European Journal of Applied Physiology (2019) 119:1127–1136
https://doi.org/10.1007/s00421-019-04102-z

ORIGINAL ARTICLE

Characterization of torque generating properties of ankle plantar flexor muscles in ambulant adults with cerebral palsy

Rasmus Feld Frisk[1,2,3] · **Jakob Lorentzen**[1,3] · **Lee Barber**[4,5] · **Jens Bo Nielsen**[1,3]

Received: 27 August 2018 / Accepted: 12 February 2019 / Published online: 18 February 2019

Abstract

Purpose Weakness of plantar flexor muscles is related to reduced push-off and forward propulsion during gait in persons with cerebral palsy (CP). It has not been clarified to what an extent altered muscle contractile properties contribute to this muscle weakness. Here, we investigated the torque generating capacity and muscle fascicle length in the triceps surae muscle throughout ankle range of motion (ROM) in adults with CP using maximal single muscle twitches elicited by electrical nerve stimulation and ultrasonography.

Methods Fourteen adults with CP (age 36, SD 10.6, GMFCS I–III) and 17 neurological intact (NI) adults (age 36, SD 4.5) participated. Plantar flexor torque during supramaximal stimulation of the tibial nerve was recorded in a dynamometer at 8 ankle angles throughout ROM. Medial gastrocnemius (MG) fascicle length was tracked using ultrasonography.

Results Adults with CP showed reduced plantar flexor torque and fascicle shortening during supramaximal stimulation throughout ROM. The largest torque generation was observed at the ankle joint position where the largest shortening of MG fascicles was observed in both groups. This was at a more plantarflexed position in the CP group.

Conclusion Reduced torque and fascicle shortening during supramaximal stimulation of the tibial nerve indicate impaired contractile properties of plantar flexor muscles in adults with CP. Maximal torque was observed at a more plantarflexed position in adults with CP indicating an altered torque-fascicle length/ankle angle relation. The findings suggest that gait rehabilitation in adults with CP may require special focus on improvement of muscle contractility.

Keywords Cerebral palsy · Torque generation · Plantar flexors · Contractile properties

Abbreviations

BM	Body mass
CI	Confidence interval
CP	Cerebral palsy
EMG	Electromyography
GMFCS	Gross motor function classification scale
MG	Medial gastrocnemius muscle
MIXED	Mixed model regression analysis
MVC	Maximal voluntary (isometric) contraction
NI	Neurological intact
ROM	Range of motion
SCI	Spinal cord injury
SE	Standard error
SD	Standard deviation
TD	Typical developed

Communicated by Nicolas Place.

Electronic supplementary material The online version of this article (https://doi.org/10.1007/s00421-019-04102-z) contains supplementary material, which is available to authorized users.

✉ Rasmus Feld Frisk
frisk@sund.ku.dk

1 Department of Neuroscience, University of Copenhagen, Blegdamsvej 3, 2200 Copenhagen N, Denmark

2 Professionshøjskolen Absalon, Roskilde, Denmark

3 Elsass Institute, Charlottenlund, Denmark

4 School of Health, Medical and Allied Sciences, Central Queensland University, Bundaberg, Australia

5 Child Health Research Centre, Faculty of Medicine, The University of Queensland, Brisbane, Australia

Introduction

Impaired gait function in individuals with cerebral palsy (CP) limits daily life activities and has been shown to reduce quality of life (Jahnsen et al. 2004b; Parkes et al. 2010; Shikako-Thomas et al. 2012; Morgan et al. 2016). One of the main determinants of human gait is the ability to produce an efficient contraction of the ankle plantar flexors at the appropriate time in late stance during the push-off phase to propel the body into propulsion (Winter 1983; Meinders et al. 1998; Neptune et al. 2001). Several studies have indicated that impaired muscle activation is a major cause of reduced forward propulsion and gait speed in adults with CP (Olney et al. 1990; Riad et al. 2012; Roche et al. 2014; Barber et al. 2017), which may contribute to increased fatigue during walking and more frequent falls (Jahnsen et al. 2004a; Morgan et al. 2016).

Altered muscle properties, including muscle atrophy, altered muscle architecture and reduced extensibility of the musculotendinous unit have also been suggested to contribute to impaired muscle function and impaired gait ability in adults with CP (Lieber and Friden 2002; Friden and Lieber 2003; Barrett and Lichtwark 2010; Riad et al. 2012; Geertsen et al. 2015; Hösl et al. 2016; Barber et al. 2017). However, there is little evidence showing a direct relation between alterations in the contractile properties of the muscle and their force generating capacity in individuals with CP. A better understanding of this may help to identify new ways to maintain gait and functional independence in this group.

Some studies have measured the ability to generate a maximal voluntary contraction (MVC) as a functional estimate of motor impairment in children (Elder et al. 2003; Stackhouse et al. 2005) and adults with CP (Ross and Engsberg 2002; Barber et al. 2012; Geertsen et al. 2015). However, MVC is not only an estimate of the force generating properties within the muscle itself, but also of the (in)ability to generate maximal neural drive to the muscle and, therefore, cannot be used to distinguish neural and non-neural components that contribute to the force generating capacity.

In contrast, maximal muscle contractions elicited by supramaximal electrical stimulation of the tibial nerve provides an objective measure of the ability of the plantar flexors muscles to generate force independent of the neural drive to the muscle (Maffiuletti et al. 2001; Maganaris et al. 2001). Supramaximal stimulation has been used previously as part of the twitch interpolation technique in both children (Stackhouse et al. 2005) and adults with CP (Hussain et al. 2014, 2017; Neyroud et al. 2017), but not with the purpose of investigating contractile properties of the muscle. Further, supramaximal stimulation in conjunction with in vivo muscle ultrasound has been used to quantify muscle fascicle length change and characterize the force-fascicle length relationship in typically developed adults and patients after stroke and spinal cord injury (SCI) (McDonald et al. 2005; Gao and Zhang 2008; Barber et al. 2013). Therefore, the present study aimed to characterize the force generating capacity of the plantar flexor muscles in adults with CP throughout ankle ROM using torque measurements from maximal muscle twitch elicited by supramaximal stimulations. Medial gastrocnemius (MG) fascicle length change was investigated to get information of how fascicle excursion contributes to the plantar flexor torque generation during maximal muscle twitch. These techniques may provide direct information about how contractile properties are involved in plantar flexor muscle activation in adults with CP. Additionally, torque generation during MVC was obtained to be able to compare to previous work. We hypothesized that adults with CP would generate lower plantar flexor torque (both during maximal muscle twitch and MVC) and that fascicle length and shortening would be reduced compared to NI adults. Additionally, it was hypothesized that adults with CP would have an altered torque–angle relation where the optimal ankle angle for generating torque would appear in a more plantar flexed ankle position.

Method

Prior to the experiments, all participants gave written consent for participation after receiving written and verbal information in accordance with the Helsinki declaration. The study was approved by the local ethics committee (H-16028528) of greater area of Copenhagen.

Participants

Fourteen adults with spastic CP (age 36 years of age, SD 10.6, range 24–58 years, sex 9 men and 5 women, body mass 65.5 kg, SD 8.22, height 171.4 cm, SD 9.9, diplegic $n=8$, hemiplegic $=6$) participated in the study and were recruited through the Danish Cerebral Palsy Organization. The participants were classified from I–III accordance to Gross Motor Function Classification Scale (GMFCS: I $n=7$, II $n=3$, III $n=4$). Exclusion criteria were: Impaired cognitive function, surgery and/or botulinum toxin treatment in the ankle joint or in surrounded muscles within 6 months prior to the experiments. 11 of the 14 participants with CP had one or more Achilles tendon elongational surgery during their childhood. Two participants had undergone Achilles surgery three times had also had surgery in hamstrings and femur correctional surgery in their teens. The three remaining participants with CP had received no previous lower limb surgery. None of the participants had received selective dorsal rhizotomy.

A control group consisted of 17 neurologically intact (NI) participants (age 36 years of age, SD 4.5, range 26–55 years, gender 5 men and 12 women, weight 64.4 kg, SD 9.4, height 169.8 cm, SD 8.7). The NI participants were recruited through the University of Copenhagen and the Elsass Institute. This group had no reported history of neurological or musculoskeletal injury that could interfere with the experiment.

The CP and NI group are not matched regarding the distribution of men and women, although this was intended. Further, the enrollment of women in the study was done without considering the menstrual or oral contraceptive status of the participants as these factors were considered not to bias the measurements systematically.

Experimental set-up

Plantar flexor torque generation was obtained from MVC and supramaximal electrical stimulation using previously defined biomechanical evaluation methods (Lorentzen et al. 2010; Willerslev-Olsen et al. 2013). Briefly, the participants were seated in a reclining non-compliant armchair with the examined foot attached to a foot plate by two straps that prevented the heel from lifting, which ensured that the exerted torque was applied to the foot plate. The most affected side was tested in the participants with CP (left: $n=6$, right: $n=8$) and the right foot was tested in the NI participants. An electro-goniometer connected to the foot plate measured the ankle joint position, and a torque-meter measured the torque exerted on the foot plate prior to and during the two types of isometric muscle contractions. The hip joint was positioned in 100° (180° representing full extension). The knee joint was in full extension during the whole experiment to facilitate contribution of the medial gastrocnemius muscle. Thus, the exerted torque is restricted to this knee position. Full extension was restricted (5°–10° limitation) in few participants with CP due to contractures. In such cases the knee joint was positioned in an as extended position as possible. The knee position was secured by velcro belts. The ankle joint was positioned in up to 8 different angles depending on the participants ROM ranging from 60° to 130° in relation to the tibial bone.

Contractile properties

Maximal voluntary isometric contractions (MVC)

Three trials of MVC were obtained in each ankle angle with 1 min of rest between each trial. The participants were thoroughly instructed to contract their plantar flexor muscles as forcefully as possible and hold the contraction for approximately 3 s. Verbal encouragement was given by the examiner to motivate the participants to generate maximal torque. The largest torque generation was used in the subsequent analysis.

Supramaximal stimulations

To obtain a torque measure that is independent of the participants' neural drive to activate the muscle, we obtained the torque generation from supramaximal nerve stimulation. Electromyography (EMG) was used to monitor the response to the electrical stimulations and to ensure that the participants were at rest before the contractions. Bipolar electrodes (Ambu Blue Sensor N-10-A/25, Ambu A/S Ballerup. Recording area 0.5 cm^2 inter-electrode distance, 2 cm) were placed over the soleus muscle because of limited space on MG due to the ultrasound probe (see description in the subsequent section). This also avoided contact between the electrodes and gel used for the ultrasound recordings. Prior to electrode placement the skin was brushed softly with sandpaper (3 M red dot; 3 M, Glostrup, Denmark). A ground electrode was placed on the distal part of tibia. EMG signals were filtered (band-pass, 5 Hz–1 kHz), amplified (2000), and sampled at 2 kHz. The supramaximal nerve stimulation was done by stimulating the tibial nerve in the popliteal fossa supramaximally, while the participants were at rest. The cathode was a ball electrode placed over the nerve in the popliteal fossa and the anode was a plate electrode placed over the patella. Stimuli were 1 ms square pulses. Single twitch electrical stimulations were given every 10 s and increased until well above the maximal M-wave and the corresponding torque could not be increased further, reflecting activation of muscle fibers of a motor unit within the motor neuron pool of the muscle (Fig. 1a, b) (Maffiuletti et al. 2001; Vieira et al. 2015). The plantar flexor torque was subsequently calculated as the peak-to-peak amplitude. Intensity of the electrical stimulation was determined by the M wave of the soleus EMG, which may be different from MG. However, soleus EMG most likely reflect the activation of MG due to volume conduction (Hutton et al. 1988). Additionally, MG's contribution to the exerted torque will be larger than soleus since the knee was positioned in full extension during the experiment (Wakahara et al. 2007).

Ultrasound measurements

Measurements of muscle architecture was recorded from MG, which represents one part of the triceps surae muscle together with lateral gastrocnemius and soleus. MG fascicle length during MVC and supramaximal stimulation were recorded using two-dimensional, B-mode ultrasound (Telemed, Vilnius, Lithuania) with a frequency range between 50 and 65 Hz. The ultrasound probe was placed over the muscle belly of MG and aligned with the longitudinal axis of the muscle and fixed with elastic adhesive tape on the skin. Hereby, it was possible to define the muscle fascicle length as a straight line between the superficial muscle fascia and the deep aponeurosis, parallel to lines of connective tissue

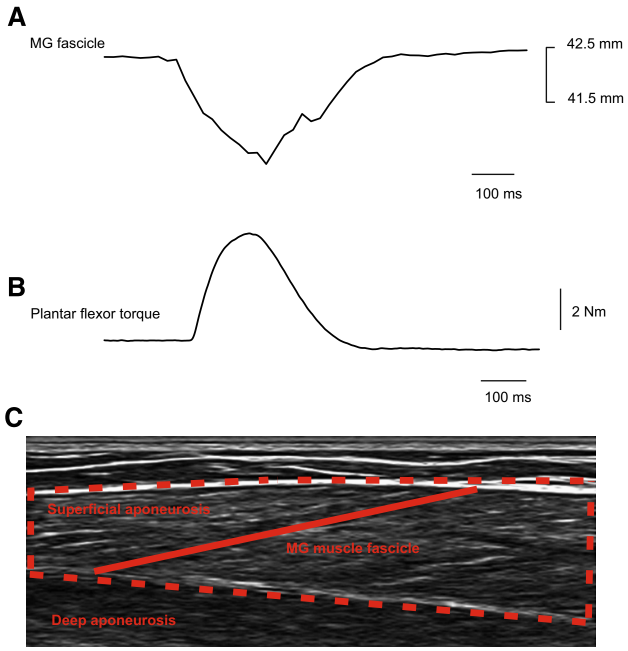

Fig. 1 The figure shows examples of original recordings of data obtained in the study. **a** The fascicle tracking illustrating the resting fascicle length and the shortening of the fascicle from the supramaximal stimulation of the tibial nerve in a neurological intact participant with the ankle joint in 90°. **b** The plantar flexor torque generated from the same supramaximal stimulation of the tibial nerve as in **a**. **c** An illustration of ultrasound recording from medial gastrocnemius muscle. The dotted square indicates the area of interest which defines the soleus muscle marked by the superficial and deep aponeurosis. The full red line defines the investigated MG muscle fascicle

visible on the image (Fig. 1c). This method has previously been shown to minimize errors when measuring fascicle length (Benard et al. 2008, 2009). MG fascicle length was determined during the MVC and supramaximal stimulations using a semi-automated fascicle tracking algorithm (Cronin et al. 2011; Gillett et al. 2013).

Statistics

Normal distribution of the outcome measurements was ensured using histograms, QQ plots and Shapiro–Wilk tests.

The plantar flexor torque from MVC and supramaximal stimulations were normalized to body mass (kg BM) and MG fascicle length measures were normalized to fibula length (cm), hereby controlling the influence of body size. All statistical analysis was performed in SAS Enterprise Guide 7.1 (SAS institute Inc., Cary, NC, USA) and figures were made in Sigmaplot 13.0. MG fascicle length and torque measurements during MVC and supramaximal stimulations were analyzed using separate mixed model regression (MIXED) analysis with ankle angle (70°–120°) and group (CP and NI) and the interaction between ankle angle and group as main effects. Using the MIXED analysis, all available data was included in the analysis in case of unbalanced datasets between the groups. Post hoc pairwise comparisons were performed using unpaired t test to detect group differences. Tukey adjustment was used in the post hoc comparisons to limit the risk of false positive results. Additionally, a sensitivity analysis was performed including the participants with CP, who had undergone Achilles tendon elongational surgery.

Relations between maximal torque generation (from both MVC and supramaximal stimulation, respectively) and fascicle shortening were analyzed using Spearman correlation coefficients. Descriptive statistics are presented as mean with standard deviations (SD) and results from the MIXED regression analysis are presented as mean differences with stand error (SE) and 95% confidence interval (CI). The level of significance was 0.05.

Results

Ankle range of motion (ROM) was reduced in the CP group. We were able to obtain data at 8 ankle angles throughout ankle ROM from 60° (maximal dorsiflexion) to 130° (maximal plantarflexion) in the NI group, but only in up to 6 ankle angles from 70° to 120° in the CP group. ROM was further reduced in 4 participants with CP ranging from 80° to 110°.

Maximal voluntary contraction (MVC) of the plantar flexor muscles

There was significant main effect of group ($F_{1,5}=21.07$, $p \leq 0.0001$), ankle angle ($F_{1,5}=13.7$, $p \leq 0.0001$) and there was a significant interaction between group and ankle angle ($F_{1,5}=2.94$, $p=0.02$). Post hoc analysis revealed significantly lower plantar flexor MVC torque for the CP group at ankle angles 70°–110° ($p \leq 0.007$, table 1A in supplementary material) compared to the NI group (Fig. 2a). There was no significant difference in plantar flexor MVC torque at 120° ($p=0.83$) between the groups. Both groups generated the largest plantar flexor MVC torque at 80°. However, the CP group generated 49.7% less torque than the NI group (mean diff: −0.28 N m/kg BM, SE 0.06, $p=0.002$, CI −0.49 to −0.07).

Torque output from supramaximal stimulation of the tibial nerve

Plantar flexor torque from supramaximal stimulation of the tibial nerve decreased from the most dorsiflexed to the most plantar flexed position in both groups (main effect of ankle angle: $F_{1,5}=51.64$, $p \leq 0.0001$) and there was a significant main effect of group ($F_{1,5}=10.66$, $p=0.003$) and

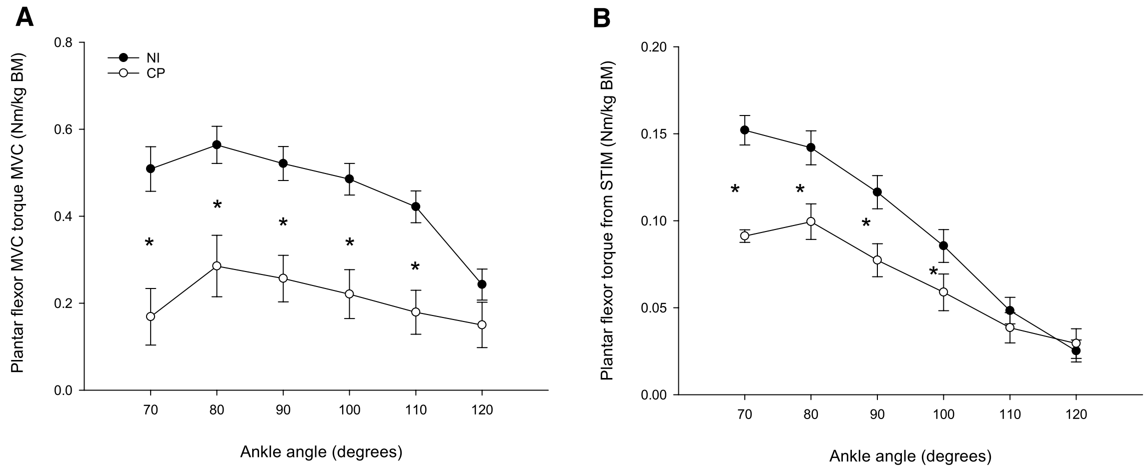

Fig. 2 Plantar flexor torque measurements throughout ankle range of motion (ROM) from dorsiflexion (70°) to plantar flexion (120°). **a** Torque generation during maximal voluntary contraction (MVC). **b** Torque generation from supramaximal electrical stimulation (STIM). Torque values are normalized to kg body mass (BM). NI (black circles) = group of neurological intact participants, CP (white circles) = group of adults with cerebral palsy. Error bars indicate standard error, asterisks indicate statistical significance

a significant interaction between group and ankle angle ($F_{1,5}=4.26$, $p=0.0013$). Post hoc analysis revealed significantly lower plantar flexor torque in the CP group at ankle angles from 70° to 100° ($p=0.03$, table 1B in supplementary material) compared to the NI group (Fig. 2b). No significant difference was observed at 110° and 120° ($p=0.28$ and 0.92, respectively, table 1B in supplementary material) between the groups. The largest difference was at 70°, where the plantar flexor torque was 45.1% lower in the CP group (mean diff −0.07 N m/kg BW, SE 0.01, $p\leq0.001$, CI −0.11 to −0.03). The largest plantar flexor torque was at 70° in the NI group, whereas the largest torque was at 80° in the CP group. The largest plantar flexor torque in the CP group was 38.7% less than the NI group (mean diff −0.06 N m/kg BW, SE 0.01, $p\leq0.0001$, CI −0.08 to −0.04).

MG fascicle length at rest

MG fascicle length declined from dorsiflexion to plantarflexion in both groups (CP: on average from 47.8 to 26.6 mm, NI 73.6 to 45.4 mm). When normalized to fibula length MG fascicle length at rest declined significantly from dorsiflexion to plantar flexion in both groups (main effect of ankle angle: $F_{1,5}=72.54$, $p\leq0.0001$) (Fig. 3a). There was also a significant main effect of group ($F_{1,5}=29.3$, $p=0.003$) showing that resting MG fascicle length was 34.2% shorter in the CP group (mean diff. −0.39 mm/cm fibula, SE 0.14, $p=0.01$, CI −0.69 to −0.1) regardless of ankle angle. Significantly lower fascicle length was confirmed in the post hoc analysis ($p\leq0.01$, table 2A in supplementary material). No effect of interaction appeared between group and

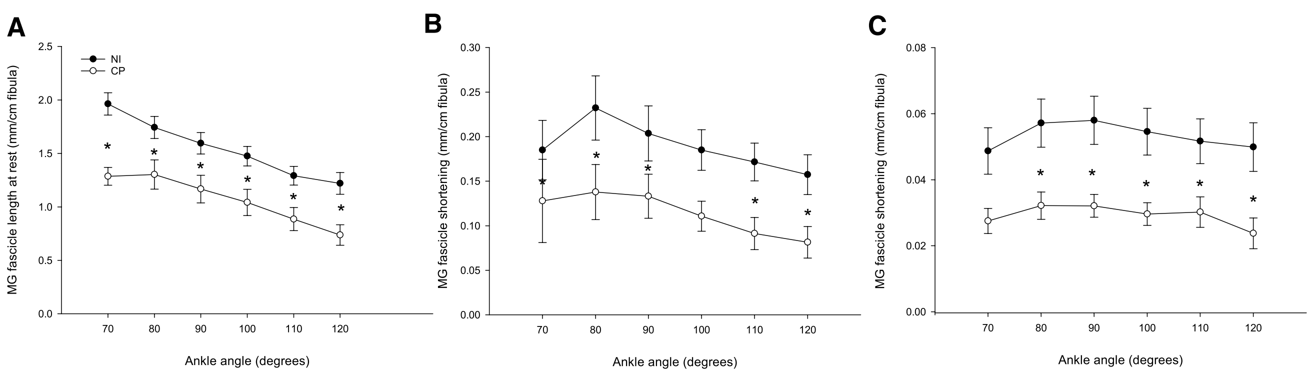

Fig. 3 **a** Medial gastrocnemius muscle (MG) fascicle length at rest throughout ankle range of motion (ROM) from dorsiflexion (70°) to plantar flexion 120 (°). **b** MG fascicle shortening during maximal voluntary contraction of the plantar flexor muscles throughout ankle ROM. **c** MG fascicle shortening during supramaximal stimulation of the tibial nerve throughout ankle ROM. MG fascicle length measures are normalized to fibula length. NI (black circles) = group of neurological intact participants, CP (white circles) = group of adults with cerebral palsy. Error bars indicate standard error, asterisks indicate statistical significance

ankle angle ($F_{1,5}=0.88$, $p=0.5$). Additionally, the difference in MG fascicle length was not influenced by the torque required to maintain the resting ankle position (main effect of group = 0.13).

MG fascicle shortening during MVC

During MVC there was significant main effect of ankle angle ($F_{1,5}=3$, $p=0.01$), and group ($F_{1,5}=7.28$, $p=0.01$) showing that MG fascicles shortened 49.6% less in the CP group compared to the NI group (mean diff. −0.08 mm/cm fibula, SE 0.04, $p=0.04$, CI −0.16 to −0.002) regardless of ankle angle. The post hoc analysis revealed that this was significant at all ankle angles ($p \leq 0.05$, table 2B in supplementary material) except at 100° ($p=0.06$). MG fascicles shortened the most at 80° but was 41.3% less in the CP group (mean diff. −0.1 mm/cm fibula, SE 0.04, $p=0.01$, CI −0.17 to −0.02) (Fig. 3b). No interaction between group and ankle angle was found ($F_{1,5}=0.17$, $p=0.97$).

MG fascicle shortening during supramaximal stimulation

No main effect of ankle angle ($F_{1,5}=0.70$, $p=0.62$) and no interaction appeared between group and ankle angle ($F_{1,5}=0.33$, $p=0.89$). However, there was a significant main effect of group ($F_{1,5}=15.60$, $p=0.0004$) showing that MG fascicle shortened 55.4% less in the CP group compared with the NI group (mean diff. −0.03 mm/cm fibula, SE 0.01, $p=0.002$, CI −0.05 to −0.01), regardless of ankle angle (Fig. 3c). The post hoc analysis revealed significantly less MG fascicle shortening at all ankle angles ($p \leq 0.01$), except at 70° ($p=0.12$, table 2C in supplementary material). As shown in Fig. 3c, there was a plateau in the fascicle shortening between 80° and 100° with no clear appearance of a peak in both groups.

Relation between maximal torque generation and MG fascicle length and shortening

The relation between MG fascicle shortening and maximal plantar flexor torque generation can be seen in Fig. 4. There was a moderate correlation between MG fascicle shortening and plantar flexor MVC torque in both groups, however, did not reach statistical significance in the CP group (CP group: $r=0.51$, $p=0.06$; NI group: $r=0.50$, $p=0.04$) (Fig. 4a). The plantar flexor torque from supramaximal stimulation was moderately correlated with MG fascicle shortening in the CP group ($r=0.56$, $p=0.06$). No correlation was found between these measures in the NI group ($r=0.26$, $p=0.32$) (Fig. 4b).

Influence of surgery

A sensitivity analysis was performed where the three participants who had received no previous lower limb surgery were excluded. The remaining subgroup who had undergone Achilles tendon elongation ($n=11$, age 35 years of age, SD 9.5, range 24–51 years, gender 7 men and 4 women, weight 66 kg, SD 7.6, height 172.5 cm, SD 10.1, diplegic $n=7$, hemiplegic $n=4$, GMFCS: I $n=4$, II $n=3$, III $n=4$) showed tendencies to lower plantar flexor torque generation during MVC and supramaximal stimulation and shorter MG fascicle length and shortening than the whole CP group. However, these were not statistically significant.

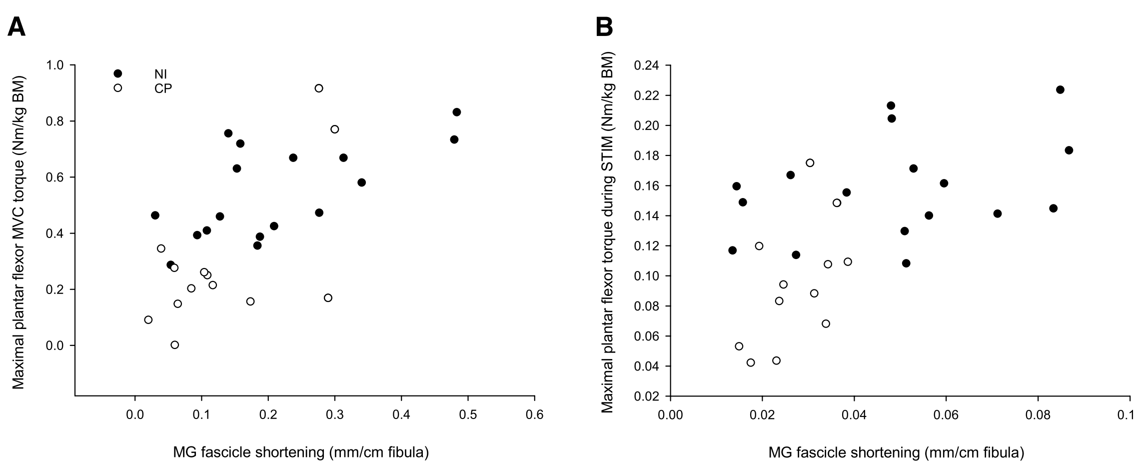

Fig. 4 **a** Relation between maximal ankle torque generation and medial gastrocnemius (MG) muscle fascicle shortening during maximal voluntary contraction (MVC). **b** The relation between maximal plantar flexor torque from maximal muscle twitch elicited by supramaximal electrical stimulation and MG fascicle shortening. Black circles represent neurological intact participants (NI). White circles represent adults with cerebral palsy (CP). Torque measurements are normalized to kg body mass (BM) and MG fascicle shortening is normalized to fibula length

Discussion

In this study, plantar flexor muscle torque and MG muscle fascicle length were simultaneously measured during maximal muscle twitches elicited by supramaximal stimulation and MVC in adults with CP and NI adults. Plantar flexor torque during both types of contractions was significantly lower and MG fascicle shortening throughout the ankle range of motion was significantly less in adults with CP compared to NI adults.

Reduced torque-generating ability in adults with CP

Our findings of reduced plantar flexor MVC torque and reduced MG fascicle shortening in adults with CP is consistent with previous findings in children and young adults with CP (Ross and Engsberg 2002; Barber et al. 2012) and confirms that adults with CP have reduced ability to generate plantar flexor torque during voluntary activation of the plantar flexor muscles (Ross and Engsberg 2002; Elder et al. 2003; Stackhouse et al. 2005; Geertsen et al. 2015; Kirk et al. 2016).

In the CP group during supramaximal stimulation maximal plantar flexor torque was reduced up to 47% across the ankle range of motion. Reduced plantar flexor torque from supramaximal stimulation across the majority of the range of motion of the ankle strongly suggests that local muscle alterations of the contractile properties of the muscle limit the torque generating ability in adults with CP. This points to muscle atrophy, with a reduction of the number of myosin and actin filaments in parallel and series, as an important cause of the reduced torque generation (Gordon et al. 1966; Shortland et al. 2002; Lieber et al. 2004).

It should be pointed out that we cannot directly infer that reduced torque generation found by supramaximal stimulations would affect muscle force generation during voluntary movements, such as gait, to the same extent. In contrast to voluntary movements, plantar flexor torque generated during maximal muscle twitches, that are evoked by supramaximal stimulations, do not take spatial and temporal summation of motor-unit activation and synergistic muscle activation into account.

The largest plantar flexor torque during supramaximal stimulation was observed at a slightly more plantar flexed position in the adults with CP (at 80°) than in NI adults (at 70°). This is in contrast to what was observed for MVC and may illustrate the importance of evaluating the contractile properties of the muscles by supramaximal stimulation rather than MVC. During MVC, adults with CP may compensate for impaired contractile properties of the muscle by activating synergist muscles (Maganaris et al. 2001). Thus, MVC torque estimates should be interpreted with caution when used to characterize the force generating capacity of the muscle in adults with CP. Using ankle torque generation from supramaximal stimulations as done in the present study, it may be possible to detect an altered torque–angle relation as compared to NI adults, which is consistent to what observed in patients after stroke and SCI (McDonald et al. 2005; Gao and Zhang 2008).

Additionally, differences in Achilles tendon length between adults with CP and NI adults also need to be taken into account concerning the force generating capacity (Kalkman et al. 2017). This may confound the torque–angle relationship, but it will not influence the torque measurements at the individual ankle joint positions. Moreover, no interaction between group and ankle angle was shown in the analysis, which makes it unlikely that differences in tendon length should have influenced our results significantly. Increased Achilles tendon length and increased tendon compliance, which have been found in adults with CP in previous studies, may affect plantar flexor MVC torque (Zhao et al. 2009; Wren et al. 2010; Barber et al. 2012), but tendon compliance appears to influence maximal muscle twitches elicited by supramaximal stimulations very little, since the contraction has a very short duration and the elicited torque is relative low compared to MVC (McDonald et al. 2005).

Reduced fascicle length at rest

The CP group had on average 34% shorter MG fascicle length at rest than the NI group. A similar reduction in resting muscle fascicle length has also been reported in previous studies in children and adults with CP (Mohagheghi et al. 2007, 2008; Barber et al. 2011; Matthiasdottir et al. 2014) using similar methods of measuring passive ankle torque throughout ankle range of motion. Reduction of fascicle length is also seen in animal studies following immobilization in shortened position (Williams and Goldspink 1973; Spector et al. 1982), and is generally assumed to be related to an adaptive reduction of the number of sarcomeres in series. There are also studies in which no reduction of muscle fascicle length has been found in children with CP (Shortland et al. 2002, 2004; Malaiya et al. 2007). However, methodological differences between studies may explain length measurement discrepancies. First, recording ankle torque throughout the range of motion during fascicle length measurement, as was performed in this study, enables a comparison of fascicle length between groups at the same torque. Comparisons of MG fascicle length with unknown applied ankle torque, when the foot/ankle complex is in a "resting position", should be interpreted with caution. Second, proper ultrasound transducer placement and stabilization have been found to be crucial for valid measurements of fascicle length (Benard et al. 2009). Ultrasound probe movement due to ineffective positioning and fixation may impact fascicle length measures as the fascicle may move out of the image plane and appear shorter. Third, subgroups of CP participants with varied functional level (GMFCS) and/or treatment history (pharmacological and surgical interventions) may also lead to contrasting results. Shortland and colleagues (Shortland et al. 2004) found no difference in MG fascicle length prior to gastrocnemius recession between children with CP and TD children, however, post-surgery MG fascicle lengths were reduced by 28% when measured in maximal dorsiflexion, which is consistent with the present findings in adults with CP. Additionally, differences in enrolled participants may also lead to contrasting findings between studies. This may be the case in the study by Hussain et al. (2017) which found longer fascicle length in men with CP with GMFCS I whereas shorter fascicle length was found in our study in which both men and women with CP with GMCS I–III was included. These differences may influence muscle size and fascicle length which consequently makes a direct comparison between the studies difficult.

Reduced fascicle shortening

The CP group had 55% less MG fascicle shortening during the muscle twitch (and 49.6% during MVC) than the NI group. When relating maximal torque generation to the corresponding fascicle shortening during supramaximal stimulation (Fig. 4), we observed reduced torque for a given shortening of the fascicles in the adults with CP. Shorter muscles (and fascicles), with less sarcomeres in series, have a smaller capacity for shortening (Lieber and Friden 2000). It would, therefore, be reasonable to consider that the shorter fascicles lengths in adults with CP reported in this study result in reduced capacity of the fascicles to shorten and reduced for generation of torque across the individual joint positions. Our results also showed that participants with the shortest resting MG fascicle length also showed the most reduced MG fascicle shortening. This may reflect an altered torque-fascicle length relation in adults with CP as a reduction of the number of sarcomeres in series resulting in a shorter muscle as has been suggested also previously (Lieber et al. 2004; Gough and Shortland 2012). If so, this may be a contributing factor for toe walking in individuals with CP, since a more plantar flexed position during gait would take advantage of the alteration in the torque–angle relation. Clinically, this may indicate that adults with CP have optimized their gait pattern where muscle fascicle shortening is executed where largest plantar flexor torque can be generated to ensure propulsion.

The reduced shortening of the muscle fascicles during supramaximal stimulation may also be related to other impairments in the function of the neuromuscular unit. Previous studies have found evidence of impaired development of the motor end-plate in children (Robinson et al. 2013) and adults with CP (Theroux et al. 2005). This is likely to reduce the available amount of acetylcholine throughout the muscle fiber, limiting maximal activation of motor units associated to the alpha motor neuron. Increased amount of connective tissue (Friden and Lieber 2003; Smith et al. 2011) and intramuscular fat (Johnson et al. 2009) may also impair shortening of the fascicles by increasing the stiffness of the muscle–tendon complex (Barrett and Lichtwark 2010; Lorentzen et al. 2010, 2017; Ranatunga 2011).

Clinical implications

Reduced contractile properties of the muscles for generating plantar flexor torque may also apply to functional motor tasks such as walking. This is supported by the findings of Barber et al. (2017) which found 10% less fascicle shortening during the push-off phase of walking accompanied with reduced ankle power production in children with CP (Barber et al. 2017). In line with this, Hösl et al. (2016) found 37% lower concentric fascicle excursion during gait in children with CP than in TD children. It is important to note that torque/power generated at the time of push-off is the sum of released torque stored in tendons (passive torque) and generated torque by muscles (active torque) (Lichtwark and Wilson 2006, 2008). The advantage of using a controlled protocol by measuring plantar flexor torque from supramaximal stimulations while tracking MG fascicle shortening, as done in the present study, is that it provides direct evidence that reduced contractile properties of the muscle reduces the torque-generating ability in adults with CP. Our findings, therefore, highlight the importance to obtain direct measures of contractile properties of the muscle to be able to address interventions to improve the contractile properties of the muscle for generating torque in adults with CP. Implementation of strength-gaining interventions have previously shown to improve muscle function and strength (Taylor et al. 2013; Kirk et al. 2016; Gillett et al. 2018) and may thus serve as an essential part of gait training in adults with CP and other persons with central nervous lesions. Although strength training generally does not appear to improve functional abilities in itself, it could be an important supplement to more functionally oriented training. Gait training with various degree of inclination may also improve the muscles excursion range, which has been shown to have positive effect on muscle stiffness and gait performance in children and adults with CP (Willerslev-Olsen et al. 2014; Hösl et al. 2016; Lorentzen et al. 2017).

Conclusion

The present study obtained torque measurements from supramaximal stimulation of the tibial nerve to characterize torque generating capacity of the triceps surae muscle throughout ankle ROM in adults with CP. Impaired contractile properties shown as reduced torque generation during maximal muscle twitch were underpinned by the observation of reduced MG fascicle length and shortening from ultrasound recordings throughout ankle ROM. Using supramaximal stimulation, we were able to detect an altered torque-fascicle length/ankle angle relation in adults with CP, which was not detected using MVC. This strongly indicates that reduced fascicle length and shortening are essential contributors to reduced torque generation in adults with CP and are in all likelihood contributing to reduced push-off while walking. Strength training interventions aiming to improve muscle strength and gait training with de- and inclination to increase muscle fascicle excursion range may be useful interventions to improve contractile properties of the plantar flexor muscles in adults with CP.

Acknowledgements We are grateful to Michaël Bertrand and Rafael Curbelo for valuable help with the experiments and to all participants. The study was supported by the Elsass foundation, Charlottenlund, Denmark. The foundation was not involved in the conduct or decision making regarding the work presented in the paper.

Author contributions JBN, JLO and RFF have conceived and designed research. JBN, JLO and RFF conducted the experiments. RFF and LB analyzed data. RFF did the statistics and wrote the manuscript in collaboration with JBN. All authors have read and approved the manuscript.

Compliance with ethical standards

Conflict of interest No conflicts of interest, financial or otherwise, are declared by the authors. All authors have contributed essentially to the work and have approved the final publication.

References

Barber L, Barrett R, Lichtwark G (2011) Passive muscle mechanical properties of the medial gastrocnemius in young adults with spastic cerebral palsy. J Biomech 44:2496–2500

Barber L, Barrett R, Lichtwark G (2012) Medial gastrocnemius muscle fascicle active torque-length and Achilles tendon properties in young adults with spastic cerebral palsy. J Biomech 45:2526–2530

Barber LA, Barrett RS, Gillett JG, Cresswell AG, Lichtwark GA (2013) Neuromechanical properties of the triceps surae in young and older adults. Exp Gerontol 48:1147–1155

Barber L, Carty C, Modenese L, Walsh J, Boyd R, Lichtwark G (2017) Medial gastrocnemius and soleus muscle-tendon unit, fascicle, and tendon interaction during walking in children with cerebral palsy. Dev Med Child Neurol 59:843–851

Barrett RS, Lichtwark GA (2010) Gross muscle morphology and structure in spastic cerebral palsy: a systematic review. Dev Med Child Neurol 52:794–804

Benard MR, Becher J, Harlaar J, Huijing PA, Jaspers R (2008) Ultrasound measurements of muscle parameters of human gastrocnemius medialis are critically determined by probe orientation. Gait Posture 28:S16–S17

Benard MR, Becher JG, Harlaar J, Huijing PA, Jaspers RT (2009) Anatomical information is needed in ultrasound imaging of muscle to avoid potentially substantial errors in measurement of muscle geometry. Muscle Nerve 39:652–665

Cronin NJ, Carty CP, Barrett RS, Lichtwark G (2011) Automatic tracking of medial gastrocnemius fascicle length during human locomotion. J Appl Physiol (1985) 111:1491–1496

Elder GC, Kirk J, Stewart G, Cook K, Weir D, Marshall A, Leahey L (2003) Contributing factors to muscle weakness in children with cerebral palsy. Dev Med Child Neurol 45:542–550

Friden J, Lieber RL (2003) Spastic muscle cells are shorter and stiffer than normal cells. Muscle Nerve 27:157–164

Gao F, Zhang LQ (2008) Altered contractile properties of the gastrocnemius muscle poststroke. J Appl Physiol (1985) 105:1802–1808

Geertsen SS, Kirk H, Lorentzen J, Jorsal M, Johansson CB, Nielsen JB (2015) Impaired gait function in adults with cerebral palsy is associated with reduced rapid force generation and increased passive stiffness. Clin Neurophysiol 126:2320–2329

Gillett JG, Barrett RS, Lichtwark GA (2013) Reliability and accuracy of an automated tracking algorithm to measure controlled passive and active muscle fascicle length changes from ultrasound. Comput Methods Biomech Biomed Eng 16:678–687

Gillett JG, Lichtwark GA, Boyd RN, Barber LA (2018) Functional anaerobic and strength training in young adults with cerebral palsy. Med Sci Sports Exerc 50:1549–1557

Gordon AM, Huxley AF, Julian FJ (1966) The variation in isometric tension with sarcomere length in vertebrate muscle fibres. J Physiol 184:170–192

Gough M, Shortland AP (2012) Could muscle deformity in children with spastic cerebral palsy be related to an impairment of muscle growth and altered adaptation?. Dev Med Child Neurol 54:495–499

Hösl M, Bohm H, Arampatzis A, Keymer A, Doderlein L (2016) Contractile behavior of the medial gastrocnemius in children with bilateral spastic cerebral palsy during forward, uphill and backward-downhill gait. Clin Biomech (Bristol Avon) 36:32–39

Hussain AW, Onambele GL, Williams AG, Morse CI (2014) Muscle size, activation, and coactivation in adults with cerebral palsy. Muscle Nerve 49:76–83

Hussain AW, Onambele GL, Williams AG, Morse CI (2017) Medial gastrocnemius specific force of adult men with spastic cerebral palsy. Muscle Nerve 56:298–306

Hutton RS, Roy RR, Edgerton VR (1988) Coexistent Hoffmann reflexes in human leg muscles are commonly due to volume conduction. Exp Neurol 100:265–273

Jahnsen R, Villien L, Egeland T, Stanghelle JK, Holm I (2004a) Locomotion skills in adults with cerebral palsy. Clin Rehabil 18:309–316

Jahnsen R, Villien L, Aamodt G, Stanghelle JK, Holm I (2004b) Musculoskeletal pain in adults with cerebral palsy compared with the general population. J Rehabil Med 36:78–84

Johnson DL, Miller F, Subramanian P, Modlesky CM (2009) Adipose tissue infiltration of skeletal muscle in children with cerebral palsy. J Pediatr 154:715–720

Kalkman BM, Bar-On L, Cenni F, Maganaris CN, Bass A, Holmes G, Desloovere K, Barton GJ, O'Brien TD (2017). Achilles tendon moment arm length is smaller in children with cerebral palsy than in typically developing children. J Biomech 56:48–54

Kirk H, Geertsen SS, Lorentzen J, Krarup KB, Bandholm T, Nielsen JB (2016) Explosive resistance training increases rate of force development in ankle dorsiflexors and gait function in adults with cerebral palsy. J Strength Cond Res 30:2749–2760

Lichtwark GA, Wilson AM (2006) Interactions between the human gastrocnemius muscle and the Achilles tendon during incline, level and decline locomotion. J Exp Biol 209:4379–4388

Lichtwark GA, Wilson AM (2008) Optimal muscle fascicle length and tendon stiffness for maximising gastrocnemius efficiency during human walking and running. J Theor Biol 252:662–673

Lieber RL, Friden J (2000) Functional and clinical significance of skeletal muscle architecture. Muscle Nerve 23:1647–1666

Lieber RL, Friden J (2002) Spasticity causes a fundamental rearrangement of muscle-joint interaction. Muscle Nerve 25:265–270

Lieber RL, Steinman S, Barash IA, Chambers H (2004) Structural and functional changes in spastic skeletal muscle. Muscle Nerve 29:615–627

Lorentzen J, Grey MJ, Crone C, Mazevet D, Biering-Sorensen F, Nielsen JB (2010) Distinguishing active from passive components of ankle plantar flexor stiffness in stroke, spinal cord injury and multiple sclerosis. Clin Neurophysiol 121:1939–1951

Lorentzen J, Kirk H, Fernandez-Lago H, Frisk R, Scharff Nielsen N, Jorsal M, Nielsen JB (2017) Treadmill training with an incline reduces ankle joint stiffness and improves active range of movement during gait in adults with cerebral palsy. Disabil Rehabil 39:987–993

Maffiuletti NA, Martin A, Babault N, Pensini M, Lucas B, Schieppati M (2001) Electrical and mechanical H(max)-to-M(max) ratio in power- and endurance-trained athletes. J Appl Physiol (1985) 90:3–9

Maganaris CN, Baltzopoulos V, Ball D, Sargeant AJ (2001) In vivo specific tension of human skeletal muscle. J Appl Physiol (1985) 90:865–872

Malaiya R, McNee AE, Fry NR, Eve LC, Gough M, Shortland AP (2007) The morphology of the medial gastrocnemius in typically developing children and children with spastic hemiplegic cerebral palsy. J Electromyogr Kinesiol 17:657–663

Matthiasdottir S, Hahn M, Yaraskavitch M, Herzog W (2014) Muscle and fascicle excursion in children with cerebral palsy. Clin Biomech (Bristol, Avon) 29:458–462

McDonald MF, Kevin Garrison M, Schmit BD (2005) Length-tension properties of ankle muscles in chronic human spinal cord injury. J Biomech 38:2344–2353

Meinders M, Gitter A, Czerniecki JM (1998) The role of ankle plantar flexor muscle work during walking. Scand J Rehabil Med 30:39–46

Mohagheghi AA, Khan T, Meadows TH, Giannikas K, Baltzopoulos V, Maganaris CN (2007) Differences in gastrocnemius muscle architecture between the paretic and non-paretic legs in children with hemiplegic cerebral palsy. Clin Biomech (Bristol, Avon) 22:718–724

Mohagheghi AA, Khan T, Meadows TH, Giannikas K, Baltzopoulos V, Maganaris CN (2008) In vivo gastrocnemius muscle fascicle length in children with and without diplegic cerebral palsy. Dev Med Child Neurol 50:44–50

Morgan P, Murphy A, Opheim A, McGinley J (2016) Gait characteristics, balance performance and falls in ambulant adults with cerebral palsy: an observational study. Gait Posture 48:243–248

Neptune RR, Kautz SA, Zajac FE (2001) Contributions of the individual ankle plantar flexors to support, forward progression and swing initiation during walking. J Biomech 34:1387–1398

Neyroud D, Armand S, De Coulon G, Sarah RDDS, Maffiuletti NA, Kayser B, Place N (2017) Plantar flexor muscle weakness and fatigue in spastic cerebral palsy patients. Res Dev Disabil 61:66–76

Olney SJ, MacPhail HE, Hedden DM, Boyce WF (1990) Work and power in hemiplegic cerebral palsy gait. Phys Ther 70:431–438

Parkes J, McCullough N, Madden A (2010) To what extent do children with cerebral palsy participate in everyday life situations?. Health Soc Care Community 18:304–315

Ranatunga KW (2011) Skeletal muscle stiffness and contracture in children with spastic cerebral palsy. J Physiol 589:2665

Riad J, Modlesky CM, Gutierrez-Farewik EM, Brostrom E (2012) Are muscle volume differences related to concentric muscle work during walking in spastic hemiplegic cerebral palsy?. Clin Orthop Relat Res 470:1278–1285

Robinson KG, Mendonca JL, Militar JL, Theroux MC, Dabney KW, Shah SA, Miller F, Akins RE (2013) Disruption of basal lamina components in neuromotor synapses of children with spastic quadriplegic cerebral palsy. PLoS One 8:e70288

Roche N, Pradon D, Cosson J, Robertson J, Marchiori C, Zory R (2014) Categorization of gait patterns in adults with cerebral palsy: a clustering approach. Gait Posture 39:235–240

Ross SA, Engsberg JR (2002) Relation between spasticity and strength in individuals with spastic diplegic cerebral palsy. Dev Med Child Neurol 44:148–157

Shikako-Thomas K, Dahan-Oliel N, Shevell M, Law M, Birnbaum R, Rosenbaum P, Poulin C, Majnemer A (2012) "Play and be happy? Leisure participation and quality of life in school-aged children with cerebral palsy." Int J Pediatr 2012:387280

Shortland AP, Harris CA, Gough M, Robinson RO (2002) Architecture of the medial gastrocnemius in children with spastic diplegia. Dev Med Child Neurol 44:158–163

Shortland AP, Fry NR, Eve LC, Gough M (2004) Changes to medial gastrocnemius architecture after surgical intervention in spastic diplegia. Dev Med Child Neurol 46:667–673

Smith LR, Lee KS, Ward SR, Chambers HG, Lieber RL (2011) Hamstring contractures in children with spastic cerebral palsy result from a stiffer extracellular matrix and increased in vivo sarcomere length. J Physiol 589:2625–2639

Spector SA, Simard CP, Fournier M, Sternlicht E, Edgerton VR (1982) Architectural alterations of rat hind-limb skeletal muscles immobilized at different lengths. Exp Neurol 76:94–110

Stackhouse SK, Binder-Macleod SA, Lee SC (2005) Voluntary muscle activation, contractile properties, and fatigability in children with and without cerebral palsy. Muscle Nerve 31:594–601

Taylor NF, Dodd KJ, Baker RJ, Willoughby K, Thomason P, Graham HK (2013) Progressive resistance training and mobility-related function in young people with cerebral palsy: a randomized controlled trial. Dev Med Child Neurol 55:806–812

Theroux MC, Oberman KG, Lahaye J, Boyce BA, Duhadaway D, Miller F, Akins RE (2005) Dysmorphic neuromuscular junctions associated with motor ability in cerebral palsy. Muscle Nerve 32:626–632

Vieira TM, Botter A, Minetto MA, Hodson-Tole EF (2015) Spatial variation of compound muscle action potentials across human gastrocnemius medialis. J Neurophysiol 114:1617–1627

Wakahara T, Kanehisa H, Kawakami Y, Fukunaga T (2007) Fascicle behavior of medial gastrocnemius muscle in extended and flexed knee positions. J Biomech 40:2291–2298

Willerslev-Olsen M, Lorentzen J, Sinkjaer T, Nielsen JB (2013) Passive muscle properties are altered in children with cerebral palsy before the age of 3 years and are difficult to distinguish clinically from spasticity. Dev Med Child Neurol 55:617–623

Willerslev-Olsen M, Lorentzen J, Nielsen JB (2014) Gait training reduces ankle joint stiffness and facilitates heel strike in children with cerebral palsy. NeuroRehabilitation 35:643–655

Williams PE, Goldspink G (1973) The effect of immobilization on the longitudinal growth of striated muscle fibres. J Anat 116:45–55

Winter DA (1983) Energy generation and absorption at the ankle and knee during fast, natural, and slow cadences. Clin Orthop Relat Res: 147–154

Wren TA, Cheatwood AP, Rethlefsen SA, Hara R, Perez FJ, Kay RM (2010) Achilles tendon length and medial gastrocnemius architecture in children with cerebral palsy and equinus gait. J Pediatr Orthop 30:479–484

Zhao H, Ren Y, Wu YN, Liu SQ, Zhang LQ (2009) Ultrasonic evaluations of Achilles tendon mechanical properties poststroke. J Appl Physiol (1985) 106:843–849