Mechanical properties of the plantarflexor musculotendinous unit during passive dorsiflexion in children with cerebral palsy compared with typically developing children

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AIM To examine the passive length–tension relations in the myotendinous components of the plantarflexor muscles of children with and without cerebral palsy (CP) under conditions excluding reflex muscle contraction.

METHOD A cross-sectional, non-interventional study was conducted in a hospital outpatient clinic. Passive torque–angle characteristics of the ankle were quantified from full plantarflexion to full available dorsiflexion in 26 independently ambulant children with CP (11 females, 15 males; mean age: 6y 11mo, range 4y 7mo–9y 7mo) and 26 age-matched typically developing children (18 females, 8 males; mean age 7y 2mo, range 4y 1mo–10y 4mo). In the children with CP, the affected (hemiplegia; n=21) or more affected (diplegia; n=5) leg was tested; in typically developing children, the leg tested was randomly selected. Gross Motor Function Classification System levels were I (n=15) and II (n=11). Care was taken to eliminate active or reflex muscle contribution to the movement, confirmed by the absence of electromyographic activity.

RESULTS There were small but significant differences between the two groups for maximum ankle dorsiflexion (p=0.003), but large and significant differences in the torques required to produce the same displacement (p<0.001). Further, the hysteresis of the average loading cycle in the children with CP was over three times that of the typically developing children (p<0.001).

INTERPRETATION We believe that the plantarflexor muscles of children with CP are stiffer and intrinsically more resistant to stretch, even though they retain near normal excursion. This increased stiffness is a non-neurally-mediated feature demonstrated by these children. The extent to which it influences function and predisposes the children to development of soft tissue contracture is unknown.

Cerebral palsy (CP) occurs in 2 to 2.5 individuals per 1000 live births1 and is associated with various movement disorders, usually with other impairments.2 Although the brain lesion is usually non-progressive, the movement disorders resulting from the lesion become more evident over time and are progressive.3 To some extent this may be due to adaptive changes occurring in the muscles.4-6 Children with CP are usually less physically active than typically developing children and their physical activity levels tend to decrease with increasing age.7 Progressive plantarflexor dysfunction is common in children with hemiplegia or spastic diplegia and may result from changes in muscle activation, myotendinous length, and stiffness.8 When a non-contracting (resting) muscle is stretched in a child with CP, the force opposing the movement is due to tension originating through the passive mechanical properties of the muscle, as well as any abnormal muscle activation evoked in spastic muscle. This spasticity is broadly recognized as a manifestation of enhanced stretch reflex activity9 and is often presented as the primary opponent to movement. The contributions of passive myotendinous stiffness to either passive or active joint motion are often unreported in children with CP.

Passive muscle stiffness refers to the ratio between the change in stretching force and the change in myotendinous length. To gain a measure of stiffness, the test must be performed without eliciting stretch reflex activity.10 Although the contribution of passive mechanical factors to overall restriction of motion is somewhat ambiguous, some studies have indicated that children with CP demonstrate increased passive stiffness in forearm and hand muscles.5,11 Given the potential for increased stiffness in plantarflexor muscles to contribute to functional disability, in, for example, walking, it is important...
to consider the passive mechanical properties of these muscles when determining the causes of ankle joint limitation and functional impairments in ambulant children with CP.

The purpose of this study was to explore the relation between plantarflexor muscle length and passive tension in ambulant children aged between 4 and 10 years with and without CP. Specifically, we tested the hypothesis that there is a difference in the torque–angle relations between children with CP and typically developing children during full-range passive ankle dorsiflexion.

**METHOD**

**Participants**

We recruited participants through the Child Assessment Centre in The Children’s Hospital at Westmead, Sydney, Australia and informed consent was obtained from each child’s parent or guardian. Comparison groups were chosen, consisting of 26 independently ambulant children with CP, and 26 age-matched, typically developing children (Table I). Using Moseley’s results, an a priori power analysis indicated that a sample size of 22 in each group would be required to detect a difference of 20% in the value of the applied torque at 10° of dorsiflexion, which seemed a minimal clinically significant difference. To ensure adequate power, an additional 20% of participants were recruited. Participants were recruited before conducting the test, based on inclusion and exclusion criteria, and their willingness to participate. Inclusion criteria for children with CP were the following: a diagnosis of spastic CP affecting one or both lower limbs; no skeletal foot or ankle deformity; and ability to walk independently on level ground (Gross Motor Function Classification System [GMFCS] levels I or II). Exclusion criteria were the following: cognitive problems that were appraised based on medical records and hindered communication or cooperation; severe affective or psychiatric impairments; unrelated musculoskeletal problems that might interfere with ankle/lower limb joint movement; botulinum toxin injection in the plantarflexor muscles in the previous 5 months; use of systemic anti-spasticity medications or phenol injections; orthopaedic soft tissue/bony surgical procedures (e.g. arthrodesis, tendon lengthening, or transplant) or neurosurgery for spasticity (e.g. dorsal rhizotomy).

The children with CP who participated in the study were classified in GMFCS levels I (57.7%) or II (42.3%). Twenty-one of the children presented with spastic hemiplegia and the remaining five with spastic-diplegic-type CP. Typically developing children were eligible for inclusion if they had no neurological, musculoskeletal, or other problems that might interfere with movement of lower limb joints.

**Design**

The study was prospective, descriptive, cross-sectional and non-interventional. It was approved by the Human Ethics Committees of the hospital and the University of Sydney in November 2006. There was no departure from the approved protocol. The study recruitment was completed over a period of 1 year 6 months.

**Procedure**

A specially constructed ankle measurement device was used, consisting of a footplate hinged to a support bracket for the lower leg, with a rotary potentiometer (Model 157, RS Australia, Sydney, Australia) aligned with the lateral malleolus, to measure ankle angular displacement (Fig. 1). The footplate and axis of rotation were adjustable to match the dimensions of the child. A 450N load cell (XTRAN S1W, Applied Measurement Australia Pty, Oakleigh, Victoria, Australia) was attached perpendicular to the footplate to measure resistance to movement. A handle was attached to allow manual oscillation of the footplate. Applied torque values were calculated from the product of the applied force and the perpendicular distance from the point of application of the force to the axis of rotation of the footplate. The load transducer measured uniaxial loading (only one component of the force), and any errors occurring if ‘off-axial’ forces had been applied would

| Table I: Characteristics of the participating children and results of the study |
|---------------------------------|---------------------------------|--------|--------|
| Age (y:mo)                      | CP group (n=26)                 | Typically developing group (n=26) | t      | p      |
| 6:11 (1:9)                      | 7:2 (1:8)                       | 0.116 | 0.913  |
| Height (cm)                     | 121.5 (12.6)                    | 121.4 (10.4) | 0.188 | 0.852  |
| Body mass (kg)                  | 24.7 (6.6)                      | 23.8 (6.6) | 0.363 | 0.718  |
| Sex ratio (male:female)         | 15:11                           | 8:18   | (r²)2=8.807 | 0.094  |
| Maximum dorsiflexion range (degrees) | 13.4 (6.7)                    | 18.9 (6.0) | 3.106 | 0.003  |
| Torque at 0° dorsiflexion (N m/kg) | 220 x 10⁻³ (90 x 10⁻³)         | 80 x 10⁻³ (40 x 10⁻³) | -6.659 | <0.001 |
| Torque at 5° dorsiflexion (N m/kg) | 290 x 10⁻³ (100 x 10⁻³)        | 100 x 10⁻³ (50 x 10⁻³) | -7.082 | <0.001 |
| Torque at 10° dorsiflexion (N m/kg) | 350 x 10⁻³ (110 x 10⁻³)        | 120 x 10⁻³ (50 x 10⁻³) | -6.847 | <0.001 |
| Stiffness, 0 to 5° dorsiflexion gradient (N m/kg/deg) | 13 x 10⁻³ (1.52 x 10⁻³) | 3.8 x 10⁻³ (1.52 x 10⁻³) | -21.823 | <0.001 |
| Hysteresis (N m/kg/deg)         | 58.9 x 10⁻³ (3.0 x 10⁻³)       | 19.3 x 10⁻³ (1.0 x 10⁻³) | -63.853 | <0.001 |
| Cumulative applied torque during loading (N m/kg) | 9.433 (3.65) | 3.92 (2.04) | -6.702 | <0.001 |
| Cumulative applied torque during unloading (N m/kg) | 4.5 (2.95) | 1.94 (2.19) | -3.551 | <0.001 |

Values are mean (SD). CP, cerebral palsy.
have been small. Nevertheless, we were vigilant in maintaining the angle of force application. High interrater and intrarater reliability has previously been demonstrated for the measurement of passive torque and ankle displacement using this technique (intraclass correlation coefficient >0.86) and the procedure has been shown to be highly responsive to change in stiffness characteristics.

Each participant lay supine with the foot placed in the ankle apparatus and positioned, by visual approximation, such that the point midway between the lateral and medial malleolus in the sagittal plane was aligned with the axis of rotation of the device. The participant’s foot was secured with Velcro straps, the knee was placed in an extended position, and light pressure was applied by the researcher’s (AA) hand above the knee over the thigh to ensure that knee position was maintained. The calf was free of contact and clear of all surfaces and structures. In the children with CP, the affected or more affected leg was tested; in typically developing children, the leg tested was randomly selected. The researcher rotated the foot passively in a sinusoidal pattern around the ankle from full plantarflexion to full available dorsiflexion (determined visually by loss of heel contact). All children were instructed to keep their legs relaxed and to avoid assisting or resisting the motion during the sinusoidal rotation.

A warm-up and familiarization process, with two or three repetitions of full range movement of the rig, was carried out before recording the data. For each child, a minimum of 10 passive stretch cycles were applied at a target frequency of 0.5Hz. This sequence was repeated two more times to ensure relaxation and compliance on the part of the participant and to avoid eliciting reflex muscle activity. This allowed at least one full cycle of passive stretching and recovery for analysis. The child’s compliance with the instruction to keep the leg relaxed and to avoid assisting or resisting the motion was confirmed by the absence of EMG activity (Spike2 software version 2.09, Cambridge Electronic Design, Cambridge, UK). During data analysis, one complete loading and unloading cycle without evidence of EMG activity was chosen for analysis, ensuring that there was no reflex contribution to total stiffness. In the event of more than one cycle meeting this criterion, selection was random.

Data processing and statistical analysis
We collected force and angle data simultaneously at a frequency of 125Hz using a 16-bit analogue-to-digital converter (DAQCard-6036E, National Instruments, Austin, Texas, USA). The application software (PhysioDAQXS version 3.0, The University of Sydney) consisted of a graphical user interface designed using Borland C++ builder. Access to the data was gained by using National Instruments callback functions to retrieve data collected by the data acquisition card. The graphical user interface supports the collection, display, and storage of data in real time. We confirmed the repeatability and linearity of the force and angle signals before data collection. We calculated the torque due to the weight of the foot-plate as a function of the angle and computed the net torque from the applied torque and footplate effect. As the children were of different ages, height, and weight, we scaled the net torque to body mass to reduce variability arising from physique.

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After processing, we ensemble-averaged the scaled torque–angle relations during loading and unloading cycles for the typically developing children and those with CP (Fig. 2). We extracted the torque values at predetermined dorsiflexion angles (0°, 5°, and 10°), the maximum passive dorsiflexion angle and the computed stiffness, as determined by the slope of the line between 0° and 5° of dorsiflexion (Fig. 3). We calculated and recorded hysteresis, expressed as the energy absorbed by the muscle–tendon unit during the cycle and enclosed within the area between the loading and unloading curves. SPSS for Windows (version 14.0, SPSS Inc., Chicago, IL, USA) was used for statistical analysis. Statistical comparison between groups was made using independent sample t-tests. Data are presented as mean (SD); the alpha level for statistical significance was set at 0.05.

RESULTS

There was no significant difference between the two groups of children for age, height, or weight. However, there was a difference in the mean angular velocity of the passive movement produced by the examiner, 60°/s (SD 20) for children with CP and 88°/s (SD 16) for typically developing children. This computed to an oscillation frequency of 0.54 Hz (SD 0.20) for children with CP and 0.70 Hz (SD 0.22) for typically developing children (t=2.74; p=0.008).

The typically developing children exhibited a significantly greater maximum range of dorsiflexion than those with CP (Table I; difference=5.5°; p=0.003); however, all participants were able to achieve dorsiflexion ranges that allowed extraction of torque values at the three nominated angles described above.

The result of averaged loading and unloading cycles, shown in Table I and Figure 2, indicate clear differences between the children with CP and typically developing children. The torque/displacement values show a regular logarithmic relation, with consistent and significant differences between the two groups for the torque required to achieve each dorsiflexion angle (0°, 5°, and 10°; Table I and Fig. 3). Significant differences were also found for the hysteresis values and for the gradient of the slope between plantigrade and 5° of dorsiflexion, as an indicator of passive myotendinous stiffness (Table I and Fig. 3).

DISCUSSION

To our knowledge, this study is the first to document the passive stiffness in children with CP using sinusoidal oscillations of the ankle joint. Our primary purpose was to examine the mechanical parameters affecting the ankle in children with CP (GMFCS levels I and II) compared with typically developing children. We used diverse variables, expressing both discrete (torque at three angles) and continuous (slope of curve, hysteresis) measures of passive stiffness of the plantarflexor muscles to analyse the data. Measurement of the passive torque–angle relations throughout dorsiflexion range is a more precise way to determine tissue stiffness than quantifying the passive range using a single point, because passive torque increases more rapidly as the joint is moved towards its maximum range.

We found significant differences between the two groups in all the passive mechanical properties of the tested ankle; thus, our results lead us to support our experimental prediction.
Torques at predetermined joint angles, ankle stiffness (slope of curve), and hysteresis were almost three times as high in the children with CP compared with the typically developing group (p<0.001). It could be argued that the additional 5.5° of ankle dorsiflexion found in the typically developing children might distort these findings; however, the torque difference between the two groups is very much greater than could be affected by such a small difference in range. Adjusting the loading and unloading cycles by scaling against total available range in each group makes no perceptible difference to the results.

The absence of EMG activity in the lower leg muscles in data from all participants supports the requirement for measurement of passive myotendinous characteristics. The average angular range and velocity of the stretch reflects the displacement and velocity that occurs during walking. Torque curves at velocities up to 70°/s show the characteristic shape of tissue stretch only. Even at stretch velocities of 120°/s, only four out of 15 people with spastic hypertonia demonstrated reflex responses in the triceps surae. We believe that the sinusoidal oscillation frequency was significantly different between groups, the children with CP, who were more likely to respond with hyperreflexia, were stretched at the lower rate, consistent with the subjective need to maintain a relaxed, passive movement.

The length of the plantarflexor muscles is difficult to measure in vivo. Therefore, indirect measures of angular displacement in response to applied torque are indicative of the force–length variables. The protocol we used also did not allow differentiation between the mechanical properties of the calf muscle and those of the Achilles tendon and other connective tissues. Nor could we exclude the contribution of other tissues, such as skin, ligament, joint capsule, and cartilage. However, these last structures generally contribute to stiffness only at the end of the dorsiflexion range. Because we measured the gradient of the loading curve between plantigrade and 5° of dorsiflexion, well short of the limit of the participants’ dorsiflexion, we believe that our values for stiffness exclude any substantive contribution of these ancillary tissues and that the myotendinous unit is the major impediment to passive ankle joint dorsiflexion. Although the intention in this study was to measure the myotendinous unit stiffness, a recent study by Zhao et al. has shown that the muscle belly, rather than the tendinous elements, offers greater resistance to passive stretching. They found that, after stroke, the calf muscles on the impaired side became stiffer and shorter, leading to a proximal shift of the midpoint between the muscle and the Achilles tendon. Therefore, on the impaired side, the Achilles tendon elongates and become less stiff than that on the unimpaired side. Ultrasonography studies have also shown that the gastrocnemius muscle has shorter, thinner fascicles, although similar pennation angles, in the parietic limbs of children with CP.

Our findings are also in agreement with previous studies involving adults with neurological impairment. Sinkjaer and Magnussen reported that non-reflex stiffness in the spastic limb of a group of patients with hemiparesis with plantarflexor muscle hypertonia was 278% greater than that seen in comparisons. Recently, Mirbagheri et al. also found that non-reflex muscle stiffness was increased in the plantarflexor muscles of individuals after stroke compared with unimpaired participants, and that the contribution of this stiffness was most pronounced with the ankle in the dorsiflexed position.

In children, non-reflex stiffness has not been widely investigated. Granata et al. studied this indirectly by measuring the electromechanical delay from the patellar tendon tap of knee extensors in children with CP. Biomechanical stiffness was found as the primary contributing factor to the significantly shorter delay in these children. In an upper limb study, Vaz et al. reported wrist flexor passive stiffness was significantly greater in children with hemiplegic CP than in typically developing children. Friden and Lieber found that flexor carpi ulnaris muscle cells were twice as stiff in biopsy material from children with CP compared with biopsies obtained from a range of muscles in able-bodied adult individuals. Booth et al. found an accumulation of type I collagen in the endomysium of the vastus lateralis muscle obtained from children with spastic CP and suggested that this increase in hydroxyproline concentration in spastic muscles may affect the muscle’s mechanical properties, contributing either directly or indirectly to the formation of contractures.

Because the muscles of the children with CP exhibit over three times as much hysteresis as those of typically developing children, less of the energy applied to the muscle to stretch it during loading is returned during unloading. The reduction in stored elastic energy may also contribute to mechanical inefficiencies during weight-bearing activities. The implication of this may be that the group with CP experience greater resistance to motion at the ankle during dorsiflexion, such as in the late stance phase of walking, than typically developing children. The impact of these manifestations of increased non-reflex stiffness on functional activity is yet to be established but would be an important topic for further research.

It was not possible to use the ankle device with more severely affected children (GMFCS levels III–V), because they often have pre-existing ankle contracture or are unable to relax sufficiently through testing. Østensjø et al. reported that children at GMFCS levels I and II had significantly less spasticity, as measured with the modified Ashworth scale, than those classified at higher GMFCS levels. We would expect to find as much or more muscle stiffness in children at levels III, IV, and V than at levels I or II, probably because of lower levels of activity and general immobility.

The findings of our study suggest that greater attention is warranted in assessing and monitoring the passive stiffness of the calf muscle in ambulant children with CP (GMFCS levels I and II). At this stage, it is not clear to what extent the increased stiffness that we have found influences function or whether it predisposes these children to later development of soft tissue contracture. It may be that the passive mechanical properties of the calf muscle imparts as much, or even more,
restriction on mobility than the neurally modulated features of the spastic muscle. It is also unclear whether increased stiffness is an adaptation to neural impairments and disuse, and at what stage in the child’s development it becomes apparent. It is possible that exercise and functional movement training to prevent adaptive muscle changes affecting stiffness and length may be an effective intervention if started early in life, but this requires further investigation.

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