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Gait training reduces ankle joint stiffness and facilitates heel strike in children with Cerebral Palsy

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Abstract.
BACKGROUND: Foot drop and toe walking are frequent concerns in children with cerebral palsy (CP). Increased stiffness of the ankle joint muscles may contribute to these problems.
OBJECTIVE: Does four weeks of daily home based treadmill training with incline reduce ankle joint stiffness and facilitate heel strike in children with CP?
METHODS: Seventeen children with CP (4–14 years) were recruited. Muscle stiffness and gait ability were measured twice before and twice after training with an interval of one month. Passive and reflex-mediated stiffness were measured by a dynamometer which applied stretches below and above reflex threshold. Gait kinematics were recorded by 3-D video-analysis during treadmill walking. Foot pressure was measured by force-sensitive foot soles during treadmill and over-ground walking.
RESULTS: Children with increased passive stiffness showed a significant reduction in stiffness following training ($P = 0.01$). Toe lift in the swing phase ($P = 0.014$) and heel impact ($P = 0.003$) increased significantly following the training during both treadmill and over-ground walking.
CONCLUSIONS: Daily intensive gait training may influence the elastic properties of ankle joint muscles and facilitate toe lift and heel strike in children with CP. Intensive gait training may be beneficial in preventing contractures and maintain gait ability in children with CP.

Keywords: Cerebral palsy, gait training, passive muscle stiffness

1. Introduction

Cerebral Palsy (CP) is the most common cause of childhood disability with an incidence of 2-3 out of 1000 live births (Koman et al., 2004; Ravn et al., 2010). Impaired gait function, which is seen to some extent in all children depending on the severity of the lesion, is one of the main factors for lack of integration with other children, participation in social activities and hence reduced quality of life (Shikako-Thomas et al., 2012). According to recent surveys the number of children with unilateral neurological signs has increased over the last decades (Ravn et al., 2010). Such children are typically ambulatory with a proximal to distal muscle gradient of the severity of muscle weakness so that foot drop and/or toe walking dominate the clinical picture in many cases (Fowler et al., 2010).

Several factors may contribute to these gait problems. In some children with foot drop and toe walking premature activation of plantarflexor muscles prior to and around the time of ground contact is seen and

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this is often assumed to be caused by spasticity (Ackman et al., 2005; Boulay et al., 2012; Koman et al., 2000). However, electrophysiological and biomechanical studies over the past three decades have failed to demonstrate any functional contribution of increased stretch reflex activity to the gait disorder observed in children with CP as well as other patients with spasticity (Dietz & Berger, 1983; Dietz & Sinkjaer, 2007; Sinkjaer et al., 1996; Willerslev-Olsen et al., 2013a). The observation that healthy children also often activate ankle plantarflexors prior to ground contact also challenges the idea that spasticity should be of major importance for the observed gait problems in children with CP (Dietz & Sinkjaer, 2007; Willerslev-Olsen et al., 2013a). There is on the other hand good evidence to suggest that reduced central drive to the ankle dorsiflexors (i.e. paresis and paralysis) plays a major role (Bland et al., 2011; Moreau et al., 2012; Willerslev-Olsen et al., 2013a; Petersen et al., 2013). Pronounced weakness in ankle dorsiflexors is thus commonly seen in children with CP (Bland et al., 2011; Moreau et al., 2012; Petersen et al., 2013) and this has been shown to be related to impaired corticospinal drive to the muscles during gait (Petersen et al., 2013). In addition, increased stiffness of the passive elastic elements in the ankle muscles has been shown to develop at an early age and cause severe functional gait problems in children with CP (Dietz et al., 1981; Willerslev-Olsen et al., 2013a, 2013b). Without treatment these muscular alterations become permanent in the form of contractures which greatly reduces the range of movement in the joint and at some point usually necessitates surgical intervention (Hagglund & Wagner, 2011). This leads us to propose that a therapeutic approach in children with CP should be aimed at strengthening the ability of lifting the toes and reducing the stiffness of the passive elastic elements around the joint in order to facilitate heel strike.

In the present study we focus on intensive home based treadmill training aimed at improving the ability to lift the toes in the end of swing and place the heel appropriately on the ground in early stance. Walking uphill has been shown to require increased activation of the ankle dorsiflexors when compared to level walking (Franz & Kram, 2012; Leroux et al., 1999; Stern & Gottschall, 2012). Furthermore, the inclination of the treadmill will force the child to walk with a greater dorsiflexion throughout stance (Franz & Kram, 2012; Leroux et al., 1999). These two effects were hypothesised to reduce ankle joint stiffness and thereby help the child to achieve a more appropriate heel strike. The children were required in this study to walk with an incline for a period of at least 30 minutes every day for one month on a treadmill with progressive incline. We report that this training effectively increased the ability of the children to lift the toes towards the end of the swing phase and place the heel on the ground. This was accompanied by a decrease in the ankle joint stiffness.

2. Methods

2.1. Participants

Seventeen children with cerebral palsy (CP) (Table 1; age 9.4 years, range 5–14; 12 boys, 5 girls; weight 32.5 ± 9.5 kg; height 138.6 ± 15.8 cm) were recruited through the Danish Cerebral Palsy Organisation (Spastikerforeningen). Thirteen children had hemiplegia (9 children were affected on the right and 4 were affected on the left side) and 4 children had diplegia (Table 1). All children had impaired gait function with some degree of foot drop and/or toe walking. The study was approved by the local ethics committee (H-B-2009-017) and all procedures were conducted within the standards of the Helsinki declaration. Prior to the experiments parents and children received written and verbal information, and consent for participation was obtained.

The severity of the CP of the children participating in the study was classified according to the Gross Motor Classification System (GMFCS (Palisano et al., 1997)). The clinical characteristics of the children are summarized in Table 1.

As a control group 17 age-matched children without CP were recruited for the study (age 8.6 years, range, range 4–15; 11 boys, 6 girls). Control children were only tested once and thus did not participate in the training intervention.

2.2. Experimental set up

Each child was involved in the study for a period of three month. Ankle joint stiffness, gait function and neurological condition were evaluated during four separate test sessions at intervals of approximately one month (Fig. 1). All test sessions took place at the Helene Elsass Center (www.elsasscenter.dk; Charlottenlund, Denmark). The first test was carried out approximately one month before gait training whereas the second test was performed the day before initiating gait training. The last two tests were made on the last day of training and one month after.

Table 1
Average information about all children with CP enrolled in the study

n	Gender (M:F)	Age (y,mo)	Weight (kg)	GMFCS	MAS	DF/PF	ROM	Reflex stiffness rest (%Tmax)	Passive stiffness rest (%Tmax)
17	10:5	9.8 (SD:2.99)	33.99 (SD:9.28)	GMFCS I:7 GMFCS II:6 GMFCS III:4	2 (SD:1.4)	3.8/2.4 (SD:0.7/1.1)	91/132 (SD:8.2/5.3)	25.85 (SD:16.22)	35.01 (SD:21.37)

CP, cerebral palsy; GMFCS, Gross Motor Function Classification System; DF, dorsiflexors; PF, plantar flexors; MAS, Modified Ashworth Scale; NI, not investigated. Muscle power in dorsiflexors and plantar flexors evaluated according to the UK Medical Research Council rating scale: 0, no contraction; 1, trace of contraction; 2, active movement through full range of motion with gravity eliminated; 3, active movement through range of motion against gravity; 4, active movement through range of motion against resistance (but weak); 5, normal. Tmax, maximal torque response measured by a supramaximal stimulation of the *n*. Tibialis at popliteal fossa.

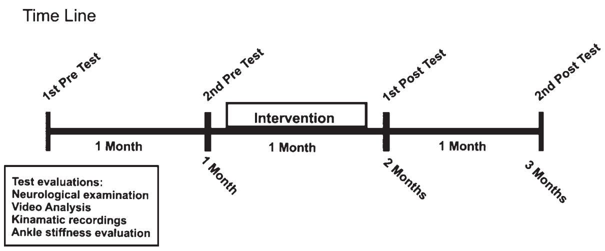

Fig. 1. Time Line. The figure illustrates the time line of the study. Each child was involved in the study for a period of three months. A total of four test sessions (1st Pre Test, 2nd Pre Test, 1st Post, 2nd Post Test) was conducted at intervals of approximately one month. The intervention period of one month was scheduled between the 2nd Pre Test and the 1st Post test.

We decided to use this design in order to use the children as their own control group rather than to have a separate group of children undergoing no intervention. This circumvents the variability in comparing data from different children and it allowed us to determine whether any effect of the intervention could be explained by simple test- retest variability.

The last test session one month after the end of the intervention was included to observe if there was any sustained improvement in gait function and/or increase in daily activity.

All four test sessions included the same measurements in the same order on every occasion and at the same time of the day. All test sessions started by the neurological examination followed by a video recording of the child's gait over ground. This was followed by evaluation of the ankle joint stiffness and finally by kinematic analysis of treadmill (level and with an incline) and over ground walking.

2.3. Neurological examination

All children were examined in the beginning of each test session by a medical doctor with 20 years of experience in neurological examination (one of the authors, JBN). All tests were performed with the children seated comfortably and relaxed on an examination couch with their legs hanging loosely over the side of the couch. Passive range of movement (ROM) in the ankle was evaluated initially by moving the foot as slowly as possible as far possible in a plantar flexion direction and then as far as possible in a dorsi flexion direction. The position of the joint was noted in both end positions. During this examination any resistance during the slow movement was also noted. The joint was then positioned in neutral position and quick stretches were applied in dorsiflexion direction to evaluate the presence of a catch and possibly clonus. Similar stretches were applied with the joint in a more plantar flexed and dorsi flexed position. Based on this evaluation a score on the modified Ashworth scale (MAS) was determined. Subsequently, the presence and possible exaggeration of Achilles tendon and patella reflexes were examined using a reflex hammer while gently supporting the foot or limb to enable relaxation. Finally, voluntary muscle force was examined for plantar flexion, dorsi flexion, knee extension, knee flexion and hip flexion. In all measurements the examiner resisted the movement with his hand and evaluated the force of contraction on the MRC scale from 1–5 (UK Medical Research Council: Table 1).

2.4. Pedometer recordings

In order to obtain an evaluation of the daily activity level of the children they were asked to wear a pedometer (Omron Walk style II; Omron Healthcare Europe, The Netherlands) as often as possible throughout the project period. The use of pedometers in the evaluation of daily activity level in children with CP has been shown to have high reliability and validity (Maher et al., 2013; Morris et al., 2013). The pedometer was placed in a pocket, in a belt around the waist or a string around the neck. All data were transferred from the pedometer every month to a PC through a USB connection. Average number of daily steps was calculated for each month before, during and after the training period. Only days in which the child had been wearing the pedometer for the whole day were included in the calculation.

2.5. Gait analysis during treadmill and over ground walking

The gait of the children was analysed by recording the leg movements using a 3-D motion capture system, by recording the pressure under the foot using pressure sensitive foot soles and by recording EMG activity from the anterior tibial muscles. The children were asked to walk on a treadmill with and without incline as well as over ground. Following 5 minutes of familiarisation in the beginning of the first test session, the children selected their own comfortable walking speed when walking without incline on the treadmill. It varied between 2 km/h and 4 km/h with a mean of 2.8 km/h ± 0.6 (Table 2). The self-chosen walking speed was used for measurements in all subsequent test sessions. All children were asked to hold on to the bars of the treadmill even though several of them were able to walk without support. The incline of the treadmill was chosen by the child by slowly increasing the incline while the child was walking. An incline at which the child felt challenged, but still able to walk without major fatigue was chosen. The only requirement was that all children should walk with an incline of at least 5%. The incline in the different children varied from 5% to 12% with a mean of 7.5% ± 2.4 (Table 2). The maximum possible incline on the treadmill used for measurements was 12%.

The children walked in their own sport shoes without shoe inserts or braces. The same shoes were used in every session. Two minute recordings were obtained during both level and with an incline walking for motion and plantar pressure analysis. For over ground walking children were asked to walk forth and back in a corridor where a five meter lane had been marked. Two minute recordings were obtained for plantar pressure analysis.

2.5.1. 3-D motion analysis

3-D kinematic data were captured using a Qualysis motion capture system (Qualisys, Gothenburg, Sweden) with six, synchronous Oqus 1 cameras operating at a sampling frequency of 200 Hz. Reflexive markers (Size 12 mm) were placed bilaterally at 1) the lateral articular line of the knees, 2) the lateral malleolus and the lateral side of the 5th metatarsal.

2.5.2. Plantar pressure measurement

Pressure sensitive soles were placed in the shoes of the child in order to record the plantar pressure from each foot during walking (F-scan wireless, Tekscan, CA Mätsystem, Sweden). The pressure sensitive soles were fitted individually for the shoes of each child and reused by that child for each of the four test sessions. Prior to each recording the system was calibrated by having the child standing relaxed while holding on to the rails of the treadmill with equal weight on the two legs.

2.5.3. Electromyographic (EMG) recording

EMG activity was recorded by two sets of bipolar electrodes (Ambu Blue sensor N,N-10-A/25. Ambu A/S Ballerup, Recording area 0.5 cm^2 inter-electrode distance, 2 cm) placed at the proximal end of the Tibialis anterior (TA) muscle on both legs. The skin was brushed softly with sandpaper (3M red dot; 3M, Glostrup, Denmark). EMG signals were sampled by a wireless EMG recording system (BTSFreeEMG; BTS Bioengineering Corp.Brooklyn, NY, USA). Data was filtered (band-pass, 10 Hz–400 Hz), sampled at 1000 Hz, and stored on a PC for off-line analysis.

Table 2a
Average information about gait ability (1st test) for the children with CP

n	Gait Speed (Average)	Gait Uphill (Average Incline)	Mean amplitude of second peak of toe lift (Average mm)	Angle (Average deg.)	Dynamic index (Heel impulse % of total)
17	2.8 km/h (SD:0.6)	7.5% (SD:2.4)	33.83 (SD:15.27)	5.47 (SD:3.27)	31.9 (SD:22.45)

Table 2b
Average information about gait ability for control children

n	Gait Speed (Average)	Gait Uphill (Average Incline)	Mean amplitude of second peak of toe lift (Average mm)	Angle (Average deg.)	Dynamic index (Heel impulse % of total)
17	3.2 km/h (SD:0.46)	8.9% (SD:3.66)	65.4 mm (SD:15.04)	8.8deg. (SD:2.2)	46.16 (SD:15.03)

2.6. Evaluation of passive and reflex stiffness

Passive and reflex mediated stiffness components of the ankle plantarflexors were objectively assessed with the use of a biomechanical and electrophysiological evaluation performed according to Lorentzen (Lorentzen et al., 2010) and Willerslev-Olsen (Willerslev-Olsen et al., 2013b). The child was seated in a comfortable chair with the most affected foot attached to a footplate. A 100° flexion in the hip, 130° flexion in the knee and 100° flexion in the ankle joint was retained during all four evaluations. The footplate could be rotated by a motor (CEM model 26) which was driven by a DC power amplifier (Brüel & Kjaer; model 2708) and could deliver maintained torques up to 80 Nm and peak torques up to 120 Nm. An electrogoniometer, connected to the foot plate, measured the angle of the ankle joint and a torque meter measured the torque exerted on the foot plate prior to and during the stretch perturbations.

Bipolar EMG recordings were obtained from two sets of electrodes (Ambu Blue sensor N,N-10-A/25. Ambu A/S Ballerup, Recording area 0.5 cm^2 inter-electrode distance, 2 cm). One pair of electrodes was placed over the soleus muscle below the gastrocnemius muscle and one pair on the belly of the TA muscle. A ground electrode was placed at the knee.

The perturbations consisted of ramp and hold dorsiflexion with amplitude of 6° at 17 different velocities between 5 and 220°/s; hold time 460 ms. Perturbations were delivered every second in a random order until ten trials per velocity were collected. Passive stiffness was calculated from the torque response at low velocities without stretch responses, whereas reflex stiffness was calculated from the torque response to the fastest perturbation (see Willerslev-Olsen et al., 2013b).

2.7. Training protocol

Only children with CP were trained. Training sessions were scheduled for 30 minutes every day for four consecutive weeks at home. All participants had a treadmill (ECO ll G6432N) delivered to their home a several days prior to initiating the training. The children were allowed to split up the 30 minutes of walking in multiple periods of shorter duration as long as 30 minutes of total walking time was achieved at the end of the day.

Speed and incline settings of the treadmill for the initial training was based on the settings for the first test session. As the child improved and became more comfortable with incline and speed, settings should be increased. Parents and children were instructed to increase primarily the incline. The children were also instructed to make an effort in placing the heel on the treadmill in early stance. The parents were instructed to encourage the child as much as possible both with respect to maintenance and progression of the training and the placement of the heel in early stance. Attention was thus focussed on walking with an incline and on facilitating activity of the dorsiflexors in a fast but controlled pace without running.

All children and their parents were handed a diary in order to record how long time the child had been training each day, the speed of the treadmill, the incline of the treadmill and what other activities they had been participating in during the day. They were asked to report in the diary if illness or if other events prevented the child from training. Moreover, they were asked to write down how they felt during the training (i.e. hard work, tired, happy or sweaty). Data summarizing the activity of the children during the training are given in Table 3.

2.7.1. Off-line data analysis

Signal processing and analysis was carried out off line. All 3-D motion and plantar pressure data were imported into Matlab (Mathworks, Massachusetts, USA) for further analysis. Two measures of foot drop were obtained from the 3-D motion analysis: 1). The amplitude of the 2nd toe lift late in the swing phase just prior to heel strike. This has been shown to provide a functionally relevant measure of foot drop in patients with spinal cord injury (Barthelemy et al., 2010). This

Table 3
Average information about daily activity and gait training

n	Step counter (Average) $n = 12$	Gait training (minutes in total)	Gait training (Average Incline Increase)	Gait Training (Velocity km/h)
17	Pre:4329 Training:6604 Post:4989	719.47 min (SD:134.57)	2.67% (SD:2.3)	3.3 km/h (SD:1.2)

measure was obtained by detection of a peak in the Z position of the toe marker larger than 4 mm within 400 ms prior to heel strike. Selected peaks were subsequently verified visually and 2) the position of the ankle joint at ground contact with respect to when the foot was flat on the ground i.e. in early stance within 150 ms after ground contact. The difference between these two angles was taken as an indication of the extent to which the child actively dorsiflexed the ankle prior to ground contact and subsequently lowered the foot.

The plantar pressure data were divided into three recording areas: Heel, midfoot and forefoot of equal size. The peak pressure in each of these areas was then averaged over all steps for the 2 minute data sets. A dynamic index was calculated as: 100*(peak pressure in heel area/peak pressure midfoot+peak pressure forefoot). This index has been shown to provide a valid measure of the extent of heel strike in children with CP (Bennett et al., 2007).

2.7.2. Statistics

Assumptions of normality of the distribution for all variables were explored through histograms and normality plots and confirmed with the Shapiro-Wilk's normality test. Paired Student T tests were used to determine significant differences in gait parameters and TA EMG activity when children walked straight and with an incline on the treadmill. One way repeated measures ANOVA was used to determine differences in measurements at the four test sessions of the following: 1) ROM, 2) MAS, 3) the amplitude of second peak of toe lift, 4) the angle of the ankle joint at heel strike with respect to the ground and 5) the dynamic index of foot pressure. Holm-Sidak postHoc test was used to determine significant differences between individual test sessions. Pearson's bivariate correlations were used to explore associations between clinical measures and gait parameters. All analyses were performed with Sigmaplot 12.5 (SYSTAT Software, San Jose, CA, USA) for Windows using two-tailed probability tests.

3. Results

Table 1 summarises the clinical characteristics of the 17 children included in the study. All children but four were able to walk without support. At the first test the children with CP chose to walk at an average speed of 2.42 km/h (Range: 1.9–3.1 km/h SD: 0.51). The 17 control children in comparison walked at a chosen speed of 3.2 km/h (range SD). The children with CP on average lifted the toes on the most affected side significantly less than the control children (Mean amplitude: 33.8 mm, SD: 15.27 compared to 65.4 mm SD: 15.04 $P = <0.001$). The angle at heel contact with respect to ground was also significantly smaller in the children with CP than in control children (5.5, SD:3.1 compared to 8.7, SD:2.2, $P = 0.001$). The children with CP also had a significantly lower dynamic index than the control children (31.9, SD: 22.45 compared to 46.16 SD: 15.03, $P = 0.037$).

The children with CP on average chose an inclination of 7.5% (Range: 5–12% SD: 2.3) when walking with an incline on the treadmill (Table 2a). Compared to level walking significantly larger EMG activity was measured from the TA muscle during incline walking (56.9 uV compared to 49.3uV). Compared to level walking when walking up an inclince the toes were lifted higher (Mean amplitude: 77.1 mm, SD: 31.41 compared to 33.8 mm SD: 15.3 $P = <0.001$) and a larger dynamic index (37.5, SD: 23.9 compared to 31.5 SD: 22.45, $P = 0.027$) was observed. Walking up an incline thus facilitated activation of the dorsiflexors in the swing phase and heel strike in early stance.

Prior to training the average number of daily steps was 4329 (Range: 574–11436, SD: 3085). This increased to 6605 (Range: 1273–13775, SD: 4105) during the training period and decreased again to 4989 (Range: 255–12577, SD: 4042) in the period after training (Table 3). There was a statistical significant difference in the number of steps during the training period as compared to the other two periods ($P = 0.002$). The children on average walked 25.7 minutes (Range: 14.4–30, SD: 4.8) daily on the treadmill. They progressively increased their walking speed from 2.4 km/h to 3.3 km/h (Range: 2–5.9 km/h, SD: 0.55, SD: 1.16), corresponding to an increase of 19% and the incline on the treadmill from 7.5% to 10% (Range: 5–12% SD: 2.3, SD: 2.76), corresponding to an increase of 25%.

All children found the training strenuous in the beginning, but all of them, nonetheless completed the full training period and reported that the training was less hard towards the end of the training period. It was generally a challenge for the families to fit the training into busy everyday life, but all found a way of integrating the training with watching television or similar. Some children even trained more than what was scheduled and several children decided to continue the training after the end of the study period. No injuries or muscular, joint or tendon complaints were observed in the study period. All families reported that the trained child showed a general increase in energy and abilities evident during their activities of daily living.

3.1. Neurological examination

The neurological characteristics at the first examination of all the children with CP included in the study are given in Table 1. All children except one scored at least 1 on the MAS scale indicating increased muscle tone. Only five of these showed exaggerated Achilles tendon or patellar reflexes. All except four showed reduced ROM in the ankle joint with a reduction of dorsi flexion ranging from 5 degrees to 30 degrees. All children showed reduced voluntary dorsi flexion force (Range: 0–4) and the majority (88.2%) showed reduced plantar flexion force (Range: 2–5).

There was a significant interaction of MAS with the different test sessions (ANOVA 1RM; $F = 3.41$, $P = 0.025$). *Post hoc* testing showed a significant reduction in MAS following training as compared to the first test session ($P = 0.035$). No other significant differences in MAS between the sessions were seen. A small but non-significant reduction of the range of passive dorsiflexion (from 91.4deg. to 88.9deg.) was also observed (ANOVA 1RM; $F = 1.4$, $P = 0.25$).

3.2. Kinematic analysis of gait on treadmill

Figure 2A-B shows kinematics analysis of one of the children when walking on the treadmill without incline during the first test prior to training (Fig. 2A.) and during the first test after training (Fig. 2B). Prior to training the child lifted the toes 23.1 mm on average towards the end of the swing phase. This increased to 36.9 mm following training. The difference in angle at heel contact with respect to the ground was 6.1deg before training and 8.5deg after training.

Kinematic data from all children are shown in Fig. 2C–F. One way RM ANOVA showed a significant interaction between tests for the amplitude of the second peak of toe lift ($F = 4.01$, $P = 0.014$). *Post hoc* analysis showed a significant difference between the first test following training when compared to the first test prior to training (Fig. 2C, D). No other combinations were significant. The significant difference in mean amplitude of second peak of toe lift between children with CP and control children which was observed prior to training, was still present following the training (*t*-test: $P = 0.002$).

A statistically significant increase was also observed in the angle at heel contact with respect to the ground after training when compared to the first test ($F = 7.97$, $P < 0.001$) (Fig. 2E, F). A significant increase was found already in the second test session prior to training ($P = 0.005$) and there was no significant difference between the measures after training and the second test prior to training. Contrary to what was observed for the first test prior to training, there was no significant difference in the angle at heel contact with respect to the ground between children with CP measured after training and the control children (*t*-test: $P = 0.5$). The increase of the angle after training was maintained also at the second test one month after training (no significant difference between first and second post training session; $P = 0.8$).

3.3. Foot pressure measurements during gait

Figure 3A-B show measurements of foot pressure during treadmill gait in one of the children before (Fig. 3A) and after (Fig. 3B) training. Before the training intervention the child placed the mid and fore foot on the ground prior to the heel, whereas this was reversed after training with the heel being placed first. The majority of pressure was measured corresponding to the mid and fore foot before training but corresponding to the heel after training. The relation between pressure on the heel and mid foot/fore foot (dynamic index) thus changed from 9.47 before training to 41.6 following the training, reflecting the larger heel impact following the training.

Similar findings were made in the majority of the other children (Fig. 3C-D). One way RM ANOVA showed a significant interaction between tests for the dynamic index ($F = 5.2$; $P = 0.003$) with significant difference between the first test after training and the first test prior to training ($P = 0.004$). Dynamic index did not differ between control children and children with CP following the training (*t*-test: $P = 0.88$), which was the case before training.

These improvements in foot clearance were transferred to over ground walking (Fig. 3E, F). Significant interaction was thus observed between tests of the dynamic index (One way RM Anova) ($F = 5.7$; $P = 0{,}002$) with a significant *post hoc* difference between the first test following training and the two tests prior to training ($P = 0{,}007$¸ $P = 0{,}003$ respectively). There was no significant difference between the two measurements of dynamic index prior to training, but a significant decline in dynamic index was found one month after training with respect to the measurement immediately after training ($P = 0.009$). Similar to what was observed for treadmill walking, no significant difference was found between dynamic index in the CP children after training and the control children (*t*-test: $P = 0.48$).

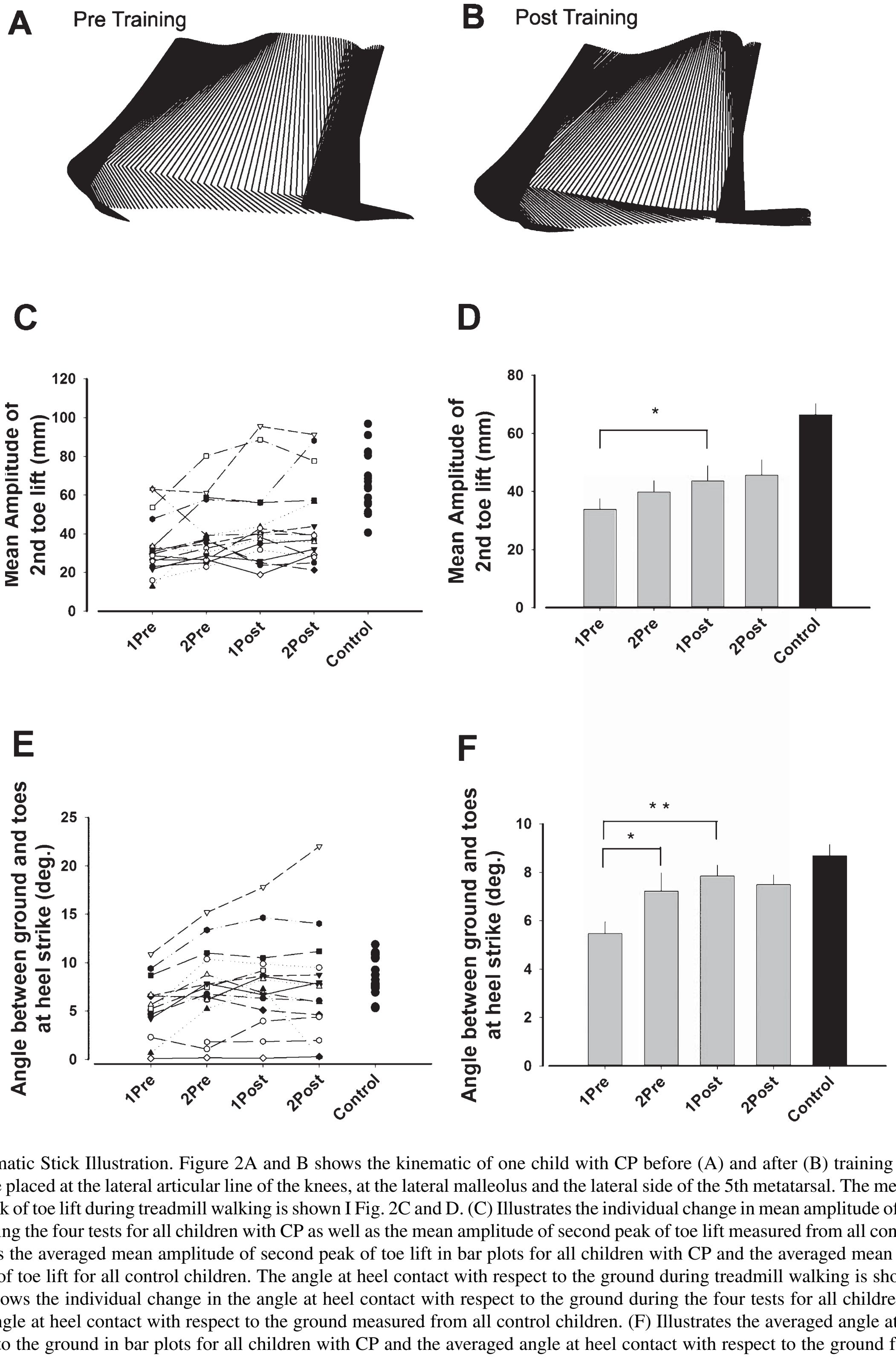

Fig. 2. Kinematic Stick Illustration. Figure 2A and B shows the kinematic of one child with CP before (A) and after (B) training intervention. Markers were placed at the lateral articular line of the knees, at the lateral malleolus and the lateral side of the 5th metatarsal. The mean amplitude of second peak of toe lift during treadmill walking is shown I Fig. 2C and D. (C) Illustrates the individual change in mean amplitude of second peak of toe lift during the four tests for all children with CP as well as the mean amplitude of second peak of toe lift measured from all control children. (D) Illustrates the averaged mean amplitude of second peak of toe lift in bar plots for all children with CP and the averaged mean amplitude of second peak of toe lift for all control children. The angle at heel contact with respect to the ground during treadmill walking is shown I Fig. 2E and F. (E) Shows the individual change in the angle at heel contact with respect to the ground during the four tests for all children with CP as well as the angle at heel contact with respect to the ground measured from all control children. (F) Illustrates the averaged angle at heel contact with respect to the ground in bar plots for all children with CP and the averaged angle at heel contact with respect to the ground for all control children. *Indicate significant difference ($P < 0.05$) **Indicate significant difference ($P < 0.01$).

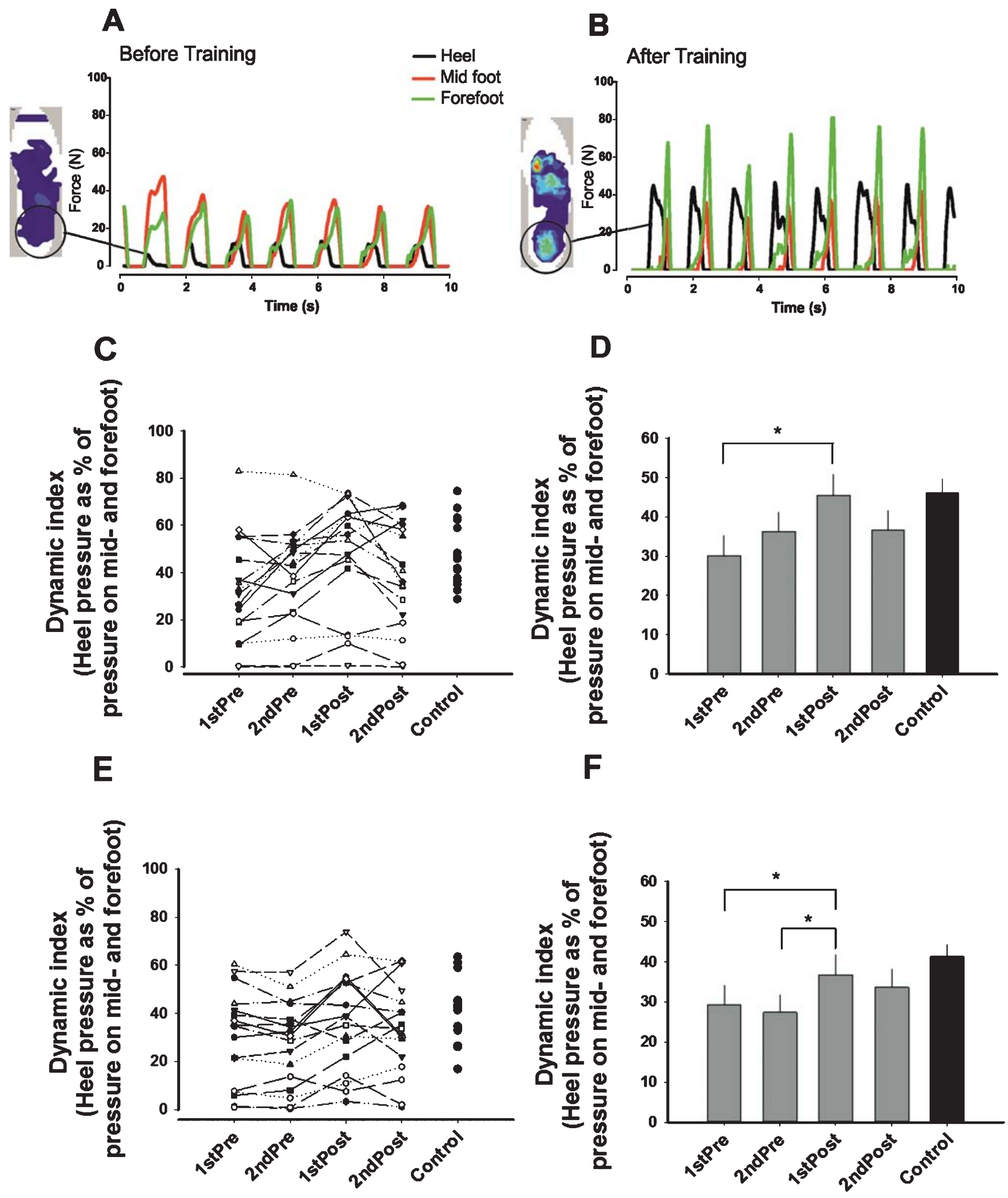

Fig. 3. Foot Pressure. The distribution of foot pressure from one child with CP before training intervention (A) and after (B) training intervention. Seven steps from the most affected leg are illustrated in the graphs. The blue coloured sole to the left of the graphs illustrate an example of the measured foot pressure in the two situations. The graphs show the distribution of the foot pressure: The black line indicates the pressure from the heel, the red line illustrate the pressure of the mid foot whereas the green line is the pressure of the forefoot. The x-axis designates the time in seconds and the y-axis the measured force in Newton. Dynamic index from all children with CP and control children are shown in Fig. 3C–F. (C) and (D) shows the dynamic index measured from level walking at the treadmill. (C) Illustrates the individual change in dynamic index during the four tests for all children with CP as well as the dynamic index measured from the control children. (D) Illustrates the averaged dynamic index in bar plots for all children with CP and the averaged dynamic index for all control children from level walking at the treadmill. (E) and (F) shows the dynamic index measured from over ground walking. (E) Illustrates the individual change in dynamic index during the four tests for all children with CP as well as the dynamic index measured from the control children. (F) Illustrates the averaged dynamic index in bar plots for all children with CP and the averaged dynamic index for all control children from over ground walking. The x-axis designate the four different test sessions as well as the recordings from the control children. The y-axis designate the heel pressure as a percentiles of the pressure on mid- and forefoot. *Indicates significant difference ($P < 0.05$).

3.4. Passive and reflex stiffness

For the children with CP passive stiffness was larger compared to control children in seven cases on the first and second test prior to training, whereas this was the case for reflex stiffness in four children. Following training passive stiffness was reduced in all 7 children (Fig. 4A, C). Despite the low number of subjects a statistically significant interaction with training was observed (1 WAY RM ANOVA; $F = 5.03$, $P = 0.01$). Statistically significant reductions were observed in the first and second test following training as compared to the first test prior to training ($P = 0.014$, $P = 0.03$). There was no statistical significance difference after training as compared to the second test prior to training. No statistical significant difference was found between the first and the second test session prior to training or between the two tests after training.

There was no statistically significant change in passive stiffness with training in children in whom passive stiffness was in the normal range prior to training. There was also no statistically significant change in reflex stiffness (1 WAY RM ANOVA; $F = 0.17$, $P = 0.92$).

3.5. Children who increase toe lift also improve heel strike following training

Children with a large mean amplitude of second peak of toe lift were found to have a large angle at heel contact with respect to the ground (Correlation coefficient = 0.6, $P = 0.02$) and a larger dynamic index (Correlation coefficient = 0.5, $P = 0.05$). Measures of toe lift and heel strike were thus well correlated in the children.

Significant correlations were also found between the increased amplitude of second peak of toe lift and the increased angle at heel contact with respect

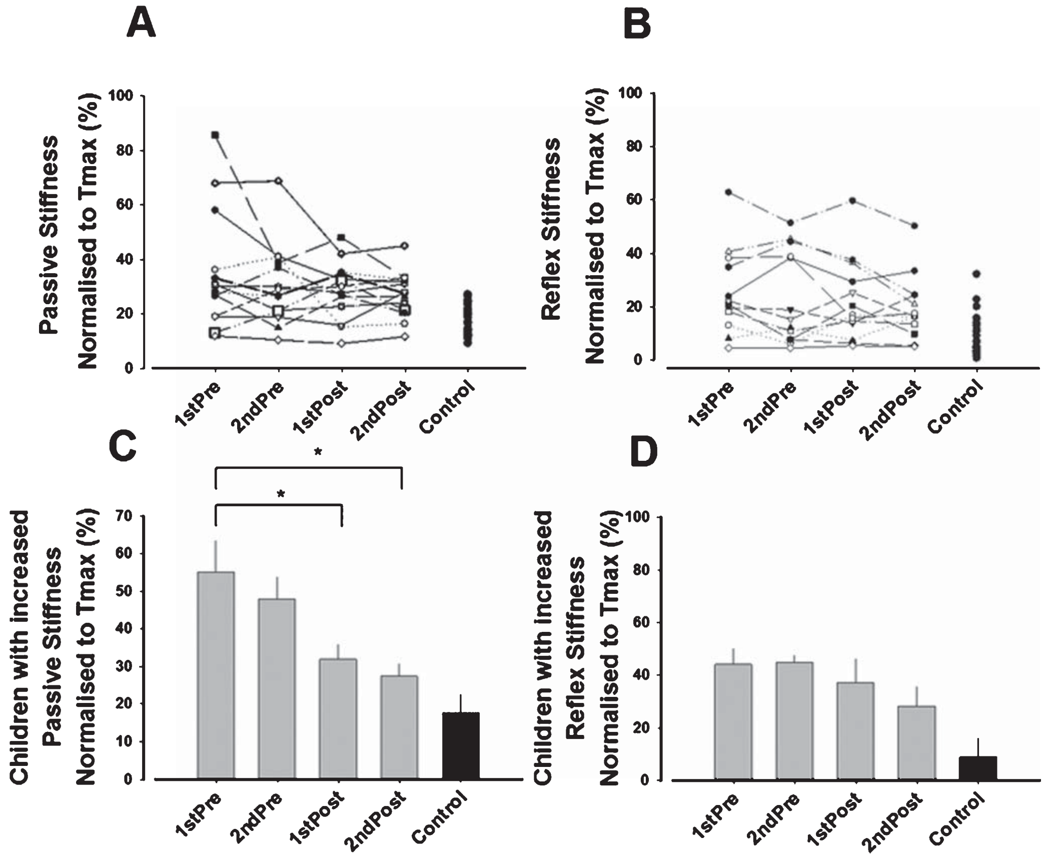

Fig. 4. Passive and Reflex stiffness. The four graphs show the passive and reflex stiffness measured from the children with CP and the control children. (A) and (B) shows the individual change in passive stiffness and reflex stiffness respectively measured in all children with CP during the four tests. All control children are illustrated in the two graphs to the far right. (C) Designate the averaged passive stiffness for the seven children with increased passive stiffness before training intervention. (D) Designate the averaged reflex stiffness for the four children with increased reflex stiffness prior to training intervention. In both (C) and (D) stiffness measured from control children is illustrated. The x-axis designate the four different test sessions as well as the recordings from the control children. The y-axis designate stiffness normalised to the maximal torque elicited by supramaximal stimulation of the tibial nerve at popliteal fossa was recorded. *Indicate significant difference ($P < 0.05$).

to the ground following training (Correlation coefficient = 0.83, $P < 0.0001$), indicating that children who showed improved toe lift also showed improved heel strike. Improved dynamic index (Correlation coefficient = 0.52, $P = 0.03$) and increased mean amplitude of second peak of toe lift (Correlation coefficient = 0.71, $P = 0.002$) following training were correlated to ankle dorsiflexion force measured during the neurological examination. This suggests that children with the least voluntary dorsiflexion force improved the least following the training.

There were no other significant correlations between any of the training induced changes. There was also no correlation between the improvements in gait and the age of the children.

4. Discussion

This study has demonstrated that intensive daily home based gait training on a treadmill with incline may effectively reduce passive muscle stiffness, increase toe lift in the end of swing phase and facilitate heel strike in children with CP. At the end of the training period children with CP reduced their foot drop and increased heel strike to such an extent that there was no significant difference between them and control children with respect to two of the kinematic measures (the angle at ground contact with respect to the ground and the dynamic index of the foot pressure distribution). Although following training the mean amplitude of the second peak of toe lift remained significantly lower in the children with CP compared to control children we did measure a very substantial and highly significant increase in this parameter. Except for the angle at ground contact with respect to the ground, no significant differences were found between the first and the second test prior to training. This indicates that the significant effects observed following the training cannot simply be explained by variability and test re-test effects, but are more likely due to the intervention. Although the increase in the angle at ground contact with respect to ground in the second test prior to training may be explained by variability, we also find it likely that it may be associated to increased daily gait activity in the children during the first month of the study. Participation in a scientific study such as this will inevitably put more focus and attention on their gait and may cause some of them to change their activity pattern. With the design of the study we cannot control for such an effect.

4.1. Passive muscle stiffness

Alterations in the passive elastic properties of muscle tendons and joints following central motor lesions were first discovered 30 years ago by Dietz (Dietz et al., 1981) and have been shown to be of major importance for gait disabilities in children with CP (Dietz & Berger 1983; Marsden et al., 2012; Willerslev-Olsen et al., 2013a, 2013b). More recent studies have shown that increased passive stiffness of the muscles around the ankle joint is difficult to distinguish from reflex mediated stiffness (spasticity) and that increased passive stiffness probably plays a more important clinical role in children with CP than what has been realised previously (Willerslev-Olsen et al., 2013b). Moreover it has been demonstrated that increased stretch reflex activity and premature activation of ankle plantar flexors prior to ground contact plays no or only a minor role for foot drop and toe walking in children with CP (Willerslev-Olsen et al., 2013a). Increased passive stiffness of the plantar flexors in contrast contributes importantly to foot drop and toe walking (Dietz & Berger, 1983; Marsden et al., 2012; Willerslev-Olsen et al., 2013a). The reduction in passive stiffness that we have observed following training may thus have contributed to the improved ability to lift the toes and place the heel on the ground in early stance. The pathological changes in muscles, connective tissue, tendons and joints responsible for increased passive stiffness have not been fully clarified, but recent studies indicate that alterations in the extracellular matrix of the muscles may be involved (Smith et al., 2011, 2009). We speculate that the present observations indicate that active contraction of the muscle in load-bearing conditions may be an efficient way of normalising this tissue. It is unknown whether passive manipulation such as stretching would have a similar effect, but the latest reviews showing no significant effects on contractures may suggest that the active contraction of the muscle may play a crucial role(Katalinic et al., 2011). Albeit, increased passive stiffness may be seen as a precursor or indicator of contractures it is important to note that the measures that we have made here of passive stiffness to some extent are probably related to contractures, but are not necessarily identical to contractures. The fact that we observed an increased ROM following training, although this was not a significant effect, may, however, indicate an effect of the training on contractures also.

It may seem surprising that we observed a significant reduction in MAS without any chances in reflex stiffness following the training. However, several studies have shown that MAS is a poor indicator of reflex mediated stiffness (spasticity), since it does not adequately distinguish passive and active components of hypertonia (Biering-Sorensen et al., 2006; Lorentzen et al., 2010). It therefore seems likely that the reduced MAS is related to the reduction in passive stiffness.

4.2. Daily activity in children with CP

Several studies have shown that children with CP are less physically active and walk significantly fewer steps per day than their healthy peers (Morris et al., 2013). This was confirmed in the present study by the observation that children walked only 4329 steps daily in the month prior to training which is much less than what is usually recommended for this age group (10,000–15,000 steps/day). The psychologist Anders Ericsson has developed the theory that around 10,000 hours of deliberate practice is required for optimal performance of many motor tasks (Ericsson et al., 1993). This corresponds to approximately 4-5 hours of practice every day in 4-5 years. If this also applies for gait during childhood it may be argued that many children with CP do not achieve a sufficient amount of gait experience until well into adulthood. Possibly part of their reduced gait ability when compared to healthy peers may therefore be explained simply by less gait experience. During the training period the children walked on average 6,600 steps daily which is still much less than recommended, but an increase of almost 50% compared to their gait activity prior to training. We find it noteworthy that this relatively short and moderate training intervention can produce changes as large as what we have observed here. There may be a great potential in activating the children even more. The lack of significant effect at follow-up test one month after training as compared to the tests before training also illustrates the importance of maintaining a relatively high level of daily activity. On average the children had a daily activity level similar to before the training during this period and it is therefore not surprising that the positive effects of the training were not maintained.

5. Conclusion

These data show that daily intensive gait training may influence the elastic properties of ankle joint muscles and facilitate toe lift and heel strike in children with CP. We propose that intensive gait training may be beneficial in reducing passive muscle stiffness, increase the daily activity level of children with CP and counteract the development of contractures. Home based treadmill training is an easy, safe and efficient way of strengthening gait ability of children with CP.

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Conflict of interest

There is no conflict of interest.

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