects on gait biomechanics. For clinical practice, individually tuning the AFO's mechanical properties to the patient's characteristics, based on an immediate evaluation of the effects on gait as assessed in the laboratory, is therefore advised.

- As the alignment of the AFO-FC is essential for adequate mechanical functioning, an evaluation of this alignment using gait analysis is advised. Although the SVA could be used as a parameter to evaluate the alignment of the AFO-FC, the use of the representation of the ground reaction force in the process of AFO alignment is emphasized in children with CP who walk with excessive knee flexion.
- Unambiguously quantifying the AFO's mechanical properties (e.g. stiffness) during the process of prescribing an AFO could facilitate orthotists and physicians in their decision-making process, and may serve as a quality check of the prescribed AFO. This will contribute to improving AFO prescription in children with CP in term.

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## Summary

Maximizing the efficacy of ankle foot orthoses in children with cerebral palsy

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Cerebral palsy (CP) is the most common cause of children's disability in Western Europe. Children with the spastic type of CP, which is the most common type of motor disorders, show impairments such as spasticity, muscle weakness, and a decreased selective motor control. These impairments lead to decreased motor function, and accordingly, these children experience gait-related problems.

The gait pattern of children with spastic CP is frequently characterized by specific gait deviations, which can be categorized into different gait types according to the classification of Becher. This thesis focusses on the children presenting with gait types 4 and 5, i.e. a gait pattern that is characterized by excessive knee flexion in (mid) stance. The excessive knee flexion during walking is usually accompanied by abnormal hip and ankle kinematics and kinetics, implying impaired biomechanical function. The gait deviations in CP are associated with an increase of energy consumption during walking. This especially applies to children with CP who walk with excessive knee flexion in stance, as these gait patterns are particularly energy consuming. To minimize the increased energy consumption, patients often decrease their walking speed. This leads to an increased walking energy cost (i.e. energy consumption per distance), reflecting poor gait efficiency. Although the nature of the association between underlying biomechanical gait deviations and the increased energy consumption in children with CP is not yet unraveled, abnormal knee and ankle kinematics and kinetics are considered key features.

To counteract the gait deviations, an ankle foot orthosis (AFO) is a commonly applied rehabilitation intervention in children with CP. AFOs apply a mechanical constraint to the ankle and foot, either to compensate for a loss of function, or to counteract an excess of function. As such, an AFO can directly control the ankle and knee joint motion, and dependent on its design, it may also affect the hip joint. Although the effects of AFOs on gait in CP have been frequently investigated, the results are inconclusive. Some studies report improvements of gait in terms of gait biomechanics, and/or efficiency, while other studies show that AFOs can have no effect or even detrimental effects in some children. Several factors may underlie the ambiguous results with regard to AFO efficacy, of which some are discussed in this thesis. The general aim of this thesis was to evaluate factors that enable an individual optimization of AFO prescription in order to maximize AFO efficacy in children with CP who walk with excessive knee flexion in stance.

Chapter II describes the protocol of the AFO-CP trial. This study aimed to optimize Ankle-Foot Orthoses for children with CP who walk with excessive knee flexion to improve their mobility and participation. One of the problems underlying the ambiguous

results within AFO research concerns the outcome at which the AFOs efficacy is being assessed. In order to prescribe a well-matched AFO, its effects should be evaluated on outcome measures at multiple domains of the International Classification of Functioning, disability and health (ICF) framework. Assessing the AFOs efficacy on multiple domains of the ICF framework could reveal mutual relations, which may give insight in the underlying working mechanisms of AFOs in CP. The protocol of the AFO-CP trial existed of an extensive evaluation of the effects of different degrees of AFO stiffness on gait-related outcome, covering all ICF domains, in children with CP who walk with excessive knee flexion in stance.

Another factor that might affect the AFOs efficacy, is the alignment of the ground reaction force with respect to the joint rotation center while walking with AFOs. This alignment is dependent on the properties of the AFO-footwear combination (AFO-FC), such as heel height. The shank-to-vertical angle (SVA) has been proposed as a relatively simple outcome parameter to quantify the alignment of an AFO-FC. In chapter III, we used an instrumented treadmill to investigate the effects of manipulations of heel height and footplate stiffness of an AFO-FC on both the SVA, and lower limb joint flexion-extension angles and net moments at midstance. To this purpose, ten healthy individuals walked with bilateral rigid AFO-FCs. We manipulated heel height in three conditions, which were controlled by an imposed SVA of 5°, 11° and 20°. These heel height conditions were combined with either a flexible, or a stiff footplate, resulting in six different walking conditions. We found that the SVA is responsive to changes in heel height, and less to changes in footplate stiffness. The increase in SVA resulted in concomitant changes in lower limb flexion-extension angles and internal net moments, especially at the level of the knee joint. As such, the results supported the potential of the SVA to serve as a control parameters for heel height manipulations of an AFO-FC in healthy adults.

The efficacy of AFOs in children with CP may also be related to an inadequate match between the AFO's mechanical properties (e.g. stiffness) and the patient's specific underlying impairments and personal characteristics. Rigid AFOs aim to shift the ground reaction force anterior to the knee joint and are therefore generally prescribed to reduce the knee flexion in children with CP who walk with excessive knee flexion in stance. The rigid AFOs however also obstruct ankle range of motion, therewith impeding ankle push-off power, which may negatively impact gait efficiency. Spring-like AFOs may enhance push-off power, and could therefore be more beneficial in terms of the gait efficiency.

In chapter IV, we investigated the mechanical properties of a spring-hinged AFO to assess its potential use in children with CP who walk with excessive knee flexion in

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stance. This AFO was manufactured with an integrated hinge of which stiffness could be varied by applying different springs, each holding specific mechanical properties. The mechanical properties of the five available springs were assessed using the Bi-articular Reciprocal Universal Compliance Estimator (BRUCE) device. The mechanical behavior of the springs was not linear, but could be described in terms of a threshold, a stiffness, range of motion, and an energy release. The stiffest spring showed the highest threshold and stiffness, which was combined with a small range of motion. We hypothesized that this spring would counteract the knee flexion most effectively. The higher energy return of the second stiffest spring was expected to enhance ankle push-off power more compared to the stiffest spring, although at the expense of higher knee flexion angles.

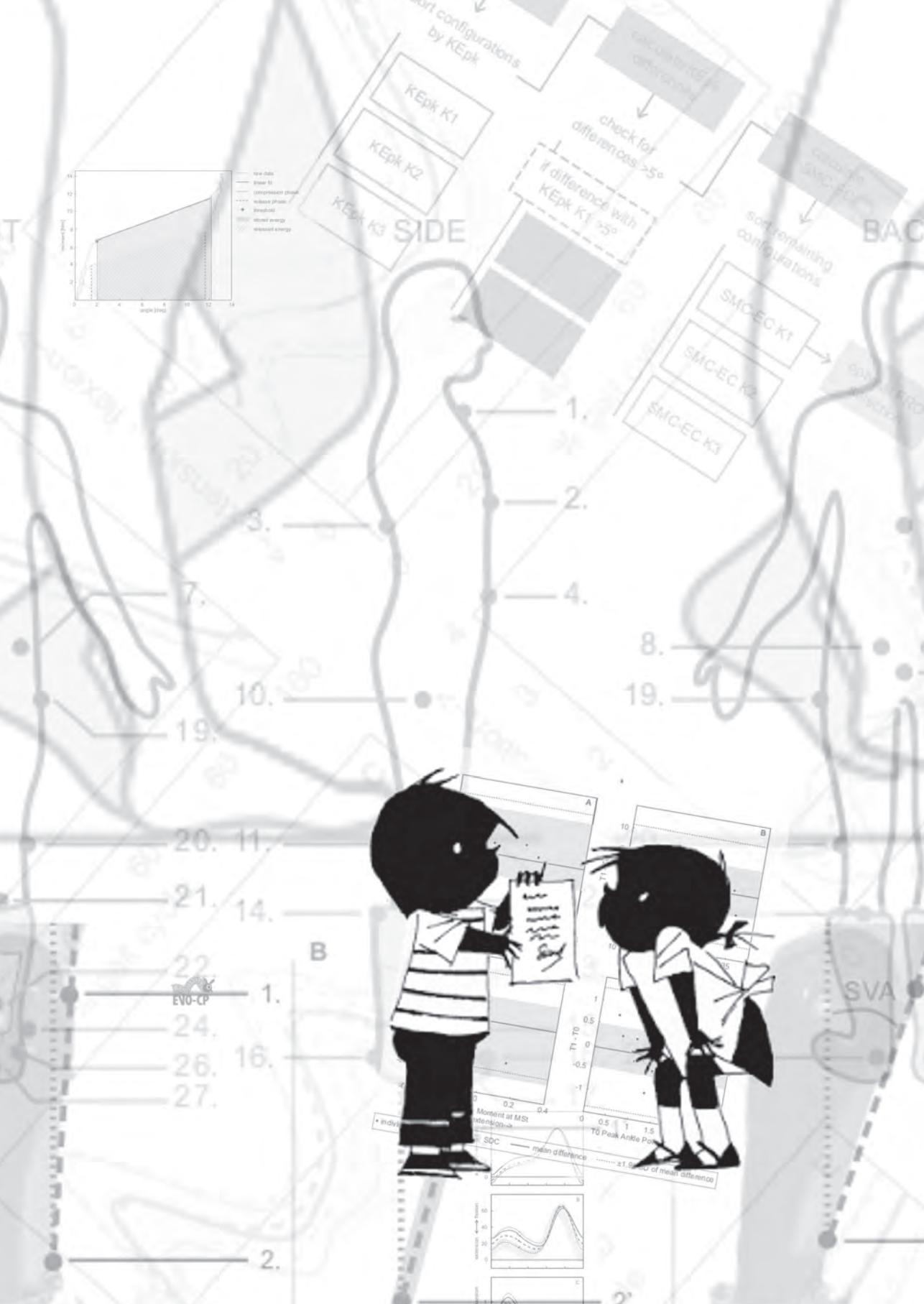
In chapter V, we evaluated the effects of different hinge settings (i.e. degrees of stiffness) of the spring-hinged AFO on gait biomechanics and gait efficiency in 15 children with CP walking with excessive knee flexion in stance. The AFO was configured in a rigid (i.e. aiming to eliminate spring-like properties and ankle range of motion), stiff (i.e. stiffest available spring) and flexible (i.e. second stiffest available spring) hinge setting. The effects of the three AFO stiffness levels were compared to walking shoes-only. The results showed that all AFOs equally reduced the knee flexion angle and internal knee flexion moment. Ankle push-off power was reduced by the rigid AFO, while remaining push-off power was preserved by the stiff and flexible AFO compared to walking shoes-only. Accordingly, ankle work was reduced by the rigid AFO, while being preserved by the two-spring like AFOs. The AFOs contribution to ankle work was smallest for the rigid AFO and comparable between the stiff and flexible AFO. Overall, the net energy cost was significantly reduced by all AFOs compared to walking with shoes-only, while no significant differences were found between AFOs. The potential benefit of spring-like AFOs on ankle kinematics and kinetics was therefore not reflected in larger energy cost reductions. These findings may suggest that, in children with CP walking with excessive knee flexion in stance, the optimal AFO stiffness that maximizes gait efficiency is primarily defined by its effects on knee kinematics and kinetics during stance, and less by its effect on ankle push-off power.

The results of chapter V were used to individually select the optimal AFO stiffness for each participant of the AFO-CP trial. In chapter VI, the effects of the stiffness-optimized AFO on the walking energy cost, knee angle and daily walking activity were investigated. The optimal AFO stiffness selection was based on the peak knee extension during single limb support (primary aim) and the walking energy cost (secondary aim) while walking with AFO. In total, 29 legs of 15 children with spastic CP were evaluated.

In eight participants, the selection lead to bilateral stiff AFO prescription. Four children were prescribed with bilateral flexible AFOs. Two participants were prescribed with one stiff, and one flexible AFO, and one participant with one rigid and one flexible AFO. After three months of wearing the optimized AFO, the net energy cost was reduced in 11 of 14 patients, with an overall energy cost reduction of 9% compared to walking shoes-only. Some children showed a reduction of more than 10%, indicating the clinical relevance of individually optimizing AFO stiffness in a subgroup of children with CP. The optimized AFO significantly reduced the knee angle, while daily walking activity was not affected at follow-up. The variety in the assignment of the optimal AFO stiffness emphasized an individual approach to AFO prescription to maximize its effects on the gait pattern and gait efficiency in children with CP who walk with excessive knee flexion in stance.

A third factor that might affect the efficacy of AFOs could be the acclimatization to an AFO. In research on the effects of AFOs on gait, an acclimatization time is generally applied. However, the applied duration of this acclimatization period in current literature varies between less than a day, and more than six weeks. The need of an acclimatization period remains therefore unclear. In chapter VII, we investigated the need of an acclimatization time for the gait pattern to adapt to a newly prescribed AFO. To this purpose, a specific set of biomechanical parameters was assessed immediately after applying a new AFO, and after four weeks of acclimatization time. Although we found some variation in our data, the results showed no significant changes in the assessed parameters after acclimatization. As such, our results suggested that, in independently walking children with spastic CP, inclusion of an acclimatization period to reliably evaluate biomechanical effects of an AFO on gait may not be needed.

In chapter VIII, the main findings of the presented studies were critically discussed, leading to clinical implications and ideas for future research. First, the exact association between changes in gait biomechanics and changes in the walking energy cost need to be explored. When this association is more clear, the potential benefit of adjustable AFOs can be optimally used to tune the mechanical function of the AFO to the specific (gait) impairments of the patient. Future research should also unravel the patient characteristics that underlie the (in)efficacy of AFOs in order to predict and improve treatment efficacy in children with CP. With regard to the clinical implications, it is advised that clinicians consider to prescribe spring-like AFOs in children with CP who walk with excessive knee flexion. However, an individual approach to AFO prescription is emphasized, which should be guided by evaluations of the AFO's effects on gait-related outcome that is significant in the context of the prescription's indication.



# Het maximaliseren van de effectiviteit van enkel voet orthesen bij kinderen met cerebrale parese

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Lopen, of wandelen, is een van de meest belangrijke activiteiten in het dagelijks leven. Hoewel lopen een gemakkelijke taak lijkt voor de meeste gezonde mensen, is het een ingewikkelde combinatie van bewegingen van verschillende delen van het lichaam. Wanneer de bewegingsvaardigheid is verminderd als gevolg van hersenschade, zoals bij kinderen met cerebrale parese (CP), wordt de complexiteit van het lopen duidelijk. Een belangrijk doel van de kinderrevalidatiegeneeskunde is het verkrijgen of behouden van de loopvaardigheid bij kinderen met CP. Een enkel voet orthese (evo) is hierbij een vaak toegepaste interventie, welke erop gericht is het lopen te verbeteren. Dit proefschrift gaat in op verschillende aspecten van het toepassen van evo's, met als doel de effectiviteit van de behandeling te verbeteren.

Om in te kunnen grijpen op de afwijkingen tijdens het lopen bij kinderen met CP, is het belangrijk om het lopen bij gezonde mensen goed te begrijpen. Het lopen wordt daarom vaak beschreven in termen van een gangcyclus en de biomechanica. De gangcyclus beschrijft een volledige stap van één been, die bestaat uit een standfase, als het been op de grond staat, en een zwaai fase, wanneer het been door de lucht naar voren zwaait. De overgang van de stand- naar de zwaai fase wordt de afzet genoemd. In de biomechanica spreekt men van de hoeken, momenten en power van gewrichten, zoals de enkel en de knie. Met een gewrichtshoek wordt de mate van strekking en buiging van een gewricht beschreven. Een moment beschrijft de hoeveelheid kracht die er op een gewricht wordt uitgeoefend, en bepaalt daarmee hoeveel kracht de spieren moeten leveren om te blijven staan. Een groot gewrichtsmoment betekent dat de spieren veel kracht moeten leveren. De power geeft aan hoeveel energie er door een gewricht gegenereerd wordt. Tijdens de afzet levert de enkel bijvoorbeeld veel power om het been voldoende snelheid te geven om naar voren te zwaaien en een stap te zetten.

Cerebrale parese (CP), of hersenverlamming, is een overkoepelende term voor schade aan de hersenen, welke is ontstaan vóór de eerste verjaardag van een kind. Door de hersenschade hebben kinderen met CP vaak symptomen zoals spasticiteit en spierzwakte. Deze symptomen kunnen leiden tot problemen bij het uitvoeren van motorische taken (bewegen), welke kunnen variëren van zeer licht tot zeer ernstig. Deze verminderde motorische vaardigheden uiten zich ook tijdens het lopen, welke te herkennen zijn aan afwijkende lichaamshoudingen. Deze afwijkingen tijdens het lopen worden vaak geclassificeerd. Grofweg bestaan er twee verschillende looptypen. Het eerste looptype wordt gekarakteriseerd door een overmatige kniestrekking. Het andere looptype is te herkennen aan overmatige kniebuiging, ook wel knieflexie genoemd. In

dit proefschrift focussen we op de kinderen die lopen met overmatige knieflexie. Deze kinderen vertonen afwijkingen in de biomechanica, zoals een te grote enkel- en kniehoek in vergelijking met normaalwaarden. Hierdoor nemen de momenten op de gewrichten toe, waardoor spieren meer kracht moeten leveren om te blijven staan. Wanneer u zelf probeert met overdreven gebogen knieën een tijd te staan of een stuk te lopen, zult u merken hoe vermoeiend dit is voor uw (boven)been spieren. Een looppatroon met overmatige knieflexie bij kinderen met CP wordt dan ook vaak geassocieerd met een verhoogd energieverbruik tijdens het lopen. Dit kan gekoppeld zijn aan beperkingen tijdens dagelijkse activiteiten en een verminderde participatie in het dagelijks leven.

Om de afwijkingen in het looppatroon tegen te gaan, krijgen kinderen met CP vaak evo's voorgeschreven. Een evo is een stevige, stijve spalk die om de voet en het onderbeen vast zit. Op deze manier wordt de enkel in een bepaalde stand gedwongen, waardoor ook de kniehoeken kunnen worden gecorrigeerd. Dit is te vergelijken met een skischoen; doordat de enkel vastgehouden wordt door de schoen, kunnen de knieën niet meer gestrekt worden zonder voorover te leunen met de bovenbenen en de romp. Ondanks dat evo's veel toegepast worden in de klinische praktijk, is er nog weinig bekend over de effectiviteit van de spalken op de loopvaardigheid van kinderen met CP. De onderzoeken die de effecten op het lopen hebben onderzocht laten bovendien wisselende effecten zien; bij een deel van de kinderen verbetert het looppatroon en/of het energieverbruik, maar bij andere kinderen hebben de evo's geen effect, of kunnen de spalken zelfs een negatieve invloed hebben op het lopen. Hoewel er nog niet veel bekend is over de oorzaken van deze wisselende effecten, zijn er wel een aantal factoren die waarschijnlijk de effectiviteit van een evo (mede) bepalen. Een aantal van deze factoren werden in dit proefschrift onderzocht. Om dit te kunnen onderzoeken, is het EVO-CP onderzoek uitgevoerd. Het protocol van dit onderzoek staat beschreven in hoofdstuk II.

Ten eerste kan de uitlijning van de evo een effect hebben op de gewrichtshoeken en -momenten tijdens het lopen. Deze uitlijning wordt bepaald door de hoek waarin de enkel wordt vastgehouden door de spalk, én de hoogte van de schoenzool. Een gezond persoon, waarbij de enkel bewegingsvrijheid heeft, kan zal de gewrichtshoeken zelf aanpassen aan de eigenschappen van een schoen. Ten slotte kunnen vrouwen op hoge hakken hun gewrichten aanpassen, waardoor een normaal looppatroon nog steeds mogelijk is. Wanneer de enkel wordt vastgehouden, zoals bij een evo, is deze aanpassing echter niet meer mogelijk (denk aan de skischoen). De eigenschappen van de combinatie van de evo en de schoen moeten daarom worden aangepast zodat de kniehoek wordt genormaliseerd. In hoofdstuk III hebben tien gezonde volwassenen met

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evo's op een loopband gelopen. Daarbij werden steeds de eigenschappen van de evo-schoen combinatie gevarieerd door middel van het aanpassen van de hakhoogte. Voor iedere hakhoogte (laag, gemiddeld en hoog) werden de effecten op de enkel, knie en heup hoeken en momenten gemeten. Uit de resultaten bleek dat de gewrichtshoeken en momenten toenamen bij het verhogen van de hak. Dit betekent dat een (te) hoge hak in de schoen een negatief effect kan hebben op de werking van de evo. Deze resultaten moeten nu verder onderzocht worden in een patiëntenpopulatie, zoals kinderen met CP.

Een tweede factor die effect kan hebben op de effectiviteit van de evo, is de stijfheid. Bij kinderen met CP die lopen met overmatige knieflexie worden er voornamelijk heel stijve evo's voorgeschreven. Door het stijve materiaal kunnen deze spalken heel goed de kniehoek en het kniemoment verbeteren. Dit betekent dat de bovenbeenspieren minder kracht hoeven te leveren, wat het energieverbruik tijdens het lopen zou kunnen verminderen. Een negatieve eigenschap van deze spalken is dat ze de bewegingsvrijheid van de enkel beperken. Hierdoor kan er geen effectieve afzet worden gegenereerd, waardoor het lopen juist meer energie kost. In het proefschrift hebben we gebruik gemaakt van een speciale gescharnierde evo, waarvan we de stijfheid konden instellen door middel van verwisselbare veren. In hoofdstuk IV hebben we de mechanische eigenschappen, zoals de stijfheid, van deze spalk bepaald door deze te meten met een speciaal ontwikkeld apparaat. Uit deze metingen kwam naar voren dat de twee stijfste veren mogelijk geschikt zouden om het lopen bij kinderen met CP te verbeteren.

In hoofdstuk V hebben we de gescharnierde evo ingesteld in een rigide (heel stijf), stijve en flexibele stand. Deze verschillende standen van het scharnier hebben we toegepast bij een groep van 15 kinderen met CP die lopen met overmatige knieflexie. De kinderen liepen steeds vier weken met elke evo, waarna de effecten op de biomechanica en energieverbruik tijdens het lopen werden geëvalueerd in het looplaboratorium. Uit de resultaten bleek dat de verbetering van de knie biomechanica hetzelfde waren voor alle evo-stijfheden. Er waren meer verschillen te zien in het enkelgewicht. De rigide evo beperkte de bewegingsvrijheid van de enkel meer dan de stijve en flexibele evo's. Bovendien was de afzet minder effectief tijdens het lopen met de rigide evo, terwijl deze met stijve en flexibele evo even effectief was als tijdens het lopen met alleen schoenen. We verwachtten dat deze verschillen zich zouden vertalen in het energieverbruik tijdens het lopen. We vonden echter geen verschil in energieverbruik tussen de verschillende evo-stijfheden. Er waren echter veel verschillen in effecten op individueel niveau, wat het belang van een individuele benadering bij een evo voorschrijf benadrukt.

In hoofdstuk VI hebben we de gegevens van het voorgaande hoofdstuk gebruikt om de optimale evo voor iedere deelnemer aan het onderzoek te selecteren. Dit hebben we gedaan aan de hand van een beslisschema. In dit schema werd eerst bepaald of de verschillende stijfheden voldoende effect hadden op de kniehoek tijdens het lopen. Bij een gelijk effect tussen evo stijfheden op de kniehoek, werd de stijfheid die resulteerde in het laagste energieverbruik geselecteerd als optimale stijfheid. De deelnemers droegen de geoptimaliseerde spalk nog eens drie maanden, waarna we de effecten op het energieverbruik, hoeveelheid stappen per dag, en de kniehoek hebben bepaald. De geoptimaliseerde spalk verbeterde de kniehoek tijdens het lopen. De optimalisatie liet ook zien dat er een verbetering in het energieverbruik tijdens het lopen behaald kan worden, hoewel dit niet voor iedereen geldt. De geoptimaliseerde spalk resulteerde niet in een verhoging van het aantal stappen gezet werd per dag.

Ten slotte hebben we in hoofdstuk VII onderzocht of kinderen met CP tijd nodig hebben om het looppatroon aan te passen aan een nieuwe evo. In onderzoek en klinische praktijk wordt vaak gebruik gemaakt van een periode van gewenning, variërend van minder dan een dag tot langer dan zes weken. Hierbij gaat met ervan uit dat de effecten van een spalk pas na een bepaalde draagtijd betrouwbaar te bepalen zijn. Echter is niet bekend of deze gewenningsperiode daadwerkelijk nodig is en/of hoe lang deze periode moet zijn. Voor het onderzoek in hoofdstuk VII hebben we bij een groep van tien kinderen met CP de effecten van een nieuwe spalk op de biomechanica tijdens het lopen gemeten direct na het toepassen van de nieuwe spalk, én vier weken later. Aangezien de effecten van de spalk direct zichtbaar waren en niet veranderde na vier weken dragen, konden we concluderen dat een periode van gewenning niet nodig is bij het voorschrijven van een nieuwe spalk. Dit is handig in de praktijk en onderzoek, aangezien we er vanuit kunnen gaan dat effecten betrouwbaar te bepalen zijn direct na het toepassen van een nieuwe spalk.

De bevindingen van dit proefschrift werden kritisch beschouwd in hoofdstuk VIII. Daaruit blijkt dat onze resultaten belangrijke handvaten biedt voor vervolgonderzoek en klinische praktijk. Enkele bevindingen van het proefschrift kunnen direct toegepast worden in de praktijk, waardoor deze een relevante bijdrage kunnen leveren aan het optimaliseren van de behandeling met evo's bij kinderen met CP in de nabije toekomst.



# Curriculum vitae & List of publications

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## CURRICULUM VITAE

Yvette Laura Kerkum was born on September 5<sup>th</sup> 1986 in Lelystad, the Netherlands. At the age of 11, she and her family moved to Zwolle where she attended the Thorbecke Scholengemeenschap. In 2004 she graduated (VWO) and in September she started to study Human Movement Sciences at the VU University in Amsterdam. Her bachelor research project on the relation between cognitive functioning and physical activity in elderly was awarded with the price for best presentation at the faculty of Human Movement Sciences. During the master's program, she performed a research internship on balance control in stroke patients at the department of Research and Development of rehabilitation center Heliomare in Wijk aan Zee. In 2010 Yvette received her Master's degree in Human Movement Sciences.

In June 2011 she started working as a PhD student at the department of Rehabilitation Medicine of the VU University medical center in Amsterdam under supervision of dr. Merel Brehm, dr. Annemiek Buizer, prof. dr. Jules Becher and prof. dr. ir. Jaap Harlaar. Yvette started the AFO-CP trial at the VUmc, aiming to improve the treatment efficacy of ankle foot orthoses in children with cerebral palsy. Yvette visited a considerable amount of conferences to present the results of her research to specialists in the field, such as physicians, physiotherapists, and orthotists. In 2013 she won the Best Poster award of the 23<sup>rd</sup> annual meeting of the European Scientific Society for Clinical Gait and Movement Analysis (ESMAC) in Glasgow, Scotland. In 2014 she was awarded the Student Scholarship to visit the 68th annual meeting of the American Academy for Cerebral Palsy and Developmental Medicine in San Diego, USA. In 2015 she received a travel grant to visit the 24th annual meeting of the European Society for Movement Analysis in Adults and Children.

From september 2015 to january 2016, Yvette worked as a researcher at the Clinical Motion Analysis Laboratory of the UZ Leuven under supervision of prof. dr. Jacques Duysens and prof. dr. Kaat Desloovere. There she performed research on the effect of asymmetric and symmetric neck reflexes on movement in children with cerebral palsy, which was initiated by the department of Kinesiology of the KU Leuven, Belgium.

## LIST OF PUBLICATIONS

### Articles

**Kerkum YL**, Buizer AI, van den Noort JC, Becher JG, Harlaar J, Brehm MA. The Effects of Varying Ankle Foot Orthosis Stiffness on Gait in Children with Spastic Cerebral Palsy Who Walk with Excessive Knee Flexion. *PLoS. One.* 2015, 10(11); e0142878.

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### **Grants and prizes**

ESMAC Travel Scholarship to attend the 24<sup>th</sup> Annual Meeting in Heidelberg, Germany; 2015.

AACPDM Student Scholarship to attend the 68<sup>th</sup> Annual Meeting in San Diego, USA; 2014.

ESMAC Poster Award at the 22<sup>nd</sup> ESMAC Annual Meeting in Glasgow, Scotland; September 2013.

ISPO-NL Travel Grant to attend the ISPO World Congress in Hyderabad, India; 2013.



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Children with spastic cerebral palsy often have problems with walking. For example, excessive knee flexion in the stance phase of gait can increase the effort to walk. Ankle foot orthoses might improve this, but scientific evidence for their effectiveness is scarce and shows limited support. We hypothesized that this is partly caused by an inadequate match between the patient's impairments and the ankle foot orthoses' mechanical properties. The studies in this thesis aimed to evaluate factors that enable an individual optimization of ankle foot orthoses to match the patients impairments. To this respect, the effects of different ankle foot orthoses stiffness levels on gait were evaluated in children with cerebral palsy who walk with excessive knee flexion in stance. In addition, effects of the ankle foot orthosis' alignment, and acclimatization to a newly prescribed orthosis were assessed. Results of our studies emphasize an individual approach to ankle foot orthosis prescription to maximize treatment efficacy.

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