

EFFECTS OF STRENGTH-TRAINING TRIALS FOR CHILDREN AND ADOLESCENTS WITH SPASTIC CEREBRAL PALSY: A SYSTEMATIC REVIEW

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Abstract

Objective

To evaluate the effects of strength-training on lower limb muscles and motor activity among children and adolescents with spastic Cerebral Palsy.

Methods

A comprehensive search of full text published studies in English was performed. Electronic databases were searched with no limit for year of publication. Terms used included: exercise, children, youth, disability, treatment, strength training, spastic cerebral palsy, muscle strength, training program, therapeutic exercise, gait training, and physical function. Reference lists of retrieved articles were also examined to identify additional studies. To be included, studies had to be: randomised controlled trials evaluating strength training outcomes on children and adolescents with spastic CP. Those with intellectual disability were excluded. Evaluation for quality: Trials were rated for methodological quality using the PEDro scale independently by the 2 researchers.

Results

Of 4 selected trials, 1 an abstract was excluded, 3 full articles were included. Trials methodological quality ranged from 6 to 8 on PEDro scale. Two studies reported significant muscle strength changes following strength training, with effect sizes ranging from d equal to 1.17 to 1.78 and another reported improved gait and perceived body image.

Conclusion

Trials demonstrate that strength-training improves strength of targeted muscle and motor functioning. More rigorous trials on larger samples should evaluate the impact of specificity in strength-training and cost-effectiveness of programs.

Key words

strength-training, spastic Cerebral Palsy, strength training, therapeutic exercise, gait training.

Introduction

Cerebral palsy (CP) is described as a chronic neurological disorder arising from a lesion on immature brain that manifests itself as, poor motor activity and postural control (Dodd, Taylor, Diane & Damiano, 2002). In view of the new paradigm shift to evidence-based practice, physiotherapists and other rehabilitation service providers want to know about the current evidence, to improve their problem-solving skills and accountability to children and adolescents with CP and their caregivers in delivering cost-efficient rehabilitation services. One such evidence should be on the effects of strength-training interventions focused at improving muscle strength, and physical functioning, such as mobility, of children with spastic CP (Mulligan, Abbot, Clayton, McKegg & Rae, 2004).

Strength-training interventions in the hospitals, community, or at school, vary in terms of their outcomes as they may either focus on strengthening a specific muscle group that is weak or general cardio-respiratory fitness. Such interventions may also be individualised strength-training programmes (ISTP) or group activities.

The result of this variability of strength training programming coupled with the rising cost of healthcare may continue to deny accessibility of interventions and opportunities, to those with disabilities, specifically children and adolescents with spastic CP. This situation promotes the

likelihood for children with spastic CP and have mobility related problems, to become susceptible to risk for, hypertension, diabetes, overweight, obesity, pressure sores, underdevelopment, declining mental health status and delay development of physical and social skills needed for independent living (Carmona, 2005). Blomquist et al. (1998) observed that children and adolescents with disabilities also become isolated from peers, meeting places and social activities due to mobility incapacity. These problems are not likely to decrease as long as, no effective intervention is available, that aim to increase physical functioning, and minimise mobility disability in individuals with disabilities (Rimmer, 1999).

Several recent empirical studies (Mulligan, et al, 2004; Eagleton, Iams, McDowell, Morrison & Evans, 2004; Blundell, Shepherd, Dean & Adams, 2003; Damiano & Abel, 1998; MacPhail & Kramer, 1995), investigated the effects of strength-training on gait parameters and energy expenditure and distances walked by children with spastic CP. In addition, only 1 review on the effectiveness of strength-training programs for people with cerebral palsy has been published, which showed strength-training could increase strength and may be improve motor activity in people with CP without causing harm (Dodd et al., 2002). However, Dodd et al. (2002) acknowledged in their review to have evaluated the results of 8 empirical

studies, 1 review and 1 randomised controlled trial, all of low methodological quality. Therefore, systematic reviews literature on strength-training interventions to improve strength and movement capability of children and adolescents with CP is lacking.

Methods

The aim of this paper is to systematically review the current evidence to identify the effects of strength-training programmes in lower limb muscles and motor activity among children and adolescents with spastic CP.

Literature search strategy

A search of electronic databases (PEDro, Cochrane, EBSCOhost, Proquest, PubMed MEDLINE, Gateway, Health Technology Assessment, CINAHL, TRIP African Healthline, Science direct, Info-Trac, GALE Academic OneFile) was performed. No limit was assigned for year of publication. Combinations of the following terms were used in all databases: exercise, trials, children, youth, disability, treatment, strength training, spastic cerebral palsy, muscle strength, training program, therapeutic exercise, physical treatment, gait training, and physical function. A manual search was also done by examining references lists of all retrieved articles to identify additional relevant studies.

The initial search yielded titles and abstracts, which were then reviewed by the 2 reviewers for: sample population (children or adolescents with spastic CP), study design (randomly assigned to intervention), type of intervention (strengthening exercise program or progressive resistance exercise program) and outcome (improved motor functions, muscle strength and energy expenditure) review criteria. 36 selected articles were retrieved in full and where not available they were purchased through the inter-library loan.

Selection criteria: All randomised controlled trials (RCTs) evaluating strength training interventions outcomes on children equal or above 5 years and adolescents below 21 years with spastic CP were included. RCTs whose sample had participants with intellectual disability were excluded. Only studies available in full text and written in English were included. Because of the paucity of published literature in this sample; studies that used exercise as an intervention to improve gait and physical functioning were also included. Studies that met the inclusion criteria are summarized in the Table 2.

Data Extraction

The methodological quality of the included studies was rated using the PEDro scale criteria independently by the 2 researchers. The two reviewers extracted

data from the studies that met the inclusion criteria using a standardised extraction form.

Data Analysis

The PEDro scale criteria used in this review for methodological rigor has 11 criteria for evaluating randomised control trials (Table 1).

Table 1: Critical appraisal tool

<ol style="list-style-type: none">1. Specified eligibility criteria2. Randomly allocated participants3. Concealed allocation4. Participants prognostic similarity at baseline5. Participant blinding6. Therapist blinding7. Assessor blinding8. >85% follow-up for at least 1 key outcome9. Intention-to-treat analysis10. Between group statistical comparison for at least one key outcome11. Point estimates and measures of variability for at least one key outcome

All studies were independently assessed by the two reviewers. Disagreement between the two reviewers' final PEDro scores was resolved by consensus. The PEDro scale primarily measures internal validity and criteria 2 through 11 were used to provide the final scores according to PEDro guidelines for clinical trials (PEDro, 2007). According to Maher, Sherrington, Herbert, Moseley and Elkins (2003) and, Bhogal, Teasell, Foley and Speechley (2005) the PEDro scale has fair to moderate reliability, validity and is extensively applied to rate the methodological quality of physiotherapy clinical trials.

Results

The initial search in the electronic databases and the manual perusal of reference lists identified 36 studies. Of the 36, only four were RCTs; one was an abstract (McCubbin & Shasby, 1985) and attempts to retrieve the full article failed, three full text articles reported results of RCTs.

Table 2 summarizes the PEDro scores of the three RCTs (Liao, Liu, Liu, & Lin, 2007; Unger, Faure & Frieg, 2006; Dodd, Taylor & Graham, 2003) that were included in this systematic review. The total scores ranged from 6 to 8 out of 10 on the PEDro scale. The study by Dodd et al. (2003), investigating the effects of strength

training programs, achieved a total score of 8/10, that by Unger et al. (2006) achieved a score of 7/10 and the study by Liao et al. (2007) investigating the effectiveness of loaded sit-to-stand

resistance exercise achieved a score of 6/10. Therefore, all three trials are of moderate to high methodological quality. A total of 72 children and adolescents (5 to 18 years old) were recruited in the 3 trials.

Table 2: Summary of methodological quality of included RCTs

Study	PEDro Criterion											Total
	1	2	3	4	5	6	7	8	9	10	11	
Liao et al (2007)	√	√	√	√	X	X	√	X	X	√	√	6
Unger 2006	√	√	√	√	X	√	√	X	X	√	√	7
Dodd et al 2003	√	√	√	√	√	X	√	√	X	√	√	8

PEDro indicators 1-11 as reflected in Table 1

At least 2 trials (Liao et al., 2007; Unger et al., 2006) had a low compliance for two criteria that have been shown to raise bias in clinical trials (blinding of participants and therapists, adequacy of follow-up (see Table 3). All included trials reported participant withdrawals for various reasons that included fear of infection with severe respiratory syndrome, surgery or sports.

Participant dropout from each trial were as follows: Unger et al. (2006) 6 participants, 3 in control group and 3 in experimental group; Dodd et al. (2003) 1 participant in control group; Liao et al. (2007) 4 participants, 2 in control group and 2 in experimental group. According to Liao et al. (2007), children who dropped out were similar at baseline between both groups but demographically different (older, taller, and heavier) from those who completed.

With regards to demographic data, Dodd et al., (2003) reported that children in the experimental group had more physical disability compared to the control group ($P=.07$) but were similar at baseline for demographic data and key outcome areas. Unger et al., (2006) on the other hand reported that demographic characteristics of the control group significantly differed from the experimental group ($P=.02$) including the distribution of impairments ($P=.03$). Important to note is that some of the participants in the experimental group had hemiplegic CP and others had diplegic CP.

All the 3 trials blinded assessors at baseline and at end of interventions. Two trials indicated it used experienced therapists, 1 in pediatric assessment and 1 in movement disorders, while the other, (Unger 2006) did not state assessor's experience. Table 3 illustrates summary of

intervention designs, outcomes assessed and results for all the trials. It also indicates the settings, the samples of participants and the duration of each trial.

Muscle Strength

Dodd et al (2003) using lower limb strengthening exercises and the study by Liao et al (2007) using loaded sit-to-stand (STS) exercises to increase lower limb muscle strength, found significant differences [$F_{[1, 19]}= 4.58, P=.046$ and $F_{1,17}=17.7, P=.001$ respectively) between the experimental and the control groups. This is demonstrated by the large calculated effect size d equal to 1.78 (95% CI, 0.68-2.73) for STS (Liao et al., 2007) after a strengthening programme of 6 weeks. However, Dodd et al. (2003) reported the effect size to be low in their trial. An effect size of $d > 1.2$ is reported elsewhere as effective for increasing muscle strength (Dodd et al., 2003).

Motor activity

Liao et al. (2007) and Dodd et al.(2003) used the Gross Motor Function Measure (GMFM) as an outcome measure while Unger et al. (2006) used a 3-dimensional gait analysis and gait questionnaire. Liao et al. (2007) found a significant difference in the GMFM goal dimensions D and E scores ($F_{1,17}=4.81, P=.02$) between participants in the experimental group and the control group at end of the intervention. Similarly, Dodd et al. (2003) found a trend towards improvement in the

experimental group GMFM dimension E measurements compared to those of the control group at the end of 6 weeks intervention.

Dimension D and E of the GMFM relates to motor activities like jumping, walking, running, and hopping (Liao et al., 2007; Dodd et al.,2003). Liao et al (2007) found a marginal effect size d equal to 1.17 [95% CI, 0.68-2.73] for the GMFM score, while it was low in the trial by Dodd et al., 2003. Using a 3-dimensional gait analysis, Unger et al. (2006) found outcomes of crouch gait differed significantly for the experimental group from the control group ($P=.05$). This is reflected in the improved crouch gait in the experimental group. Unger et al. (2006) and Liao et al. (2007) also evaluated gait parameters: both found no significant change in gait speed had occurred at the end of their strength training programs. No trial reported adverse effects such as reduced physical functioning or joint range of motion.

Energy expenditure

Only 1 trial, Liao et al. (2007) measured the Physiological Cost Index (PCI) as an outcome. Their analysis yielded a moderate effect size d equal to 1.34 (95% CI, .32-2.25) of PCI at the end of the intervention, reflecting a beneficial reduction in energy expenditure after loaded STS strength training program among the participants.

Perception of body image and competence

Only 1 trial, Unger et al. (2006) investigated perception. Perception of body image scores of experimental group

differed significantly from the control group ($P=.01$), but there was no change in perception of competence between groups.

Table 2. Summary of interventions design features

Study	Setting	Sample	Design	Outcome assessed	Intervention	Duration	Results
Liao et al., 2007	Children's homes	20 Children (5 to 12 years old) with spastic diplegia CP	RCT	GMFM outcomes at baseline and at 6 weeks. Gait speed, -1-RM load STS exercise -Isometric strength Knee extensor - Physiological cost index	Loaded STS exercise 3 times a week Vs regular Physical Therapy: passive range of motion exercises, positioning, balance, functional and neuro-developmental training	6 weeks	GMFM goal dimension scores ($F_{1,17}=4.81, P=.02^*$), effect size of 1.17 (95% CI, 0.68-2.73.) -1-RM STS($F_{1,17}=17.7, P=.001^*$) effect size of 1.78 (95% CI, 0.68-2.73) and -PCI ($F_{1,17}=8.04, P=.005^*$) effect size of 1.34 (95% CI, .32-2.25). Follow-up = 80% - No significant difference in gait speed , and isometric knee extensor strength between the groups
Unger et al., 2006	School for children with special needs	31 spastic CP learners (13-18years old). 21 in intervention group, and -10 in control group	RCT	-Gait analysis, and - gait parameters and perceptions of body image and functional competence	I.D. circuit format strength training program (exercises for upper & lower limb & Trunk)	8 weeks	- measures of crouch gait differed significantly for the experimental group from the control group ($P=.05$) -Perception of body image scores of experimental group differed significantly from the control group ($P=.01$). -Follow-up = 83.78% -No change for stride length, velocity and frequency (cadence). -No significant difference in perception of competence between both groups
Dodd et al., 2003	A hospital	21 children and adolescents (8-18 years old) with spastic diplegia CP	RCT	Strength Dimension D & E of the GMFM timed stair test at own pace - -strength of ankle plantar flexors - knee extensors - hip extensors	Strengthening exercise 3 times week (targeted for -ankle plantar-flexor, Knee extensor -hip extensors)	6 weeks	-At 6 weeks experimental group LL strength significantly differed from that of controls ($F_{1, 19}= 4.58, P=.046$), & at 18 months ($F_{1,16}=6.25, P=.041$). -at 6 weeks experimental group GMFM dimension E measurements showed a trend of improvement (standing, running, jumping, and faster stair climbing) compared to those of the control group. -No significant interaction effect for individual muscle groups -21 completed baseline and six-week testing. 1 left due to surgery. Follow-up = 95.23%

- Significance was calculated at 5% level [$P<.05$] in each study. Abbreviations: CP: Cerebral Palsy, GMFM: Gross Motor Function Measure, RM: Repetition Maximum, STS: Sit-To-Stand, RCT: Randomised Controlled Trial, PCI: Physiological Cost Index, I.D: Individually Designed, LL: Lower limb.

Discussion

The purpose of this systematic review was to determine the effectiveness of strength training on lower limb function of children and adolescents with cerebral palsy. One systematic review (Dodd et al., 2003) has found that strength-training could improve physical functioning in people with cerebral palsy without causing any adverse effects.

Empirical studies (Eagleton et al., 2004; Blundell et al., 2003; Damiano & Abel, 1998; MacPhail & Kramer, 1995) have shown that strength-training improves functional outcomes (gait speed, muscle strength and physical functioning) and gait cadence (Damiano & Abel, 1998). However, very few trials have adequately assessed their effects, on muscle and motor activity outcomes, in children and adolescents with spastic CP.

Among the trials included in this systematic review, 1 trial (Liao et al., 2007) estimated that the strength effect sizes were significantly different between groups at the end of the strength training program, and of sufficient threshold to increase muscle strength. However, Dodd et al. (2003) found this were low in their trial.

There is reason to suspect bias in the results in the former trial given the fact that it had not controlled three key confounding factors (scored 1/3 for blinding, and 20% dropout) compared to the latter (Dodd et

al., 2003), that had less than 5% dropout rate and scored 2 out of 3 on blinding. Other than this methodological bias, the results apparently indicate a standardised STS (90° hips flexion, 105° knee flexion and 15° ankles dorsiflexion) outlined by Liao et al. (2007) may yield more effectiveness than squatting and stairs climbing (Dodd et al., 2003) in increasing lower limb muscle strength.

This finding may have clinical implications on the specificity of applied treatment techniques, intended for higher client outcomes, and improving accountability of therapist to clients and other stakeholders. From the perspective of this review, another major limitation of the trial by Liao et al. (2007) was the failure to standardize the frequency of intervention for all participants entered in the trial and the enthusiasm caregivers.

A significant finding is the effect of increases of muscle strength which appears to improve motor efficiency. This is reflected in changes on GMFM goal dimensions analysis (Liao et al., 2007; Dodd et al., 2003). Motor efficiency sustainability in spastic CP children is a controversial area of knowledge among clinical therapists.

This finding may be clinically relevant. However, the trials are too small to allow interpretation of their results. The use of diverse tools to assess clinical outcomes

may have some implication of transferability of the trial results to clinical settings. The study by Unger et al (2006) used a 3-D gait analysis video recording system which may only be available in specialised research laboratory settings, meaning this test may be difficult to replicate in a clinical setting compared to the other 2 trials which assessed gait using the GMFM goal dimensions. A major criticism of all the trials is that none assessed the cost of their trial relative to clinical practice. In the era of decreasing healthcare funding from states, any new research must address the issue of costs of interventions, stating the cost-effectiveness.

The selection of outcome measures in all trials (Liao et al., 2007; Unger et al., 2006; Dodd et al., 2003) is questionable, as all the studies disregarded the notion of specificity of their training. In all cases the selected outcomes do not directly relate to the interventions and may never have changed following such interventions. For example: Liao et al. (2007) used loaded STS for lower limb strengthening, yet measured gait speed, and energy expenditure as outcomes; Dodd et al. (2003) used exercises targeted at improving strength of extensors of hips and knees and ankle plantar-flexors, but measured for change in motor activities like walking cadence, running and jumping. Similarly, Unger et al. (2006) applied a combination of resistances, to the upper and lower limbs to promote

muscle strength, but measured gait (length, velocity and cadence) and perceptions of body image and functional competence as outcomes. Basic strength training exercises focused on strengthening specific postural muscle groups especially in the lower limbs may not necessarily increase gross motor functioning measured as outcomes in these studies. Specific task-oriented activity training may have been more appropriate, were it integrated in trials to facilitate integration of strength, with balance and coordination components, that normalize dynamic movements.

This systematic review advances three reasons as the basis to question the statistical analysis used in the 3 trials as follows:(1) the fact that 2 trials had inadequate follow-up; (2) it was acknowledged by Unger et al. (2006) and Dodd et al. (2003) that participants were allowed to participate in sports and other forms of physiotherapy rehabilitation during the intervention period, yet this were not analysed as covariates; and (3) social contact between the experimental group and the control group might have occurred in all the trials, either at the laboratory during assessments, school or in the community. If this happened no full-proof treatment controls could have been achieved. Thus, no-true control, could have introduced bias in these trials. Finally, none of them explicitly used intention-to-treat analysis. This could have improved the interpretability of the results.

Conclusion

In conclusion, the trials reviewed were of moderate-to-high methodological quality. Although they had design flaws, such as, using very small samples, which limit the interpretation of their results, missed to demonstrate that strength-training improves strength of targeted muscle and motor functioning. More rigorous trials on larger samples should evaluate the impact of specificity in strength-training on muscle and motor activity as well as cost-effectiveness of programs.

Implications for practice

Available evidence is insufficient to support the use of strength-training programs for specific muscle groups, in the rehabilitation of children and adolescents with CP.

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