There is little evidence on the effect of upper limb strengthening in children with cerebral palsy

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CLINICAL SCENARIO: A growing body of evidence supports the use of strengthening in children with cerebral palsy (CP). Muscle weakness is a secondary consequence of cerebral palsy and impacts not only on a child's ability to move but on their ability to participate fully in activities of daily living. It is well documented that children with CP do not move as much as their typically developing peers and as such their muscles do not just atrophy but fail to develop normally (Damiano, 2008). Strengthening programs applied to children with CP are based on guidelines developed for children without disability. These guidelines, applied to lower limb strengthening in children with CP, are effective and safe, and evidence suggests that strengthening programs can improve motor activities as well as strength. Investigation of outcomes following upper limb strengthening programs is warranted. Clinical best practice guidelines are also required to guide clinicians currently implementing these programs.

FOCUSSED CLINICAL QUESTION: What is the evidence that upper limb strengthening is effective in increasing active upper limb range of motion, upper limb function and performance of activities of daily living in children with CP?

SUMMARY of Search, 'Best' Evidence' appraised, and Key Findings:

- Only three studies (pre/post) of upper limb strengthening were found (one in combination with electrical stimulation) (Lee et al., 2009, O'Connell et al., 1995; Vaz et al. 2008). Consequently the search was extended to include studies involving the lower limb.
- Two systematic reviews of strengthening, mostly pertinent to the lower limb were located and included in this CAT (Dodd et al., 2002; Verschuren et al., 2008).
- Some improvement in wrist strength was achieved with combined electrical stimulation and isometric resistance, but no functional gains (Vaz et al, 2008).
- Upper limb strength and wheelchair propulsion improved following progressive upper limb resistance training (O'Connell & Barnhart, 1995).
- Muscle strength, bulk and motor function improved following a novel strengthening technique (Lee et al, 2009).
- Two systematic reviews provided some evidence that strength training may be effective in improving lower limb strength (Dodd et al, 2002; Verschuren et al, 2008) but there was limited evidence for other outcomes.
- Due to the paucity of evidence on upper limb strength training in CP, we located literature regarding strength training for typically developing children. We used this literature to develop best practice guidelines in CP
- Functional outcomes of strength training have yet to be adequately evaluated

CLINICAL BOTTOM LINE: There is scant evidence on the effect of upper limb strength training in children with CP. Strength training appears to be safe and does not increase spasticity. .

Limitation of this CAT: This critically appraised paper (or topic) has been peer-reviewed by three other independent people.

SEARCH STRATEGY:

Terms used to guide Search Strategy:

- **P**atient/Client Group: Cerebral palsy, children, paediatrics
- **I**ntervention (or Assessment): Strengthening, strength training, resistance training or weight training in combination with upper limb, upper extremity, arm, hand, fingers, shoulder or elbow.
- **C**omparison: nil
- **O**utcome(s): nil

Databases and sites searched	Search Terms	Limits used
CINAHL EMBASE MEDLINE OTseeker PEDro Cochrane Library	Strengthening, strength training, resistance training or weight training upper limb, upper extremity, arm, hand, fingers, shoulder or elbow	Preschool child 2 – 5 years or child 6 to 12 years or adolescence 13 to 18 years

INCLUSION and EXCLUSION CRITERIA

- Inclusion: Studies that reported on upper limb outcomes of strengthening programs in children with CP. Only two studies relevant to upper limb strengthening were located. Consequently, criteria were broadened to include studies of lower limb strengthening in children with CP.
- Exclusion: Studies where strengthening was only one component of the intervention or where the study was already included in one of the systematic reviews included in the CAT.

RESULTS OF SEARCH

Searches were conducted in July 2008. One additional study was also located in early 2009 (Lee et al, 2009) Three studies relevant to upper limb strengthening and two systematic reviews of strength training and exercise programs predominantly of the lower limb which met the inclusion and exclusion criteria were located and included in the CAT. The included five studies, deemed to represent best evidence for this CAT are listed in Table 1 and summarised in Tables 3 to 6. Three other studies were identified but not included in this CAT as they were included in the two systematic reviews or were not sufficiently specific to the topic of upper limb strengthening to warrant inclusion are listed in Table 2.

Table 1: Summary of Study Designs of Articles retrieved

Study Design of Articles Retrieved	Number Located	Author (Year)	Reason for inclusion
Systematic review	2	Dodd, Taylor & Damiano (2002) Verschuren, Ketelaar, Takken, Helders & Gorter (2008)	Two articles representing highest level of evidence mostly on lower limb strengthening in CP.
Pre test / Post test, single group design	2	O'Connell & Barnhart (1995) Vaz et al (2008)	Two articles specific to upper limb strengthening in CP.
Case study (n=1)	1	Lee, You, Lee, Oh & Cha (2009)	Strength training used with a child with CP.

Table 2: Potential articles located but excluded

Author (Year)	Study Design of Articles Retrieved	Reason for exclusion
Verschuren, Ketelaar, Gorter, Helders, Cuno, Uiterwaal & Takken (2007)	RCT	Strengthening was only one component of the intervention
Darrah, Wessel, Nearingburg & O'Connor (1999)	Pre test / Post test design	Strengthening was only one component of the intervention
Lockwood (1996)	Pre test / Post test design	Mean age of participants was 29.5 years.

SUMMARY OF BEST EVIDENCE

Table 3: Summary and appraisal of Study 1 (Vaz et al, 2008)

Aim/Objective of the Study: Evaluate the effects of resistive exercises coupled with electrostimulation on wrist flexor and extensor strength, passive stiffness and hand function in children with hemiplegic cerebral palsy.

Study Design: Single group pre test/post test design

Setting: Brazil

Participants: Nine children with spastic hemiplegic cerebral palsy aged 7 – 11 years (mean age 9yrs 1 mth), 5 participants were female and 5 had left hemiplegia. A convenience sample was used. All participants used their hemiplegic hand as an assist (MACS Level I = 1, MACS level II = 8). All could actively extend their wrist to 30° extension but used hand predominantly in flexio n.

Intervention Investigated: 24 sessions (3 sessions/week for 8 weeks) of wrist muscle strengthening (extended wrist range) aided by electrostimulation (FES). Electrodes were placed over bulk of wrist flexors and extensors and distally next to wrist. FES intensity increased gradually until visible contraction was obtained (300μs pulse width, 30pps frequency, rise and decay of 3 seconds and ON time of 5 seconds). Therapy was provided by physical therapists who offered maximum resistance against wrist flexion and extension with the wrist positioned in the extended range only, according to each child's capability. Movements did not occur in neutral or flexion ranges. All children received intervention (ie no control group or phase)

Outcome Measures
- Measures were taken at baseline and one week after completion of intervention. Data collectors unknown
- Isometric strength tests of wrist flexors and extensors – performed in wrist 30° flexion and 30° extension and neutral. Limb was sta bilised during assessment and a 5 second isometric contraction was measured by Microfet-2 dynamometer.
- Passive stiffness assessment measured by Biodex System 3 pro isokinetic dynamometer plus EMG monitoring.
- Manual dexterity measured using 3 tasks from Jebson-Taylor Hand Function Test (tasks adapted as too hard for some children to complete) Score range (0-120 seconds)
- Wrist angle during manual activity measured from 3 D positional data using a Qualysis ProReflex MCU movement analysis system. Participants had to pick up 3 items.

Main Findings: At baseline the isometric strength of both extensors and flexors was significantly lower in the extended wrist position than in the neutral and flexed position.
- Extensor strength increased significantly in the extended and neutral positions but remained lower than in the flexed position.
- Flexor strength increased significantly in the extended position but did not change in the neutral or flexed position. The strength in the extended position remained lower than in the neutral or flexed positions.
- No changes were noted in passive stiffness, the wrist angle used for function or in hand function.
- All children continued to function with their wrist in a mean of 21° of wrist flexion following the intervention (this result has not been found in LL studies where various strengthening programs have resulted in a change in the angular position of the ankle and/or knee).

Original Authors' Conclusions*:* The study hypothesis was realised, following a specific intervention protocol the participants' demonstrated increased strength in wrist flexors and extensors in the extended wrist position and neutral position. Strength in extension remained lower than other positions. Functional gains were not noted and in order to generalise the study outcomes to improved function, the authors concluded that specific task training would be necessary. The authors commented that available evidence indicates there is not always a direct link between impairment and activity levels and thus decreasing the original impairment does not always lead to improved function.

Critical Appraisal:

Validity: There is no justification for sample size or any information about selection of participants except general exclusion criteria. Participants were MACS levels I and II thus were more able children with hemiplegia. Sample size was small and study participants were not randomly selected. Little information is provided on treatment with no detail on length of sessions, number of resistance repetitions or the reliability of the maximum resistance provided by physical therapist. No information was provided on session attendance or follow-up attendance. Follow-up measures were completed only 1 week following treatment and no longitudinal follow-up data were provided regarding maintenance of gains made. Data collectors and their blinding status were not described. A strength of the study is that the test retest reliability of all outcome measures was established as high (ICC = 0.81 to 0.99) in a pilot study.

Interpretation of Results: Statistically significant differences were identified following intervention for wrist extension in the extended and neutral positions and flexors in the extended position (using paired t-tests and $p<0.05$). However, the 95%CI overlapped between baseline and post-intervention and the lower limits of the CI for the differences between means was quite small (eg 0.96N, 1.81N, 1.42N). This suggests to us, that in combination with a lack of measurement of functional outcomes, the changes are not clinically important. No attempt was made by the authors to provide guidance on the clinical importance of the magnitude of the changes.

Summary/Conclusion: This study reported statistically significant increases in muscle strength in targeted but weaker wrist muscles in the extended position following intensive resistive strengthening exercise coupled with electrostimulation. However confidence in these results is weakened following consideration of the overlapping 95% confidence intervals. It is not possible to identify the specific contribution the strength training component of the intervention made to these findings. The results indicating increases in strength are consistent with the literature where gains are found following 8–12 week strength training programs (Vehrs, 2005b).

The increase in strength however did not translate to functional gains or improved position of the wrist during functional activity. The literature recommends that strengthening exercises are carried out over the entire joint range of motion (Council on Sports Medicine and Fitness, 2008). Adherence to this principle, which may be difficult for children with CP, may have influenced results. As few details of the actual intervention are provided it is not possible to use this study to draw clinical applications from the study.

Table 4: Summary and appraisal of Study 2 (O'Connell & Barnhart, 1995)

Aim/Objective of the Study: To determine the effects of an 8-week upper body concentric resistance training programme on wheelchair propulsion in children with cerebral palsy and spina bifida.

Study Design: Pre test/post test design **Setting**: USA

Participants: Six children, 3 with spastic CP (2=quad, 1-diplegia) and 3 with spina bifida (Lesions – T8-12), aged 4 to 16 years (mean = 10yrs) with wheelchair as their primary means of mobility.

Intervention Investigated: Children completed 3 sets of six-repetition maximum (6RM) antigravity, progressive resistance, strengthening exercises using cuff weights, dumbbells or barbells in eight upper limb movements/muscle groups, 3 times per week for 30mins in a supervised circuit situation.

Outcome Measures: Measures were completed by the authors (physiotherapists) at baseline and during the final, 8th week of training. Two wheelchair propulsion tasks: i) 50m wheelchair dash (time to complete, measures strength/explosive power), ii) 12min test (distance travelled, measures muscular and cardiovascular endurance). Strength was also measured (6RM, in kg in eight upper limb muscle groups).

Main Findings: Distance covered in 12 mins increased significantly from 259.56m (SE 77.41) at baseline to 334.7m (SE 70.62) at week 8, a change of 29% (146m; p=0.031). Strength increased significantly (6RM) for all 8 muscle groups (p=0.018 to 0.031). Fifty metre dash time decreased by a non-statistically significant 20.2% (approx 27.78 secs).

Original Authors' Conclusions*:* Progressive resistance exercise training improves muscle strength and wheelchair performance in selected children with disabilities. Consideration should be given to using resistance training to enhance and maintain wheelchair propulsion abilities.

Critical Appraisal:

Validity

As this is a pre-post study without control group, it cannot be determined whether results are related to the strength training programme or other variables. The convenience sample size (n=6) was very small, with only 3 children with cerebral palsy, the population of interest for this CAT. These factors restrict the generalisability of the results. The outcome measures were well described and although no psychometric properties were reported they are well-referenced. Likewise, the intervention and setting was adequately reported. It appears all participants had complete follow up but attendance of training sessions was not reported. A longer term follow-up would be desirable to determine the maintenance or otherwise of the strengthening programme.

Interpretation of Results: The results do suggest important improvements in wheelchair propulsion over 12 minutes and a trend for improvements in time to travel 50m. Strength improved significantly. The authors did not comment on the clinical significance of the increase in mean distance covered in 12 mins (146m). It is unclear whether this would translate to a meaningful reduction in wheelchair travel time in children's daily lives.

Summary/Conclusion: This upper limb strengthening programme enhanced the ability of this small group of wheelchair-using children to propel their wheelchair. The muscles strengthened were specific to the task of wheelchair propulsion and the outcome measure was specific to the goal of strengthening (wheelchair propulsion). This is one of few examples where strengthening has led to enhancement of functional ability. The intervention had some of the key, identified features of strengthening programmes: progressive resistance, completed three times a week over 8 weeks, with supervision to ensure correct technique. These results need to be treated cautiously due to the small sample size.

Table 5 Summary and appraisal of Study 3 (Lee at al, 2009)

Aim/Objective of the Study: Evaluate Comprehensive Hand Repetitive Intensive Strengthening Training (CHRIST) on morphological changes in muscle size and motor improvement in a young child with CP.

Study Design: Single case study, pre-test/post-test design **Setting**: South Korea.

Participant: 4.9 year old female with right hemiplegic CP.

Intervention Investigated: The participant completed a 5 week course of CHRIST involving slow arm stepping on a treadmill whilst child was suspended in prone above the treadmill. CHRIST swing phase involved maximal elbow and wrist extension and 160° shoulder flexion; and the arm stance phase inv olved weight bearing through elbow onto palm and MCP heads with shoulder in 90° flexion. Intervention was provided for 60 minutes per day (10 minutes of exercise and 3 minutes rest), 5 days per week for 5 weeks. Neuromuscular Electrical Stimulation (NMES) was provided to targeted muscles (extensor carpi radialis and triceps) for first 2 weeks to evoke maximum possible wrist and elbow extension. NMES discontinued after 2 weeks as full elbow extension achieved.

Outcome Measures: Measures were completed at baseline and following intervention (5 weeks post baseline) by an OT and radiologist blinded to the study. Upper extremity tests included: modified Wolf Motor Function Test (WMFT) to assess functional upper limb gross motor skills; modified Jebsen-Taylor Hand Function test to assess fine motor skills and a modified Pediatric Motor Activity Log (PMAL) to assess amount of use and quality of movement during activities of daily living. Ultrasound imaging of the right extensor carpi radialis and triceps muscles at rest and during maximum contraction measured cross sectional area of the muscles. Muscle strength was measured using a hand held dynamometer.

Main Findings:

	MAS	WMFT	Jebsen		PMAL		ECR	Triceps
			Right	Left	AOU	QOU		
Pre-CHRIST	2	2.3	83.3	30.13	1.05	1.3	5.56	7.78
Post-CHRIST	0	3.6	67.74	27.73	2.6	3.3	12.23	17.29

MAS: Modified Ashworth Scale (0-4), WMFT: modified Wolf Motor Function Test (modification to scale not specified) Jebsen: modified Jebsen-Taylor Hand Function Test (timed in seconds), PMAL: modified Pediatric Motor Activity Log (modification to scale not specified), AOU: amount of use, QOU: quality of use, ECR: extensor carpi radialis muscle.

Original Authors' Conclusions:

The study used percentage change to argue that CHRIST, an innovative neurorehabilitation technique can improve muscle strength, muscle bulk and motor function in the upper limb of a child with hemiplegic cerebral palsy. These results cannot be generalised to all children with cerebral palsy and additional investigation with a larger cohort is required.

Critical Appraisal:

Validity: As this is a pre-post-test case study without any control comparisons, it cannot be determined whether study results are solely related to the strengthening programme or other variables including the NMES. This restricts the generalisability of the results. Inadequate description of the study participant also restricted the generalisability of the results, for example, the status of upper limb function, cognitive ability and motivation of the child were not given. Further, the feasibility of this intensive intervention in other health systems, and which presumably would require a motivated child and family, needs consideration.

There was inadequate description of how each of the clinical tests had been modified for this study. In addition the authors claimed each of the measures had established validity and reliability, however no research investigating the psychometric characteristics of these modified versions of the tests in paediatric CP have been published. There was no evidence of reliability or validity of the isometric muscle strength measured using the dynamometer. Caution needs to be used when interpreting all the results of this study. A strength of this study was use of blinded assessors to measure outcomes.

The intervention was adequately described. The intervention was only provided for 5 weeks, a period somewhat shorter than the 8 to 10 weeks recommended in the strengthening literature, although intervention was more intensive than is recommended in the same literature. The impact of NMES on the outcomes cannot be determined. Longer term follow-up would be desirable to determine the maintenance or otherwise of the CHRIST programme.

Interpretation of Results: The results do suggest positive changes, not only in muscle size/bulk and in the strength of the targeted muscles, but also in functional upper limb use following the intervention. In particular there was an increase in the amount and quality of use of the affected upper limb. An additional benefit described was the development of a protective extension reaction in the affected upper limb following the treatment. Caution needs to be used when interpreting these results due to the limitations of the outcome measures noted above and also that the size of the scale for each measure was not provided. This is required in order to allow decisions to be made about the clinical significance of the results.

Summary/Conclusion: The study results indicate the CHRIST may improve muscle strength and functional use of a hemiplegic upper limb in a child with cerebral palsy. The generalisability of these results is limited. The long term impact of such an intensive intervention is not clear due to a lack of longitudinal follow-up data.

Table 6. Summary and appraisal of Study 4 (Dodd et al, 2002)

Aim/Objective Systematic Review: To determine whether strength training produces beneficial outcomes for people (adults and children) with CP.

Study Design: Systematic review

Methods of the Review
Search strategy: Relevant databases were searched to 2000. Search terms were selected to represent cerebral palsy, exercise, strength and physical training. Reference lists of were scanned and related links of PubMed explored. Non-English language articles were excluded.

Inclusion criteria: Articles which measured the outcome of upper limb or lower limb strength training or progressive resistance exercise programmes in adults, children or adolescents with cerebral palsy were eligible for inclusion. Two reviewers determined eligibility of articles and also rated methodological rigour using the PEDro scale. Articles scoring less than 3 were excluded from the review. Review articles were eligible for inclusion if they identified primary studies and detailed criteria for selecting, and assessing the quality of studies. The numbers of review authors extracting data from included studies were not specified.

Main findings: Ten studies and 1 review formed the basis of this systematic review.

The included review article evaluated the effects of progressive resistance exercise in children with CP. It concluded that there was a low level of evidence supporting strengthening exercises in children with CP for increasing strength, no detrimental effects and unclear effects on function.

Only one of the 10 studies was an RCT, the remainder were case series (total n for diagnosis of CP = 114, range = 5-30; age range = 4 to 47 years; PEDro score median = 4; range = 3 to 6). This review calculated effect sizes with 95% confidence intervals to compare results between studies. These results were graphically presented but the numerical data were not specified.

Impairment level outcomes
Eight of the 10 included studies reported increases in strength including the RCT (n=30, PEDro score = 6/10). The two studies that reported no difference after strengthening were poorly powered with a sample size of n=6. Few studies evaluated spasticity or ROM but those that did inferred no negative outcomes resulting from strengthening.

Activity level outcomes
Four of the 10 studies evaluated activity level outcomes. These activities included Gross Motor Function Measure scores, walking speed and wheelchair propulsion. The effect sizes appeared smaller than those for strength related outcomes and only 3 of the 7 variables measured reached significance. None of the studies evaluated outcomes at the level of participation.

Exercise program characteristics
There was very wide variability in both participant characteristics (eg age, severity, topographic classification of CP) and characteristics of the strength training

programmes. The training was generally performed 3 times per week over 6 to 10 weeks and there were mechanisms to adjust resistance. However, the muscle groups trained, the intensity and duration of the programme, equipment, exercise detail, method to determine amount of resistance and outcome measures were highly heterogeneous. Six of the programmes were individually administered, one was a group programme and the remainder were not specified. Three were conducted in combined home/clinic setting and one in a community gym.

Studies of Upper Limb Strengthening
This review did not attempt to separate studies to evaluate the outcomes of upper limb strengthening. Four of the studies completed upper limb strengthening, one was the RCT and one other evaluated both upper and lower limb strengthening (total n for diagnosis of CP = 62, range = 3-30; age range = 7 to 47 years; PEDro score median = 4; range = 3 to 6). Three of the 4 studies reported substantial increases in strength. Only one study examined activity level outcomes. This study which includes only 3 (of 6) children with CP evaluated a programme to increase wheelchair mobility. Favourable improvement was reported for a wheelchair endurance test and a non-significant trend for time to decrease over a 50m wheelchair dash. See Table 3 for full details of this study.

Original Authors' Conclusions:
When the studies included in this systematic review were viewed collectively there was an indication that strength training may have positive strength benefits for people with CP. Due to the methodological weaknesses and heterogeneity of participants and programme characteristics, however, there was insufficient evidence to make definitive recommendations. There appears to be a smaller, less clear effect of strength training on activity level outcomes and no studies measured participation. There was no evidence that strength training increased spasticity or contractures in people with CP. Similar conclusions can be drawn when the studies which included upper limb strengthening are examined.

Critical Appraisal: The methods of this review were carried out appropriately for systematic reviews. Strengths of the review were that two reviewers completed quality rating although it is not clear whether this also occurred for data extraction. The review authors calculated effect sizes and 95% CI to allow for comparison of the size of the treatment effect across studies. A meta-analysis was not possible due to the heterogeneity of study characteristics and particularly outcome measures.
Most of the lower level studies and the RCT reported positive outcomes for strength. There was a trend for improvement in activity level outcomes. No study reported an increase in spasticity as a result of strengthening. Specific outcomes for children were not able to be identified from this review.

Conclusions of CAT authors: This systematic review, published in 2002 (searches conducted in 2000) suggested there was a low level of evidence that strength training may be effective in increasing strength in people with CP. Although a trend existed (on the basis of case series) there was limited evidence for the effects of strengthening on activity level outcomes. The strengthening programmes were highly heterogeneous, except for the frequency of programmes (whether individual or group based) were completed 3 times per week over 6 to 10 weeks and the load/resistance was adjusted.

Table 7 Summary and appraisal of Study 5 (Verschuren et al 2008)

Aim/Objective of Systematic Review:
To determine i) what exercise programmes focusing on lower-extremity muscle strength, cardiovascular fitness or a combination have been studied and their effects in children with CP, ii) the outcome measures used in included studies and iii) the methodological quality of the studies.

Study Design: Systematic review

Methods of the Review
Search strategy. Relevant databases were searched to September 2006. Search terms were selected to represent cerebral palsy, exercise, the lower extremity and clinical trials.

Inclusion criteria: Articles which measured the outcome of interventions were eligible for inclusion. The authors expected to find a substantial number of articles which were not randomised controlled trials (RCT) thus precluding meta-analysis. This review therefore was a descriptive review. Inclusion criteria also specified studies where participants were children or adolescents with cerebral palsy, the intervention was exercise focusing on lower extremity strengthening, cardiovascular fitness or a combination, and outcome measures were across the domains of the International Classification of Functioning, Disability and Health. Excluded were studies reported in doctoral dissertations, books or conference proceedings and studies where participants included children with other diagnoses as well as cerebral palsy.
Data extraction and rating of methodological quality was completed by three reviewers for each article. Included trials were classified as those addressing lower-extremity strength training, aerobic training or mixed. There were no articles addressing anaerobic training. The outcome measures used in the included studies were classified according to the ICF. Quality was rated using the PEDro scale and each article was graded according to the system developed by the American Academy for Cerebral Palsy and Developmental Medicine.

Main findings:
There were 20 included studies. Eleven studies addressed strength training, 5 aerobic training and 4 evaluated mixed training interventions. *As strength is the focus of this CAT only material pertaining to the evidence about strength training will be reported.*

The authors of the review reported that the 11 articles addressing strength training in the lower limb consisted of 5 Level 1 RCTs (PEDro score median = 7, range = 5 to 8) and 6 level IV studies (before and after case series without a control group). The median PEDro score across all 11 studies was 3 (range 0 to 8). It is important to note however that one of the trials which the review authors report as being an RCT (McBurney) reports qualitative information gathered from only the strengthening (not the control) group of an RCT and thus does not comprise Level I evidence. The evidence from this study will therefore be not included in the discussion of RCT evidence below.

The total number of participants was 170 (range 5 to 39) and the age range was 6 to 20 years. The classification of CP of the participants was not reported in this review. Exercise frequency for all studies was 3 times per week over varying periods (6 weeks

= 7 studies, 8 weeks = 3 studies, 9 months = 1 study). Two programmes used group intervention and the remainder were individually supervised.

All studies reported results in the body function and structure domain, 8 in the activity domain and one at the level of participation. The domains evaluated at the body function and structure level included: various measures of strength (n=7 studies), spasticity/hypertonicity (n=3), energy expenditure (n=4), VO_2 measure (n=1) and self perception (n=2). The Gross Motor Function Measure (n=5), various walking and stair climbing tests (n=3) and gait analysis (n=2) were used at the activity level of the ICF. A semistructured interview developed for the purpose of measuring participation in school, leisure, family and social events was used in one study.

Of the RCTs one reported small improvements in one measure of muscle strength (combined) for the strength training group when compared with the control group (Dodd 2003). In another RCT enhancements of body image and upright posture were reported (Unger 2006). Dodd 2004 and Patikas 2006 reported no differences between groups except a decrease in specific areas of self perception (scholastic competence and social acceptance) in the strengthening group (Dodd 2004). The Level IV studies generally reported increased strength from pre-test to post-test, no difference in spasticity and mixed results for energy expenditure and function (eg walking, stair climbing). See Table 8 for a summary of these results.

Original Authors' Conclusions: Methodological quality and level of evidence of included studies (5 RCTs and 6 pre-post studies) was low, however the review authors concluded that children with CP may benefit from lower extremity muscle strengthening programmes. More evidence is needed however to determine whether training makes substantial or sustained impact on performance of daily activities, participation, self-competence and quality of life.

Exercise studies varied greatly in the type of training programme evaluated. Based on the existing evidence, a strength training programme is suggested to be three sessions a week for a minimum of 6 weeks. There is little evidence to guide other components such as mode, intensity, setting, supervision, duration and type of activities in the programme. These findings are similar to three previous systematic reviews on the topic.

Although no studies compared aged related response, based on the broad age range of children across the various studies, review authors concluded that strength seems to be trainable in children of all ages. Longer term gains from strengthening are unknown. Review authors also concluded that outcome measures should be selected which are specific to the focus of the intervention and there is also a need to determine whether strengthening impacts on participation in children with cerebral palsy.

The reason for discrepancies in the effect of strengthening on aspects of self-perception is unclear, but perhaps related to the mode of programme delivery. Dodd et al. (2004) for instance, delivered intervention individually to children whereas Unger et al. (2006) reported increases in self perception following group based programmes.

Critical Appraisal: The methods of this review were carried out appropriately for systematic reviews. Strengths of the review were three reviewers completing quality rating and data extraction and the efforts to grade the quality and strength of evidence for each study. The exclusion of research published in dissertations and conference abstracts may have resulted in valuable evidence being missed. There was no mention of contacting authors or experts, reviewing reference lists of located papers or whether non-English language research was eligible for inclusion.

Although the authors reported that 5 RCTs were included, one study by McBurney et al. (2003) reported data obtained from the experimental group alone therefore the data cannot be classified as Level I evidence. The results of this study of 11 children used interviews of parental perception of outcomes. In comparison with the RCTs included, this study reported more positive results so consideration by the reviewers of this study as an RCT may have biased their conclusions drawn from RCT evidence.

Pooling of data for meta-analysis as none of the RCTs used common outcome measures. There was no attempt by the review authors to appraise the results of each trial in terms of the magnitude and significance of the results, thus no ability to determine the precision of the results.

Most of the Level IV papers but few of the RCTs reported positive outcomes for strength. No study reported an increase in spasticity as a result of strengthening.

The authors' conclusions, that children with cerebral palsy may benefit from strength training is largely drawn from lower level studies.

Conclusions of CAT Authors: Although this review covered studies of children with CP, the focus was on lower limb strengthening. It was included in this CAT due to the paucity of specific research targeting upper limb strength training, with the intent of cautiously extrapolating results to guide clinical decision-making relating to upper limb strengthening. The review provided a low level of evidence suggesting strengthening may be effective in increasing strength. The evidence for activity level outcomes is inconclusive and there is insufficient evidence for participation related outcomes.

One common thread amongst the studies was that the exercise regime most frequently evaluated was completed 3 times per week over a minimum of 6 weeks.

Table 8: Summary of results of RCTs on the effect of strengthening from Verschuren et al systematic review

RCT	Statistically significant change in strengthening group vs control (Yes/No)			
	Strength	Spasticity	Motor function	Self perception
Dodd 2003	No Yes for combined muscle strength		No	
Dodd 2004	No			Decrease
Patikas 2006		No	No	
Unger 2006			Yes - upright posture No - gait	Yes

IMPLICATIONS FOR PRACTICE, EDUCATION and FUTURE RESEARCH

This CAT explored the evidence for strengthening as a clinical intervention for increasing functional use of the upper limb in children with CP. Five publications were included and appraised.

The evidence is scant. The conclusion drawn from examining the literature, which is derived predominantly from studies of the lower limb, is that there is level V evidence (Oxford Centre from Evidence-based Medicine Levels of Evidence, 2009) suggesting that strengthening programmes may increase strength in children with cerebral palsy. There was no adverse increase in spasticity arising from strength training. There is insufficient evidence to draw conclusions about the effects of strength training on activity or participation level outcomes. Further, there was little definitive information guiding the assessment of upper limb strength or specific strength training programme development. Implementation of upper limb strengthening programmes, therefore, should be carefully evaluated with a view to capturing strength related outcomes and the impact of strengthening on relevant functional outcomes.

The American Academy of Pediatrics (AAP; Council of Sports Medicine and Fitness, 2008) and the American College of Sports Medicine (ACSM; Faigenbaum & Micheli, 2002) publish policy statements guiding strength training in children. General consensus on the definition of strength training, also referred to as resistance training, is a systematic programme of exercises aimed at increasing an individual's ability to resist or exert force (Council on Sports Medicine and Fitness, 2008; Verhs, 2005b). Strength training is often part of a general fitness programme aimed at developing health, fitness, wellbeing and performance.

Drawing together information from AAP and ACSM, other expert recommendations (Vehrs, 2005a & 2005b) and the findings from the articles included in this CAT, we identified the following issues for consideration in recommending and designing upper limb strengthening programmes for children with CP.

- The recommendations for strengthening programmes which accompany general fitness programmes, state that all muscle groups should be exercised including the core (abdominals, lower back, and gluteals; and the flexibility of quads and hamstrings should be assured) (Council on Sports Medicine and Fitness, 2008). However, when considering children with CP, it is unclear whether all muscle groups should be considered for inclusion in an upper limb strengthening programme or whether strengthening should be focused on particular muscle groups which demonstrate relative weakness. Should weak antagonist muscles be targeted within an affected limb, for instance, or should an entire hemiplegic upper limb be the focus of strengthening? The specific issue of grip strength, assumed to be a critical component of upper limb functional use has not been addressed in the literature but needs to be considered.
- The muscles and movements targeted by a strengthening programme should be specific to the movement patterns required for achieving functional goals. (ACSM).
- Strength training programmes have been delivered in individual or group settings. It is unclear about the relative effectiveness of these modes of delivery. Group programmes should be organised into similar age groups (Verhs, 2005b)
- Programmes may start approx 7 to 8 years of age (AAP), although the ACSM also states that children who are able to accept and follow directions and who are ready for organised sports could undertake strengthening programmes (Verhs, 2005b).

This age guideline may be reconsidered for younger children when the upper limb is the sole focus of the strengthening programme.

- Proper technique is mandatory. No load or resistance should be used until the correct technique of each exercise is mastered (Council on Sports Medicine and Fitness, 2008; Verhs, 2005b). Fatigue should be avoided as it may result in incorrect technique and injury. Light loads which are carefully increased over time can be added when technique is correct and can be maintained. Single joint exercises should be learnt well at the onset of the programme after which multiple joint exercises can be added which may improve coordination and skill. Children may take some time to learn correct technique. This may be especially so with children with CP where motor control is variable and experimentation may be required to determine appropriate exercises.
- The guidelines state that exercises should be completed through the full range of motion at each joint (Council on Sports Medicine and Fitness, 2008) to develop strength through the full range and prevent loss of flexibility (Verhs, 2005b) and this principle can be applied to children with CP as much as possible.
- If both single and multiple joint exercises are being completed once a strength programme is well underway, it is recommended that multiple joint exercises should be completed before single joint exercise, large muscle groups be exercised before small muscle groups and complex movements before less complex movements (Verhs, 2005b).
- The guidelines recommend a 10 to 15 min warm up and cool down associated with any strength training programme (Council on Sports Medicine and Fitness, 2008, Verhs, 2005b). As this recommendation is related to multiple muscle groups, it may be amended for upper limb programmes.
- The guidelines recommend that workouts should be 20-30 mins long (Council on Sports Medicine and Fitness, 2008) (this is for a full body strengthening programme), but may be amended for upper limb programmes.
- The recommendations regarding numbers of sets, repetitions and frequency varies slightly across the different guidelines. Generally the recommendations suggest strength training programmes should include 2 to 3 sets of repetitions (ie 8 to 15 repetitions), 2 to 3 times per week on non-consecutive days (no more than 4 days per week). The programme should continue for at least 8 weeks.
- A period of rest should occur between each set for a particular muscle to allow recovery (Verhs, 2005b)
- The guidelines suggest that the load (resistance) should be added gradually as strength improves. It is recommended that weight can be added in 5% to 10% increments (Council on Sports Medicine and Fitness, 2008, Verhs, 2005b) when 8 to 15 reps are mastered (Council on Sports Medicine and Fitness, 2008) or when 20 repetitions become easy (Vehrs, 2005b). Recommendations regarding the minimum starting weight need further investigation for children. We located a recommendation for adults which suggested that a weight of approximately 85% of maximal strength (3-6 sets of 4-8 reps) would increase strength and muscle size. The relevance of this in children is not known (ACSM).
- The guidelines state that strengthening machines may be too large and have weight increments too large for children (Council on Sports Medicine and Fitness, 2008). Free weights, although they may require better balance and control, are portable and may be more practical. For children with weakness alternate means such as rice filled plastic bags, weighted cuffs, Theraputty and Theraband/elastic tubing (the resistance increases further into the range, can add grips to reduce the likelihood of letting go) should also be considered. Factors such as individual

children's motor control, balance, coordination, stability of grasp etc should be considered when determining the means of providing resistance. Body weight can be carefully explored as a means of providing resistance depending in the relationship between each individual's strength and body weight (Vehrs, 2005b).

- Movements during strengthening exercises should be slow and controlled (Ashmore, 2003). Explosive, rapid movement is not recommended (Council on Sports Medicine and Fitness, 2008). The sports of power and weight lifting are distinct from strengthening programmes (Council on Sports Medicine and Fitness, 2008)
- An exercise should cease if pain is experienced. Exercises which cause pain, including into the next day should be excluded the next session.
- Touching the muscles which should be contracting during each exercise may reinforce muscle groups exercising. (Ashmore, 2003)
- Efforts should be made to make the strength programme fun (Verhs, 2005b; Ashmore, 2003) use a variety of interesting exercises/activities/tasks (ACSM), use charts of progress and allow child choice in type and order of exercises.
- Strength gains are lost approx 6 weeks after training is discontinued (Council on Sports Medicine and Fitness, 2008).

Other points of interest

- Safety and supervision of strengthening programmes are critical (Council on Sports Medicine and Fitness, 2008). Strengthening programmes however are safe if the recommendations and guidelines are followed (Verhs, 2005b)
- All guidelines recommend that a medical evaluation is completed prior to commencing formal strength training (Verhs, 2005b)
- Pre-pubertal children will not get larger muscles but will recruit more muscle fibres to fire with each contraction (Council on Sports Medicine and Fitness, 2008).
- Strengthening and endurance are important for developing bone mineral density and a number of other health indicators (Verhs, 2005a). Strengthening may also increase strength of tendons and ligaments and the stability of joints as well as motor coordination and performance. (Verhs, 2005a) Increasing bone mineral density may decrease the risk of developing osteoporosis later in life. To achieve this long term effect, training is most beneficial during growth, that is, for girls before 16 years of age and before 18 years for boys.
- It is unknown as to whether increasing upper limb strength using a strengthening programme will improve the functional abilities of children with CP. Two of the upper limb programmes (n=1; 3) included in this CAT did report functional gains (Lee et al., 2009; O'Connell & Barnhart, 1995) but a third study did not (n=9; Vaz et al., 2008).
- <u>Outcome Measurement</u>: The literature gives little guidance in definitive outcome measurement following upper limb strength training. Grip strength was commonly measured using a range of dynamometry. There was no uniformity in other assessments used but rarely were they standardised for children with cerebral palsy. Future research should use objective measures to determine whether any alterations in strength are associated with meaningful functional changes. Tools such as the Assisting Hand Assessment (Krumlinde-Sundholm, Holmefur & Eliasson, 2005), The Melbourne Assessment of Unilateral Upper Limb Function (Randall, Johnson & Reddihough, 1999) and the Pediatric Motor Activity Log (Wallen, Pont, Bundy & Ziviani, 2009) have been developed for use with children with cerebral palsy and should be utilised to assess functional upper limb change.

The Canadian Occupational Performance Measure (Law et al, 2005) and Goal Attainment Scaling (Kiresuk, Smith & Cardillo, 1994) should also be used to determine any changes in the ability to complete important activities of daily living.

CONCLUSIONS

The literature reviewed does not provide answers to the clinical question of this CAT. Active range of movement was not measured in the studies reviewed, there is scant and inconclusive information about the effects of strength training on upper limb function and change in the ability to complete activities of daily living has not been explored.

There is evidence in the literature that strength training should be and is increasingly becoming part of an overall health and fitness strategy for children and adolescents. This focus on strength training is also a current theme in paediatric cerebral palsy literature, although the scientific evidence is not definitive in terms of clinical application. Strength training does not appear to have adverse effects when undertaken by people with CP. Many of the documented health benefits seen in the typically developing paediatric population could also benefit children with cerebral palsy. Some of the benefits could include: improved bone mineral density; increased joint stability; improved motor coordination; increased muscle fibre recruitment and overall improvements in cardio respiratory and musculoskeletal health, endurance and well being.

The available literature, particularly policy statements from the American Academy of Pediatrics and the American College of Sports Medicine, clearly outlines safe guidelines for strengthening programmes with children, but there is little reference to these in the literature on upper limb strengthening in children with cerebral palsy. Research in this area should, where possible, incorporate safe, best-practice strengthening guidelines in order to determine their relevance to children with cerebral palsy. There are also other issues, particular to children with CP and outlined above, that should be considered in developing upper limb strength training programmes in this population. Future research should also focus on functional upper limb outcomes at the International Classification of Functioning, Disability and Health levels of activity and participation (WHO, 2001).

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