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ABSTRACT

Background: Children with spastic cerebral palsy gradually lose muscle extensibility but the interplay between the muscular and neurological components of the condition is unclear especially in the pathophysiology of equinovalgus gait.
Aim: This study aimed to quantify the muscular and neurological disorders in young children with unilateral cerebral palsy, and to investigate the role of the peroneus longus (PL) in equinovalgus gait.
Design, setting and population: This was an observational study with prospective assessments of 31 children (median age: 2.9 years, range: 2–6) from outpatient clinic in a tertiary teaching hospital.
Methods: Clinical measures of plantar flexor extensibility (X_{V1}), stretch response (X_{V3}), and active ankle dorsiflexion angle (X_A) were obtained as well as walking velocity and electromyography of tibialis anterior (TA), gastrocnemius medialis (GM) and PL during walking.
Results: We found reduced extensibility of the triceps surae on the paretic side (effect size $r = 0.73$, $p < 0.001$ for soleus and $r = 0.68$, $p < 0.001$ for gastrocnemius) and a correlation between reduced triceps surae extensibility and earlier stretch response ($\rho = 0.5$, $p = 0.004$). During the swing phase, there was major co-contraction between TA and GM/PL, and significantly larger activation of PL compared to GM ($r = 0.46$, $p = 0.011$). Both GM and PL activation decreased with age.
Conclusions: Our results suggest gradual deterioration of the muscular disorder and a link between the muscular and neurological disorders, although plantar flexor co-contraction improved with age. The PL was more activated than the GM and may be considered an intervention target to treat equinovalgus gait.

1. Introduction

Cerebral palsy refers to a group of disorders that originate from a static lesion to the brain before or shortly after birth, which leads to a lack of spinal cord development (Graham et al., 2016). The prevalence of cerebral palsy has remained stable at around 2.5 per 1000 births (McIntyre, 2018) although there is a trend towards an increase in disorders primarily affecting one side of the body (e.g. unilateral spastic cerebral palsy, USCP) instead of both sides (Himmelmann and Uvebrant, 2014).

Children with USCP often present with equinus gait (Perry et al., 1992) whereby the ankle is plantar flexed at initial contact and for most of the stance phase of gait. Equinus gait may be attributed to the overactivity and/or reduced extensibility of the plantar flexors, and to the

weakness of the dorsiflexors. Indeed, neurological (e.g. hypertonia and weakness due to reduced downstream command) and muscle (e.g. reduced length and weakness due to reduced muscle volume) disorders are concomitant and difficult to disentangle.

With respect to the control of the ankle during gait, the neurological disorder has three main components: (i) reduced central command of tibialis anterior (Elder et al., 2003), (ii) co-contraction of ankle agonists and antagonists (Gracies, 2005; Geertsen et al., 2018), (iii) hypertonia of the plantar flexors expressed both as a spastic stretch reflex, tested at rest (Ross and Engsberg, 2002) and as dystonic postures (Baude et al., 2019).

The muscle disorder has two main components: (i) reduced muscle length and/or increased stiffness which worsen with growth and may lead to reduced extensibility first, then contractures (Gough and Shortland, 2012; Willerslev-Olsen et al., 2018), (ii) muscle weakness due to reduced growth in volume and cross-sectional area (Noble et al., 2014).

The respective responsibilities between the muscle and neurological disorders for equinus gait at an early age are still uncertain (Willerslev-Olsen et al., 2013; Hägglund and Wagner, 2011). The Five-Step Assessment (FSA) of spastic paresis applied to the plantar flexors proposes to use clinical examination measurements as proxy for the overall extensibility of the muscle-tendon unit, the threshold of their passive spastic stretch reflex, and the balance between agonists and antagonist muscles during attempts at active ankle dorsiflexion (Gracies et al., 2010). The first aim of this study was to investigate the muscle and neurological disorders around the ankle, utilizing measurements from the Five-Step Assessment.

The second aim was to investigate the role of the peroneus longus in equinovalgus gait. The peroneus longus (PL) muscle contributes to ankle valgus and plantar flexion (Boulay et al., 2012). In early primates, the role of the PL was to support gripping with feet equipped with a detached thumb. The role of the PL has changed following adoption of bipedalism and modifications to the foot anatomy. The PL contributes to the longitudinal and transverse stiffness of the arch of the foot. Synergistic actions of the tibialis anterior (TA) and the PL increase the arch of the foot (Keith, 1928). The combination of weakness of TA and overactivity of PL may lead to equinovalgus gait first and, with time, to midfoot break (Young et al., 1990).

2. Material and methods

This was a cross-sectional study of patients attending the gait clinic of the pediatric orthopedy department at the Hôpitaux Universitaires de Marseille (AP-HM, Marseille, France) between March 2009 and November 2014.

Assessment and data collection were prospective. All children who presented to the gait clinic were assessed for the following inclusion and exclusion criteria. Inclusion criteria were (1) a confirmed diagnosis of unilateral cerebral palsy with a gross motor function classification system (GMFCS) 1 or 2 assessed by BC (MD) and CB (MD), (2) under 6 years of age, and (3) presence of equinovalgus gait, defined as initial ground contact with the hallux and/or the head of the first metatarsal and assessed using multiple angles video footages (Sup. Fig. 1). Equinovalgus foot contact was judged by two independent assessors CB (MD) and GA (PT) with over 10 years of experience in the clinical assessment of children with cerebral palsy. Exclusion criteria were (1) prior surgical interventions or toxin injections to any of the lower limb muscles, (2) severe fixed equinovalgus contracture defined as a maximum passive ankle angle of dorsiflexion (knee flexed) of 10° plantar flexed, and (3) lower limb length discrepancy greater than 1 cm.

The study was approved by the local ethics committee (Aix-Marseille University, 2014-11-05-03) and parents provided written informed consent prior to their children participating in the study.

Clinical examination included the first three technical steps of the Five Step Assessment (FSA, (Yelnik et al., 2010) of spastic paresis, all measurements utilized a goniometer. Ankle joint range of movement measurements targeting the soleus (knee flexed) and the gastrocnemius-soleus complex (GSC, knee extended) were performed with the patient in the supine position and ensuring that no foot pronation occurred. The maximum passive stretch, termed extensibility, was quantified by X_{V1}, the angle of arrest under slow stretching. The ankle angle was measured between the fibula and the posterior half of the external border of the foot. The threshold for the spastic stretch response of the muscles was quantified by X_{V3}, the angle of catch or clonus under rapid passive stretching. The balance between the maximal activation of the dorsiflexors and the passive and active resistance of the plantar flexors was quantified by X_A, the ankle angle measured while the children performed a maximum voluntary ankle dorsiflexion, in the knee extended and knee flexed positions.

We provide the reliability of the clinical examination measurements in supplementary materials. The standard error of the measurements were 2.3° or less (supplementary materials). The clinical examination measurements were compared to that of typically developed children published by Mudge et al. with the closest age range (4–7 years old, n = 20) (Mudge et al., 2014).

Video and dynamic EMG recordings were performed as the children walked barefoot at self-selected speed back and forth a 10 m long walkway for 2 min (Sup. Fig. 1). One video camera was located half the length of the walkway, looking perpendicular to the sagittal plane. Stride length and walking speed were extracted using the software Kinovea, following calibration using an object with known length in the field of view. Walking speed measurements were determined for a representative gait cycle. Non-dimensional walking speed was calculated using the subject's height as the characteristic length (Hof, 1996).

The EMG activity of the gastrocnemius medialis (GM), peroneus longus (PL) and tibialis anterior (TA) was recorded as per SENIAM recommendations for non-invasive surface EMG in young children (Boulay et al., 2012). The electrodes were maintained in place using elastic tape so the children could move freely. EMG data were collected wirelessly (Wave wireless EMG, Cometa, Italy) at 2000 Hz. The EMG signal was amplified (×1000), filtered (10 Hz high-pass filter, 500 Hz low-pass filter), and rectified.

A series of movement were performed with the patients lying supine to verify there was no crosstalk (synchronized onset) between the signals recorded by the GM and PL electrodes. The presence of timing differences was verified during voluntary ankle plantar flexion (knee extended) for the GM, ankle plantar flexion with forefoot pronation for the PL and active dorsiflexion for the TA.

Reference EMG signals for the GM and PL muscles were recorded with the patients standing on tiptoes for 5 s with their hands gently resting on a table to maintain balance. Reference EMG signal for the TA was recorded with the patients performing active dorsiflexion. The peak voltage and RMS over 500 ms around the peak were measured using the EMG Easy Report© software (MerloBioengineering, Italy; Sup. Fig. 3).

EMG data was collected during walking back and forth the length of the gait laboratory for trials of 2 min. The foot contact and foot off events were identified using four footswitches per foot, taped under the hallux, the first and fifth metatarsal heads, and the heel (Cometa, Italy). The RMS amplitude of the EMG signal from the muscles was averaged for the overall swing phase (SW) and in three equal thirds of the swing phase (SW1, SW2 and SW3). The SW, SW1, SW2, and SW3 activation percentages for GM, PL and TA muscles were calculated as the ratio between the averaged RMS signal during each phase and the RMS signal from the same muscle during standing on tiptoes (for GM and PL) or active ankle dorsiflexion while standing (for TA).

2.1. Statistical analysis

Non-parametric tests were used throughout the study because most variables were not normally distributed, as tested with the Shapiro-Wilk test. We used the median, interquartile range (IQR), min and max to summarize the data. We used the Wilcoxon-Kolmogorov test (α level for significance = 0.05) to investigate paired differences between data from

the paretic and non-paretic sides, and paired differences of EMG activation percentages in swing between gastrocnemius and peroneus longus. We used spearman rank correlation tests between variables. All statistical analysis was conducted in R, (R Core Team. R, 2014).

3. Results

Thirty-one children met the inclusion–exclusion criteria and agreed to participate in the study. The subjects were between 2 and 6 years of age, between 79 and 112 cm tall, and weighed between 9 and 22 kg. Gait speed range was 0.2 to 1.1 $m.s^{-1}$ and non-dimensional speed range was 0.06 to 0.36 (Table 1).

The soleus and GSC were less extensible (as measured by X_{V1}) on the paretic compared to the non-paretic side (Table 1, effect size $r = 0.73$ $CI_{95\%}$ [0.64,0.81] $p < 0.001$ for soleus i.e. knee flexed, effect size $r = 0.68$ $CI_{95\%}$ [0.55,0.81] $p < 0.001$ for GSC i.e. knee extended). Average X_{V1} for soleus and GSC muscles was negatively correlated with patient age ($\rho = -0.34$ $CI_{95\%}$ [80.67,0.00] p 0.061, Fig. 1B) but not on the non-paretic side (Fig. 1A). Compared to age-matched values from typi- cally developing children (Mudge et al., 2014), the soleus was lessextensible on both sides (X_{V1} knee flexed) but only on the paretic side forthe GSC (X_{V1} knee extended).

Average stretch response angle for soleus and GSC (X_{V3}) strongly and positively correlated with average extensibility (X_{V1}) (ρ 0.5 $CI_{95\%}$ [0.23,0.79] p 0.004, Table 2 and Sup. Fig. 2). Stretch response angle was not significantly correlated with patient age although there was a negative trend (Table 2 and Fig. 1C).

Data regarding the active range of dorsiflexion (X_A) was obtained in 11 children only (n = 11, 5 girls, mean[range] age: 3.7[2.5–6], height: 0.98 m[0.83–1.12], weight: 15 kg (Willerslev-Olsen et al., 2018; Noble et al., 2014; Willerslev-Olsen et al., 2013; Hägglund and Wagner, 2011; Gracies et al., 2010; Boulay et al., 2012; Keith, 1928; Young et al., 1990; Yelnik et al., 2010; Mudge et al., 2014; Hof, 1996; R Core Team. R, 2014). X_A in the knee flexed position was positively and strongly correlated with soleus extensibility, X_{V1} (n = 11, $\rho = 0.70$ $CI_{95\%}$ [0.32,1.17] $p = 0.017$). However, there was no correlation between X_A and X_{V1} with knee extended.

No clinical examination variable had a significant correlation with non-dimensional speed.

Electromyographic data could not be collected in one child (n 30). An average of 97 gait cycles were collected and analysed (range:40–153). All three muscles were active during the swing phase (Fig. 2), which is contrary to normal gait for GM and PL (Perry et al., 1992). Activation % of the GM was smaller than that of the PL during theentire swing phase (r 0.46 $CI_{95\%}$ [80.73, 0.19] p 0.011). Most ofthe difference occurred in the first two thirds of the swing phase, with non-significant difference in the last third of the swing phase (Fig. 2).

Activation percentages of the GM and PL in swing both negatively correlated with age ($\rho = -0.59$ $CI_{95\%}$ [−0.89, −0.34] $p < 0.001$ for GM and $\rho = -0.42$ $CI_{95\%}$ [−0.86, −0.07] p = 0.02 for PL, Table 2). GM activation percentage in swing positively correlated with extensibility of the soleus and GSC ($\rho = 0.41$ $CI_{95\%}$ [0.06,0.83] $p = 0.023$ with X_{V1} knee flexed, $\rho = 0.48$ $CI_{95\%}$ [0.15,0.80] p = 0.007 with X_{V1} knee extended, Table 2). However, after adjusting for age (Liu et al., 2018) only GM activation and GSC extensibility remained correlated (ρ 0.40 $CI_{95\%}$ [0.05,0.65] p 0.026).

Activation percentage of the TA in swing was large (Fig. 2) alongside that of the GM and PL muscles (median(IQR) was 81%(40%) for TA,

Table 1

A) Clinical characteristics. B) Clinical examination results. X_{V3} was not measured for the NP side (no spasticity) and only X_{V1} was available for the TD group. * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$, the effect size (r) and [CI_{95}] is mentioned if $p < 0.05$C) Activation percentage in swing phase (SW, SW_1, SW_2, SW_3) of gastrocnemius medialis (GM), peroneus longus (PL) and tibialis anterior (TA). X_{V1} in age matched typically developing children was defined as 119° (knee flexed) and 112.5° (knee extended) (Mudge et al., 2014).

(A) Clinical characteristics

	Subjects	*Paretic side*	*Gender*
	n = 31	20 R/11L	20M/11F

	Age (years	*Weight (Kg)*	*Height (m)*	*BMI (Kg/m^2)*	*Gait velocity (m/s)*	*Non-dimensional speed*
Median	2.89	14.00	0.95	14.88	0.73	0.23
IQR	1.11	2.87	0.10	1.77	0.24	0.08
Min	2.00	9.00	0.79	11.11	0.20	0.06
Max	6.00	22.00	1.12	21.97	1.10	0.36

(B) Clinical examination results

	Knee Flexed						*Knee extended*					
	Non Paretic		Paretic				Non Paretic		Paretic			
degree	X_{V1}	X_A	X_{V1}	X_{V3}	X_{V1}-X_{V3}	X_A	X_{V1}	X_A	X_{V1}	X_{V3}	X_{V1}-X_{V3}	X_A
n	31	11	31	31	31	11	31	11	31	31	31	11
Median	120	110	110	100	10	95	110	100	100	90	15	80
IQR	5	13	13	5	10	8	8	8	10	10	8	8
Min	105	105	100	90	0	90	100	80	95	70	70	70
Max	135	125	125	120	25	105	125	115	120	115	100	100
NP vs P			**** 0.73 [0.64,0.81]*			*** 0.89 [0.87,0.90]*			**** 0.68 [0.55,0.81]*			*** 0.89 [0.87,0.91]*
vs TD	*p = 0.898*		**** 0.70 [0.53,0.88]*				*** 0.56 [0.26,0.86]*		**** 0.83 [0.72,0.93]*			

(C) Activation % in swing phase (SW, SW1, SW2, SW3) of gastrocnemius medialis (GM), peroneus longus (PL) and tibialis anterior (TA), strides number mean: 153, SD: 98

n = 30	GM paretic				PL paretic				TA paretic			
	SW	SW_1	SW_2	SW_3	SW	SW_1	SW_2	SW_3	SW	SW_1	SW_2	SW_3
Median (%)	43%	25%	26%	62%	59%	38%	48%	75%	81%	107%	56%	47%
IQR (%)	26%	14%	17%	37%	32%	21%	25%	46%	40%	57%	39%	30%
Min (%)	19%	7%	8%	29%	19%	13%	18%	17%	37%	30%	28%	23%
Max (%)	98%	51%	65%	154%	109%	73%	95%	141%	195%	256%	167%	157%

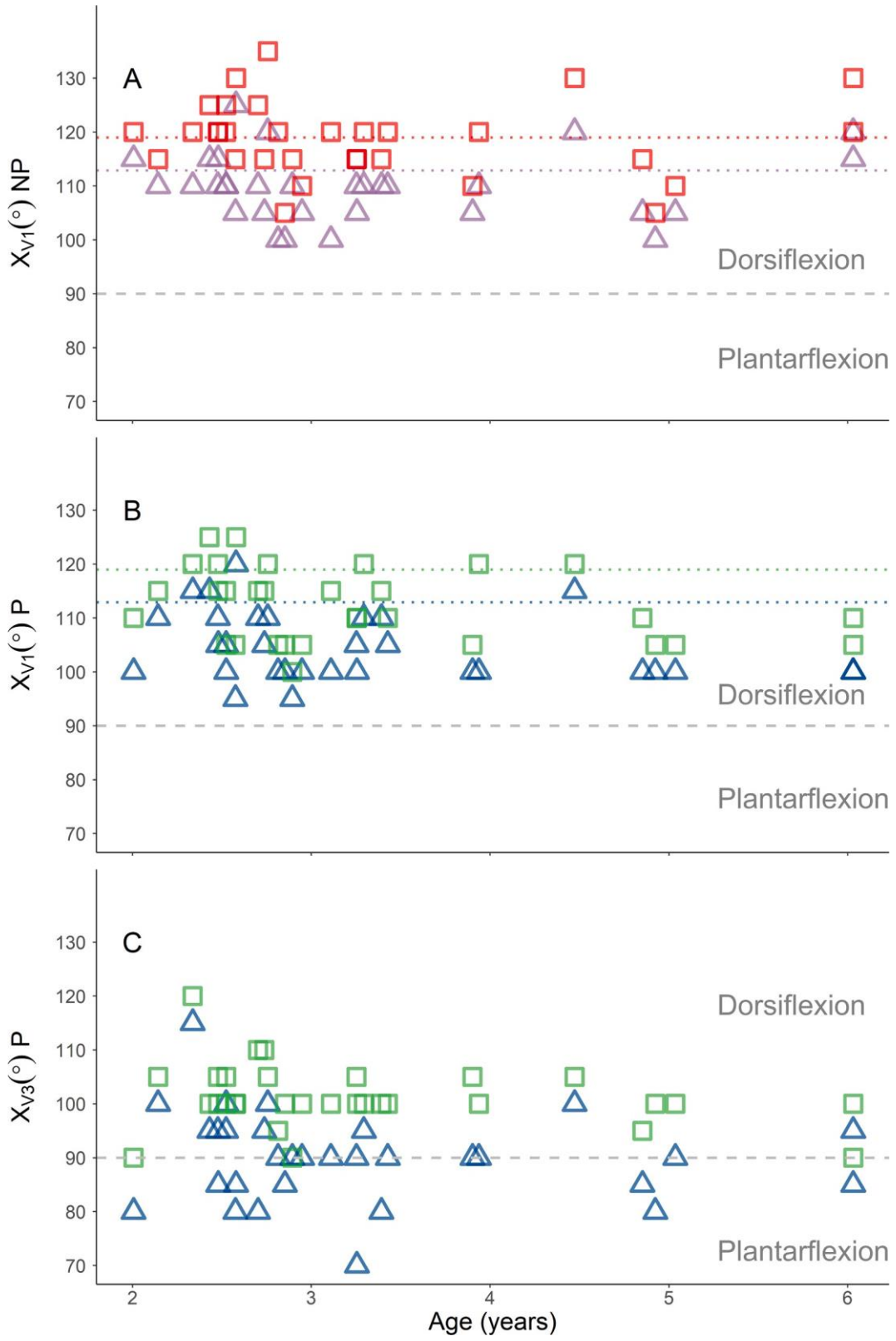

Fig. 1. Triceps surae extensibility and passive stretch reflex with age. (A) Extensibility (X_{V1}) of the gastrocnemius-soleus complex (purple triangles) and in the soleus muscle (red squares) on the non-paretic side. (B) Extensibility (X_{V1}) of the gastrocnemius soleus complex (blue triangles) and in the soleus muscle (green squares) on the paretic side. (C) Threshold for the spastic stretch response X_{V3} of the gastrocnemius soleus complex (blue triangles) and in the soleus muscle (green squares) on the paretic side. The dashed lines correspond to plantar grade = 90° and the dotted lines (A and B) correspond to the values from typically developing children aged 4–7 years old (Mudge et al., 2014). The Spearman correlation coefficients were (A) extensibility non-paretic side, gastrocnemius soleus complex (GSC), ρ = — 0.18 (p = 0.34, *n* = 31); soleus, ρ = — 0.22 (p = 0.22, *n* = 31) (B) extensibility paretic side, GSC: ρ = — 0.35 (p = 0.054, *n* = 31); soleus, ρ = — 0.33 (p = 0.07, *n* = 31); (C) high speed extensibility paretic side, GSC: ρ = — 0.21 (p = 0.25, *n* = 31); soleus: ρ = -0.27 (p = 0.14, *n* = 31).

Table 2
Correlations between age, non-dimensional walking speed, clinical examination parameters and activation % of the gastrocnemius medialis, peroneus longus and tibialis anterior in swing (paretic side). XV1, XV3, and XA were measured knee flexed (KF) or knee extended (KE). Spearman rank correlation * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$, Partial Spearman's rank correlation, adjusting for age † $p < 0.05$, only performed for the line corresponding to GM activation in swing.

	Age	ND Speed	$X_{V1\ KF}$	$X_{V1\ KE}$	$X_{V3\ KF}$	$X_{V3\ KE}$	$X_{A\ KF}$	$X_{A\ KE}$	GM % in swing	PL % in swing
ND Speed	0.23									
$X_{V1\ KF}$	−0.33	−0.19								
$X_{V1\ KE}$	−0.35	−0.33	**0.84** ***							
$X_{V3\ KF}$	−0.27	−0.19	**0.40** *	**0.54** **						
$X_{V3\ KE}$	−0.21	−0.12	**0.38** *	**0.41** *	**0.51** **					
$X_{A\ KF}$	0.14	0.44	**0.70** *	0.49	−0.1	0.21				
$X_{A\ KE}$	0.42	−0.3	0.14	0.03	0.04	0.6	0.48			
GM % in swing	**0.59** ***	−0.16	**0.41** *	**0.48** **	0.18	0.09	0.34	−0.24		
PL % in swing	**0.42** *	−0.04	0.28	0.25	0.36	0.32	−0.44	−0.25	**0.42** *	
TA % in swing	−0.3	−0.2	0.3	0.35	**0.45** *	0.27	−0.08	−0.25	**0.58** ***	**0.62** ***

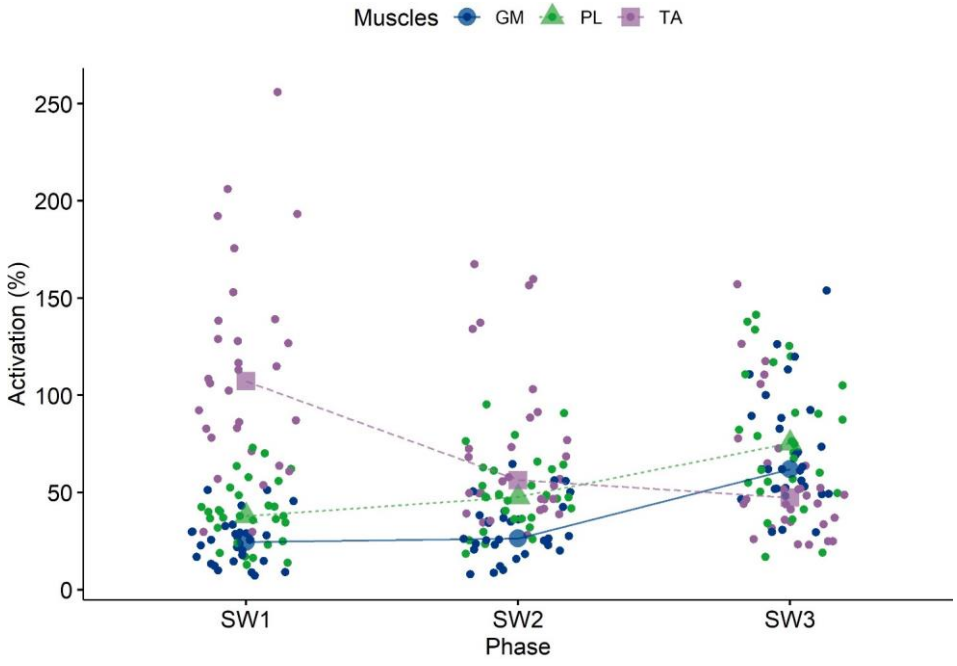

Fig. 2. Activation percentages (i.e. compared to a reference exercise) during the three parts of the swing phase (SW1, SW2, SW3) on the paretic side for gastrocnemius medialis (GM), peroneus longus (PL) and tibialis anterior (TA). The reference exercise was standing on tiptoes for GM and PL, and active ankle dorsiflexion while standing for TA. The mean and the distribution of individual data points are presented for each part.

43%(26%) for GM and 59%(32%) for PL, Table 1). Activation percentage of TA was also positively correlated with activation % of the GM and PL muscles (Table 2) which indicates a high level of co-contraction in swing. Contrary to the GM and PL, activation percentage of TA in swing had not correlation with age.

4. Discussion

Children with cerebral palsy present with primary neurological disorders accompanied by secondary musculo-skeletal disorders that may worsen over time. Clinical examination measurements may be performed routinely to quantify the neurological and musculo-skeletal disorders and monitor change with age or with treatments. In particular, the Five Step Assessment of spastic paresis utilizes measurements designed to estimate maximum passive stretch of the plantar flexors, termed extensibility (X_{V1}), the threshold of the spastic stretch reflex of the plantar flexors (X_{V3}) and the balance between the force generated by the activation of the dorsiflexors and the passive and active (because of co-contraction) resistance of the plantar flexors (X_A). We investigated these three measurements as well as surface electromyography of the tibialis anterior, gastrocnemius medialis, and peroneus longus during gait in a cohort of young children (median age: 2.8, IQR: 1.1, min:2, max:6) with unilateral spastic cerebral palsy presenting with equinovalgus gait.

Our first aim was to investigate the prominence of the neurological and muscle disorders in this young, cohort before any botulinum toxin or surgery. We found that both soleus and gastrocnemius-soleus complex (GSC) had reduced extensibility (X_{V1}) on the paretic side compared to the paired non-paretic side (strong effect size r between 0.63 and 0.89 depending on the specific variable) and compared to typically developing peers (strong effect size r=0.70 and 0.83). On the non-paretic side, the GSC extensibility was significantly smaller than typically developing peers (moderate effect size r 0.56, p=0.002). Extensibility tended to decrease with age on the paretic side but not on the non-paretic side (Fig. 1). Overall, these results indicate altered extensibility for both the GSC and the soleus at an early age.

We found that the threshold for the spastic stretch response (X_{V3}) correlated with extensibility (ρ=0.5, p =0.004), whereby less extensible muscles experienced lower thresholds for the stretch reflex. This result illustrates what we term spastic myopathy, a relationship between altered muscle properties, in particular reduced extensibility, and stretch-sensitive forms of muscle overactivity including spastic stretch

reflexes (Gracies, 2015). This mechanism may be due to an increased sensitivity of the spindles intertwined with stretched and/or stiffer muscle fibres (Maier et al., 1972). The relationship between the threshold of the spastic stretch response and age was not significant, although a negative trend may exist ($\rho = -0.26$, $p = 0.16$, Fig. 1). These results are in broad agreement with those of Linden et al. who, in a longitudinal study, observed an increase of spasticity with age until 5 years old followed by a decrease after 5 years old (Lindén et al., 2019).

The X_A measurement is informative to assess the capacity of ankle dorsiflexor activation to overcome passive and active resistance from the plantar flexors; however, this assessment was difficult to obtain in many of these young children. We were able to collect the measurement on one third of the children only. The main issue was difficulty to perform the active dorsiflexion and poor selective motor control, i.e. large co-contraction of GM/PL with TA, especially in the knee extended position (see supplementary video). In the knee flexed position, X_A positively and strongly correlated with soleus extensibility ($\rho = 0.70$, $p = 0.017$), strongly suggesting that shorter soleus muscle produced excessive resistance to the active dorsiflexion of the ankle by TA and thus determined active movement capacities for a large part. However, we found no correlation between X_A and GSC extensibility. It may be that increased inhibition of TA due to stretched GSC muscles (i.e. stretch-sensitive paresis) prevent dorsiflexion of the ankle (Vinti et al., 2015) or that the measurement of X_A was not reliable because active dorsiflexion with the knee extended was too difficult to perform for these young children. In addition, the smaller sample size for the X_A variable reduced statistical power.

Overall, the correlations between the three clinical measurements highlight the bidirectional relationship between the neurological and muscle disorders, a vicious cycle whereby the original neurological disorder causes muscle adaptations (Willerslev-Olsen et al., 2018) that, in turn, aggravate the neurological disorder (Gracies, 2005).

Our second aim was to investigate the role of the peroneus longus (PL) in equinovalgus foot contact during gait. We found large activation percentages of both the PL and the gastrocnemius medialis (GM) during the swing phase (median: 58% IQR: 32% for PL and median: 43% IQR: 25% for GM), where PL was more activated than the GM (effect size $r = 0.46$, $p = 0.011$), especially during the first two thirds of the swing phase.

These activation percentages are inappropriate since plantar flexors are not active during swing in normal gait (Perry et al., 1992). Only minimal inertial forces are applied to the swinging foot so there is no biomechanical role for the plantar flexors during swing. Patterned activation of the plantar flexors due to the activation of other muscles during swing (i.e. lack of selective motor control) may be one explanation for our observations. We found that activation percentage of TA was also large in swing (Fig. 2). Children in this cohort thus displayed high levels of co-contraction of ankle agonist (TA)-antagonist (GM and PL) muscles during swing. Lorentzen et al. argued that co-contraction may be a beneficial neurological behaviour in supporting ankle stability until maturation of the feedforward neural control of toe-walking (i.e. equinus gait) which is impaired in children with cerebral palsy (Lorentzen et al., 20192019). However, during the swing phase of gait the foot is not loaded, and joint stability may not be as critical as the ability to clear the ground by dorsiflexing the foot. In this cohort of young children, negative correlation between GM/PL activation in swing and age may thus indicate some improvement in selective motor control with age. One explanation why activation was maximal in the last third of the swing phase might be pre-activation of the plantar flexors before landing in equinus (Fig. 2). Another explanation may be increased motoneuronal excitability of plantar flexors related to the gastrocnemius stretch that occurs as the knee re-extends at late swing and the related excitatory afferent discharges (Marque et al., 2001).

We believe that large activation of TA in the first third of the swing phase (median: 107%, IQR: 57%) may be an attempt by the nervous system to compensate for passive resistance from and co-contraction of the plantar flexors rather than the other way around (increased co-contraction due to increased activation of TA).

This study has limitations. We lacked concurrent three-dimensional gait analysis data to discuss further the relationship between muscle activation timing and kinematics at the knee, ankle, and foot. We took a pragmatic approach to calculating muscle activation percentages given the difficulty in recording maximum voluntary contraction reference EMG in young children with cerebral palsy. Some children displayed activation percentages of the GM and PL muscles during swing larger than 100% (max: 154% for GM and 141% for PL, both in SW3) which indicates that our reference exercise (standing on tiptoes) did not always elicit maximum voluntary contraction. This was also the case for TA, with maximum activation of 256% in SW1 for one child (Fig. 2) compared to the activation during maximum ankle dorsiflexion while standing. We note that the knee is flexed, and flexing, in SW1 while the knee was extended in our reference exercise for TA. The fact that activation percentages were often greater than 100% for TA in SW1 further illustrate the reduced maximum activation (or inhibition) of TA with the knee extended position, as in our reference exercise (i.e. with increased stretch of the triceps surae) compared to the knee flexed position.

5. Conclusion

This study found age-related changes in opposite directions for muscle and neurological disorders in a cohort of young children with USCP and equinovalgus gait between two and six years of age. Reduced plantar flexor extensibility on the paretic side worsened with age, while inappropriate plantar flexor co-contraction during swing improved with age.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Appendix A. Supplementary data

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