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Original article

Ankle foot orthoses in cerebral palsy: Effects of ankle stiffness on trunk kinematics, gait stability and energy cost of walking

P. Meyns [a,1], Y.L. Kerkum [b,*,1], M.A. Brehm [c], J.G. Becher [b], A.I. Buizer [b], J. Harlaar [b,d]

[a] Rehabilitation Research (REVAL), Faculty of Rehabilitation Sciences, Hasselt University, Agoralaan, Building A, BEC3590, Hasselt, Belgium
[b] Amsterdam UMC, Vrije Universiteit Amsterdam, Department of Rehabilitation Medicine, Amsterdam Movement Sciences, de Boelelaan 1117, Amsterdam, the Netherlands
[c] Amsterdam UMC, University of Amsterdam, Department of Rehabilitation, Amsterdam Movement Sciences, Meibergdreef 9, Amsterdam, the Netherlands
[d] Department of Biomechanical Engineering, Delft University of Technology, Mekelweg 2, 2628 CD, Delft, the Netherlands

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ABSTRACT

In children with cerebral palsy (CP), rigid ventral shell ankle-foot orthoses (vAFOs) are often prescribed to reduce excessive knee flexion in stance and lower the energy cost of walking (ECW). However, how vAFOs affect ECW is a complex issue, as vAFOs may have an impact on lower limb biomechanics, upper body movements, and balance. Besides, the vAFO's biomechanical effect have been shown to be dependent on its stiffness around the ankle joint. We examined whether vAFO stiffness influences trunk movements and gait stability in CP, and whether there is a relationship between these factors and ECW. Fifteen children with spastic CP were prescribed vAFOs. Stiffness was varied into a rigid, stiff and flexible configuration. At baseline (shoes-only) and for each vAFO stiffness configuration, 3D-gait analyses and ECW-tests were performed. From the gait analyses, we derived trunk tilt, lateroflexion, and rotation range of motion (RoM) and the mediolateral and anteroposterior Margins of Stability (MoS) and their variability as measures of gait stability. With the ECW-test we determined the netEC. We found that wearing vAFOs significantly increased trunk lateroflexion (Wald $\chi 2 = 33.7$, $p < 0.001$), rotation RoM (Wald $\chi 2 = 20.5$, $p < 0.001$) and mediolateral gait instability (Wald $\chi 2 = 10.4$, $p = 0.016$). The extent of these effects partly depended on the stiffness of the vAFO. Significant relations between trunk movements, gait stability and ECW were found $r = 0.57–0.81$, $p < 0.05$), which indicates that trunk movements and gait stability should be taken into account when prescribing vAFOs to improve gait in children with CP walking with excessive knee flexion.

1. Introduction

Walking ability in children with spastic cerebral palsy (CP) is often reduced due to motor impairments like spasticity and muscle weakness, caused by brain malformation or damage during early development [1]. These motor impairments can lead to gait deviations, such as excessive knee flexion during stance [2]. Interventions in these children are primarily aimed at improving knee extension during stance, as a flexed knee gait pattern is prone to deteriorate over time, reflected by the development of knee flexion contractures [3]. In addition, children with CP show highly elevated energy cost of walking [4,5].

Rigid ventral shell Ankle-Foot Orthoses (vAFOs) are commonly prescribed in CP to improve knee extension during stance [6], and lower the elevated energy cost of walking (ECW) [6,7]. However, rigid vAFOs are known to impede the ankle range of motion, therewith reducing ankle push-off power considerably compared to walking without vAFOs [7]. Previous research in CP that compared vAFOs of different stiffness's showed that more compliant vAFOs enhance ankle push-off power by 70–100% compared to rigid vAFOs, while still adequately improving the knee extension angle and external knee extension moment [7]. However, all vAFOs resulted in a similar mean ECW reduction (of ±10%) compared to walking with shoes-only, which suggests that multiple biomechanical parameters may influence ECW when wearing vAFOs [7].

* Corresponding author. Research and Development, OIM Orthopedie, Balkendwarsweg 5, 9405 PT, Assen, the Netherlands.
E-mail address: y.kerkum@oim.nl (Y.L. Kerkum).
[1] Meyns P & Kerkum YL are joint first authors. They both contributed equally to this work.

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Abbreviations

3D-gait analysis	Three dimensional gait analysis
AP_MoS	Anteroposterior margins of stability
CP	Cerebral palsy
ECW	Energy cost of walking
GMFCS	Gross motor function classification system
ML_MoS	Mediolateral margins of stability
MoS	Margins of stability
netEC	Net energy cost
sdAP_MoS	Standard deviation of the anteroposterior margins of stability
sdML_MoS	Standard deviation of the mediolateral margins of stability
(v)AFO	(ventral shell) ankle-foot orthosis
VCO_2	carbon dioxide production
VO_2	oxygen uptake

Previous studies on trunk movements in CP [8,9], showed increased trunk motion in the frontal and sagittal plane while walking with AFOs compared to walking without. Swinnen et al. also found increased trunk rotation as a result of wearing AFOs [9] and suggested that this could be a compensatory strategy for the reduced ankle push-off power. Moreover, the altered trunk movements could be induced by the biomechanical constraints caused by the AFOs [9]. As abnormal trunk movements during walking are known to be significantly associated to increased joint work in CP [10], therewith implying higher energy cost levels, the vAFO's effect on trunk movements may mediate its effect on ECW.

On the other hand, abnormal trunk movements may also be a reflection of decreased gait stability [8,9,11], which has been previously reported in CP [12]. Moreover, the impeding effect of a vAFO on ankle motion may further challenge gait stability as less ankle motion has been associated with lower gait stability in healthy adults [13]. As such, it is hypothesized that vAFOs may improve lower limb kinematics and kinetics, although at the expense of increased trunk movements, possibly induced by gait instability, which in turn, may affect ECW. In other words, the effect of vAFOs on trunk movement and/or gait stability might comprise the beneficial effects of the orthoses on lower limb biomechanics and related improvements in ECW. Considering that the vAFO's effect on lower limb kinematics and kinetics has been shown to be dependent on its stiffness [7], various vAFO stiffness levels with different biomechanical constraints (e.g. allowing more or less ankle range of motion) might also affect trunk movements and gait stability, and thus ECW. However, his has never been investigated in children with CP.

The aim of our study was to examine the effect of vAFO stiffness on ECW, trunk movements and gait stability compared to walking with shoes-only in children with CP who walk with excessive knee flexion. In addition, we investigated the relationship between ECW, trunk movements, and gait stability for the investigated vAFO stiffness conditions. We hypothesized that the effect of vAFOs on ECW was mediated by gait stability and trunk movements, and that this mediating effect is dependent on vAFO stiffness.

2. Material and methods

2.1. Participants

Data used in this study were collected in the AFO-CP trial, which has been previously described [14]. Participants in the AFO-CP trial were recruited from the rehabilitation department of the Amsterdam University Medical Centre, location VUmc, Amsterdam, The Netherlands. Children diagnosed with spastic CP, aged 6–14 years old were included. Children had to be able to walk independently, i.e. Gross Motor Function Classification System (GMFCS) level I-III [15], with a barefoot gait pattern characterized by excessive knee flexion in stance (i.e. more than 10° knee flexion in midstance). Children with ankle plantar flexion contractures, knee flexion contractures and/or hip flexion contractures of more than 10° were excluded, as these would have obstructed the intended effect of the vAFOs [6].

The study protocol of the AFO-CP trial was approved by the institutional review board (Medisch Ethische Toetsingscommissie) of the VUmc (ABR number NL37910.029.11) and registered at the Dutch trial register (NTR3418). Before inclusion, both parents of all participants and participants above 12 years old provided written informed consent. The study was performed in accordance to the Declaration of Helsinki.

2.2. Procedures

Children were prescribed with a full-carbon prepreg vAFO with a full-length rigid footplate and an integrated ankle hinge (Neuro Swing®, Fior&Gentz, Germany). In this hinge, different springs could be inserted, which allowed the vAFO's stiffness and ankle range of motion to be varied [16] (see Fig. 1). For each participant,

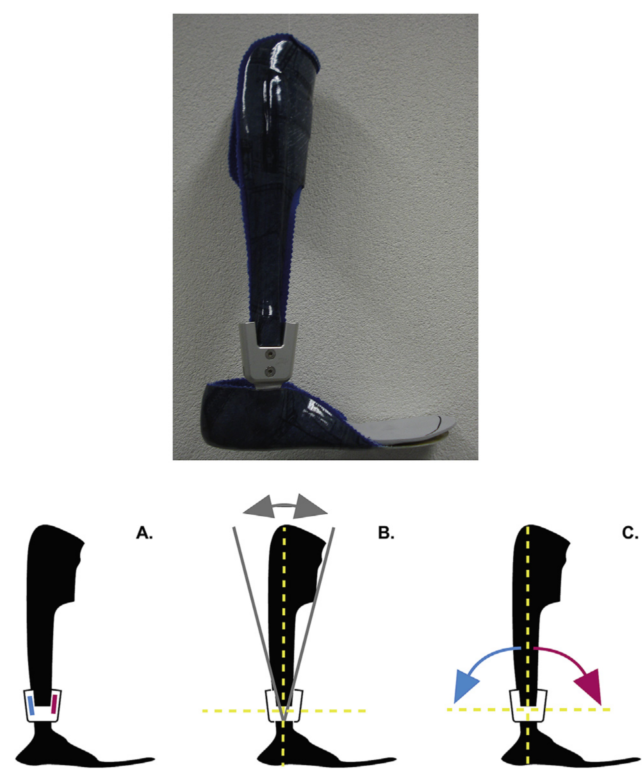

Fig. 1. The prepreg vAFO with full-length rigid footplate and integrated ankle hinge, which was custom-made for all participants (n = 15). The lower panel shows a schematic representation of the adjustments that can be made using the hinge. The hinge allows to (A) vary ankle stiffness towards dorsal and plantar flexion by inserting different springs, (B) adjust the alignment of the ventral shell with respect to the vAFO's foot part and (C) change the ankle range of motion towards dorsal and plantar flexion, although stiffer springs allow less range of motion. *Figure from Kerkum YL, Buizer AI, van den Noort JC, Becher JG, Harlaar J, Brehm M-A (2015) The Effects of Varying Ankle Foot Orthosis Stiffness on Gait in Children with Spastic Cerebral Palsy Who Walk with Excessive Knee Flexion. PLoS ONE 10(11): e0142878.* https://doi.org/10.1371/journal.pone.0142878.

three vAFO stiffness configurations toward dorsiflexion were applied [7] (block randomized): mean (SD) stiffness's were: rigid [3.8 (0.7)Nm·deg^{-1}], stiff [1.6 (0.4)Nm·deg^{-1}], and flexible [0.7 (0.2) Nm·deg^{-1}]. Stiffness towards plantar flexion was: mean (SD) rigid [4.6 (1.3)Nm·deg^{-1}], stiff [0.12 (0.2)Nm·deg^{-1}], flexible [0.11 (0.1) Nm·deg^{-1}]. The rigid vAFO allowed no ankle range of motion, while the others allowed dorsiflexion of mean (SD) stiff [6.6 (1.1) deg], flexible [11.8 (1.0)deg] and plantarflexion of mean (SD) [stiff 14.3 (1.8)deg], flexible [13.7 (2.5)deg]. The vAFO was worn in sneakers with flat flexible soles. This vAFO-footwear combination was tuned according to a clinical protocol, i.e. optimal ground reaction force alignment in midstance and terminal stance and maximal knee extension in terminal stance) [17]. After acclimatizing to the vAFO for 4–6 weeks, the efficacy was evaluated with a ECW-test and 3D-gait analysis. Afterwards, the hinge was set into the next stiffness configuration and the procedure was repeated. The ECW-test and 3D-gait analysis were also performed for shoes-only walking, while participants wore their own shoes.

2.3. Measurements

ECW was assessed with a 6-min rest-test, followed by a 6-min walk-test at comfortable speed on a 40-m indoor oval track. During the rest-test and walk-test, breath-by-breath oxygen uptake (VO_2) and carbon dioxide production (VCO_2) were recorded using a portable gas analysis system (Metamax 3B, Cortex Biophysik, Germany). A description of the procedure has been previously published [7].

For 3D-gait analyses, participants were instructed to walk over a 10m-walkway at a comfortable speed. Kinematic data were collected using an OptoTrak3020 motion capture system (Northern Digital, Waterloo, Canada). Technical clusters of three markers were attached to the trunk, pelvis, thighs, shanks and feet, and anatomically calibrated by probing 32 bony landmarks [18]. The technical cluster of the trunk was attached to the sternum. The cervical vertebra 7, thoracic vertebra 8, jugular notch, xhypoid process and right and left acromion served as bony landmarks to define the local reference frame of the trunk, with the Y-axis pointing upwards. Segment movements were tracked (sample frequency: 100 Hz). Measurements were repeated until a minimum of three successful steps of both legs were recorded (i.e. no missing marker data during a step of a single leg on the force plate).

2.4. Data-processing

2.4.1. ECW and walking speed

Breath-by-breath VO_2 and VCO_2 values from the third to sixth minute of both the rest- and walk-test were used to calculate the mean steady-state energy consumption values ([J·kg^{-1}·m^{-1})] [19]. Mean walking speed [m·min^{-1}] was measured over the same time frame of the walk test. Net energy cost (netEC) [J·kg^{-1}·m^{-1}] was than calculated as: (steady-state energy consumption during walking − steady-state energy consumption during rest)/walking speed.

2.4.2. Trunk range of motion

For 3D-gait analyses, optoelectronic marker data were analyzed using custom-made software (Bodymech, www.bodymech.nl) based on MATLAB R2011a (The Mathworks, Natick, USA). Initial contact and toe-off in the gait cycle were determined using foot angular velocity [20]. Joint and segment kinematics were calculated according to ISB anatomical frames [18]. Trunk range of motion in three planes [deg] was calculated as the angle of the trunk with respect to the pelvis and averaged over the walking trials (Fig. 1), and included mean trunk tilt (sagittal plane), lateroflexion (frontal plane) and rotation (transverse plane) range of motion.

2.4.3. Gait stability

Gait stability was quantified using the Margins of Stability (MoS), which is based on the inverted pendulum model. In the inverted pendulum model, a person is considered stable when the vertical projection of the body centre of mass is kept within the base of support in a static situation. MoS extends the inverted pendulum model of stability in static situations to dynamic situations as it takes into account the velocity of the centre of mass. It is specifically developed as a measure of dynamic stability [21], and has often been used to predict gait stability in different populations including children with cerebral palsy [11]. MoS was calculated as the distance between the edge of a person's base of support and the 'extrapolated' Center of Mass [22] (i.e. the sum of the position of the Centre of Mass and an extrapolation based on the dynamics of an inverted pendulum model [21]). A larger MoS indicaties increased stability. For each step, the step width (to reflect the mediolateral base of support) and MedioLateral MoS (ML_MoS) and AnteriorPosterior MoS (AP_MoS) were calculated for the most affected side. The corresponding standard deviations were calculated as a measure of variability in gait stability (sdML_MoS and sdAP_MoS), where more variability indicates less stable gait [23].

2.4.4. Lower limb biomechanics

Peak knee extension during single support, internal knee moment at the timing of peak knee extension in single support, and the peak ankle push-off power were determined as described in our former study for all three stiffness levels [7].

2.5. Statistics

In this study we aimed to elucidate whether the effect of different vAFO stiffness levels on ECW is compromized by altered trunk movements and/or gait stability compared to walking with shoes-only. Therefore, we first examined the effect of different vAFO stiffness levels on ECW, trunk movements and gait stability (Aim1) with Generalized Estimation Equation analyses. The within-subject factor was 'conditions' (i.e. shoes-only, rigid vAFO, stiff vAFO and flexible vAFO). Exchangeable correlation structures were assumed. Walking speed was added as covariate, as walking speed affects joint kinematics and kinetics [24].

To further elucidate whether trunk movements and/or gait stability may have a relation to ECW (and thus evaluate the possible compromising effect) we investigated the correlation between ECW, trunk movements, and gait stability for all investigated vAFO stiffness levels (Aim2) with Pearson's correlations.

Finally, we aimed to examine the impact of trunk movements and gait stability on ECW compared to the previously investigated aspects that affect ECW such as lower limb kinematics (Aim3). As such, multiple regression analyses were performed for each stiffness condition, with netEC as dependent variable. Trunk motion and gait stability parameters that significantly correlated with netEC were included in the model, as were peak knee extension during single support, internal knee moment at the timing of peak knee extension in single support, and the peak ankle push-off power. Backward elimination was used ($p \geq 0.05$) to define parameters explaining netEC.

Statistical analyses were performed using SPSS v20 (SPSS Inc, Chicago, USA), with alpha-level 0.05.

3. Results

3.1. Participants

Fifteen children with spastic CP (11 boys, 4 girls) were included in the study. Participant characteristics are presented in Table 1. The effects of all three vAFO configurations were evaluated in 13 participants. One child refused to wear the rigid vAFO, hence in this child only the flexible and stiff vAFOs were evaluated. Another child showed too much foot deformation within the flexible vAFO leading to pressure ulcers, therefore only the stiff and rigid vAFOs were evaluated in this child.

3.2. The effect of varying vAFO stiffness on ECW (Aim1)

All vAFO conditions decreased the mean (SD) netEC approximately by 10% ($p = 0008$) compared to shoes-only (Table 2); from 6.1 (1.7) $J \cdot kg^{-1} \cdot m^{-1}$ for shoes-only, to 5.6 (1.5), 5.4 (1.2) and 5.5 (1.1) $J \cdot kg^{-1} \cdot m^{-1}$ for flexible, stiff and rigid vAFOs respectively.

3.3. The effect of varying vAFO stiffness on trunk movements (Aim1)

Trunk lateroflexion range of motion was increased by on average 4–6° ($p < 0.001$) for all vAFO conditions compared to shoes-only (Table 2; Fig. 2). Decreasing vAFO stiffness increased trunk lateroflexion range of motion, but was not significantly different between vAFO conditions (Table 2; Fig. 2). Trunk rotation range of motion was increased by 2–7°($p < 0.001$) compared to shoes-only. It increased with increasing stiffness being significantly different between the flexible and rigid vAFO (Table 2; Fig. 2). Wearing a vAFO did not significantly affect trunk tilt range of motion ($p = 0.095$); Table 2; Fig. 2.

3.4. The effect of varying vAFO stiffness on gait stability (Aim1)

Step width was similar between all walking conditions ($p = 0.870$) (Table 2). ML_MoS decreased gradually with increasing vAFO stiffness ($p = 0.016$) (Table 2). sdML_MoS increased with increasing vAFO stiffness ($p = 0.032$), but only the rigid vAFO stiffness showed a significantly higher sdML_MoS compared to shoes-only (Table 2). No effects were found for different stiffness configurations on gait stability, besides ML_MoS, which was significantly different ($p < 0.05$) between the rigid and flexible vAFO.

3.5. The possible masking effect of trunk movements and gait stability on ECW (Aim2)

No significant relations were found between netEC and trunk range of motion for shoes-only walking. The correlation coefficients between netEC and trunk tilt range of motion, and lateroflexion range of motion were significant for each vAFO stiffness condition, except for the tilt range of motion while walking with rigid vAFOs. Correlation coefficients were strongest for trunk lateroflexion range of motion ($\rho = 0.78–0.81$). Trunk rotation range of motion did not correlate significantly with netEC (Table 3).

The strength of the (negative) correlation coefficients between netEC and ML_MoS increased with increasing vAFO stiffness, but were not statistically significant. Also no relation was found between netEC and AP_MoS. Significant relations were found between netEC and sdML_MoS for the flexible and rigid vAFO (Table 3).

3.6. The impact of trunk movements and gait stability on ECW (Aim3)

Following the results from the correlation tests, trunk lateroflexion range of motion, trunk tilt range of motion, ML_MoS, sdML_MoS, together with peak knee extension during single support, internal knee moment at the timing of peak knee extension in single support, and peak ankle push-off power were included in our multiple regression analysis to explore the impact of these parameters on netEC. For shoes-only, peak ankle power explained 43% of the netEC, with lower ankle power resulting in higher netEC (Table 4). For walking with different vAFOs, trunk lateroflexion range of motion explained most of the netEC (61–75%) (Table 4).

4. Discussion

Our study in children with CP walking in a flexed knee gait

Table 1
Patient characteristics.

		Value
Number of participants		15
Gender	male/female	11/4
GMFCS	I/II/III	2/11/2
Age	years: mean (SD)	10 (2)
Weight (baseline)	kg: mean (SD)	37.2 (9.0)
Height (baseline)	cm: mean (SD)	141 (9.0)
Selective motor control[a]	good/moderate/poor	11/3/1
vAFO use	unilateral/bilateral	1/14
Hip extension RoM[b]	degrees: median [min max]	10 [0 20]
Knee extension RoM	degrees: median [min max]	0 [−10 0]
Popliteal angle	degrees: median [min max]	55 [45 70]
Ankle dorsiflexion (flexed knee) RoM	degrees: median [min max]	10 [0 25]
Ankle dorsiflexion (extended knee) RoM	degrees: median [min max]	0 [-10 10]
Hamstrings spasticity[c]	spasticity scale: [0/1/2/3]	[10/1/3/0]
Soleus spasticity[c]	spasticity scale: [0/1/2/3]	[13/1/0/1]
Gastrocnemius spasticity[c]	spasticity scale: [0/1/2/3]	[13/1/0/1]

Note that range of motion and spasticity scores are provided for the most affected leg.
Abbreviations: *GMFCS = Gross Motor Function Classification Scale, vAFO = ventral shell ankle foot orthosis, RoM = range of motion.*
[a] According to the modified Trost test (Smits et al., 2010 Dev Neurorehabil; Voorman et al., 2007 Arch Phys Med Rehabil).
[b] Hip extension was measured with the participant in prone position. The other measures were performed with the participant in supine position (van den Noort et al., 2010 Arch Phys Med Rehabil; Scholtes, Dallmeijer & Becher 2007 Amsterdam, Ponsen & Looijen BV, p.29–64).
[c] Spasticity was tested according to the Spasticity Test Protocol (Scholtes, Dallmeijer & Becher 2007 Amsterdam, Ponsen & Looijen BV, p.29–64; van den Noort et al., 2010 Arch Phys Med Rehabil; Scholtes, Dallmeijer & Becher 2007 Amsterdam, Ponsen & Looijen BV, p.29–64).

Table 2
Results of generalized estimation equation analyses for walking ability, lower limb biomechanics, trunk range of motion and gait stability.

			Condition				Statistics		
			Shoes (n = 15)	**Flexible (n = 14)**	**Stiff (n = 15)**	**Rigid (n = 15)**			
			mean (SD)	mean (SD)	mean (SD)	mean (SD)	Wald χ2	p	
Walking ability[a]									
Speed	6MWT	[m·min-1]	58.6 (11.3)	58.8 (7.4)	57.5 (8.4)	57.8 (8.0)	1.53	0.675	
ECW	6MWT	[J·kg-1·m-1]	6.1 (1.7)	5.6 (1.5)	5.4 (1.2)	5.5 (1.1)	11.8	0.008	sh-r ◆; sh-s ◆; sh-f ◆;
Lower limb biomechanics[a]									
Peak knee extension (PKE)	single support	[deg]	22.7 (8.7)	18.4 (9.3)	18.1 (8.6)	16.7 (10.0)	31.7	<0.001	sh-r §; sh-s §; sh-f ◆
Internal knee extension moment	timing PKE	[Nm·kg-1]	0.02 (0.18)	−0.09 (0.18)	−0.13 (0.18)	−0.21 (0.23)	24.6	<0.001	sh-r §; sh-s ◆; sh-f ◆; r-f ◆
Ankle power	peak	[W·kg-1]	1.49 (0.71)	1.43 (0.53)	1.21 (0.43)	0.73 (0.30)	91.0	<0.001	sh-r §; r-s §; r-f §
Trunk kinematics									
Tilt	RoM	[deg]	9.1 (3.2)	10.0 (2.3)	8.5 (3.2)	10.1 (2.9)	6.38	0.095	
Lateroflexion	RoM	[deg]	16.7 (6.0)	22.6 (7.8)	21.9 (8.5)	20.5 (7.9)	33.7	<0.001	sh-r §; sh-s §; sh-f §
Rotation	RoM	[deg]	11.9 (4.4)	13.4 (5.9)	15.0 (6.2)	18.4 (4.9)	20.5	<0.001	sh-r §; sh-s ◆; r-f §
Gait stability									
Step width		[m]	0.13 (0.07)	0.14 (0.07)	0.13 (0.05)	0.13 (0.05)	0.72	0.870	
ML_MoS		[m]	0.028 (0.022)	0.016 (0.021)	0.014 (0.025)	0.011 (0.019)	10.4	0.016	sh-r ◆; r-f ◆
AP_MoS		[m]	0.150 (0.058)	0.150 (0.048)	0.149 (0.051)	0.159 (0.066)	0.57	0.903	
sdML_MoS		[m]	0.018 (0.011)	0.021 (0.009)	0.022 (0.008)	0.026 (0.014)	8.83	0.032	sh-r ◆
sdAP_MoS		[m]	0.135 (0.059)	0.136 (0.044)	0.1330.044)	0.143 (0.040)	1.07	0.784	

◆ $p < 0.05$.
§$p < 0.001$.
Abbreviations: ECW, walking energy cost; 6MWT, 6-min walk test; RoM, range of motion; ML, medio-lateral; AP, antero-posterior; MoS, Margins of Stability; SD, standard deviation; sh, shoes-only; r, rigid AFO; st, stiff AFO; f, flexible AFO.
[a] Data derived from Kerkum et al. (PLoS One. 2015 Nov 23; 10 (11):e0142878), which was conducted on the same dataset.

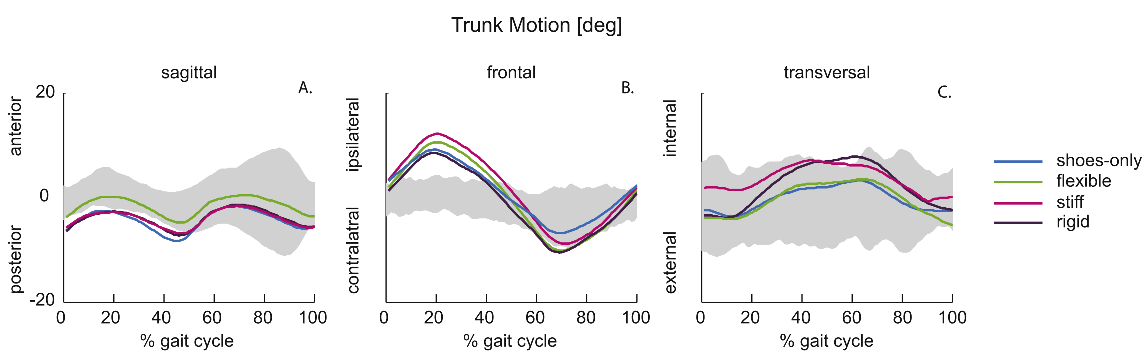

Fig. 2. Mean trunk motion of all participants during walking with shoes-only (n = 15) and the vAFO with different stiffness configurations (flexible [n = 14], stiff [n = 15] and rigid [n = 14]). Different panels show trunk motion in (A) sagittal plane (tilt), (B) frontal plane (lateroflexion) and (C) transversal plane (rotation) for different walking conditions. Shaded areas indicate reference data (i.e. normal walking). While the movement patterns of the trunk do not change, trunk lateroflexion and rotation range of motion increase while wearing vAFOs (Table 2).

pattern showed that wearing vAFOs significantly improved netEC, while trunk lateroflexion and trunk range of motion rotation increased, and mediolateral gait stability reduced compared to walking with shoes-only. Our hypothesis was partly comfirmed, i.e. a higher vAFO stiffness increases the trunk movements, as trunk rotation range of motion increased with increasing stiffness, but this was not significantly different between vAFO stiffness levels. Also, we found a strong positive relation between trunk lateroflexion range of motion and netEC for all vAFO conditions, which was comparable between all vAFO conditions, while no relation between (the smaller) trunk motions and netEC was found for shoes-only walking. This was supported by our regression analysis, indicating that trunk lateroflexion range of motion is the most important parameter regarding changes in netEC while wearing vAFOs. Altogether, our results show that all vAFOs reduced netEC, likely by improved lower limb biomechanics, while affecting trunk range of motion and gait stability, thereby possibly negatively impacting on netEC. This implies that the effect of vAFOs on ECW might be mediated by its effects on trunk movements and gait stability.

Trunk lateroflexion and rotation range of motion increased while wearing vAFOs, compared to shoes-only. This is in accordance to findings in another study, showing that wearing an AFO can increase the trunk frontal angular velocity during gait in CP [8]. Nevertheless, trunk rotation and lateroflexion were not affected similarly in the different walking conditions in our study. Namely, trunk lateroflexion was impacted by vAFO use but not by vAFO stiffness, while trunk rotation appeared to be impacted by vAFO

Table 3
Pearson's correlation results between netEC and trunk range of motion and gait stability (ρ(p)).

	Condition			
	Shoes-only	Flexible vAFO	Stiff vAFO	Rigid vAFO
Tilt RoM	0.24 (0.343)	**0.68 (0.007)**	**0.68 (0.005)**	0.28 (0.341)
Lateroflexion RoM	0.47 (0.078)	**0.78 (0.001)**	**0.81 (< 0.001)**	**0.78 (0.001)**
Rotation RoM	−0.14 (0.615)	−0.13 (0.657)	0.13 (0.639)	−0.13 (0.649)
ML_MoS	−0.06 (0.832)	−0.37 (0.197)	−0.39 (0.155)	−0.53 (0.051)
AP_MoS	−0.41 (0.135)	−0.17 (0.564)	−0.14 (0.611)	−0.19 (0.520)
sdML_ MoS	0.33 (0.224)	**0.57 (0.033)**	0.34 (0.217)	**0.76 (0.002)**
sdAP_MoS	−0.22 (0.440)	0.14 (0.633)	−0.01 (0.960)	−0.16 (0.583)

Statistical significant correlations are presented in bold & italics.
Abbreviations: ML, medio-lateral; AP, antero-posterior; MoS, Margins of Stability; sd, standard deviation; RoM, range of motion.

stiffness. Trunk rotation increased with increasing vAFO stiffness, and was 5° (significantly) greater for the rigid versus the flexible vAFO. We also showed that ankle push-off power was reduced by the rigid vAFO in this study population (Table 2). Yet, our findings suggest that the increased trunk rotation acts as a compensation for the limited push-off power, induced by the (rigid) vAFO. This is in accordance to a study in able-bodied individuals [25], showing that a decreased push-off power leads to increased trunk rotation. The change in trunk lateroflexion while wearing a vAFO could be related to reduced gait stability [11,26], likely by restricting the possibility to use the ankle strategy to maintain a stable gait.

Accordingly, our results show that wearing vAFOs leads to a more unstable gait, in terms of mediolateral stability (Table 2). Specifically, we found that increasing vAFO stiffness gradually decreased mediolateral gait stability, and gradually increased variability of mediolateral gait stability (Table 2). The increase in mediolateral gait stability was a result of increased trunk lateroflexion as the step width did not change with walking condition (Table 2). The increase in gait instability is in accordance with previous literature indicating that wearing hinged AFOs [27] does not improve standing balance in spastic bilateral CP. As increasing vAFO stiffness decreased mediolateral gait stability, it seems that stiffer vAFOs further obstruct the use of the ankle to remain stable in children with CP. There are several mechanisms described in literature to stabilize gait; foot placement (where the foot is positioned; larger and or wider steps), ankle strategy (to apply an active muscle moment around the ankle of the stance foot), and the counter-rotation mechanism (altering the angular momentum of segments around the center of mass to change the direction of the ground reaction force). Given that foot placement was unchanged between walking conditions and the vAFOs obstructed the ankle strategy, participants in our study, most likely used the counter-rotation mechanism (sometimes named the hip strategy) for stability. Even though it has been described that children with CP show an increased dependency on proximal strategies (i.e. counter-rotation mechanism) to maintain stable [27], the ankle strategy does appear to be an important strategy in the current studied sample, given that gait instability increased even though vAFOs only limit ankle movements.

In our previous study on the same data set, we showed that the vAFOs are associated with improved ECW, which was primarily attributed to improved knee kinematics and kinetics [7](Table 2). The current results show that the vAFO's effect on ECW seems to be influenced by its effect on trunk movements and gait stability. Specifically, when walking with shoes-only, neither gait instability nor trunk movements were related to ECW. When wearing vAFOs, however, we found that larger trunk movements (lateroflexion and tilt range of motion) and a more unstable gait (higher sdML_MoS) were related to higher ECW levels. As trunk lateroflexion might be induced by gait instability, it seems that the vAFO's negative effect on gait stability has an important role in its relation to ECW in these children.

Additionally, our regression analysis showed that ankle push-off power is only important in relation to ECW when walking shoes-only (Table 4), even though compliant vAFOs allow similar push-off power compared to walking shoes-only (Table 2). Although these results should be interpreted with caution as our data set is too limited for such analyses, these findings could be of importance in relation to AFO prescription and evaluation. Previously, an individual approach to optimize vAFO stiffness characteristics based on lower limb kinematics has been shown to have promising effects on ECW in CP [28]. Our results suggest that the vAFOs' effects on gait stability and trunk movements might be as relevant as lower limb biomechanics in relation to the ECW reduction. As vAFOs improved ECW compared to shoes-only (Table 2), it seems that normalization of the lower limb kinematics and kinetics by the vAFO may have a greater effect on ECW than the effect of increased trunk movements and gait instability induced by the vAFO. At the same time, it can be hypothesized that the effect of vAFOs on ECW might be mediated by its effects on trunk movements and gait stability.

We did not assess head and arm movements during gait. Hence, the CoM was estimated based on the position of the pelvis and trunk, which may have affected MoS calculations. Furthermore, this study was performed on a small group of CP children and restricted to those walking with excessive knee flexion in stance. Besides, the possible effects of footplate stiffness have not been taken into account. However, all vAFOs were manufactured with a rigid footplate. Although the results should be interpreted with caution, our results are indicative for the effects of vAFOs on whole body kinematics, gait stability and the relation to ECW, and are of great value for clinical practice. More research, preferably in a larger sample of children with CP, is however necessary to confirm our hypotheses. Also, effects of changes in the vAFO-shoe combination to allow more mediolateral movement, and varying the footplate stiffness could be investigated to study the influence on gait stability and

Table 4
Results of the multiple regression analyses using a backward elimination procedure ($p > 0.05$), with netEC as dependent variable.

		R^2	F	β	*p*
Shoes-only	*Peak ankle power*	0.43	9.52	−0.65	.009
Rigid	*Trunk lateroflexion RoM*	0.75	16.1	0.52	.020
	sdML_MoS most affected			0.45	.036
Stiff	*Trunk lateroflexion RoM*	0.65	24.6	0.81	>.0.001
Flexible	*Trunk lateroflexion RoM*	0.61	18.8	0.78	.001

Trunk lateroflexion range of motion), trunk tilt range of motion, sdML_MoS (variability of medio-lateral Margins of Stability), peak knee extension during single support, internal knee moment at the timing of peak knee extension, and peak ankle push-off power [7] were initially included.

ECW.

In conclusion, the results of this study indicate that although wearing vAFOs improves ECW, they also (negatively) affect trunk lateroflexion range of motion, trunk rotation range of motion and gait stability in children with CP walking in a flexed knee gait pattern. The extent of these effects are partly depended on the stiffness of the vAFO. Altogether, our results indicate that clinicians should consider other parameters than lower limb biomechanics alone when prescribing and evaluating the efficacy of vAFOs in CP. As trunk lateroflexion range of motion reflects gait stability, we suggest to consider this parameter in the process of improving the individual's gait pattern and ECW with vAFOs.

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Declaration of competing interest

YK is currently employed by OIM Orthopedie, the company that manufactured all orthoses for the study. However, at the time the study was conducted, she was employed by Amsterdam UMC (as a PhD-student) and had no (financial) relation to OIM in any way.

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