

RESEARCH ARTICLE | *Control of Movement*

# Toe walking in children with cerebral palsy: a possible functional role for the plantar flexors

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**Beyaert C, Pierret J, Vasa R, Paysant J, Caudron S.** Toe walking in children with cerebral palsy: a possible functional role for the plantar flexors. *J Neurophysiol* 124: 1257–1269, 2020. First published September 2, 2020; doi:10.1152/jn.00717.2019.—Equinus and toe walking are common locomotor disorders in children with cerebral palsy (CP) walking barefoot or with normal shoes. We hypothesized that, regardless of the type of footwear, the plantar flexors do not cause early equinus upon initial foot contact but decelerate ankle dorsiflexion during weight acceptance (WA). This latter action promoted by early flat-foot contact is hypothesized to be functional. Hence, we performed an instrumented gait analysis of 12 children with CP (Gross Motor Function Classification System class: I or II; mean age: 7.2 yr) and 11 age-matched typically developing children. The participants walked either barefoot, with unmodified footwear (4° positive-heel shoes), or with 10° negative-heel shoes (NHSs). In both groups, wearing NHSs was associated with greater ankle dorsiflexion upon initial foot contact, and greater tibialis anterior activity (but no difference in soleus activity) during the swing phase. However, the footwear condition did not influence the direction and amplitude of the first ankle movement during WA and the associated peak negative ankle power. Regardless of the footwear condition, the CP group displayed 1) early flattening of the foot and ample dorsiflexion (decelerated by the plantar flexors) during WA and 2) low tibialis anterior and soleus activities during the second half of the swing phase (contributing to passive equinus upon foot strike). In children with CP, the early action of plantar flexors (which typically decelerate the forward progression of the center of mass) may be a compensatory mechanism that contributes to the WA's role in controlling balance during gait.

**NEW & NOTEWORTHY** Adaptation to walking in negative-heel shoes was similar in typically developing children and children with cerebral palsy: it featured ankle dorsiflexion upon initial contact, even though (in the latter group) the soleus was always spastic in a clinical examination. Hence, in children with cerebral palsy, the early deceleration of ankle dorsiflexion by the plantar flexors (promoted by early flattening of the foot, and regardless of the type of footwear) may have a functional role.

adaptability; cerebral palsy; equinus; foot; pathophysiology

## INTRODUCTION

Cerebral palsy (CP) is a group of disorders with permanent, activity-limiting impairments in movement and posture that are attributed to damage to the developing fetal or infant brain (Bax et al. 2005; Graham et al. 2016). The motor disorders in CP result from lesions to the central nervous system (including the upper motor neurons) and the reorganization of central functions (Graham et al. 2016).

Toe walking (defined as an absence of the first heel rocker or as failure of the heel to make contact with the floor at the onset of stance) is a common motor disorder in hemiplegic and diplegic children with CP (Armand et al. 2006; Galli et al. 1999; Rodda and Graham 2001; Winters et al. 1987). Thus, children with CP usually first strike the ground with the foot flat or with the forefoot. This initial contact (IC) is immediately followed by a second rocker: ankle dorsiflexion, where the ankle angle is defined as the foot-tibia angle (Galli et al. 1999; O'Byrne et al. 1998; Perry and Burnfield 2010; Rodda and Graham 2001; Winters et al. 1987). During the initial years of gait in children with CP, the ankle angle at IC usually corresponds to plantar flexion (i.e., equinus) but can evolve to dorsiflexion (associated with prolonged knee flexion) if gait deteriorates into “crouch gait” (Galli et al. 1999; O'Byrne et al. 1998; Rodda and Graham 2001; Winters et al. 1987). In children with CP, equinus upon IC is usually associated with activity of the gastrocnemius and soleus muscles at the end of the swing phase and the beginning of the stance phase (Patikas et al. 2007; van der Krogt et al. 2010). Although equinus is initially only “dynamic,” constant equinus develops over time due to contracture of the gastrocnemius or the gastrocnemius-soleus complex (Graham et al. 2016; Horsch et al. 2019; Nordmark et al. 2009). Dynamic equinus is generally believed to be due to premature (i.e., pre-IC) overactivity of the plantar flexors, caused by hyperexcitable stretch reflexes (spasticity, according to Lance's definition) in these muscles (Graham et al. 2016; Lance 1980; Winters et al. 1987). However, the existence and/or functional significance of exaggerated stretch reflexes during walking in spastic patients has been questioned for decades (Dietz et al. 1981; Dietz and Sinkjaer 2007; Hodapp et al. 2007; Sinkjaer and Magnussen 1994). Recently, reflex measurement studies of children with CP failed to evidence exaggerated sensory inputs to the ankle plantar flexors during the swing phase of walking (Willerslev-

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Olsen et al. 2014). In contrast, toe walking in children with CP and in typically developing (TD) children is characterized by feedforward control of the ankle muscles, although some differences in the electromyographic (EMG) pattern are observed (Lorentzen et al. 2019). This finding suggests that toe walking is part of an adaptive process (Lorentzen et al. 2019).

At the beginning of the second rocker phase of gait, the plantar flexors 1) decelerate the ankle dorsiflexion and the trunk's forward progression and 2) accelerate the trunk upward for body support (Neptune et al. 2001; Perry and Burnfield 2010). It is noteworthy that the plantar flexors can exert a direct action on the tibia and an indirect action on segments above the tibia only when the anterior part of the foot lever arm is touching the ground—i.e., either when the foot is flat on the ground or in the absence of heel contact (Neptune et al. 2001; Perry and Burnfield 2010). In toe walking, plantar flexors exert an action from the IC onward. In the first weeks or months of walking, toddlers commonly display toe walking; this is followed by a gait pattern with an early second rocker phase (Forsberg 1992; Sutherland et al. 1980). Both patterns are associated with early activity of the plantar flexors (Chang et al. 2006; Sutherland et al. 1980). The plantar flexors exert negative ankle power during WA, and this phenomenon is significantly more intense in children under the age of 4 than in older children (Cupp et al. 1999; Samson et al. 2011, 2013). The early action by plantar flexors during WA in early childhood has functional benefits, since it helps to decelerate the body and stabilize forward gait (Kuo and Donelan 2010; Samson et al. 2011) while the child is learning to 1) generate and control propulsive forces (Bril et al. 2015) and 2) increase the dimensionless step length and frequency (Vaughan et al. 2003). Children with CP typically have a low dimensionless step length (O'Malley et al. 1997) and a toe-walking pattern that is associated with early activation of the plantar flexors in the stance phase (Berger et al. 1982; Colborne et al. 1994; Wong et al. 1999). Thus, from the beginning of the stance phase, the plantar flexors decelerate the ankle dorsiflexion (Buckon et al. 2004; Cimolin et al. 2007; Davids et al. 1999) and decelerate the body's center of mass more than is seen in controls (Kurz et al. 2010). In other words, toe walking allows the plantar flexors to make a significant contribution to the negative work exerted by the leading leg joints during the weight acceptance (WA) phase of gait (Worthen-Chaudhari et al. 2014). This can 1) prevent joint collapse and provide vertical support (Liu et al. 2006; Winter 1980) and 2) decelerate the body's center of mass (Donelan et al. 2002; Kuo and Donelan 2010).

Thus, a combination of early equinus and toe walking in children with CP walking barefoot might reflect an adaptation of the foot-ankle kinematics that enable the plantar flexors to contribute early to power absorption during WA rather than being the result of spastic excessive activation of the plantar flexors during the swing phase. We reasoned that asking children with CP to walk either barefoot or in shoes with different sole inclinations would change foot kinematics (relative to TD children) during the swing phase and WA. This change would allow early ground contact (regardless of whether this is forefoot or fore-shoe contact) and thus early ankle power absorption. Thus, the primary objective of the present study was to determine whether or not children with CP have a specific foot-ankle pattern (compared with TD children), based on the plantar flexors' action at WA and regardless of the footwear condition. We further hypothesized that 1) this specific foot-ankle pattern would

include (again, relative to TD children) early flat-foot or flat-shoe ground contact at WA; 2) this ground contact would be favored by less intense tibialis anterior (TA) activity and the absence of concomitant higher soleus activity during the second half of the swing phase; and 3) the plantar flexors would exert early, more intense negative ankle power to decelerate ankle dorsiflexion. The study's secondary objective was to characterize foot-ankle adaptation to different footwear conditions in children with CP and in TD children. In the children with CP, maintaining early flat-foot or flat-shoe contact during WA would be associated with early ankle plantar flexion when walking barefoot or walking with conventional, positive-heel shoes (PHSs), and with early ankle dorsiflexion when walking with negative-heel shoes [NHSs, i.e., shoes in which the heel is thinner than the front of the shoe (Fig. 1)]. Indeed, placing an NHS flat on the ground requires ankle dorsiflexion, i.e., motor adaptation with higher TA activity during the swing phase. However, this adaptive ankle dorsiflexion in the swing phase and upon IC would be prevented if equinus is due to overactive plantar flexors during the swing phase. We hypothesized that in both children with CP and TD children, wearing NHSs would induce 1) ankle dorsiflexion in the swing phase and upon IC, and 2) greater TA activity [but no change in soleus activity] during the swing phase—as previously reported in healthy adults (Li and Hong 2007)]. Thus, in the NHS condition (versus barefoot and PHS conditions), ankle dorsiflexion in the swing phase and upon IC would shift from plantar flexion to dorsiflexion in children with CP and, to an even greater extent, in TD children.

## PATIENTS AND METHODS

**Participants.** Twelve children with CP (8 boys and 4 girls; mean  $\pm$  SD duration of pregnancy: 32  $\pm$  6 wk; mean age: 7.2  $\pm$  1.7 yr; mean body weight: 23  $\pm$  3.9 kg; mean leg length: 58  $\pm$  6 cm) and 11 age-matched TD children (7 boys and 4 girls, mean  $\pm$  SD age: 7.7  $\pm$  2.3 yr; mean body weight: 23.7  $\pm$  4.3 kg; mean leg length: 64  $\pm$  8 cm) participated in the study. The children with CP were diplegic ( $n = 8$ ) or hemiplegic ( $n = 4$ ). The main inclusion criteria were as follows: age between 4 and 12 yr, presence of triceps surae spasticity in a clinical examination, no or minimal contracture of the triceps surae (defined as maximal ankle dorsiflexion of 5° or more with the knee extended), and the ability to stand upright with heel contact on a board inclined at an angle of 15°. The main exclusion criteria were the requirement for a walking aid, minimal hip flexion above 20° in a clinical examination, pain in the lower legs when standing or walking, botulinum toxin injections in the previous 6 mo, and lower limb surgery in the previous 12 mo. Between 7 and 42 mo before inclusion, 5 of the 12 children with CP had already received one or two botulinum toxin injections in the gastrocnemius ( $n = 5$ ) and the hamstring muscles ( $n = 2$ ). Another child with CP had undergone bilateral femoral derotation osteotomy 22 mo before inclusion. In the TD group, the additional exclusion criteria were a history of neurological or orthopedic disease, a history of lower limb surgery, and pain in the lower legs when standing or walking. The children's parents gave their written, informed consent, and (depending on age) the children also gave their verbal informed consent. The study was approved by the local institutional review board (*CPP Est III*, Nancy, France; reference: 10.10.01).

**Experimental procedures.** After the clinical examination, a three-dimensional (3D) gait analysis (including an EMG recording) was performed. Participants were asked to walk at a self-selected speed under three footwear conditions: barefoot, wearing PHSs, and wearing NHSs (in that order). In each condition, the children were asked to walk several times along a 12-m track with embedded force sensors. All the kinematic and kinetic data reported here were averaged over three gait

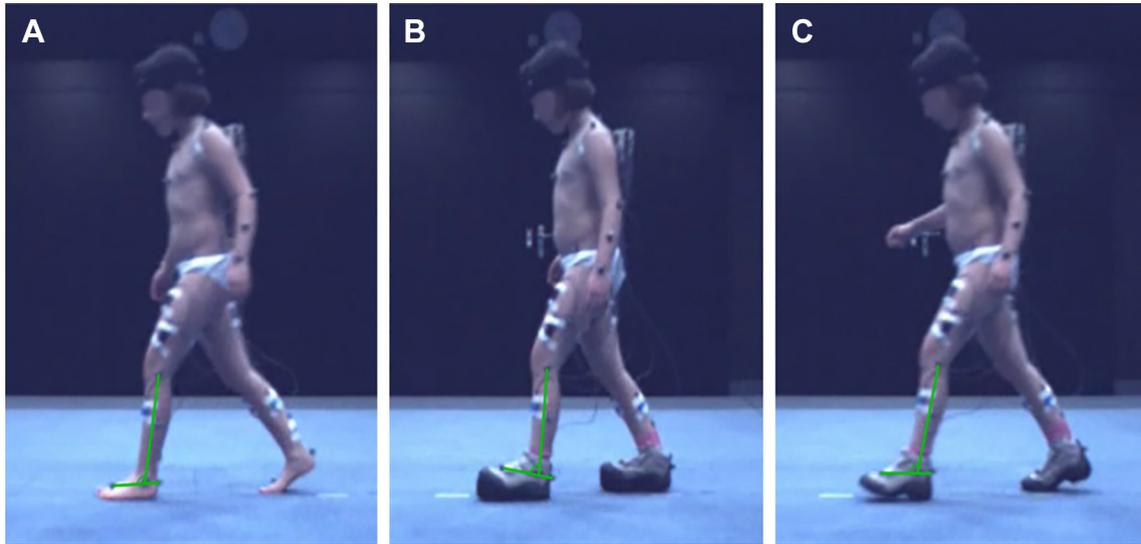


Fig. 1. Left foot initial contact for a left hemiplegic child with cerebral palsy while walking barefoot (A), with negative-heel shoes (NHSs,  $-10^\circ$ ; B), or with positive-heel shoes (PHSs,  $+4^\circ$ ; C). The green lines indicate the foot-tibia angle. Note that the bare foot or NHS was flat when it touched the ground, which is associated with ankle plantar flexion and dorsiflexion in barefoot and NHS conditions, respectively.

cycles. The three gait cycles were selected after a familiarization period during which the children walked up and down the track at least twice in the barefoot and PHS conditions and at least five times in the NHS condition. We studied the affected limb in hemiplegic children with CP, the limb with the more spastic soleus in diplegic children with CP, and the right limb in TD children. Unfortunately, the PHS condition was not performed in one of the children with CP.

**Shoes used in the study.** The same commercially available hiking boots (with a  $4^\circ$  positive instep) were worn in the PHS condition and (after modification) in the NHS condition. The modification consisted in creating a  $10^\circ$  negative instep, i.e., the boot's heel was thinner than the effective sole at the front of the boot (Fig. 1). The boot had been modified by a pedorthist using light foam (Chaussures Orthopediques G'ssell, Nancy, France).

**Functional scores and evaluation of soleus spasticity.** In the CP group, three functional scales were assessed: 1) the Gross Motor Function Classification System for classifying children with cerebral palsy on the basis of their abilities or limitations in gross motor function (levels I to V, where level I is best) (Palisano et al. 1997); 2) the Gillette Functional Assessment Questionnaire, for evaluating walking abilities in all community settings and over various terrains (ranging from 1, nonambulatory, to 10, best ambulatory) (Novacheck et al. 2000); and 3) the Functional Mobility Scale over three distinct distances (ranging from 1 to 18, where 18 is best) (Graham et al. 2004). In the CP group, soleus spasticity was assessed on the Tardieu scale (Gracies et al. 2010). Passive ankle dorsiflexion at slow and fast velocities was measured while the knee was flexed at  $90^\circ$ . Two variables were assessed: the Spasticity Angle X (i.e., the difference between the arrest angle at slow speed and the catch-and-release (clonus) angle at fast speed) and the Spasticity Grade Y [an ordinal variable that grades the intensity (gain) of the muscle's response to fast stretching] (Gracies et al. 2010).

**Three-dimensional kinetic and kinematic gait analyses.** The ground reaction force and the kinetic variables were computed from recordings on three 3D force platforms (OR6 and BP series, AMTI, Watertown, MA) integrated into the 12-m walking track. The two smaller (OR) platforms were placed together in the direction of gait, and a larger (BP) platform was placed alongside them. Kinetic data were sampled at 1,000 Hz and then synchronized with kinematic data. Kinematic data were recorded using a nine-camera instrumented 3D gait analysis system (the Vicon Nexus 1.8.5 from Vicon Motion Systems Ltd, Oxford

Metrics, Oxford, UK). Reflective markers were placed on the skin and shoes, according to a standard gait model (the Vicon Plug-in Gait model) (Baker et al. 2017; Louey and Sangeux 2016). The marker trajectories were sampled at 100 Hz and filtered with a Woltring filter (mean square error:  $20 \text{ mm}^2$ ). The markers were labeled, and the standard gait model was applied. In particular, the model described the foot as a rod defined by a marker at the heel ("Heel") and at the dorsal surface of the forefoot ("Toe"); these markers are aligned with the sole of the foot in the sagittal plane, in the barefoot, PHS, or NHS conditions (Baker et al. 2017; Louey and Sangeux 2016). Ankle dorsiflexion and plantar flexion were calculated as the foot's angular rotation around the lateral axis of the tibia (Nair et al. 2010).

**Electromyographic recordings.** The EMG activity was recorded using bipolar electrodes (AMBU Blue sensor N, N-10-A/25, AMBU, Ballerup, Denmark; recording area  $0.5 \text{ cm}^2$ ; interelectrode distance 2 cm) placed over the TA and the soleus muscle according to the Surface Electromyography for the Non-Invasive Assessment of Muscle guidelines (Freriks et al. 1999). Since the gastrocnemius and soleus muscles have similar levels of activity during the swing phase in children with CP (Patikas et al. 2007; van der Krogt et al. 2010), the single-joint soleus muscle was chosen for the present study. Electromyographic activity was synchronized with the 3D gait data using Vicon Nexus software (Vicon Motion Systems Ltd). The EMG signals were band-pass filtered (25 Hz to 1 kHz), amplified ( $\times 2,000$ ), sampled at 1 kHz, and stored on a personal computer for offline analysis. Next, the EMG records were rectified and low-pass filtered at 10 Hz (using a fourth-order Butterworth filter) to create a linear envelope. An average profile was constructed for each participant over five gait cycles for each condition, and then a grand average was calculated for all participants. The EMG activity was normalized against the maximum activity in the barefoot condition.

**Variables of interest.** The following time-distance variables were analyzed: dimensionless walking speed, dimensionless stride length [both normalized using Hof and Zijlstra's method (Hof and Zijlstra 1997)], and the duration of the swing phase as a percentage of the gait cycle (%GC).

During the swing phase (i.e., between foot-off and foot IC), the following kinematic and EMG variables were analyzed: peak ankle dorsiflexion (in  $^\circ$ ) at the midpoint of the swing phase (i.e., in the middle of three swing phases of equal duration), ankle dorsiflexion upon IC (in  $^\circ$ ), and the difference between these two variables. We also measured the

Table 1. Time-distance variables in the TD and CP groups

|  | TD                   |                    | CP                 |                      | F-Value            |                    | P-Value         |                 | F-Value             |                         | P-Value                   |                           |
|--|----------------------|--------------------|--------------------|----------------------|--------------------|--------------------|-----------------|-----------------|---------------------|-------------------------|---------------------------|---------------------------|
|  | Barefoot<br>(n = 11) | TD<br>PHS (n = 11) | TD<br>NHS (n = 11) | Barefoot<br>(n = 12) | CP<br>PHS (n = 11) | CP<br>NHS (n = 12) | Group<br>Factor | Group<br>Factor | Condition<br>Factor | Condition<br>Factor     | Group*Condition<br>Factor | Group*Condition<br>Factor |
| Gait speed<br>(dimensionless)          | 0.45 (0.07)          | 0.47 (0.11)        | 0.47 (0.08)        | 0.48 (0.07)          | 0.44 (0.10)        | 0.46 (0.09)        | <i>0.0</i>      | <i>0.889</i>    | <i>0.5</i>          | <i>0.598</i>            | <i>1.9</i>                | <i>0.157</i>              |
| Swing phase duration<br>(% gait cycle) | 40 (2)               | 40 (3)             | 40 (3)             | 42 (1)               | 40 (2)             | 41 (3)             | <i>1.5</i>      | <i>0.238</i>    | <i>1.3</i>          | <i>0.297</i>            | <i>0.8</i>                | <i>0.472</i>              |
| Stride length<br>(dimensionless)       | 1.61 (0.17)          | 1.87 (0.18)        | 1.86 (0.34)        | 1.53 (0.18)          | 1.69 (0.23)        | 1.72 (0.27)        | <i>2.6</i>      | <i>0.122</i>    | <b><i>9.6</i></b>   | <b><i>&lt;0.001</i></b> | <i>0.5</i>                | <i>0.591</i>              |

Values are mean (SD). CP, cerebral palsy; TD, typically developing. *F*-values and *P*-values are all italicized and additionally bolded when statistically significant.

knee flexion at IC (in °). The integrated EMG activity of the TA and the soleus (units: %peak×%GC×10<sup>-1</sup>) were measured during the first half and second half of the swing phase. Indeed, during the first half of the swing phase, the dorsiflexors (primarily the TA) lift the foot out of plantar flexion for foot clearance; maximum dorsiflexion of the ankle and minimum plantar flexion occur approximately at the middle of the swing phase (Perry and Burnfield 2010). During the second half of the swing phase, the action of the dorsiflexors and plantar flexors will influence the foot's kinematic pattern during the WA phase, from IC onward (Perry and Burnfield 2010).

During the WA phase [defined as the period of combined initial power absorption activity around the lower limb joints (Worthen-Chaudhari et al. 2014)], we measured the amplitude (in W/kg) and the time point (in %GC) of the peak ankle negative total power associated with the ankle's first sagittal movement. In TD children, the first ankle movement is characterized by movement toward plantar flexion from IC to the first maximum ankle plantar flexion (or minimum ankle dorsiflexion). In children with CP, the first ankle movement is usually characterized by a movement toward dorsiflexion from IC to the first maximum ankle dorsiflexion (Lin et al. 2000; Winters et al. 1987), as observed in the CP group (Fig. 3B). We measured the amplitude (in °) and direction (dorsiflexion or plantar flexion) of the first ankle movement, the ankle dorsiflexion angle at the end of the first ankle movement (in °), and the first ankle movement's time point (in %GC). The type of foot contact with the ground (heel-only, flat-foot, or forefoot) upon IC was assessed in a lateral view (Fig. 1), and the most frequent type of contact during the analyzed gait cycles was recorded for each child. The time of first flat-foot contact with the ground (in %GC) was calculated from the trajectories of the Heel and Toe markers.

Data were processed using Vicon Nexus software (version 1.8.5), Vicon Polygon software (version 3.5.2; both from Vicon Motion Systems Ltd), and MATLAB R2017 software (MathWorks, Inc., Natick, MA). The shadedErrorBar Matlab function [Rob Campbell (2020). raacampbell/shadedErrorBar (<https://github.com/raacampbell/shadedErrorBar>), GitHub. Retrieved July 3, 2020] was used in the figures representing the kinematic, kinetic, and electromyographic variables (Figs. 3 and 4).

**Statistical analyses.** Quantitative data were expressed as the mean ± SD. We performed a repeated-measures analysis of variance with a general linear model, "group" as the between-participant factor (TD group; CP group), and the "footwear condition" (barefoot; PHS; NHS) as the within-participant factor. Tukey's honestly significant difference test was used for post hoc comparisons, when necessary. The threshold for statistical significance was set to  $\alpha = 0.05$ . Pearson's correlation was calculated, to probe the relationship tested between muscle activity and ankle kinematic variables. All statistical analyses were performed using Statistica software (version 13, Tibco Software Inc., Palo Alto, CA).

## RESULTS

**Functional scores and soleus spasticity.** All 12 children in the CP group had good motor function and high levels of

mobility. Two were graded I according to the Gross Motor Function Classification System, and 10 were graded II. The mean ± SD Gillette Functional Assessment Questionnaire score was 8.8 ± 1.4 and the mean ± SD Functional Mobility Scale was 16.3 ± 2.5. The soleus muscle was spastic, with mean ± SD spasticity angle X of 16° ± 8 and a spasticity grade Y of 1 (*n* = 3) or 2 (*n* = 9).

**Time-distance variables.** There were no significant differences between the two groups or between the three conditions with regard to the walking speed and swing phase duration (Table 1). Likewise, there were no significant group×condition interactions. These results are important because walking speed influences kinematic, kinetic, and EMG signals (Schwartz et al. 2008). Stride length was significantly influenced only by the condition, with a greater distance in both the NHS and PHS conditions than in the barefoot condition.

**The WA phase.** Left-side IC by a left-side hemiplegic child in each of the three conditions is illustrated in Fig. 1. It is noteworthy that 1) in the barefoot condition, flat-foot IC was made (i.e., with plantar flexion); 2) in the NHS condition, the front and back of the boot contacted the ground together (with ankle dorsiflexion); and 3) in the PHS condition, the IC was made by the heel (with plantar flexion).

During WA, all the children in the TD and CP groups placed their foot flat on the ground. However, the groups differed markedly with regard to the foot kinematics and the associated ankle kinetics (effect of group, *P* < 0.001 for all comparisons, and with all three conditions taken together) (Table 2, Figs. 2 and 3). Thus, in the CP group, the first flat-foot contact occurred early (at 3 ± 3%GC, on average) and was generally preceded by flat-foot contact with ankle plantar flexion (−2 ± 9°) upon IC. The first movement of the ankle was ample (16 ± 6°, moving toward dorsiflexion), ended at 14 ± 4%GC, and was associated with a high peak negative ankle power (−1.8 ± 1.0 W/kg) at the middle (7 ± 3%GC) of the first ankle dorsiflexion movement. Knee flexion upon IC was higher in the CP group (21 ± 9°) than in the TD group (8 ± 6°). In contrast, heel strike with ankle dorsiflexion (+9 ± 8°) upon IC was most common in TD children. The first flat-foot contact occurred at 12 ± 3%GC. The first ankle movement was small (9 ± 4°, moving toward plantar flexion), stopped early (at 5 ± 2%GC), and was associated with a low peak negative ankle power (−0.4 ± 0.3 W/kg) at the middle (2 ± 2%GC) of the first ankle plantar flexion. Thus, the mechanical effects of negative ankle power were completely different in the TD versus CP groups. In the TD group, negative ankle power decelerated the ankle's plantar flexion before the foot flattened on the ground. In the CP group, negative ankle power exerted a deceleration of ankle dorsiflexion and

Table 2. Knee and ankle sagittal kinematics, foot ground contact patterns, and ankle total power during the WA phase, in the TD and CP groups

|  | TD                   |                 |                   | CP                             |                  |                   | F-Value         |                   | P-Value             |                     | F-Value                       |                               | P-Value |  |
|--|----------------------|-----------------|-------------------|--------------------------------|------------------|-------------------|-----------------|-------------------|---------------------|---------------------|-------------------------------|-------------------------------|---------|--|
|  | Barefoot<br>(n = 11) | PHS<br>(n = 11) | NHS<br>(n = 11)   | Barefoot<br>(n = 12)           | PHS<br>(n = 11)  | NHS<br>(n = 12)   | Group<br>Factor | Group<br>Factor   | Condition<br>Factor | Condition<br>Factor | Group×<br>Condition<br>Factor | Group×<br>Condition<br>Factor |         |  |
| IC knee flexion (°)                              | 6 (5)                | 5 (6)           | 11 (5)            | 21 (6)                         | 21 (10)          | 21 (10)           | <b>23.4</b>     | <b>&lt; 0.001</b> | 2.5                 | 0.098               | 3.1                           | 0.054                         |         |  |
| IC ankle DF (°)                                  | 5 (3)                | 7 (9)           | 16 (7)            | -8 (8)                         | -4 (8)           | 6 (6)             | <b>20.6</b>     | <b>&lt; 0.001</b> | <b>33.4</b>         | <b>&lt; 0.001</b>   | 0.2                           | 0.844                         |         |  |
| IC ground contact (heel, flat, forefoot)         | 10 heel<br>1 flat    | 11 heel         | 10 heel<br>1 flat | 3 heel<br>5 flat<br>4 forefoot | 8 heel<br>3 flat | 2 heel<br>10 flat |                 |                   |                     |                     |                               |                               |         |  |
| First flat-foot contact (% Gait cycle)           | 11 (1)               | 15 (3)          | 9 (2)             | 3 (2)                          | 5 (3)            | 1 (2)             | <b>133.2</b>    | <b>&lt; 0.001</b> | <b>28.4</b>         | <b>&lt; 0.001</b>   | 1.8                           | 0.178                         |         |  |
| First ankle movement (°) (positive if toward DF) | -6 (2)               | -9 (5)          | -5 (5)            | 15 (7)                         | 17 (6)           | 16 (6)            | <b>183.4</b>    | <b>&lt; 0.001</b> | 0.7                 | 0.509               | 3.2                           | 0.054                         |         |  |
| Ankle DF at the end of first ankle movement (°)  | -1 (4)               | -2 (7)          | 11 (6)            | 7 (7)                          | 13 (11)          | 22 (9)            | <b>22.1</b>     | <b>&lt; 0.001</b> | <b>37.7</b>         | <b>&lt; 0.001</b>   | 2.0                           | 0.144                         |         |  |
| End of first ankle movement (% GC)               | 5 (1)                | 8 (2)           | 4 (1)             | 14 (4)                         | 17 (5)           | 11 (2)            | <b>95.0</b>     | <b>&lt; 0.001</b> | <b>26.1</b>         | <b>&lt; 0.001</b>   | 0.8                           | 0.452                         |         |  |
| WA peak ankle power (watts/kg)                   | -0.4 (0.2)           | -0.7 (0.4)      | -0.2 (0.2)        | -1.3 (0.8)                     | -1.9 (1.0)       | -2.1 (1.1)        | <b>29.2</b>     | <b>&lt; 0.001</b> | 2.9                 | 0.067               | 2.7                           | 0.084                         |         |  |
| WA peak ankle power occurrence (% GC)            | 2 (2)                | 3 (1)           | 2 (1)             | 7 (2)                          | 10 (3)           | 6 (2)             | <b>49.0</b>     | <b>&lt; 0.001</b> | <b>12.1</b>         | <b>&lt; 0.001</b>   | 2.9                           | 0.065                         |         |  |

Values are mean (SD). CP, cerebral palsy; DF, dorsiflexion; IC, initial contact; TD, typically developing; WA, weight acceptance phase. *F*-values and *P*-values are all italicized and additionally bolded when statistically significant.

anterior tibia tilt (since the foot was already flat on the ground soon after IC).

The footwear condition significantly altered the foot kinematics when considering the two groups together (effect of condition,  $P < 0.001$  for all comparisons) (Table 2, Figs. 2 and 3). The magnitude of ankle dorsiflexion upon IC was significantly greater in the NHS condition ( $11^\circ \pm 8$ ) than in the barefoot condition ( $-2 \pm 9^\circ$ ;  $P < 0.001$ ) and the PHS condition ( $2 \pm 10^\circ$ ,  $P < 0.001$ ); this was also true at the end of the first ankle movement ( $17 \pm 9^\circ$ ,  $P < 0.001$  versus  $3 \pm 7^\circ$  and  $6 \pm 12^\circ$ , respectively). The first flat-foot contact occurred significantly earlier in the NHS condition ( $5 \pm 12\%$ GC) than in the barefoot condition ( $7 \pm 4\%$ GC,  $P = 0.005$ ) and the PHS condition ( $10 \pm 6\%$ GC,  $P < 0.001$ ). The same was true for the end of the first ankle movement ( $7 \pm 4\%$ GC for the NHS condition versus  $10 \pm 5\%$ GC for the barefoot,  $P = 0.01$ , and  $12 \pm 6\%$ GC for the PHS,  $P < 0.001$ ). Furthermore, the first flat-foot contact ( $P < 0.001$ ), the end of the first ankle movement ( $P < 0.001$ ), and the peak ankle power ( $7\% \pm 4$  GC versus  $5 \pm 3\%$ GC;  $P = 0.008$ ) occurred significantly later in the PHS condition than in the barefoot condition. However, the footwear factor did not significantly alter the direction and amplitude of the first ankle movement during WA or the associated peak ankle power. It should be noted that the footwear conditions was associated with different IC modes (but similar knee flexion angles) in the CP group only. Whereas IC for the TD children usually involved the heel in all three footwear conditions, the children with CP mostly displayed flat-foot and forefoot contact in the barefoot condition, heel contact in the PHS condition, and flat-foot contact in the NHS condition (see Table 2). We did not observe significant interactions between group and condition factors for any of the other variables during WA.

**The swing phase.** During the swing phase, the foot is usually repositioned for IC; this characterizes the end of the

swing phase and the beginning of the stance phase. As was seen for ankle dorsiflexion upon IC, the peak ankle dorsiflexion at midswing was significantly lower in the CP group than in the TD group ( $0 \pm 9^\circ$  versus  $+9 \pm 8^\circ$ , respectively;  $P < 0.001$ ) when all the conditions were pooled (Table 3, Figs. 2 and 3). The significant main effect of condition and a post hoc analysis indicated that peak ankle dorsiflexion at midswing was also significantly higher in the NHS condition ( $12 \pm 8^\circ$ ) than in the barefoot condition ( $-1 \pm 9^\circ$ ;  $P < 0.001$ ) and PHS condition ( $3 \pm 6^\circ$ ;  $P < 0.001$ ) when the two groups were pooled. We did not observe any significant interactions between group and condition. The difference between ankle dorsiflexion upon IC and peak ankle dorsiflexion at midswing was much the same in the CP and TD groups (CP:  $-2 \pm 4^\circ$  versus TD:  $0 \pm 4^\circ$ ,  $P = 0.14$ ) and between the footwear conditions (NHS:  $-1 \pm 3^\circ$ ; barefoot:  $-1 \pm 3^\circ$ ; PHS:  $-1 \pm 5^\circ$ ,  $P = 0.82$ ) but there was a significant interaction between the group and the footwear condition factors ( $P = 0.005$ ). Thus, from midswing to IC in the PHS condition, the ankle displayed plantar flexion in the CP group ( $-4 \pm 5^\circ$ ,  $P = 0.025$ ) but dorsiflexion in the TD group ( $2 \pm 5^\circ$ ) (Fig. 2). In summary, ankle dorsiflexion upon IC was mainly related to peak ankle dorsiflexion at midswing—apart from in the PHS condition, where the children with CP (but not the TD children) added a significant plantar flexion movement than that contributed to the ankle plantar flexion upon IC (CP:  $-4 \pm 8^\circ$  versus TD:  $+7 \pm 9^\circ$ ) and early flat-shoe ground contact upon IC (CP:  $5 \pm 3\%$ GC versus TD:  $15 \pm 3\%$ GC).

The integrated EMG activities of the TA and soleus were recorded for all children in the TD group ( $n = 11$ ) but for only 7 of the 12 children in the CP group (Fig. 4); the other 5 children (generally the youngest) were afraid of the EMG electrodes. In the CP group, TA activity did not differ significantly from that in the TD group during the first half of the swing phase (CP:  $110 \pm 35$  versus TD:  $92 \pm 30$ ;  $P = 0.14$ , see Table 3) but was significantly lower than in the TD group during the second half of

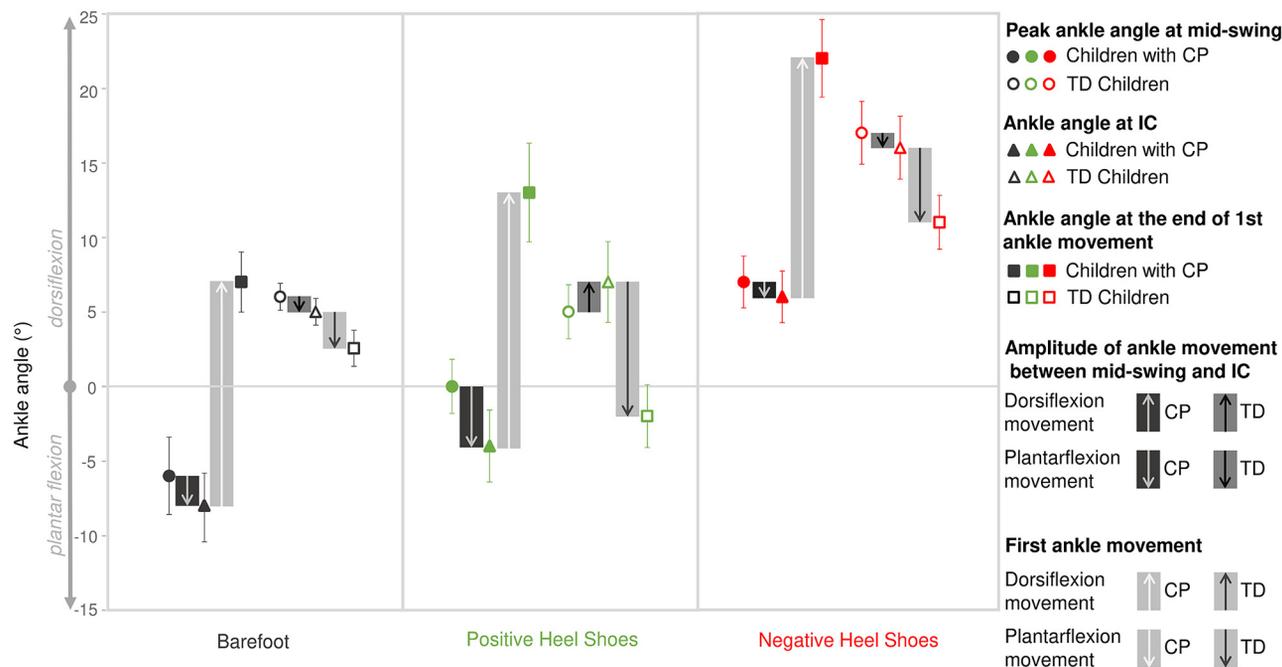


Fig. 2. Ankle angles at midswing (measured at the peak), at initial contact (IC), and at the end of the first ankle movement during weight acceptance and amplitudes of the dorsiflexion / plantarflexion ankle movements between these time markers in children with cerebral palsy (CP) and in typically developing (TD) children. The mean peak ankle angle at midswing (circle symbols), the mean ankle angle at IC (triangle symbols), and the mean ankle angle at the end of the first ankle movement (square symbols) are depicted with solid symbols for the CP group ( $n = 12$ ) and with open symbols for the TD group ( $n = 11$ ) in the barefoot condition (black symbols, *left*), positive-heel-shoe condition (PHS, green symbols, *middle*), and negative-heel-shoe condition (NHS, red symbols, *right*). Error bars represent  $\pm 1$  standard error. Amplitudes of the dorsiflexion (upward arrow) or plantarflexion (downward arrow) ankle movements are indicated by dark gray bands for the movement between midswing peak and IC and by light gray bands for the movements between IC and the end of the first ankle movement.

the swing phase (CP:  $41 \pm 11$  versus TD:  $90 \pm 40$ ;  $P = 0.001$ ). In contrast, soleus activity was significantly lower in the CP group during the first half of the swing phase (CP:  $13 \pm 8$  versus TD:  $40 \pm 33$ ;  $P = 0.018$ ) but did not differ significantly during the second half of the swing phase (CP:  $26 \pm 14$  versus TD:  $24 \pm 11$ ,  $P = 0.63$ ). It should be noted that at the middle of the second half of the swing phase, TA activity in the CP group was at its lowest (considering the whole gait cycle) but was quite high in the TD group (Fig. 4A). The TA activity then increased at the end of the second half of the swing phase; a peak occurred at WA in both groups but was lower in the CP group (Fig. 4A). The soleus activity during the second half of the swing phase was similar in the two groups, with a low level for the first part of this period, an increase at the end of the period, and a peak at WA (which was higher in the CP group) (Fig. 4B). The peak ankle dorsiflexion at midswing was not correlated with the TA or soleus EMG activity during the first half of the swing phase ( $r = 0.21$ ,  $P = 0.15$  and  $r = 0.22$ ,  $P = 0.14$ , respectively). However, ankle dorsiflexion upon IC was significantly correlated with TA activity ( $r = 0.475$ ,  $P < 0.001$ ) but not with soleus activity during the second half of the swing phase ( $r = 0.055$ ,  $P = 0.7$ ).

The effect of footwear condition factor was only significant for TA EMG activity in both halves of the swing phase (Table 3). Thus, TA activity was significantly higher in the NHS condition than in the barefoot or PHS conditions during the first half of the swing phase (NHS:  $119 \pm 41$  versus barefoot:  $92 \pm 17$ ,  $P = 0.003$ ; and PHS:  $86 \pm 27$ ,  $P < 0.001$ ) and the second half of the swing phase (NHS:  $90 \pm 52$  versus barefoot:  $60 \pm 22$ ,  $P < 0.001$ ; and PHS:  $64 \pm 36$ ,  $P = 0.001$ ). Furthermore, the group  $\times$  condition

interaction was significant for the TA activity in the second half of the swing phase. In the TD group, greater TA activity was observed in the NHS condition than in either of the other two footwear conditions (barefoot:  $P < 0.001$ , PHS:  $P = 0.005$ ). Furthermore, the mean TA activity value in the NHS condition was significantly greater in the TD group than in the CP group ( $P = 0.004$ ).

## DISCUSSION

Our present results showed that the CP and TD groups displayed highly different foot-ankle patterns from the swing phase to WA, regardless of the footwear condition. The TD children mainly displayed heel strike with ankle dorsiflexion upon IC; this was preceded by biphasic TA activity and low soleus activity during the swing phase and followed by short, small, decelerated ankle plantar flexion and late flat-foot or flat-shoe ground contact. As we hypothesized, the children with CP all displayed early flat-foot or flat-shoe ground contact at WA. This contact was preceded by low TA and soleus activities during the second half of the swing phase and was associated with negative ankle power that decelerated the ankle dorsiflexion.

The footwear condition significantly modified certain aspects of ankle kinematics but did not influence the direction and amplitude of the first ankle movement during WA and the associated peak negative ankle power. As we had hypothesized, wearing NHSs was associated with a non-disease-specific increase (i.e., observed in both CP and TD groups) in ankle dorsiflexion at midswing and upon IC, and an increase in TA activity (but no change in soleus activity) during the swing phase. Children with

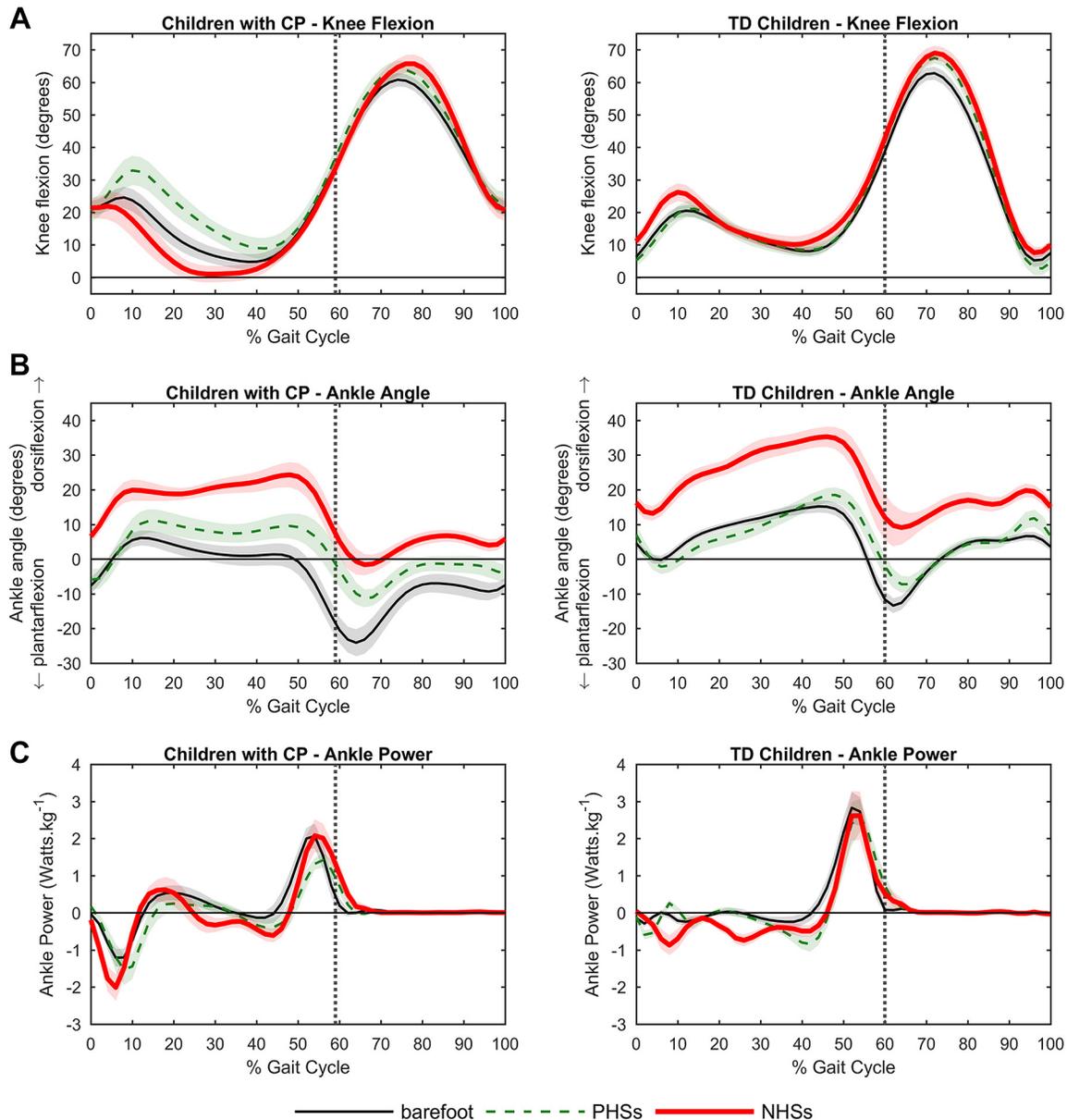


Fig. 3. Mean knee flexion (A), ankle dorsiflexion/plantarflexion angle (B), and ankle total power (C) profiles in children with cerebral palsy (CP) and in typically developing (TD) children. Mean signals (represented by the curves)  $\pm$  1 standard error (represented by shaded areas) during the gait cycle are depicted on the left side for the CP group ( $n = 12$ ) and on the right side for the TD group ( $n = 11$ ). Black solid lines, green dashed lines, and red thick lines indicate the barefoot, NHS, and PHS conditions, respectively. Vertical dotted lines indicate the average toe-off event in the CP and TD groups. NHS, negative-heel shoes; PHS, positive-heel shoes.

CP adapted to NHSs in the same way that TD children did, but achieved flat-foot contact or heel strike, respectively. In addition, in the CP group, wearing PHSs was associated with an additional ankle plantar flexion movement before IC, which contributed to early flat shoe contact during WA. Below, we consider 1) the two typical foot-ankle patterns in the CP and TD groups that were maintained regardless of the footwear condition, 2) the adaptation of foot kinematics to the footwear condition, and 3) the plantar flexors' functional role with regard to the typical foot-ankle pattern seen during WA in children with CP.

*The different foot-ankle patterns in CP and TD groups in a pooled analysis of the footwear conditions.* The TD children displayed a typical gait pattern for their age; the first heel rocker was heel strike with ankle dorsiflexion upon IC and was

followed by a short, decelerated ankle plantar flexion and then by late flat-foot ground contact (Perry and Burnfield 2010). The TA activity during the swing phase comprised two components, which were related to two locomotor subtasks: 1) swinging the limb forward with toe clearance, and 2) placing the foot on the ground (Ting et al. 2015). In the first half of the swing phase, dorsiflexion of the foot was required for clearance at midswing. In the second half of swing phase, TA activity that peaked during WA, maintained ankle dorsiflexion for IC, decelerated ankle plantar flexion during WA, and thus avoided excessively sudden flat-foot ground contact (Agostini et al. 2010; Houx et al. 2014; Perry and Burnfield 2010).

The children with CP displayed on average an equinus at IC, a second rocker immediately after IC, with early flat-foot or flat-



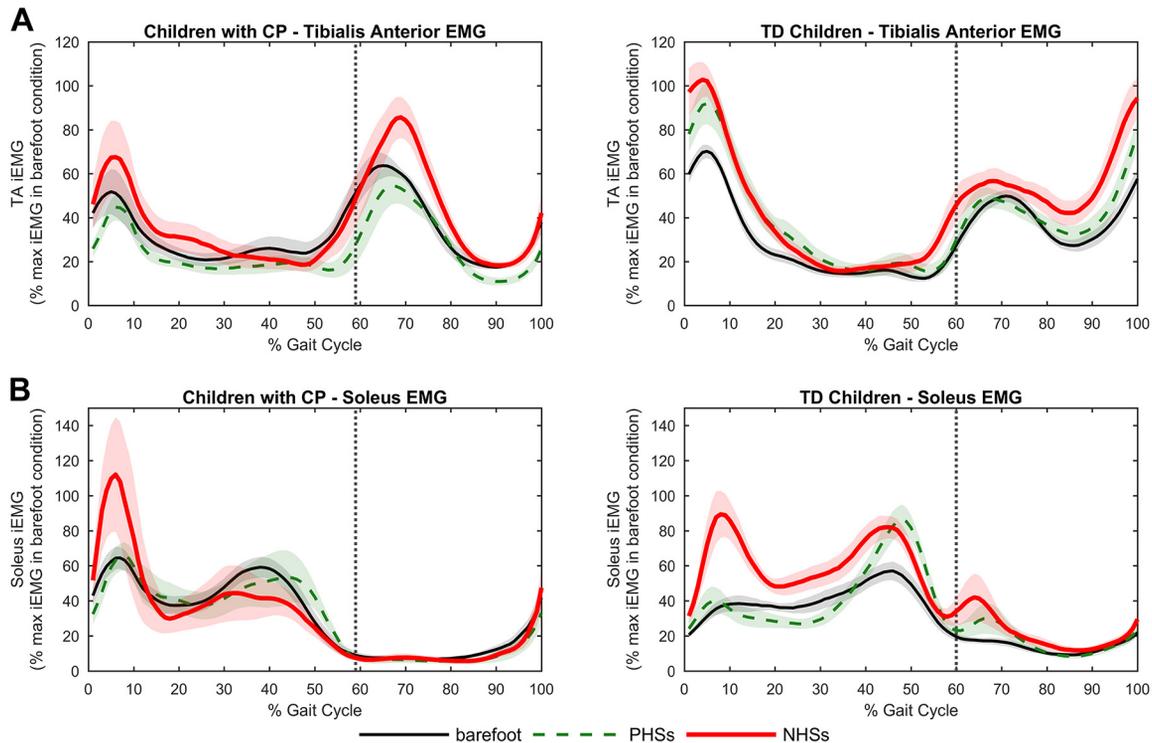


Fig. 4. Integrated EMG (iEMG) profiles for the tibialis anterior (TA) (A) and soleus (B) muscles in children with cerebral palsy (CP) and in typically developing (TD) children. Mean iEMG signals (represented by the curves)  $\pm$  1 standard error (represented by shaded areas) during the gait cycle are depicted on the left side for the CP group ( $n = 7$ ) and on the right side for the TD group ( $n = 11$ ). Black solid lines, green dashed lines, and red thick lines indicate the barefoot, NHS, and PHS conditions, respectively. Vertical dotted lines indicate the average toe-off event in the CP and TD groups. max iEMG, peak of integrated EMG; NHS, negative-heel shoes; PHS, positive-heel shoes.

continuously updated from multisensory feedback and is used to forward commands (such as motor module commands) and thus control body position in space (Sousa et al. 2012; Ting et al. 2015). Toe clearance is achieved by both TD children and children with CP when they are required to adapt their gait under various ecologic conditions, such as stepping over an obstacle (Law and Webb 2005), walking uphill (Hösl et al. 2016), or wearing NHSs (the present study). Gait adaptability during the swing phase probably involves a feedforward mechanism, as suggested by studies in which the removal of a resistance force or a weight previously applied to one leg provoked aftereffects in both TD children and children with CP (Damiano et al. 2017; Tang et al. 2019). The motor adaptability observed during the swing phase might involve somatosensory inputs from the lower limb in general and the dorsum of the foot in particular (Howe et al. 2015). In children with CP, the motor control process probably takes account of common somatosensory impairments of the lower limbs (Wingert et al. 2009; Zarkou et al. 2020) and even poor prediction of the sensory consequences of movement (Nielsen et al. 2020).

It is frequently suggested that equinus upon IC is due to dysfunctional, spastic overactivity of the plantar flexors before foot strike (Graham et al. 2016; Lance 1980; Winters et al. 1987). However, in children with CP, reflex measurements during walking have failed to evidence accentuated sensory inputs to ankle plantar flexors during the swing phase (Willerslev-Olsen et al. 2014). Although the soleus was found to be spastic in all the children with CP studied here, its activity during the first half of the swing phase was at its lowest when the muscle was stretched

during rapid, ample dorsiflexion. Furthermore, soleus activity during the second half of the swing phase increased slightly before IC (with a peak at WA) but was not significantly higher than in the TD group and was not preceded by soleus stretching (ankle plantar flexion) (Fig. 3B). When considering the plantar flexors, one can note that the soleus muscle was more stretched at midswing and upon IC in the NHS condition than in the two other conditions; this was due to higher ankle dorsiflexion. Likewise, the gastrocnemius muscle was more stretched at least upon IC in the NHS condition for the same reason, given that knee flexion upon IC was essentially the same in all three conditions. Thus, in the NHS condition, the plantar flexors did not oppose the higher ankle dorsiflexion observed upon IC. Therefore, our data do not support a causal relationship between any spastic overactivity in the soleus and equinus upon IC.

In a pooled analysis of the two groups, we found that the footwear condition significantly influenced the time point of the first flat-foot contact. Compared with the barefoot condition, this time point was later in the PHS condition and earlier in the NHS condition. However, the modalities of foot-to-ground contact upon IC differed only in the CP group; heel contact was most common in the PHS condition (contributing to later first flat-foot contact), and flat-foot contact was most common in the NHS condition (contributing to early first flat-foot contact). Interestingly, in the PHS condition, the CP group compared with the TD group added a significant ankle plantar flexion movement from midswing to IC (Fig. 2); this was likely to reduce the delay in the first flat-foot contact caused by wearing a PHS.

Overall, the footwear condition significantly altered foot-ankle kinematics in both groups. However, these changes related to the shape of the shoe did not affect the direction and amplitude of the first ankle movement (Fig. 2), which was associated with the ankle's negative power during WA.

*An adaptive, functional role of the plantar flexors early in stance.* During WA, the negative power exerted by the leading limb decelerates the body's center of mass (Donelan et al. 2002; Kuo and Donelan 2010). In typical gait in adults with heel strike, the negative power and work of the leading limb during WA are mostly exerted by the eccentric vasti (up until the first peak in knee flexion), whereas the negative work exerted by the plantar flexors is low and begins after the foot has been placed flat on the ground (Ellis et al. 2014; Neptune et al. 2004; Worthen-Chaudhari et al. 2014). In early childhood, the plantar flexors are frequently active in the terminal swing phase and throughout WA (Chang et al. 2006; Sutherland et al. 1980); this activity is associated with either toe walking in the first weeks or months of walking, or an early second rocker phase later on (Forssberg 1992; Sutherland et al. 1980). Thus, net power absorption exerted by the plantar flexors at the ankle joint is present during WA and significantly more intense in children under the age of 4 than in older children or adults (Cupp et al. 1999; Samson et al. 2011, 2013). This early-onset negative power at the ankle joint exerts a decelerating effect and a stabilizing effect (Samson et al. 2011); it helps to decelerate the body and stabilize forward gait (Kuo and Donelan 2010) while the child is learning to generate and control propulsive forces (Bril et al. 2015). Plantar flexor activity during WA is also notably involved in adaptations to walking down stairs or downhill, during which the leading leg exerts high negative work to decelerate progression of the body (Holm et al. 2010; McFadyen and Winter 1988; Protopapadaki et al. 2007). Whereas early action by plantar flexors during WA has functional benefits in early childhood or under certain ecological conditions, its origin in children with CP is still subject to debate.

Since the plantar flexors' eccentric action during WA helps to decelerate the center of mass in the forward and downward directions and thus to stabilize gait (Kuo and Donelan 2010; Kurz et al. 2010; Neptune et al. 2001), this biomechanical effect may also help to maintain balance in the anteroposterior plane during gait by children with CP, who generally have difficulty balancing during gait (Bruijn et al. 2013; Hsue et al. 2009). In particular, the plantar flexors' action at the beginning of WA (reflected by the abrupt rise in anterior acceleration of the center of pressure in the leading leg in children with CP) might counter the high forward and downward decelerations of the body's center of mass (Hsue et al. 2009). It is noteworthy that the negative ankle power that slows the body lasted for longer and was more than four times more intense in the CP group than in the TD group; in the latter, the negative ankle power merely slows the flattening of the foot.

In the context of stroke (also an upper motor neuron syndrome), IC usually involves forefoot strike or flat-foot strike in equinus. As seen in children with hemiplegic CP, this occurs with an anterior shift of the center of pressure at IC (Nolan et al. 2015), together with plantar flexor activity from IC onward (Clark et al. 2010) and ankle negative power that decelerates ankle dorsiflexion (Farris et al. 2015). Furthermore, this equinus foot-ankle pattern is not due to a primary motor dysfunction because adaptation (marked dorsiflexion) can occur upon IC

when ascending stairs to ensure appropriate foot strike (Novak and Brouwer 2013). In individuals with stroke [who also displayed poor dynamic stability during gait (Devetak et al. 2019)], the early braking action of the triceps surae might be a compensatory mechanism that stabilizes posture and forward progression (Beyaert et al. 2015). This hypothesis might explain (at least in part) the early triceps surae action typically observed under balance-challenging conditions (e.g., walking on a slippery surface) (Cappellini et al. 2010; Fong et al. 2005) or in certain balance-challenging diseases [such as cerebellar ataxia (Martino et al. 2014; Mitoma et al. 2000) and Duchenne muscular dystrophy (Alkan et al. 2017; Gaudreault et al. 2010)]. These observations suggest that the triceps surae action early in WA might be a nonspecific compensatory mechanism that helps to control center of mass acceleration in the sagittal plane under balance-challenging conditions or in a disease setting. Further studies are needed to specify the relationship between early triceps action and body balance characteristics during gait under various (patho)physiological conditions.

In conclusion, children with CP displayed a specific foot-ankle pattern, including early flat-foot or flat-shoe ground contact at WA. This ground contact was 1) favored by low TA activity (rather than high soleus activity) during the second half of the swing phase and 2) associated with the exertion of early negative ankle power by the plantar flexors to decelerate ankle dorsiflexion under all footwear conditions. Children with CP adapted to NHSs in the same way that TD children did, i.e., with greater ankle dorsiflexion at midswing and upon IC, and higher TA activity during the swing phase. In the NHS condition, soleus activity in children with CP was lowest when the muscle was rapidly lengthening during dorsiflexion in the swing phase and was not significantly greater than in TD children—despite evidence of spasticity in the soleus when not walking. These findings suggest that in children with CP walking barefoot, equinus upon IC is not caused by spasticity of the soleus. We suggest that the plantar flexors' early action during WA is allowed by early flattening of the foot, which in turn is allowed by equinus upon IC when walking barefoot or with PHSs. The plantar flexors' early action during WA decelerates the forward progression of the body's center of mass; this may help to compensate for poor trunk control and balance during gait in children with CP (Heyrman et al. 2014; Hsue et al. 2009). Further studies (including therapeutic trials) are needed to test this hypothesis. In particular, treatments that improve trunk and balance control during gait might decrease the need for early plantar flexor activation during WA.

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#### DISCLOSURES

No conflicts of interest, financial or otherwise, are declared by the authors.

## AUTHOR CONTRIBUTIONS

C.B., R.V., and J.Paysant conceived and designed research; C.B. and J. Pierret performed experiments; C.B. and J.Pierret analyzed data; C.B., J.Pierret and S.C. interpreted results of experiments; C.B., J.Pierret and S.C. prepared figures; C.B., J.Pierret and S.C. drafted the manuscript; C.B., J. Pi., R.V., J.P.A. and S.C. edited and revised the manuscript; C.B., J.Pierret, R.V., J.Paysant and S.C. approved the final version of the submitted manuscript.

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