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

Yvette Kerkum
Jaap Harlaar
Annemieke Buizer
Josien van den Noort
Jules Becher
Merel-Anne Brehm

Gait and Posture, conditionally accepted
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·kg⁻¹·m⁻¹] were assessed to individually select the optimal stiffness. After three months, net EC, daily walking activity [strides·min⁻¹] 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·kg⁻¹·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° (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.
Effectiveness of stiffness-optimized AFOs

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[1] and elevated walking energy cost levels, reflecting poor gait efficiency[2]. Interventions in these children therefore primarily aim to reduce knee flexion to prevent deterioration, which could improve gait efficiency[3] 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[4] in stance and improve gait efficiency[3,5]. 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[6]. Considering the diversity in underlying impairments within CP, the varying effects of AFOs on gait efficiency as reported in the literature[3,5] might be partly explained by an inadequate match between the patient’s impairments and the AFO’s mechanical properties, including its ankle stiffness[7,8].

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[3]. The AFO’s properties however also obstruct ankle range of motion, therewith impeding ankle push-off power and negatively impacting gait efficiency[5,10]. Applying a more compliant, spring-like AFO may enhance push-off power and subsequent gait efficiency[7,11], 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[12].

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[8]. Such an optimization requires an extensive evaluation of the effects of AFOs on multiple gait-related outcomes[9]. Brehm et al.[14] 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[14,5]. 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[13]; Dutch National Trial Register no. NTR3418), consisting of two repeated measurements; at baseline (T0), walking with shoes only, and at 12–20 weeks follow-up (T2), walking with the optimized AFO. Additional measurements were performed to provide data for the optimal AFO stiffness selection (T1).

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[16] 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[4].
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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.
Chapter VI

**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[^12]. 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[^13].

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[^17].

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[^18]. 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⁻¹] 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 devices¹⁹.

Data processing

To calculate walking energy cost, breath-by-breath VO₂ and VCO₂ 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 [m·min⁻¹] was measured over the same time frame of the walk test. Accordingly, the net energy cost (EC) [J·kg⁻¹·m⁻¹] was calculated as \((ECSwalk - ECSrest)/walking speed\). Net EC values were normalized to calculate the net non-dimensional energy cost²⁰, 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 thresholds²¹: 0 strides·min⁻¹ (SR0), 1 to 15 strides·min⁻¹ (SR1-15), 16 to 30 strides·min⁻¹ (SR16-30), 31 to 60 strides·min⁻¹ (SR31-60), and >60 strides·min⁻¹ (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). 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 frames¹⁸. 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 calculated²². Using force plate data and inverse dynamics, peak ankle power generation (AP) [W·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.[13]

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 (T2) on net EC, walking speed, daily walking activity, and biomechanical gait parameters were compared to walking with shoes-only (T0) 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>
<thead>
<tr>
<th>Table 6.1 Participant’s demographic and disease characteristics at baseline (n=15).</th>
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<tbody>
<tr>
<td>age [yrs]</td>
</tr>
<tr>
<td>weight [kg]</td>
</tr>
<tr>
<td>height [cm]</td>
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<tr>
<td>sex [boy/girl]</td>
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<tr>
<td>GMFCS [I/II/III]</td>
</tr>
<tr>
<td>(most) affected side [right/left]</td>
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<tr>
<td>selective motor control[a] [good/moderate/poor]</td>
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</tbody>
</table>

[a]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.[31]

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>
<thead>
<tr>
<th>angle of interest&lt;sup&gt;a&lt;/sup&gt;</th>
<th>muscles</th>
<th>RoM</th>
<th>spasticity scale&lt;sup&gt;b&lt;/sup&gt;</th>
</tr>
</thead>
<tbody>
<tr>
<td>hip extension</td>
<td>[+ = extension]</td>
<td>10 [0 20]</td>
<td>n/a</td>
</tr>
<tr>
<td>knee extension</td>
<td>[+ = extension]</td>
<td>0 [-10 0]</td>
<td>n/a</td>
</tr>
<tr>
<td>popliteal angle</td>
<td>Hamstrings</td>
<td>55 [45 70]</td>
<td>[10/1/3/0]</td>
</tr>
<tr>
<td>ankle dorsiflexion (flexed knee)</td>
<td>Soleus</td>
<td>10 [0 25]</td>
<td>[13/1/0/1]</td>
</tr>
<tr>
<td>ankle dorsiflexion (extended knee)</td>
<td>Gastrocnemius</td>
<td>0 [-10 0]</td>
<td>[13/1/0/1]</td>
</tr>
</tbody>
</table>

<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⁻¹, 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⁻¹.
**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° 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 (T0; shoes-only), and follow-up (T2; optimized ankle foot orthosis).

<table>
<thead>
<tr>
<th></th>
<th>T0</th>
<th>T1</th>
<th>T2</th>
<th>T2-T0&lt;sup&gt;b&lt;/sup&gt;</th>
<th>Z</th>
<th>p</th>
</tr>
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<tbody>
<tr>
<td><strong>gait efficiency</strong></td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>speed [m·min&lt;sup&gt;-1&lt;/sup&gt;]&lt;sup&gt;a&lt;/sup&gt;</td>
<td>61.7 [14.2]</td>
<td>58.2 [8.1]</td>
<td>61.6 [29.0]</td>
<td>-0.1</td>
<td>-0.03</td>
<td>1.000</td>
</tr>
<tr>
<td>net EC&lt;sup&gt;a&lt;/sup&gt; [J·kg·m&lt;sup&gt;-1&lt;/sup&gt;]&lt;sup&gt;b&lt;/sup&gt;</td>
<td>5.8 [2.1]</td>
<td>4.8 [1.5]</td>
<td>5.3 [2.1]</td>
<td>-0.5</td>
<td>-1.48</td>
<td>0.077</td>
</tr>
<tr>
<td><strong>daily walking activity</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>total [strides/day]</td>
<td>3986 [2579]</td>
<td>n/a</td>
<td>2513 [3715]</td>
<td>-1473</td>
<td>-0.09</td>
<td>0.966</td>
</tr>
<tr>
<td>SR1-15 [strides/day]</td>
<td>984 [1082]</td>
<td>n/a</td>
<td>976 [567]</td>
<td>-16</td>
<td>-0.45</td>
<td>0.700</td>
</tr>
<tr>
<td>SR16-30 [strides/day]</td>
<td>946 [924]</td>
<td>n/a</td>
<td>963 [1245]</td>
<td>2</td>
<td>-0.71</td>
<td>0.520</td>
</tr>
<tr>
<td>SR31-60 [strides/day]</td>
<td>1692 [1955]</td>
<td>n/a</td>
<td>836 [1949]</td>
<td>-855</td>
<td>-0.36</td>
<td>0.765</td>
</tr>
<tr>
<td>SR&gt;60 [strides/day]</td>
<td>0 [90]</td>
<td>n/a</td>
<td>18 [99]</td>
<td>18</td>
<td>-1.54</td>
<td>0.148</td>
</tr>
<tr>
<td><strong>gait biomechanics</strong></td>
<td></td>
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<td></td>
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<td></td>
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</tr>
<tr>
<td>KE&lt;sup&gt;a&lt;/sup&gt; [deg]</td>
<td>22.0 [11.7]</td>
<td>14.3 [15.2]</td>
<td>19.6 [17.2]</td>
<td>-2.4</td>
<td>-2.44</td>
<td>0.006</td>
</tr>
<tr>
<td>AP [W·kg&lt;sup&gt;-1&lt;/sup&gt;]&lt;sup&gt;b&lt;/sup&gt;</td>
<td>1.6 [0.9]</td>
<td>1.3 [0.7]</td>
<td>1.2 [0.5]</td>
<td>-0.4</td>
<td>-1.87</td>
<td>0.064</td>
</tr>
</tbody>
</table>

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

<sup>b</sup>Difference in percentage between T2 and T0 was calculated as: \((T2-T0)/(\text{Average of T0 and T2})\)*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 0 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\([5,23,24]\), some of which also used walking energy cost assessments to select the most beneficial AFO\([5,25]\). 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\([24]\), 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\([8,11]\).

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\([3,5]\). 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
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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[13]. 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 [22,26]. The SVA of 20° as found in our study was much larger compared to findings in other literature[26], 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[27]. 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[3,5,23].

In our study, the AFOs were used for a median [IQR] of 8.8 [5.0] hours·day⁻¹, which is comparable to Wren et al.[28]. Despite this relatively intensive use, daily walking activity did not increase with the optimized AFO. Wren et al.[28] 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\cite{29}, 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\cite{30}.

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.
Effectiveness of stiffness-optimized AFOs

REFERENCES


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⁻¹], stiff [mean (SD) 1.6 (0.4) Nm-deg⁻¹], and flexible [mean (SD) 0.7 (0.2) Nm-deg⁻¹]. The stiffness was measured using the Biarticular Reciprocal Universal Compliance Estimator (BRUCE) device, according to a standard protocol[12]. 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[13]. 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.