Muscle strength and walking ability in Diplegic Cerebral Palsy: Implications for assessment and management

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1. Introduction

Children with spastic Cerebral Palsy (CP) have smaller [1,2] and weaker muscles [3,4] than healthy children. Muscle strength correlates with gait [5,6] and motor function [7] and strength is more highly related to function than spasticity [6]. Spastic muscles respond positively to strength training and strength gains have been shown to be similar or greater than those reported in the healthy population [8]. Various studies have reported significant improvements in gait following strengthening programmes in terms of temporal parameters [7,9,10] and Gross Motor Function Measure (GMFM) Dimension E [7,10–12]. McPhail and Kramer [8] found a direct correlation between the strength gained and functional improvement, but significant changes in gait kinematics following strength training appear difficult to achieve and have rarely been reported [11]. These strengthening studies, however, presume weakness is present, and reports specifically documenting the extent and distribution of muscle weakness in children with CP remain very limited. In Spastic Diplegia (SD), the most prevalent diagnostic category of CP, muscle strength has been quantified at the knee joint [13], and two lower limb strength profiles have been undertaken and compared with controls [3,4].

The purpose of this study was to examine the extent and distribution of weakness in lower limb muscles in SD/CP children and to relate this to ambulatory function. The intention was to help progress our understanding of which muscle groups play an important role in achieving independent ambulation, and may also serve as a basis for planning management and improving functional outcomes.

2. Patients

The participants in this study were 50 children (28 boys, 22 girls) with an established diagnosis of SD/CP and mean age of 11 years 7 months (range 7 years 1 month–16 years 9 months) who walked with or without walking aids. Using the Gross Motor Function Classification System (GMFCS) [14] 14 children were...
classified as level I, 26 children as level II and 10 children as level III. Patients had no orthopaedic surgery or Botulinum toxin in the year preceding the study and, at the time of assessment, were on the waiting list for single event multi-level surgery (SEMLS). A group of 15 healthy children (seven boys, eight girls) with no known neurological or orthopaedic problems and mean age 11 years 1 month (range 7 years–15 years 1 month) were recruited as controls. Children were excluded below the age of 5 years, since it was assumed they could not consistently perform isometric strength testing, and over the age of 18 years.

3. Methods

Each CP child underwent a clinical examination including range of motion measurements and manual muscle testing of the lower limbs. Maximal isometric muscle strength was measured in all children in six muscle groups in each leg using a digital myometer (MIE Medical Research Ltd., Leeds, UK) sampling at 50 Hz. The myometer was calibrated prior to each assessment. Muscle groups measured were: the hip flexors, extensors and abductors, the knee flexors, and the knee extensors at both 30° and 90° of flexion. Each child was tested in standardised positions (Table 1) and muscle groups tested sequentially. The positions were based on the existing strength testing literature [15] in gravity neutral positions as possible. The child was instructed to hold onto the couch in both sitting and lying, and pelvic stabilisation was used to minimise compensatory movements. A combination of fixed and hand-held techniques were used; fixed dynamometry was used for the knee extensors where normal values were expected to exceed the upper limb strength of the assessor using a hand-held technique. Maximal isometric contractions were performed using a ‘make test’, which maintains the muscle at a constant length to minimise eliciting a reflex during the test [16]. The technique was clearly explained and a practice trial undertaken at the start of the session to familiarise the child with the technique. Three trials were administered and recorded for each muscle group and strong verbal encouragement was used during the trial to achieve maximum effort.

We have previously undertaken a reliability study of the above protocol in 5 children with SDECP/Spastic Diplegia and 5 healthy children [17]. The same assessor repeated testing of all six muscle groups over three visits held at least one week apart. Two assessors each repeated testing of three muscle groups over three visits held at least one week apart. The protocol demonstrated very good intra- and inter-rater reliability for the isometric strength testing in the healthy group (ICC > 0.81) and good reliability for the CP group (ICC > 0.61). The rate of maximal force development and maximal voluntary contraction force was reduced in the CP children, which is consistent with the literature [18].

3.1. Data analysis

Isometric muscle strength was expressed as torque (Nm) by multiplying the force measurements by the lever arm distance. This was the distance from the estimated joint centre of rotation to the point of application of the force, measured with a tape measure (Table 1). The peak value for each muscle group was normalised to body mass (Nm/kg) and used for analysis. Differences in mean muscle strength values between the CP and control groups were analysed using an independent t-test (p < 0.05). Differences in both mean muscle strength values and mean joint angles according to walking ability (GMFCS level), were analysed with one-way analysis of variance (ANOVA) and Tukey post hoc testing (p < 0.05) using SPSS version 16 (SPSS Inc., Chicago, IL).

Not all data could be obtained. The maximum force limit of the MIE myometer is 1000 N but hip extensor strength sometimes exceeded the upper limb strength of the assessor (~450 N). Where this occurred, in 8/30 control limbs and in 2/100 CP limbs, a value of 450 N was ascribed for analysis. Some children had insufficient hip abductor strength to register a reading on the myometer. This occurred in 11/100 CP limbs and a value of 0 N was ascribed for analysis. It was not possible to measure the strength of the knee extensors at 90° in 26/100 CP limbs due to knee flexion contractures and these were excluded from the analysis.

4. Results

Mean joint angles and mean maximal normalised strength values for all test positions in all CP children versus controls, as well as CP children according to GMFCS level, are shown in Table 2. Muscle strength in the CP children was significantly less than controls in all muscle groups with the exception of the hip extensors. Fig. 1 shows the strength profile of the CP children with mean strength values presented as a percentage of control values. Muscle strength of the CP children ranged from 43% to 90% of control values depending on the muscle group. The knee extensors measured at 30° of flexion was the relatively weakest muscle group. With worsening ambulatory level from GMFCS levels I to III strength values decreased incrementally in all muscle groups.

<table>
<thead>
<tr>
<th>Figure</th>
<th>Muscle group</th>
<th>Subject position</th>
<th>Dynamometer position</th>
<th>Lever arm distance</th>
</tr>
</thead>
<tbody>
<tr>
<td><img src="image1.png" alt="Figure" /></td>
<td>Hip flexors</td>
<td>Supine</td>
<td>Hip and knee flexed to 90°. Thigh cuff around distal thigh, proximal to femoral condyles. Dynamometer held so hip flexion is resisted</td>
<td>Greater trochanter to thigh cuff</td>
</tr>
<tr>
<td><img src="image2.png" alt="Figure" /></td>
<td>Hip extensors</td>
<td>Supine</td>
<td>Hip and knee flexed to 90°. Thigh cuff around distal thigh, proximal to femoral condyles. Dynamometer held so hip extension is resisted</td>
<td>Greater trochanter to thigh cuff</td>
</tr>
<tr>
<td><img src="image3.png" alt="Figure" /></td>
<td>Hip abductors</td>
<td>Supine</td>
<td>Hip and knee flexed to 30° in near neutral abduction. Thigh cuff around distal thigh, proximal to femoral condyles. Dynamometer held so hip abduction is resisted</td>
<td>Greater trochanter to thigh cuff</td>
</tr>
<tr>
<td><img src="image4.png" alt="Figure" /></td>
<td>Knee flexors</td>
<td>Sitting</td>
<td>Hip and knee flexed to 90°. Ankle cuff around distal leg, proximal to malleoli. Dynamometer held so that knee flexion is resisted</td>
<td>Lateral femoral condyle to ankle cuff</td>
</tr>
<tr>
<td><img src="image5.png" alt="Figure" /></td>
<td>Knee extensors 90°</td>
<td>Sitting</td>
<td>Hip and knee flexed to 90°. Ankle cuff around distal leg, proximal to malleoli. Dynamometer fixed to a stable point so knee extension is resisted</td>
<td>Lateral femoral condyle to ankle cuff</td>
</tr>
<tr>
<td><img src="image6.png" alt="Figure" /></td>
<td>Knee extensors 30°</td>
<td>Sitting</td>
<td>Hip flexed to 90° and knee to 30°. Ankle cuff around distal leg, proximal to malleoli. Dynamometer fixed to a stable point so knee extension is resisted</td>
<td>Lateral femoral condyle to ankle cuff</td>
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(© ©) Fixed point. (➡) Direction of active muscle contraction. (➡) Direction of resistance.
while joint contractures increased (Fig. 3 and Table 2). ANOVA analysis revealed a significant difference in muscle strength between GMFCS levels in four out of six muscle groups and post hoc analysis showed this difference to be between GMFCS levels I and II, and levels I and III in the four muscle groups and between levels II and III in two muscle groups. The greatest strength reduction between independent (GMFCS level I) and dependent walkers (GMFCS level III) was in the hip abductors (61%) and knee extensors at 30° (45%).

Table 2
Mean (SD) joint angles (°) and normalised muscle torque values (Nm/kg) of all CP children versus controls and for CP children according to GMFCS. ‘n’ represents number of legs in each group. Bold type indicates significant difference between CP and control children.

<table>
<thead>
<tr>
<th></th>
<th>Controls (n = 30)</th>
<th>CP (n = 100)</th>
<th>p value</th>
<th>GMFCS I (n = 28)</th>
<th>GMFCS II (n = 52)</th>
<th>GMFCS III (n = 20)</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mean age (range)</td>
<td>11.1 (7.0–15.1)</td>
<td>11.7 (7.1–16.9)</td>
<td></td>
<td>12.1 (8.6–16.9)</td>
<td>11.6 (7.1–15.4)</td>
<td>11.2 (7.9–15.1)</td>
<td></td>
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<tr>
<td>Joint angles (°)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hip flexion contracture</td>
<td>7 (8)</td>
<td>6</td>
<td>0.02</td>
<td>12^{ab}</td>
<td>0.06</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Knee flexion contracture</td>
<td>4 (14)</td>
<td>0</td>
<td></td>
<td>4</td>
<td>10^{a}</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Popliteal angle</td>
<td>65 (17)</td>
<td>57</td>
<td></td>
<td>65</td>
<td>77^{ab}</td>
<td></td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Normalised torque (Nm/kg)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
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<td></td>
</tr>
<tr>
<td>Hip flexors</td>
<td>1.05 (0.19)</td>
<td>0.51 (0.21)</td>
<td>&lt;0.001</td>
<td>0.55</td>
<td>0.47</td>
<td>0.17</td>
<td></td>
</tr>
<tr>
<td>Hip extensors</td>
<td>2.64 (0.51)</td>
<td>2.37 (0.83)</td>
<td>0.10</td>
<td>2.62</td>
<td>2.18</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hip abductors</td>
<td>1.05 (0.29)</td>
<td>0.61 (0.23)</td>
<td>&lt;0.001</td>
<td>0.79</td>
<td>0.53^{bc}</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Knee flexors</td>
<td>1.26 (0.25)</td>
<td>0.93 (0.28)</td>
<td>&lt;0.001</td>
<td>1.05</td>
<td>0.89^{c}</td>
<td></td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Knee extensors at 90°</td>
<td>1.82 (0.37)</td>
<td>1.50 (0.54)</td>
<td>0.03</td>
<td>1.78</td>
<td>1.46^{c}</td>
<td></td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Knee extensor at 30°</td>
<td>1.27 (0.17)</td>
<td>0.56 (0.30)</td>
<td>&lt;0.001</td>
<td>0.77</td>
<td>0.45^{c}</td>
<td></td>
<td>&lt;0.001</td>
</tr>
</tbody>
</table>

p value indicates level of significance for the ANOVA analysis between all three GMFCS levels.

- ^a^ Significant difference between GMFCS levels I and III.
- ^b^ Significant difference between levels II and III.
- ^c^ Significant difference between GMFCS levels I and II.

5. Discussion

The degree and distribution of weakness in children with CP/SD has not been well documented. The finding of lower limb weakness in all muscle groups tested, when compared with controls, is in agreement with the two existing strength profiles in Diplegic CP [3,4]. We measured a range of 43–90% of control values depending on the muscle group. It is however difficult to observe distinct trends in the extent or pattern of weakness between this and the existing two strength profiles, due to differences in the age range, muscles measured, testing position, technique and analysis. In particular, these three studies highlight that muscle strength is position dependent. The optimal position for measuring muscle strength is a gravity-neutral position to eliminate the effect of the weight of the limb, which will vary subject to subject, and to facilitate the measurement of subjects with sub-gravity strength. In all three studies the only muscles measured in identical gravity-neutral positions were the hip abductors and knee extensors at 90° and these show remarkably similar readings when expressed as a percentage of control values (Fig. 2). Eek et al. [19] chose to measure the hip flexors and knee flexors in both anti-gravity and gravity-neutral positions, and the hip extensors in two different anti-gravity positions, but then combined the two measurements for each muscle group in the analysis. They reported significantly higher values for controls in the hip flexors in sitting (anti-gravity) versus supine lying (gravity neutral), significantly higher values for the knee flexors in sitting (gravity neutral) versus prone lying.
The hip extensors are a particularly problematic group to measure accurately. Crompton et al. [20] have shown it is possible to measure this muscle group reliably in supine lying but not in prone. We deliberately chose supine lying with the hip and knee flexed as a gravity-neutral position, and to isolate gluteus maximus from the hamstrings. Hip extensor strength in this position is considerably greater than in prone lying [20]. This may be due to some additional contribution from the hamstrings and possibly knee extensors working in concert. Frigo et al. [21] suggest co-contraction of the knee extensors and hamstrings may be a well co-ordinated compensation for hip extensor weakness, rather than a pathological action. It is, however, interesting to note that the gluteus maximus in Lampe et al.’s [1] study of hemiplegic subjects were close in volume to the unaffected side. The alternative use of anti-gravity positions in prone, with and without isolating gluteus maximus, by Wiley and Damiano [3] and Eek et al. [4], respectively, restricted the number of patients who could achieve this starting position due to weakness. All three studies may therefore have over estimated the strength of the hip extensors due to these difficulties.

We found an incremental reduction in strength in all muscle groups with increasing walking difficulty from GMFCS levels I to III (Fig. 3) as did Eek and Beckung [22]. Mean ages and age ranges were similar across GMFCS levels with no significant difference in age between GMFCS levels. The greatest drop in strength between independent ambulators (GMFCS level I) and those dependent on walking aids (GMFCS level III) was in the hip abductors (61%) and the knee extensors at 30° (45%). Eek et al.’s data also showed the greatest drop in strength between GMFCS levels I and III to be in the hip abductors as well as the hip extensors. They did not however measure the knee extensors at 30°. These proximal muscle groups are key groups in maintaining dynamic stability during gait. Throughout stance muscles contract when body alignment creates an external moment which threatens the weight-bearing stability of the limb and trunk. In the sagittal plane the moments that must be controlled are those that induce hip flexion, knee flexion and ankle dorsiflexion. In the coronal plane the effects of gravity predominantly induce an external hip adductor moment which threatens the stability of the pelvis and trunk. These two studies highlight a significant loss of strength in the hip extensors, knee extensors and the hip abductors with increasing GMFCS classification and these are key muscle groups associated with sagittal and coronal plane stability during independent walking. Targeting key muscle groups in strength training programmes to maximise walking ability is an important consideration during routine therapy treatment, during rehabilitation following surgery, as well as targeted sparing of muscle strength during surgery [23].

Multiple impairments contribute to motor deficits in CP. We did not evaluate muscle strength at the ankle which we found difficult due to impairment of selective voluntary motor control distally combined with foot deformity in some children. Crompton et al. [20] have shown large variability when measuring the ankle plantarflexors in CP children and testing of these muscles with hand-held dynamometry is unreliable. Furthermore, in healthy children we found the plantarflexors difficult to manually stabilise due to the short lever arm and very high force generating capacity. This is a limitation of this study since CP children demonstrate significant power deficits in the ankle plantarflexors during gait. Wiley and Damiano did use hand-held dynamometry but included children who were unable to achieve a neutral ankle position [3], while Eek et al. built a special device to stabilise the dynamometer and measure the plantarflexors [19]. Both found the ankle dorsiflexors to be one of the weakest groups in SDCP but it would be helpful to separate out the influence of selective control and foot deformity. Objective assessment of selective voluntary motor control has been a challenge but Fowler’s recently validated ‘Selective Control Assessment of the Lower Extremity’ (SCALE) is a promising new tool [24].

In the 1990s there was a resurgence of interest in strength training in children with CP and initial studies concentrated on establishing whether it was possible to strengthen muscles in this group of patients. Progressive strength training has since been shown to result in moderate to large gains in both strength and function but relatively mild changes in gait [25]. Only recently have studies begun to emerge which target resistance strengthening to specific muscle groups. Engsberg and Ross [11] and McNee et al. [26] focused solely on the distal musculature. Engsberg and Ross [11] randomised 12 CP/SD children to a 12 weeks of isotonic strength training of the dorsiflexors, or plantaflexors, or dorsi and plantarflexors and evaluated gait and motor function. McNee et al. [26] evaluated a 10-week resistance programme for the plantarflexors on 13 hemiplegic and diplegic children and looked at the effect on in muscle volume, gait and motor function. Eek et al. [4]
assessed whether an 8-week strength training programme could specifically influence the gait pattern and gait function of 16 CP/SD children by training four lower limb muscle group chosen on the basis of strength testing and gait abnormality (kinematics and kinetics). Damiano et al. [27] examined whether 8 weeks of progressive strength training of the hip and knee extensors could diminish the degree of crouch and internally rotated gait in 8 children with spastic Diplegic CP. So far the outcomes of these more focused studies remain equivocal, with significant improvements in strength or muscle volume in all studies, significant improvement in motor function in two [4,11] out of four studies, significant improvement in just one kinematic parameter in one study [11], and no significant improvement in temporal parameters in any of the four studies. The absence of improvement in gait kinematics in Eek et al.’s [4] study was perhaps not surprising given the variability in target muscles which were strengthened across subjects. However, this was the only study to additionally measure kinetics which did show significant improvements in hip extensor moments and ankle power. Their subjects included independent ambulators only (GMFCS levels I and II), while the remaining studies included independent ambulators as well as those at a lower functional level using assistive devices (GMFCS levels I–III). Damiano et al. [27] undertook a sub-group analysis comparing independent ambulators with those with assistive devices. The children who were more functional showed a small improvement in crouch in comparison with an appreciable worsening in the children who required assistive devices to walk. This still did not reach statistical significance but the numbers were small. We have also found that GMFCS level II subjects respond significantly better to strength training than GMFCS level III [28]. Mockford and Caulton’s [25] recent systematic review of strength training in CP found more conclusive results than other reviews [29,30] by deliberately limiting the clinical heterogeneity of CP to ambulatory children and adolescents and excluding upper limb studies, adults and non-ambulatory subjects. They concluded that function and gait improve more in younger than older subjects. We need to consider how to further improve the outcomes of future strength studies by reducing the heterogeneity of CP by not only focusing strength training to target muscles but evaluating results by subgroups such as functional or GMFCS level and age.

This study confirms that ambulant children with CP, even those who are mildly affected, have significant lower limb weakness when compared with controls. We found an incremental drop in strength in all muscle groups with increasing walking difficulty, from GMFCS levels I to III. The greatest drop in strength between independent ambulators and those needing assistive devices was in the hip abductors and knee extensors at 30° which are key muscle groups in sagittal and coronal plane walking stability. This has implications in targeting strength training to maximise functional outcomes. The study highlights important considerations in the assessment of muscle strength to reflect true readings of individual muscle groups and to facilitate comparison of strength outcomes across studies.

Conflict of interest statement

None of the authors reports a conflict of interest.

References