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A Comparison of Three Different Methods to Analyze Ankle Plantarflexor Stiffness in Children with Spastic Diplegia Cerebral Palsy

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Abstract

Objective—To compare three different methods of measuring plantarflexor stiffness in children with spastic diplegia cerebral palsy (CP) and children without disability.

Design—Case-control study

Setting—Human Performance Laboratory

Participants—A retrospective analysis was conducted with one hundred twenty-one children (mean age 8.4 years) with spastic diplegia (CP group) and forty-eight children (mean age 9.7 years) with typical development (TD group).

Intervention—Not applicable

Main Outcome Measures—An isokinetic dynamometer was used to measure ankle plantarflexor stiffness at 10°/s using three methods; 1) End-range method which applied a linear slope to the end of the torque-angle curve, 2) Set-range method which applied a linear slope from 30–10° plantarflexion and 3) a Linear method which applied a slope only to the linear portion of the curve.

Results—Two-way ANOVA revealed significant main effects for group and stiffness method. The End-range method showed no significant difference between groups for plantarflexor stiffness ($p=.62$), the Set-range method showed the CP group with 120% greater stiffness than the TD group ($p<.046$) and the Linear method showed the CP group with 35% greater stiffness than the TD group ($p<.001$).

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Conclusions—The Linear method appeared to resolve the issues with the previous methods; applying a linear slope to a nonlinear curve or applying a linear slope to the same range of motion for each child regardless of their range limitations. It is clear that children with CP have limited range of motion; therefore stiffness occurs earlier in the range than would be expected for a typically developing child. Using the Linear method, children with CP were 35% stiffer in the ankle plantarflexors than typically developing peers.

Keywords

Cerebral palsy; ankle; muscle; passive stretching

Common impairments in children with cerebral palsy include spasticity, stiffness and limited range of motion. Stiffness (i.e., ratio of muscle resistance to muscle length change) has been directly related to muscle tissue changes associated with prolonged spasticity and has been considered the primary cause for contracture formation and limited range of motion in children with cerebral palsy (CP).^{1,2} The amount of stiffness perceived by the clinician, when stretching a spastic muscle, may be due to both reflex and non-reflex components. Reflex muscle stiffness is due to the stretch reflex-mediated response associated with spasticity. Non-reflex or passive muscle stiffness is associated with the viscoelastic properties of the tissues surrounding a joint. Given the velocity dependent nature of spasticity, when a spastic muscle is stretched very slowly the resistance is more likely due to passive stiffness and when a spastic muscle is stretched quickly the resistance is more likely due to reflex stiffness or spasticity. Stiffness historically has been associated with contracture formation in children with CP, yet the literature on stiffness is controversial and the methods used to determine stiffness are variable.

Much of the literature in this area pertains to muscles of the upper extremity. According to Friden and Lieber, 2003, people with CP have greater muscle stiffness than normal either due to titin (a large protein found in muscle tissue) or excessive connective tissue in the extracellular matrix surrounding muscle fibers.³ They tested stiffness of individual muscle fibers in various extracted specimens of upper extremity muscles and found spastic muscle fibers in children with CP were two times as stiff as nonspastic muscle fibers. However, when the same group of investigators later tested a bundle of muscle fibers with the extracellular matrix intact they surprisingly found less stiffness compared to nonspastic bundles.⁴ The investigators discussed possible links between increased muscle fiber stiffness and inferior quality and lower viscosity of the extracellular matrix. In children with CP, spastic muscle fibers developed increased stiffness to compensate for the disorganized extracellular matrix or given the increased stiffness of the muscle fibers the extracellular matrix compensated by having inferior consistency. Vaz et al, 2006 supports the findings of increased stiffness in CP.⁵ They measured stiffness of the wrist flexors from 0–60° extension and reported significantly greater stiffness (tested at 30°/s) in the wrist flexors of 11 children with hemiplegic CP compared to typically developing children. However, work by Smeulders et al, 2004 contradicts this finding.⁶ They tested 14 patients with wrist flexion deformity intra-operatively and found that the limited wrist extension range of motion was not due to increased passive stiffness of the wrist flexors. They concluded the cause of the contracture was not due to intramuscular stiffness, but may be due to the linkage of connective tissue between adjacent muscles.

In the lower extremity, there are very few reports of stiffness in cerebral palsy. Damiano, 2001 measured stiffness of the hamstrings and quadriceps at 30°/s in a group of 23 subjects with CP and found that stiffness was not significantly greater than able-bodied values.⁷ However Alhusaini, 2010 reported significantly greater stiffness of the ankle plantarflexors

(tested at 60°/s from 0–5° dorsiflexion range) in a group of 26 children with CP compared to typically developing children.⁸

Ankle muscle stiffness has been measured in other diagnoses with similar controversy. Sinkjaer, 1994 measured ankle stiffness in nine individuals with a stroke using a 4° perturbation at approximately 10° plantarflexion, and reported passive stiffness of the spastic ankle plantarflexors to be 94% greater than the “non-spastic” contralateral leg and 278% greater than able-bodied individuals.⁹ However, Galiana et al, 2005 studied ankle stiffness in eleven individuals with a stroke using a pseudo-random sequence for a 5° perturbation at various ankle ranges and speeds and reported that the majority, 7/11 patients had no difference in ankle stiffness compared to control subjects and the 4/11 with increased stiffness could be attributed to an increased reflex gain associated with spasticity.¹⁰

Some of the controversy with regard to stiffness may be in the method of measurement and interpretation of the value. Many of the studies were conducted over a set range of motion for both spastic and non-spastic groups which may be problematic.^{5,8,9} Additional controversy comes with determining the slope of the often non-linear stiffness curve.^{7,11} Methods used to determine stiffness are variable and thus may influence the results considerably. Therefore, the purpose of this investigation was to compare three different methods of measuring plantarflexor stiffness in children with CP and children without disability.

Methods

A retrospective analysis was performed on initial data collected on 121 children (mean age 8.38 y, SD 4.1, range 4–18; 65 males, 56 females; weight 63.8 lbs, SD 34.8) with spastic diplegia (CP group) and 48 children (mean age 9.69 y, SD 3.5, range 4–16; 22 males, 26 females; weight 84.5 lbs, SD 40.9) with typical development (TD group). The children with CP had been referred to the Human Performance Laboratory and were at least one-year post orthopedic surgery, 6 months post botulinum toxin and had no history of spasticity altering surgeries (Baclofen pump, selective dorsal rhizotomy) prior to testing. Children with CP were independent ambulators, some with assistive devices, Gross Motor Function Classification System (GMFCS) levels I–III (Table 1). Individuals in GMFCS level I ambulate independently but may have limitations in more advanced gross motor skills, level II ambulate independently but have limitations walking outdoors and in the community and level III ambulate with an assistive device. All of the children in the TD group (a convenience sample of healthy comparisons) and the children in the CP group were participating in various ongoing studies in the Laboratory, however only the initial data collection was used in this analysis. The studies were approved by the Washington University Internal Review Board. All children or parents (when appropriate) signed an informed consent.

The general methods used in this investigation have been described elsewhere for the ankle.^{13,14} They will be briefly summarized here. The children sat on an isokinetic dynamometer with their ankle joint axis aligned with the center of the lever arm according to manufacturer guidelines (Figure 1). The amount of knee range was not standardized and was typically between 20–40° flexion. Ankle dorsiflexion and plantarflexion range of motion limits were established. Maximum dorsiflexion was determined by the testing physical therapist as the point where the child’s heel began to lift out of the device. To measure stiffness of the ankle plantarflexors, the child was instructed to remain as relaxed as possible as the ankle joint was rotated passively at 10°/s from maximum plantarflexion to the child’s maximum dorsiflexed position thereby stretching the ankle plantarflexor muscles (Figure 2). Although data were collected at both ankles, only the right ankle was used for analysis.

Torque-angle data were processed to partial out the effects of gravity and minimize acceleration and machine dynamic responses.

Stiffness was calculated using three different methods. The first method we used was similar to Gadjosik et al, 2007.¹¹ Stiffness was calculated using the final 20° of range of motion of the torque-angle curve (**End-range method**) for each child (Figure 3A). The second method (**Set-range method**) similar to Alhusaini, Vaz and Sinkjaer, calculated stiffness over a set range of motion for both groups.^{5,8,9} All of the children in our data set were able to stretch from 30° plantarflexion to 10° plantarflexion thus, this 20° set range was used to measure stiffness (Figure 3B). Lastly, we developed a modified version of previous methods to calculate end range stiffness that used only the linear portion of the end of the curve (**Linear method**) for each child (Figure 3C).

With the Linear method each child's slope was analyzed separately using a custom Matlab program. Using simple analytical methods, the program calculated the first and second derivatives of the torque-angle data. The program used these derivatives to estimate the "change point" or the point on the graph where the basic shape of the curve changed from nonlinear to linear (Figure 4). Theoretically, the points in the linear portion of the curve should have large first derivatives, because the slope is greatest in this portion. Also, the points should have small second derivatives, because the slope is nearly constant in this portion. This "change point," or the first point in the linear portion of the curve, was estimated by finding the initial data point where the first derivative was within 50% of its maximum value and the second derivative was within 50% of its minimum value. These thresholds were found through an iterative process until the majority, greater than 50% of the remaining data points, also fit these criteria. Once this "change point" was established, the first derivatives beyond this point were averaged and this average was used to estimate the slope of the linear portion at the end of the torque-angle curve. A custom graphical user interface permitted the investigator to check the validity of the change point and perform a manual correction. The "change point" was manually chosen for 5/169 or only 3% of the total participants in this study.

Dorsiflexion end range of motion during the stiffness test was also recorded. A two-way ANOVA with pairwise comparisons was used to analyze the 2 groups and 3 stiffness methods with significance set at $p < 0.05$. An independent t-test was used to determine differences for dorsiflexion range of motion for the two groups.

Results

The stiffness value means and standard deviations for the groups and each stiffness method are presented in Table 2. The ANOVA revealed main effects for group and stiffness method on stiffness values with no significant interaction between stiffness method and group (Table 3). Simple main effects for the CP group revealed a significant difference between the stiffness methods with pairwise comparisons showing significant differences $p < .001$ between each of the three methods. For the TD group there was also a significant difference between the stiffness methods however the pairwise comparison revealed the End-range method and the Linear method were not significantly different $p = .77$.

Comparisons for each stiffness method based on group revealed the End-range method was not a significantly different between the CP and TD groups ($F(1,501) = .25, p = .62$). There was a significant difference between groups for the Set-range method ($F(1,501) = 4.00, p = .046$) and the Linear method ($F(1,501) = 10.74, p = .001$). With the Set-range method, the CP group had stiffness values that were 120% greater than the TD group. With the Linear method, the CP group had 35% greater plantarflexor stiffness than the TD group. A

significant difference was found between the groups for dorsiflexion (df) end range of motion ($p < .001$). The CP group mean was 3.0° df (SD 9.8°) and the TD group mean was 14.1° df (SD 7.1°).

Discussion

The purpose of this investigation was to compare three methods of measuring stiffness; two methods commonly found in the literature and a modified previous method. The contrasting result using the three separate methods on the same data set was remarkable.

Study Limitations

This was a retrospective analysis of children with CP and typical development that had ankle passive resistance measured in our laboratory which permitted a large number of participants compared to other stiffness studies, however this type of analysis also presents limitations because the group selection was not age-matched. In addition the positioning of children in the isokinetic device was set according to manufacturer guidelines and not ideal for ankle stiffness. The knee angle was not held constant and varied between 20 – 40° flexion. In the seated position where the child could easily see their ankle and be comfortable with passive stretching, hamstring tightness made it impossible to position the knee in full extension to test the stiffness of only the gastrocnemius muscle. As a result stretch of the ankle plantarflexors in this study included both the soleus and gastrocnemius muscles. Maintaining a consistent knee extension angle, thereby isolating the stretch to the gastrocnemius muscle group may have increased the amount of stiffness found in our participants.

An additional limitation was lack of electromyography (EMG) during stiffness measures. Other investigators measured stiffness at $30^\circ/\text{s}$ and $60^\circ/\text{s}$ when EMG was quiet so passive stiffness was confirmed.^{9,15} We measured passive stiffness at $10^\circ/\text{s}$, a very slow speed, but without quiet EMG confirmation a reflexive component of stiffness may have contributed to the increased passive resistance measured for the CP group.

The majority of children with spastic diplegia CP in this study were high functioning independent ambulators (GMFCS level I and II). It is not clear if plantarflexor stiffness would be very different for children with hemiplegia or more severe forms of CP.

The advantage of using the End-range method is simply the ease of use; it can be quickly calculated in any common spreadsheet using the slope formula (slope of the line, or $\text{rise/run } m = y_2 - y_1 / x_2 - x_1$). While applying a linear slope to a selected end range of the torque-angle curve is easy, this method essentially ignores the shape of the curve. Figure 3A clearly showed the slopes for each child (CP and typically developing) were relatively parallel even though the CP slope was actually slightly steeper at end range than the TD slope. As expected from the parallel slopes, the results using the End-range method showed no significant difference between the groups. Use of the End-range method resulted in applying a linear slope to data that were often nonlinear and thus, was not a good fit for much of the data.

The second method used to measure stiffness, the Set-range method, was the most common method found in the literature and the most problematic if there is limited range of motion.^{5,8,9} Using a set range for each child, regardless of their range of motion limitations, resulted in measuring stiffness earlier in the range for the TD group compared to closer to the end range for the CP group (Figure 3B). The best example of this is to consider the resistance a clinician would perceive while stretching a typically developing child's ankle to a neutral position compared to a child with CP. The typically developing child usually has

15° of additional dorsiflexion range left so the clinician feels very little resistance to stretch at neutral but in the child with CP the clinician is closer to the child's end range so the resistance is much greater. Comparing the resistance felt at neutral between the two children is analogous to using the Set-range method. Not surprisingly, using this method, the results showed a significant difference in stiffness between groups. Similar to the End-range method, the Set-range method is easy to calculate in any spreadsheet. However, the use of the Set-range method will consistently show the CP group having greater ankle plantarflexor stiffness than the TD group since children with CP often have limited range of motion. This causes a shift in the torque-angle curve toward plantarflexion resulting in earlier stiffness where the slack in the tendon is taken up earlier in the range of motion.

The Linear method represented stiffness toward the end range of passive stretch, but unlike the end-range method, the stiffness slope was only applied to the linear portion of each child's torque-angle curve. Our results showed that there was no difference between the Linear method and End-range method for the TD group (similar curve consistency for the last 20 degrees), however for the CP group there was a significant difference (inconsistent curves, likely showing greater slope values at the very end range). The Linear method appeared to resolve the issues with the previous methods; applying a linear slope to a nonlinear curve and applying a slope to the same range of motion for each child regardless of their individual range of motion limitations. The only downside of this method was that a custom Matlab program was needed to automate the processing of the data. The results using the Linear method showed a significant difference in plantarflexor stiffness between groups and appeared to quantify the slight difference in steepness of the slopes noted in Figure 3C.

Using the Linear method, the CP group had 35% greater plantarflexor stiffness compared to the TD group. While significantly different, this finding is considerably less than the 242% greater ankle plantarflexor stiffness between groups reported by Alhusaini⁸ in children with CP and the 285% reported by Sinkjaer⁹ in persons with a stroke. These investigators used a set-range method (Alhusaini 0–5° dorsiflexion; Sinkjaer 4° perturbation at 10° of plantarflexion) to analyze plantarflexor stiffness which likely contributed to the considerable difference between groups. Similar results occurred when we applied the Set-range method (30–10° plantarflexion) to our data set (120% greater plantarflexor stiffness for the CP group compared to the TD group). The Set-range method may answer a question about stiffness at a particular functional range such as 0–5° dorsiflexion for gait, but it does not characterize the actual passive stiffness of each child's plantarflexors.

Determining stiffness toward the end range using the Linear method produced the highest stiffness mean values compared to the other methods. Stiffness toward the end range has been associated with intrinsic or passive stiffness¹⁶ and may be due to stretching multiple physiologic tissues including muscle, tendon, joint capsule and skin. Tightness in one or all of these tissues is the likely reason for the limited range of motion found in children with CP. Determining stiffness earlier in the midrange of motion is less indicative of passive stiffness and, according to Mirbagheri et al, 2008, may be more associated with spasticity.¹⁶ *In vitro* testing has shown that the joint capsule contributes 47%, tendon 10%, muscle 41% and skin 2% to midrange stiffness and that end-range stiffness has much greater contributions from the tendon.¹⁷ However, with intact tissue there is no way to parcel out the relative contribution of each tissue to stiffness or where in the range each tissue contributes significantly to the stiffness value.

The only other group of investigators to directly measure stiffness in CP was Damiano and colleagues, who found no significant difference between CP and able-bodied values for hamstring or quadriceps stiffness measured at 30°/s.⁷ While the CP group consistently

demonstrated greater stiffness values than the able-bodied group, the difference was not significant. The method they used to measure stiffness was to apply a slope to the isokinetic portion of the torque-angle curve. The isokinetic portion of a curve (when rotation stays constant at 30°/s) is typically most of the torque-angle curve during a passive test. This may have resulted in applying a linear slope to a nonlinear curve similar to the end-range method discussed earlier. Consistent with our findings using the End-range method they found no significant difference in stiffness between groups.

Children with CP clearly have limited range of motion during passive stretch. This resulted in a shift in the torque-angle curve toward plantarflexion; therefore stiffness occurred earlier in the range of stretch than would be expected for a typically developing child. This perception of early resistance gives the clinician a sense of excessive stiffness in children with CP compared to TD. Lieber's findings that spastic muscles develop tension at significantly shorter lengths than those taken from subjects without spasticity appear consistent with the findings in this study.

The variability of stiffness, using the Linear method, in the CP group was great ranging from 0.05 to 1.25 Nm/° with 51 children (42%) having stiffness values less than the TD mean (0.26 Nm/°). This finding supports the concept that some children with CP have a great deal of stiffness and others have very little stiffness in the ankle plantarflexors. Similar variability in stiffness values were reported in individuals with spinal cord injury.¹⁸ Future research possibilities include exploring the variability in stiffness and the relationship between stiffness and functional limitations or impairments in children with CP.

Conclusions

Comparing three different methods to determine plantarflexor stiffness showed remarkably different results. The Linear method may resolve the problems found with the methods commonly used in the literature; applying a linear slope to a nonlinear curve and applying a slope to a fixed range of motion for each child regardless of their individual range of motion limitations. It is clear that children with CP have limited range of motion; therefore stiffness occurs earlier in the range than would be expected for a typically developing child. While our results, using the Linear method, showed the amount of stiffness was 35% greater in children with CP compared to children with typical development, the difference was not as large as often reported in the literature.

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List of abbreviations

CP	Cerebral Palsy
TD	Typical Development
GMFCS	Gross Motor Function Classification System
EMG	Electromyography

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Figure 1.
Isokinetic device setup.

Figure 2.

This stiffness figure includes the 10°/s torque-angle curves for a child with CP (thick line) and TD (thin line). The passive stretch begins with the ankle in full plantarflexion and ends with the ankle in full dorsiflexion. The child with TD has greater dorsiflexion range of motion compared to the child with CP.

Figure 3.

Using the same children in Figure 2. Determining ankle plantarflexor stiffness using three different methods: A: End-range method – applying stiffness slope to the end 20° ankle range for each child shows nearly parallel stiffness slopes or no difference between children. B: Set-range method – applying stiffness slope from 30–10° plantarflexion for both children shows an obvious difference in stiffness slopes between children. C: Linear method – applying the stiffness slope to the linear portion of each child's curve showing a slightly steeper slope for the child with CP.

Figure 4. Using the same children in Figure 2. Linear method shows the Matlab “change point” where the curve is no longer linear for each child.

Table 1

Gross motor function classification system (GMFCS) distribution for the CP group.

GMFCS	
Level I	47
Level II	44
Level III	30

Table 2

Stiffness value means for each group and each stiffness analysis method.

Stiffness method	Group	Mean stiffness values Nm/° (SD)
End-range method	CP	0.24 (0.14)
	TD	0.22 (0.12)
Set-range method	CP	0.11 (0.11)
	TD	0.05 (0.04)
Linear method	CP	0.35 (0.24)
	TD	0.26 (0.17)

Table 3
Two-way ANOVA for group and stiffness method with Simple Main Effects for Stiffness method within Group.

Source	Sum of Squares	DOF	Mean Square	F	p
Stiffness method	3.641	2	1.821	68.699	.000
Group	.295	1	.295	11.137	.001
Stiffness method × group	.102	2	.051	1.931	.146
Stiffness method within group (CP)	3.604	2	1.802	68.000	.000
Stiffness method within group (TD)	1.185	2	.592	22.349	.000
Error	13.277	501	.027		
Total	42.293	507			