Journal of Electromyography and Kinesiology 49 (2019) 102353

Contents lists available at ScienceDirect

Journal of Electromyography and Kinesiology

journal homepage: www.elsevier.com/locate/jelekin

Reliability outcomes and inter-limb differences in ankle joint stiffness in children with unilateral cerebral palsy depend on the method of analysis

C. Boulard[a,b,*], V. Gautheron[a,b], T. Lapole[a]

[a] Univ Lyon, UJM Saint-Etienne, Laboratoire Interuniversitaire de Biologie de la Motricité, EA 7424, F-42023 Saint-Etienne, France
[b] Department of Pediatrics Physical Medicine and Rehabilitation, University Hospital of Saint-Etienne, Faculty of Medicine, France

ARTICLE INFO

Keyword:
Cerebral palsy-ankle joint-stiffness-reliability

ABSTRACT

Children with cerebral palsy (CP) present increased passive ankle joint stiffness, measured as the slope of the torque-angle curve relationship. However, large discrepancies in results exist among studies, likely because of various methodologies used. The purpose of this study was to determine the influence of different calculation methods on the outcomes and their inter-session reliability in children with unilateral CP (UCP).

Thirteen children (mean age: 9.8 years) with spastic UCP underwent passive ankle mobilization at 2°/s on both legs using a dynamometer, on two occasions separated by one week. Passive ankle joint stiffness was calculated as the slope of the torque-angle curve using linear regression on three different relative ranges of torque (i.e. 30%–100%, 20–80% and 50–90% of maximal torque for method 1, 2 and 3, respectively) for both the paretic and non-paretic legs.

Inter-session reliability was significantly lower on paretic leg (mean CV = 13.8%, ICC = 0.62) when compared to non-paretic leg (mean CV = 6%, ICC = 0.85), and method 3 presented lower reliability outcomes (mean CV = 11.7%, ICC = 0.75) than methods 1 (mean CV = 7.5%, ICC = 0.78) and 2 (mean CV = 6.6%, ICC = 0.79). Paretic values (0.24 Nm/°) were not different from the non-paretic leg (0.25 Nm/°), although significantly higher when considering the same angular sector (0.18 Nm/°).

Passive ankle joint stiffness measurement can be reliably performed in children with UCP using method 1 and 2 while method 3 should be avoided. The non-paretic leg may be used for comparison with the paretic leg when taking into account differences in maximal dorsiflexion angle between legs.

ClinicalTrials.gov Identifier: NCT02960932.

1. Introduction

Cerebral Palsy (CP) refers to motor control alterations after brain damage occurring perinatally (Rosenbaum et al., 2007), and has a prevalence of 1.7 per 1000 live births in Europe (Sellier et al., 2016). The most common form of CP is spastic CP resulting from alterations to the pyramidal system that leads to a velocity-dependent increase in tonic stretch reflexes with exaggerated tendon jerks (Lance, 1980). Spastic CP is well known to induce several functional motor impairments, especially in lower limb muscles, that may appear during growth and alter quality of life. For instance, muscles contractures, defined a decrease in muscle belly length (Lieber and Fridén, 2018) associated with an increase in passive muscle stiffness (Farmer and James, 2001), have been frequently reported for plantarflexor (PF) muscles in spastic CP (Mathewson and Lieber, 2015, Švehlík et al., 2013). As a consequence, ankle joint range of motion has been reported to decrease dramatically during growth in children with CP (Hägglund and Wagner, 2011, Švehlík et al., 2013). Moreover, due to the important functional role of the PF muscles in vertical support and forward propulsion during walking (Perry et al., 1974), their alteration may induce gait impairments such as equinus gait (Wren et al., 2010, Švehlík et al., 2010). Equinus foot deformity is one of the most common problems leading to gait and functional limitations in children with CP (Goldstein and Harper 2001, Švehlík et al., 2010, Wren et al., 2010).

Passive ankle joint stiffness (PAJS) has been classically assessed by establishing the torque-angle relationship during passive dorsiflexion (DF) which may be considered as the ‘gold standard’ for mechanical assessment of muscle-tendon properties (Moseley et al., 2001, Harlaar et al., 2000, Tardieu et al., 1982). PAJS is then calculated as the slope of the torque-angle relationship using a linear fit (Alhusaini et al., 2010,

* Corresponding author at: UJM Saint-Etienne, Laboratoire interuniversitaire de biologie de la motricité, 10 chemin de la Marandinière 42270 Saint-Priest En Jarez, France.

E-mail address: clement.boulard@univ-st-etienne.fr (C. Boulard).

https://doi.org/10.1016/j.jelekin.2019.102353
Received 7 April 2019; Received in revised form 23 July 2019; Accepted 22 August 2019
1050-6411/

Barber et al., 2011, Kruse et al., 2017, Ross et al., 2011, Theis et al., 2016). This has led to reports of increased PAJS in children with CP when compared to typically developing (TD) peers (Alhusaini et al., 2010, Barber et al., 2011, de Gooijer-van de Groep et al., 2013, Ross et al., 2011), although others did not evidence differences between groups (Kruse et al., 2017, Ross et al., 2011). Moreover, large discrepancies have been reported for the magnitude of increased stiffness in children with CP among studies, ranging from 35% (Ross et al., 2011) to 225% (Alhusaini et al., 2010). Such discrepancies could be due to differences in methodology and/or heterogeneity of the sample of tested children.

First, the choice of the angular sector for slope calculation has been reported to largely influence the results and their interpretation (Ross et al., 2011). Since all the aforementioned studies used different angular [i.e. 0–5° for Alhusaini et al. (2010), −30 to −10° and last 20° of DF for Ross et al. (2011)] or torque [30–100% for Barber et al. (2011), 20–80% for Theis et al. (2015, 2016) and 50–90% of maximal passive torque for Kruse et al. (2017)] sectors for slope calculation, comparison between studies may be hazardous and clear recommendations should be provided to ensure accuracy of future studies. Here, we propose to investigate the influence of PAJS calculation method on reliability outcomes and interpretation that can be made from the results. We believe that this may help clinical decision making such as to perform either conservative (e.g. casting, splinting, stretching) or surgical treatment.

Secondly, most studies included children with both bilateral and unilateral CP (UCP) notwithstanding that the rate of PF growth and strength differs among them (Barber et al., 2016, Wiley and Damiano, 1998). Moreover, children with UCP are more prone to developing fixed equinus deformity during growth because of a frequent combination of a true equinus pattern (Goldstein et al., 2001, Winters et al., 1987) and dorsiflexors muscle weakness (Cobeljic et al., 2009). Altogether, this may potentially influence passive stiffness. Accordingly, it would be of interest to specifically investigate PAJS in children with UCP in a clinical routine. Comparing the paretic and non-paretic legs could be of high interest for the clinician in a longitudinal follow-up during growth and to evaluate the impact of therapeutic strategies such as stretching program (Theis et al., 2015), home based exercises program (Kruse et al., 2018) or gait training (Hösl et al., 2018; Willerslev-Olsen et al., 2015). Before that, it is crucial to characterize the reliability of the measurement.

Therefore, the purpose of this study was to compare in children with UCP paretic and non-paretic legs PAJS and to further determine the influence of different calculation methods on the outcomes and their inter-session reliability. Because of differences in ankle range of motion during passive stretch between both paretic and non-paretic legs, increased stiffness may occur earlier in the range of absolute stretch for the spastic than non-spastic leg (Ross et al., 2011). An additional aim of this study was to further investigate the inter-limb difference when stiffness is calculated on the same absolute or relative angular sector.

2. Method

Thirteen children (7 females, 6 males) with spastic UCP (Table 1) participated in this study, which was approved by the local ethics committee (CPP Sud-Est, number 2016-A01558-43) and conducted according to the declaration of Helsinki. Parents and children gave their written informed consent prior to inclusion in the study. Children (aged 5–12 years) with spastic UCP (i.e. Tardieu scale ≥ X2, V2) were recruited provided they were able to understand and follow simple verbal instructions, had no botulinum toxin injections or cast applications in the last three months, and had less than 10° of knee flexion contracture. Children were excluded if they had any surgical procedures performed on the lower limbs.

Each child was tested twice at a 1-week interval and at the same time of the day by the same experimental investigator. Paretic and non-paretic limbs were assessed in a random order in the first session and the same order was kept for the second session.

Table 1
General characteristics of the tested group; Data are presented as mean (SD) *(Nb: number, GMFCS: Gross Motor Function Classification System, DF: dorsiflexion).*

n	13
Sex (M/F)	6/7
Paretic side (right/left)	6/7
Age (Y:Mo)	9.8 (1.8)
Weight (kg)	32.6 (9.6)
Height (cm)	137.8 (13.5)
Tibia length on paretic leg (cm)	32.9 (4)
Tibia length on non-paretic leg (cm)	33.9 (4.1)
GMFCS Level, n	I; n = 13
Modified Ashworth Scale	2.46 (0.77)
Tardieu Scale	1.2 (0.86)
Boyd scale	1.23 (0.44)
Maximum DF angle paretic side (°)	2.1 (6.5)
Maximum DF angle non-paretic side (°)	12.7 (3.8)
Nb Botulinum toxin injection	6.5 (2)
Nb weeks ankle casting	6.5 (5.6)
Nb session physiotherapy per week	1.5 (0.6)
Night splinting (Yes/No)	11/2
Gait orthoses (Yes/No)	10/3

After determining their Gross Motor Function Classification System (GMFCS) level, spasticity level on soleus and gastrocnemius muscles on the paretic limb was assessed with the Modified Ashworth and Tardieu Scales, and selective motor control was assessed with the Boyd scale. Passive maximal DF angle was then measured manually for both limbs with the knee extended and in supine position using a manual goniometer. After that, children were installed in the prone position (with hip and knee at 0° extension (i.e. fully extended) and with the ankle joint positioned at 25° of ankle PF) on an isokinetic dynamometer (Con-Trex®, Medimex, France). The axis of the dynamometer was aligned with the presumed axis of rotation of the ankle. The foot was firmly fixed to the dynamometer and the thigh was fixed to the dynamometer with a Velcro™ attachment. Experimental condition of ankle joint positioning was presented Fig. 1. Torque-angle relationships were then recorded during passive DF performed at 2°/s between 25° of ankle PF and the pre-determined maximal DF angle of the ankle. To ensure that muscles were totally relaxed during passive movements, EMG activity was recorded using dry-surface electrodes (Delsys DE 2.1, Delsys Inc, Boston, MA, USA) placed on the gastrocnemius lateralis and tibialis anterior muscles. Sampling frequency for angle, torque and EMG signals was 2000 Hz. A total of 5 passive loading/unloading cycles was performed to account for a possible conditioning effect (Maganaris, 2003) and the last loading cycle (i.e. DF phase) was retained for joint stiffness analysis since no significant changes have been reported to occur between the fourth and the fifth cycles (Maïsetti et al., 2012).

Considering the variety of methodological approach used in the literature to calculate PAJS, stiffness was considered in the present study as the slope of the linear regression between passive torque and ankle angle in the range between (1) 30–100% (method 1, Barber et al., 2011), (2) 20–80% (method 2, Theis et al., 2015 and 2016) and (3) 50–90% of maximal passive torque (method 3, Kruse et al., 2017) as previously performed by others. To account for differences in range of motion between paretic and non-paretic legs, PAJS was calculated for the non-paretic leg not only on the relative range defined above ($PAJS_{rel}$) but also on the same absolute angular sector as for the paretic one ($PAJS_{abs}$).

2.1. Statistical analysis

Coefficients of determination (R^2) of the torque-angle relationships were calculated for linear fitting. As recommended by Hopkins (Hopkins, 2000), inter session reliability of the PAJS were assessed for

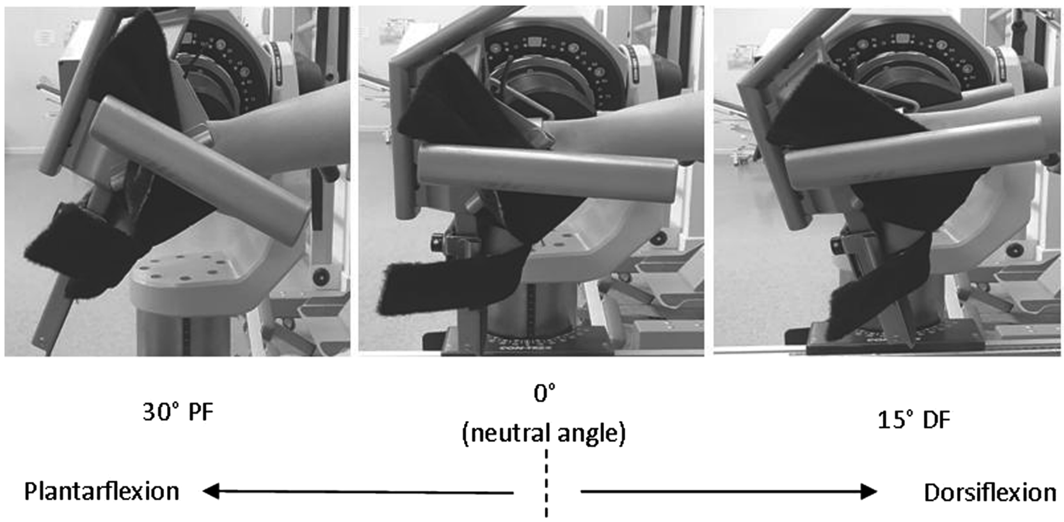

Fig. 1. Experimental condition of ankle positioning on the isokinetic dynamometer (PF: plantarflexion, DF: dorsiflexion).

each method (i.e. 30–100%, 20–80% and 50–90%) and both legs (i.e. non-paretic leg and paretic leg on both the same absolute and relative angular sector as for the paretic one) using the intra-class correlation coefficient (ICC), standard error of the mean (SEM) and coefficient of variation (CV). The ICC provides a measure of relative reliability showing the degree to which individuals maintain their position in a sample with repeated measurements (Hopkins, 2000). According to Fleiss (Fleiss, 1981), ICC > 0.75 represents excellent reliability; an ICC between 0.74 and 0.40 represents moderate to good reliability and an ICC < 0.40 represents poor reliability. The SEM scores were also calculated to quantify absolute reliability. Thus, a low SEM value means strong reliability and vice-versa (Hopkins, 2000). The CV (standard deviation (SD) / mean) was calculated to represent the degree to which repeated measurements vary for individuals (Hopkins, 2000). Reliability was considered as good for CV < 10% and moderate for CV 10–20%. All variables passed the normality test (Kolmogorov–Smirnov normality test) and homogeneity of variance verification (Levene's test). A paired *t*-test was used to compare maximal DF angle between paretic and non-paretic legs. Three-way ANOVAs were performed to determine the effect of slope calculation method (i.e. method 1, 2 and 3), sessions (i.e. 1 and 2) and tested limb (i.e. paretic leg, non-paretic leg, and non-paretic leg within the same absolute angular sector as for the paretic leg) for PAJS as well as coefficients of determination (R2) of the torque-angle relationships. Two-ways ANOVAs were performed to determine the effect of slope calculation method and tested limb for reliability outcomes (CV). When the ANOVA identified significant effects, post-hoc Fisher testing was performed. Data are presented as mean ± SD. Statistical significance was set at $p < 0.05$.

3. Results

The characteristics of the population are shown in Table 1. Children with UCP presented significantly decreased maximal ankle DF angle ($p < 0.0001$) (Table 1).

Averaged torque-angle relationships obtained for the whole group during both first and second testing sessions are presented for the paretic legs in Fig. 2.

Relationships were well fitted by the linear regression. Mean R^2 ranged between 0.94 (i.e. paretic limb, session 1, 50–90%) and 0.97 (i.e. non-paretic leg within the same absolute angular sector as for the paretic leg, session 2, 20–80%). There were no main effect of session ($p = 0.3$) or tested limb ($p = 0.15$). However, there was a main effect of the calculation method ($p < 0.001$) with post-hoc testing only revealing a trend for greater R^2 for method 2 (mean R^2 of 0.97) than method 3 (mean R^2 of 0.95) ($p = 0.077$). Moreover, there was a method × tested limb interaction effect ($p < 0.0001$). Post-hoc testing demonstrated higher R^2 value for method 2 (mean R^2 of 0.96) when compared to method 3 (mean R^2 of 0.94) on the paretic leg ($p < 0.001$). Similarly, higher values were reported for method 1 and 2 (mean R^2 of 0.97 for both methods) when compared to method 3 (mean R^2 of 0.94) on the non-paretic leg within the same absolute angular sector as for the paretic leg ($p < 0.001$ and 0.0001, respectively).

Using method 2, Fig. 3 illustrates for the whole group the comparison of $PAJS_{rel}$ and $PAJS_{abs}$ measurements between paretic and non-paretic legs. Table 2 presents stiffness measurement values as well as their inter-session reliability outcomes.

When considering PAJS values, the three-ways ANOVA (limbs × sessions × methods) revealed:

- a significant main effect of method ($p < 0.0001$): post-hoc testing revealed significantly lower PAJS values using method 2 (mean stiffness of 0.19 Nm/°) when compared to method 1 (mean stiffness of 0.23 Nm/°) ($p = 0.009$) and to method 3 (mean stiffness of 0.26 Nm/°) ($p < 0.0001$) while the difference was almost significant between the latter two ($p = 0.06$).
- a significant method × tested limb interaction effect ($p = 0.009$): post-hoc testing revealed that stiffness values were significantly higher for the paretic leg when compared to non-paretic PAJSabs whatever the method used for calculation ($p < 0.001$ with method 1, $p = 0.007$ with method 2, $p < 0.0001$ with method 3). No significant difference was found between paretic leg and non-paretic leg PAJSrel values whatever the method used for calculation ($p = 0.69$, 0.86 and 0.40 with method 1, 2 and 3, respectively).
- no interaction effect involving the factor session (session × method: $p = 0.56$; sessions × limbs × methods: $p = 0.42$).

Regarding the inter-session CVs, two-ways ANOVA revealed significant main effect of method ($p = 0.001$). Post-hoc testing demonstrated significantly higher CV values for method 3 (mean CV of 11.7%) when compared to method 2 (mean CV of 6.6%) ($p = 0.03$), and a non-significant difference when compared to method 1 (mean CV of 7.5%) ($p = 0.07$). CVs were not different between methods 1 and 2 ($p = 0.69$). Two-ways ANOVA also revealed a significant main effect of the tested limb ($p < 0.001$). Higher CV values were reported for the paretic limb (mean CV of 13.8%) when compared to the non-paretic one (mean CV of 7%) ($p = 0.04$) and to the non-paretic leg within the same absolute angular sector as for the paretic leg (mean CV of 5%) ($p = 0.01$). There was no difference between CV values of the latter two

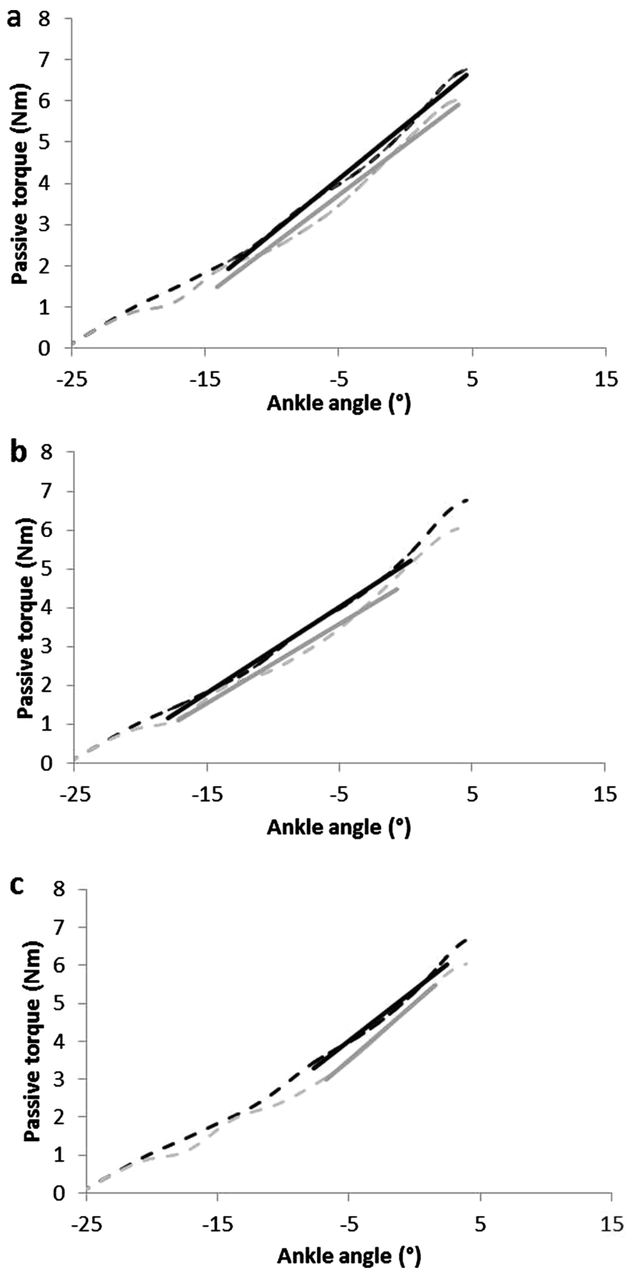

Fig. 2. Averaged torque-angle relationships between 25° of plantarflexion and maximal dorsiflexion angle on the paretic leg for both first (black line) and second (grey line) testing sessions. Ankle joint passive stiffness (dashed lines) was calculated as the slope of the torque-angle curve from 30% to 100% (a), 20% to 80% (b) and 50 to 90% (c) of maximal passive torque.

($p = 0.53$). Finally, there was no significant method × tested limb interaction effect ($p = 0.08$).

4. Discussion

In the present study, we investigated in children with UCP inter-limb difference in PAJS through torque-angle curve measurements. We further investigated how the methodological approach (i.e. the selected angular sector for stiffness calculation) may influence PAJS values of both limbs as well as their reliability outcomes. Our results suggest that the three tested method for slope calculation (i.e. 30–100%, 20–80% and 50–90% of maximal passive torque for methods 1, 2 and 3, respectively) led to similar results. For instance, there was no inter-limb difference when considering $PAJS_{rel}$ while values were greater for the paretic limb when compared to non-paretic $PAJS_{abs}$. Moreover, measurements were reported to be reliable, although lower reliability was observed for the third method as well as for the paretic limb.

All the tested method for slope calculation were well fitted by a linear fit, whatever the considered limb (R^2 ranged from 0.94 to 0.97). This confirms that linear regression is appropriate to calculate PAJS as the slope of the torque-angle curve. Yet, method 3 was significantly less fitted by the linear regression. Moreover, we report mean PAJS values in agreement with previous reports in children with CP with similar characteristics than in the present study (Ross et al., 2011).

Based on CV and ICC outcomes, PAJS reliability was considered as good for the non-paretic leg whatever the method used for calculation (mean CV = 7% and 5%, mean ICC = 0.84 and 0.86 for $PAJS_{rel}$ and $PAJS_{abs}$, respectively) and moderate for the paretic leg (mean CV = 13.8% and mean ICC = 0.62). For instance, we reported significant higher CV values for the paretic leg, in agreement with findings on adults with hemiplegia (Harlaar et al., 2000). While this may be the result of spasticity (Harlaar et al., 2000), we used a very low angular velocity (2°/s) to avoid reflex activation. While the reason for such difference in reliability between limbs remains unknown, this may be the consequence of misalignment of the axes of the ankle and dynamometer and/or motion of the knee during testing (Barber et al., 2011). These methodological considerations may have predominantly affected measurements on the paretic leg because of its likely smaller Achilles tendon moment arm (Kalkman et al., 2017) and foot size. Consequently, correct ankle joint positioning and correct alignment of the dynamometer were complicated on the paretic leg. Nonetheless, despite lower reliability was found on the paretic leg, we reported CV values (12.4%, 9.7% and 19.2% for method 1, 2 and 3 respectively, table 1) largely lower than the decreased ankle joint stiffness reported after interventions such as long-term stretching program (−832%, Theis et al., 2015), or gait training (~40%, Willerslev-Olsen et al., 2015). This suggests that performing such measurements in the clinical field may still make sense to evaluate the benefits of various treatment strategies. Our results also demonstrated good inter-session reliability for methods 1 (mean value for CV = 7.5% and ICC = 0.78) and 2 (mean value for CV = 6.6% and ICC = 0.79) for all the tested limbs, as it was for method 3 on the non-paretic leg when considering both the absolute or relative angular sector (mean value for CV = 8% and ICC = 0.82). Conversely, lower and moderate reliability was found when using method 3 on the paretic leg (CV = 19.2% and ICC = 0.6).

Using a lower limit of 20% of maximal torque is thought to avoid the toe region at the lower end of the stiffness curve (Abellaneda et al., 2009). As such, the three tested methods should not have been influenced by this phenomenon. Moreover, an upper limit of 80% of maximal torque is thought to minimize the contribution of passive elastic structures such as ligaments, connectives tissues and skins at the upper end of the curve (Abelladena et al., 2009). EMG responses of PF muscles may also tend to rise significantly from 80% of maximal torque (Mcnair et al., 2001). Nonetheless, these influences may not explain the lower reliability observed for method 3 (50–90% of maximal torque) because reliability was still good for method 1 calculating slope of the torque-angle curve up to 100% of maximal torque. Alternatively, we suggest that the lower reliability may be the result of the restricted range of torque (i.e. 40% for method 3 vs 70% and 60% for methods 1 and 2, respectively) for slope calculation that may be more sensitive to inter-session variability, especially for the paretic leg. Taking together, we suggest to choose the method 2 (i.e. 20–80% of maximal torque) given that our findings present better reliability outcomes for the paretic leg and avoid the final portion of the torque-angle curve (i.e. last 20% of maximal torque).

To the best of our knowledge, this study is the first to compare PAJS between paretic and non-paretic legs in children with UCP. Whatever the method used for slope calculation, we found no difference in PAJS between paretic and non-paretic legs when considering the same relative angular sector. While this is to our knowledge the first study to

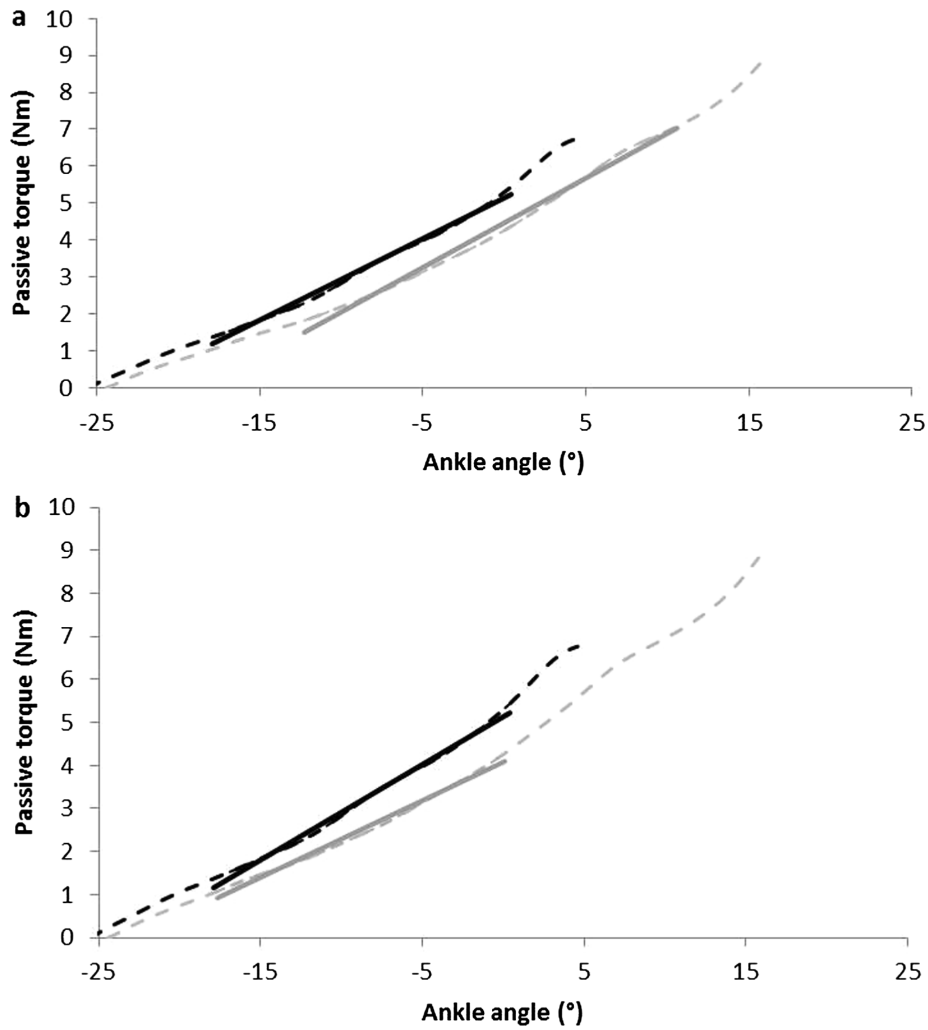

Fig. 3. Averaged torque-angle relationships between 25° plantarflexion and maximal dorsiflexion angle on the paretic (black line) and non-paretic (grey line) legs. Data from both testing sessions are averaged. Ankle joint passive stiffness was calculated as the slope of the torque-angle curve (dashed lines for paretic and non-paretic leg, respectively) from 20% to 80% of maximal passive torque in the same relative range of torque (a) and in the same absolute range of angle (b).

specifically compare both legs in children with UCP, this finding is in agreement with a recent study of Kruse et al. (2017) demonstrating using the third method we presented no difference in PAJS between children (mean age: 11.9 years) with CP and TD peers. Conversely, Barber et al. (2011) reported significant differences between CP young adults with CP (mean age: 18 years) and TD peers using method 1. Discrepancies between studies may likely be the consequence of differences in clinical status (e.g. CP severity) and age of participants. In

Table 2
inter-session reliability outcomes for passive ankle joint stiffness (PAJS, in Nm/°) on paretic leg ($PAJS_{rel}$) and non-paretic leg on its relative ($PAJS_{rel}$) and absolute ($PAJS_{abs}$) angular sector calculated with three different methods. *(ICC: intra-class correlation coefficient, CV: coefficient of variation, SEM: standard error of measurement).*

Inter-session reliability		Mean (SD)				
Method	Side leg	Session 1	Session 2	ICC	CV (%)	SEM
Method 1	**$PAJS_{rel}$ paretic leg**	0.24 (0.09)[d]	0.26 (0.06)[d]	0.57	12.41[b]	0.05
	$PAJS_{rel}$ non-paretic leg	0.25 (0.05)	0.25 (0.06)	0.84	6.5	0.02
	$PAJS_{abs}$ non-paretic leg	0.18 (0.003)	0.18 (0.03	0.93	3.48	0.01
Method 2	**$PAJS_{rel}$ paretic leg**	0.20 (0.06)[c,d]	0.21 (0.04)[c,d]	0.69	9.72[b]	0.03
	$PAJS_{rel}$ non-paretic leg	0.20 (0.03)[c]	0.20 (0.04)[c]	0.86	5.36	0.01
	$PAJS_{abs}$ non-paretic leg	0.15 (0.02)[c]	0.16 (0.02)[c]	0.83	4.59	0.01
Method 3	**$PAJS_{rel}$ paretic leg**	0.26 (0.13)[d]	0.30 (0.11)[d]	0.6	19.21[a,b]	0.07
	$PAJS_{rel}$ non-paretic leg	0.29 (0.06)	0.30 (0.08)	0.82	9.15[a]	0.03
	$PAJS_{abs}$ non-paretic leg	0.19 (0.06)	0.19 (0.04)	0.82	6.84[a]	0.02

[a] Significant higher CV compared to method 2.
[b] Significant higher CV compared to $PAJS_{rel}$ non-paretic leg and $PAJS_{abs}$ non-paretic leg.
[c] Significant lower PAJS values compared to method 1 and 3.
[d] Significant higher $PAJS_{rel}$ on the paretic leg compared to $PAJS_{abs}$ on the on-paretic leg.

the present study, lack of inter-limb difference in PAJS despite reduced range of motion in the paretic side is in agreement with the notion that non-paretic leg in children with UCP cannot be considered as an “healthy” leg (Wiley et al., 2008). It has accordingly been reported in hemiparetic post-stroke adults no inter-limb difference in PAJS while the paretic leg presented significantly higher passive stiffness when compared to TD adults (Freire et al., 2017). Moreover, this is consistent with findings of altered structural properties observed on both paretic and non-paretic medial gastrocnemius muscles in children with UCP when compared to TD Peers (Obst et al., 2017).

When taking into account inter-limb difference in maximal DF angle (i.e. by calculating slope of the torque-angle curves on a same absolute angular sector for both legs), PAJS was significantly increased in the paretic leg when compared to contralateral one whatever the method used. Our results demonstrated 37%, 28% and 45% inter-limb differences for method 1, 2 and 3 respectively. We assume that comparing both legs on the same angular sector on legs with significantly different maximal DF angle led to a comparison of different portions of the torque-angle curve. Consequently, the slope of the curve should be systematically higher on the limb with significantly lower maximal DF angle (Ross et al., 2011). Nonetheless, this method allowed us to quantify increased resistance to stretch of the PF muscle-tendon unit of the paretic leg when compared to the non-paretic one, just as the clinician would perceive it while manually mobilizing the ankle joint of both legs through the same angular range (Ross et al., 2011). This increased stretch resistance at a given ankle angle is further supported by the higher gastrocnemius medialis stiffness reported on the paretic leg when compared to the contralateral one at neutral angle (i.e. 0°) using Shear Wave Elastography in children with UCP (Lee et al., 2016). Such an inter-limb difference in PAJS could be due to a mismatch between bone and muscle length development in the paretic leg during growth (Pingel et al., 2017), with decreased gastrocnemius muscle length (Malaiya et al., 2007). Although PAJS does not necessarily reflect muscular (i.e. gastrocnemius and soleus) or tendinous (i.e. Achilles Tendon) tissues behaviors during passive mobilization in children with CP (Bar-On et al., 2018), musculo-tendinous unit, and especially muscle belly in children with CP, is considered as the primary contributor to joint passive stiffness (Blazevich, 2018, Lieber et al., 2017). Moreover, PAJS can be reliably measured using a simple dynamometer, as confirmed by the present study, while measurements at muscle and/or tendon level would require a complex experimentation design using ultrasonography in addition to dynamometry. Accordingly, PAJS may be measured in a clinical routine as a valuable and convenient parameter (1) to give useful objective parameters for clinical follow-up of children with CP during growth, and (2) to evaluate the impact of therapeutic strategies. This may be especially relevant for children with UCP who are more likely to develop ankle contracture and/or to be less responsive to botulinum toxin injection than children with bilateral CP (Hastings-Ison et al., 2016).

5. Conclusions

The present results suggest that the non-paretic leg in children with UCP may serve as a “control” limb during the clinical follow-up, provided that the comparison with the paretic leg is performed on the same absolute angular sector. We also suggest that PAJS should be assessed using method 2, i.e. 20–80% of maximal passive torque.

Declaration of Competing Interest

None.

Acknowledgements

Clément Boulard performed the planning, neurological examination and biomechanical measurement of the children. Clément Boulard and Thomas Lapole performed all analyses and discussion. Clément Boulard wrote the first draft of the manuscript, which was read and commented on by all the other authors.

This study was funded by several grants from ALLP (Association Lyonnaise de Logistique Post-Hospitalière, France), SFEHRE (Société Francophone d'Etude et de Recherche sur le Handicap de l'Enfant, France) and R4P (Réseau Régional de Rééducation et de Réadaptation en Rhône-Alpes, France), which are French scientific societies supporting research into childhood disabilities.

These foundations have no financial interest in the publication of the paper. The authors have stated that they had no interests which might be perceived as posing a conflict or bias. The authors thank Callum Brownstein for English revision of the manuscript and Rodolphe Testa for helpful in data processing.

References

Abellaneda, S., Guissard, N., Duchateau, J., 2009. The relative lengthening of the myotendinous structures in the medial gastrocnemius during passive stretching differs among individuals. J. Appl. Physiol. 106, 169–177. https://doi.org/10.1152/japplphysiol.90577.2008.

Alhusaini, A.A., Crosbie, J., Shepherd, R.B., Dean, C.M., Scheinberg, A., 2010. Mechanical properties of the plantarflexor musculotendinous unit during passive dorsiflexion in children with cerebral palsy compared with typically developing children: Calf Muscle Stiffness in Children with CP. Dev. Med. Child Neurol. 52, e101–e106. https://doi.org/10.1111/j.1469-8749.2009.03600.x.

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. https://doi.org/10.1016/j.jbiomech.2011.06.008.

Barber, L.A., Read, F., Lovatt Stern, J., Lichtwark, G., Boyd, R.N., 2016. Medial gastrocnemius muscle volume in ambulant children with unilateral and bilateral cerebral palsy aged 2 to 9 years. Dev. Med. Child Neurol. 58, 1146–1152. https://doi.org/10.1111/dmcn.13132.

Bar-On, L., Kalkman, B.M., Cenni, F., Schless, S.-H., Molenaers, G., Maganaris, C.N., Bass, A., Holmes, G., Barton, G.J., O'Brien, T.D., Desloovere, K., 2018. The relationship between medial gastrocnemius lengthening properties and stretch reflexes in cerebral palsy. Front. Pediatr. 6. https://doi.org/10.3389/fped.2018.00259.

Blazevich, A.J., 2018. Adaptations in the passive mechanical properties of skeletal muscle to altered patterns of use. J. Appl. Physiol. https://doi.org/10.1152/japplphysiol.00700.2018.

Cobeljic, G., Bumbasirevic, M., Lesic, A., Bajin, Z., 2009. The management of spastic equinus in cerebral palsy. Orthop. Trauma 23, 201–209. https://doi.org/10.1016/j.mporth.2009.05.003.

de Gooijer-van de Groep, K.L., de Vlugt, E., de Groot, J.H., van der Heijden-Maessen, H.C., Wielheesen, D.H., van Wijlen-Hempel, R.M.S., Arendzen, J., Meskers, C.G., 2013. Differentiation between non-neural and neural contributors to ankle joint stiffness in cerebral palsy. J. NeuroEngineering Rehabil. 10, 81. https://doi.org/10.1186/1743-0003-10-81.

Farmer, S.E., James, M., 2001. Contractures in orthopaedic and neurological conditions: a review of causes and treatment. Disabil. Rehabil. 23, 549–558.

Fleiss, J.-L., 1981. The measurement of interrater agreement. Stat. Methods Rates Proportions 2.

Freire, B., Dias, C.P., Goulart, N.B.A., de Castro, C.D., Becker, J., Gomes, I., Vaz, M.A., 2017. Achilles tendon morphology, plantar flexors torque and passive ankle stiffness in spastic hemiparetic stroke survivors. Clin. Biomech. 41, 72–76. https://doi.org/10.1016/j.clinbiomech.2016.12.004.

Goldstein, M., Harper, D.C., 2001. Management of cerebral palsy: equinus gait. Dev. Med. Child Neurol. 43, 563–569.

Hägglund, G., Wagner, P., 2011. Spasticity of the gastrosoleus muscle is related to the development of reduced passive dorsiflexion of the ankle in children with cerebral palsy: A registry analysis of 2,796 examinations in 355 children. Acta Orthop. 82, 744–748. https://doi.org/10.3109/17453674.2011.618917.

Harlaar, J., Becher, J.G., Snijders, C.J., Lankhorst, G.J., 2000. Passive stiffness characteristics of ankle plantar flexors in hemiplegia. Clin. Biomech. Bristol Avon 15, 261–270.

Hastings-Ison, T., Blackburn, C., Rawicki, B., Fahey, M., Simpson, P., Baker, R., Graham, K., 2016. Injection frequency of botulinum toxin A for spastic equinus: a randomized clinical trial. Dev. Med. Child Neurol. 58, 750–757. https://doi.org/10.1111/dmcn.12962.

Hopkins, G., 2000. Measures of reliability in sportsmedicine and science. Sports Med. 30, 1–15.

Hösl, M., Böhm, H., Eck, J., Döderlein, L., Arampatzis, A., 2018. Effects of backward-downhill treadmill training versus manual static plantarflexor stretching on muscle-joint pathology and function in children with spastic Cerebral Palsy. Gait Posture 65, 121–128. https://doi.org/10.1016/j.gaitpost.2018.07.171.

Kalkman, B.M., Bar-On, L., Cenni, F., Maganaris, C.N., Bass, A., Holmes, G., Desloovere, K., Barton, G.J., O'Brien, T.D., 2017. Achilles tendon moment arm length is smaller in children with cerebral palsy than in typically developing children. J. Biomech. 56, 48–54. https://doi.org/10.1016/j.jbiomech.2017.02.027.

Kruse, A., Schranz, C., Svehlik, M., Tilp, M., 2018. The effect of functional home-based

strength training programs on the mechano-morphological properties of the plantar flexor muscle-tendon unit in children with spastic cerebral palsy. Pediatr. Exerc. Sci. 1–10. https://doi.org/10.1123/pes.2018-0106.
Kruse, A., Schranz, C., Svehlik, M., Tilp, M., 2017. Mechanical muscle and tendon properties of the plantar flexors are altered even in highly functional children with spastic cerebral palsy. Clin. Biomech. 50, 139–144. https://doi.org/10.1016/j.clinbiomech.2017.10.019.
Lee, S.S.M., Gaebler-Spira, D., Zhang, L.-Q., Rymer, W.Z., Steele, K.M., 2016. Use of shear wave ultrasound elastography to quantify muscle properties in cerebral palsy. Clin. Biomech. 31, 20–28. https://doi.org/10.1016/j.clinbiomech.2015.10.006.
Lieber, R.L., Fridén, J., 2018. Muscle contracture and passive mechanics in cerebral palsy. J. Appl. Physiol. https://doi.org/10.1152/japplphysiol.00278.2018.
Lieber, R.L., Roberts, T.J., Blemker, S.S., Lee, S.S.M., Herzog, W., 2017. Skeletal muscle mechanics, energetics and plasticity. J. NeuroEngineering Rehabil. 14. https://doi.org/10.1186/s12984-017-0318-y.
Maganaris, C.N., 2003. Force-length characteristics of the in vivo human gastrocnemius muscle. Clin. Anat. 16, 215–223. https://doi.org/10.1002/ca.10064.
Maïsetti, O., Hug, F., Bouillard, K., Nordez, A., 2012. Characterization of passive elastic properties of the human medial gastrocnemius muscle belly using supersonic shear imaging. J. Biomech. 45, 978–984.
Malaiya, R., McNee, A., Fry, N.R., Eve, L., Gough, M., Shortland, A.P., 2007. The morphology of the medial gastrocnemius in typically developing children and children with spastic hemiplegic cerebral palsy. J. Electromyogr. Kinesiol. 17, 656–663.
Mathewson, M.A., Lieber, R.L., 2015. Pathophysiology of muscle contractures in cerebral palsy. Phys. Med. Rehabil. Clin. N. Am. 26, 57–67. https://doi.org/10.1016/j.pmr.2014.09.005.
Mcnair, P.J., Dombroski, E.W., Hewson, D.J., Stanley, S.N., 2001. Stretching at the ankle joint: viscoelastic responses to holds and continuous passive motion. Med. Sci. Sports Exerc. 354–358.
Moseley, A.M., Crosbie, J., Adams, R., 2001. Normative data for passive ankle plantarflexion–dorsiflexion flexibility. Clin. Biomech. Bristol Avon 16, 514–521.
Obst, S.J., Boyd, R., Read, F., Barber, L., 2017. Quantitative 3-D ultrasound of the medial gastrocnemius muscle in children with unilateral spastic cerebral palsy. Ultrasound Med. Biol. 43, 2814–2823. https://doi.org/10.1016/j.ultrasmedbio.2017.08.929.
Perry, J., Hoffer, M.M., Giovan, P., Antonelli, D., Greenberg, R., 1974. Gait analysis of the triceps surae in cerebral palsy. A preoperative and postoperative clinical and electromyographic study. J. Bone Joint Surg. Am. 56, 511–520.
Pingel, J., Bartels, E.M., Nielsen, J.B., 2017. New perspectives on the development of muscle contractures following central motor lesions: Muscle contractures following central motor lesions. J. Physiol. 595, 1027–1038. https://doi.org/10.1113/JP272767.
Rosenbaum, P., Paneth, N., Leviton, A., Goldstein, M., Bax, M., Damiano, D., Dan, B., Jacobsson, B., 2007. A report: the definition and classification of cerebral palsy April 2006. Dev. Med. Child Neurol. Suppl. 109, 8–14.
Ross, S.A., Foreman, M., Engsberg, J.R., 2011. Comparison of 3 different methods to analyze ankle plantarflexor stiffness in children with spastic diplegia cerebral palsy. Arch. Phys. Med. Rehabil. 92, 2034–2040. https://doi.org/10.1016/j.apmr.2011.06.025.
Sellier, E., Platt, M.J., Andersen, G.L., Krägeloh-Mann, I., De La Cruz, J., Cans, C., Surveillance of Cerebral Palsy Network, 2016. Decreasing prevalence in cerebral palsy: a multi-site European population-based study, 1980 to 2003. Dev. Med. Child Neurol. 58, 85–92. https://doi.org/10.1111/dmcn.12865.
Švehlík, M., Leistritz, L., Kraus, T., Zwick, E.B., Steinwender, G., Linhart, W.E., 2013. The growth and the development of gastro-soleus contracture in cerebral palsy. Gait Posture 38, S12. https://doi.org/10.1016/j.gaitpost.2013.07.032.
Švehlík, M., Zwick, E.B., Steinwender, G., Kraus, T., Linhart, W.E., 2010. Dynamic versus fixed equinus deformity in children with cerebral palsy: how does the triceps surae muscle work? Arch. Phys. Med. Rehabil. 91, 1897–1903.
Tardieu, C., Huet de la Tour, E., Bret, M.D., Tardieu, G., 1982. Muscle hypoextensibility in children with cerebral palsy: I. Clinical and experimental observations. Arch. Phys. Med. Rehabil. 63, 97–102.
Theis, N., Korff, T., Mohagheghi, A.A., 2015. Does long-term passive stretching alter muscle–tendon unit mechanics in children with spastic cerebral palsy? Clin. Biomech. 30, 1071–1076. https://doi.org/10.1016/j.clinbiomech.2015.09.004.
Theis, N., Mohagheghi, A.A., Korff, T., 2016. Mechanical and material properties of the plantarflexor muscles and Achilles tendon in children with spastic cerebral palsy and typically developing children. J. Biomech. 49, 3004–3008. https://doi.org/10.1016/j.jbiomech.2016.07.020.
Wiley, M.E., Damiano, D.L., 1998. Lower-Extremity strength profiles in spastic cerebral palsy. Dev. Med. Child Neurol. 40, 100–107.
Willerslev-Olsen, M., Petersen, T.H., Farmer, S.F., Nielsen, J.B., 2015. Gait training facilitates central drive to ankle dorsiflexors in children with cerebral palsy. Brain 138, 589–603. https://doi.org/10.1093/brain/awu399.
Winters, T., Gage, J., Hicks, R., 1987. Gait patterns in spastic hemiplegia in children and young adults 69, 6.
Wren, T.A., Cheatwood, A.P., Rethlefsen, S.A., Hara, R., Perez, F.J., Kay, R.M., 2010. Achilles tendon length and medial gastrocnemius architecture in children with cerebral palsy and equinus gait. J. Pediatr. Orthop. 30, 479–484.

Clément Boulard is a PhD student and physiotherapist in the Pediatric Physical Medicine and Rehabilitation Department of Saint-Etienne, France. His thesis focuses on mechanical properties of spastic plantarflexors muscles in children with CP, with special interest to the methodological approaches of measurement and the impact of treatment strategies.

Vincent Gautheron is currently an associate professor, hospital practitioner, and is in charge of the Pediatric Physical Medicine and Rehabilitation Department of Saint-Etienne, France. He has been working for 30 years with several French academic society on research in pediatric disability.

Thomas Lapole is currently an associate professor in the department of sport sciences at the University of Saint-Etienne (UJM)-France. His researches are focused about the effects of repeated tendon vibration on neuromechanical properties of muscles and the changes in these parameters according to age, training, disuse or disease.