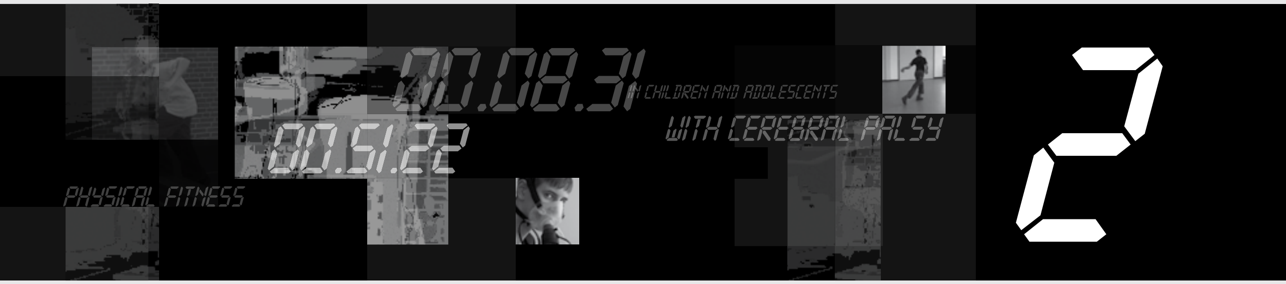

EXERCISE PROGRAMS FOR CHILDREN WITH CEREBRAL PALSY: A SYSTEMATIC REVIEW OF THE LITERATURE

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Abstract

The aim of this literature review regarding all types of exercise programs focussing on cardiovascular fitness (aerobic and anaerobic capacity) and/or lower extremity muscle strength in children with cerebral palsy (CP), was to address the following questions: (1) what exercise programs focusing on muscle strength, cardiovascular fitness or a combination are studied and what are the effects of these exercise programs in children with CP? (2) what are the outcome measures that were used to assess the effects of the exercise programs? (3) what is the methodological quality of the studies?

We systematically searched the literature in electronic databases up to October 2006 and included a total of 20 studies that were evaluated.

The methodological quality of the included trials was low. However, it appears that children with CP may benefit from improved exercise programs that focused on lower extremity muscle strength, cardiovascular fitness or a combination. The outcome measures used in most studies were not intervention-specific and often only focused on the International Classification of Function, Disability and Health (ICF) body function and activity level. There is a need to determine the efficacy of exercise programs to improve the daily activity and participation level of children with CP and increase their self-competence or quality of life.

Introduction

Cerebral palsy (CP) describes a group of disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain.[1] The motor disorders of CP are often accompanied by disturbances of sensation, cognition, communication, perception, and/or behavior, and/or by a seizure disorder.[1] Because of the impairments, many children and adolescents*[2] with CP have at least difficulty with activities such as walking independently, negotiating stairs, running or navigating safely over uneven terrain.[3] Improving one's ability to walk or to perform other functional activities are often the primary therapeutic goals for children with CP.[4]

Exercise refers to planned structured activities involving repeated movement of skeletal muscles that result in energy expenditure and seeks to improve or maintain levels of physical fitness above the intensity of activities of daily living.[5] Exercise in children with CP has often been avoided because of the concern about the negative effect of such effort on muscle spasticity and children's movement patterns.[6] Several factors have contributed to a recent shift in perspective about the use of exercise in children with CP. Studies evaluating the effect of exercise on children with CP reported no adverse effect on patterns of movement,[7,8] flexibility[8,9] or spasticity.[10] These findings have influenced current practice.

Most exercise programs for children with CP are primarily designed for the lower extremity. The most common functions of the lower extremity tend to be gross motor activities that involve repetitive, reciprocal, coordinated motions of both extremities in order to move through space and that often require little conscious effort once under way.[11] There has been an increased interest in developing and implementing exercise programs that improve the cardiovascular fitness (aerobic and anaerobic capacity) and/or lower extremity muscle strength of children with CP.

Two systematic reviews have been published that examined the effects of strengthening in the CP population.[12,13] To date, there is no systematic review that examined all types of exercise programs focusing on cardiovascular fitness (aerobic and anaerobic capacity) and/or lower extremity muscle strength in children with CP.

The purpose of the present paper was to systematically review the literature regarding exercise programs in children with CP to address the following questions: (1) what exercise programs focusing on lower extremity muscle strength, cardiovascular fitness or a combination are studied and what are the effects in children with CP? (2) what are the outcome measures that were used to assess the effects of the exercise programs? (3) what is the methodological quality of the studies?

* Childhood generally spoken refers to the period 2 to 12 years of age, and adolescence refers to the period 13 to 21 years of age.[2] In this review children and adolescents are referred to as children.

In many systematic reviews, a meta-analysis is performed, statistically combining the results of the various studies into a single estimated effect size. However, meta-analysis has been described specifically for randomized controlled trials. We expected most of the studies to be observational studies, a situation in which the use of meta-analysis is generally not recommended.[14] Therefore, a qualitative systematic review on the effects of all types of exercise programs focusing on cardio-vascular fitness (aerobic and anaerobic capacity) and/or lower extremity muscle strength in children with CP was performed.

Method

Search strategy

The following electronic databases were searched from their respective inceptions to October 2006: MEDLINE, PubMed, EMBASE, CINAHL, Sports Discus, Cochrane, PEDro. Search terms included subject headings and text words based on (I) cerebral palsy; (II) exercise (in combination with strength, fitness, working capacity, aerobic power, anaerobic power, endurance, cardiorespiratory physical training or program); (III) lower extremity; (IV) clinical trials. Inclusion criteria were: (1) children and adolescents with CP, (2) intervention (exercise programs focusing on lower extremity muscle strength, cardiovascular fitness or a combination) and (3) outcome (measurement of change in body function and structure, activity or participation). Exclusion criteria were: (1) doctoral dissertations, (2) reports published in books, (3) reports published in conference proceedings and (4) studies that included children with CP as well as children with other diagnoses.
Titles and available abstracts of all items identified by the electronic searches were scrutinized by one author (OV).

Data extraction

Included papers were read in full by 3 (arbitrarily chosen out of a sample of 5 for each paper) independent reviewers with their background in pediatric physical therapy, exercise physiology or rehabilitation. They all recorded details of the study design, practice setting, participants, interventions, outcome measures, results and conclusions on a data extraction form. Any disagreements or discrepancies were resolved through discussion and checking the original papers. Where key information was not reported, efforts were made to contact the authors in order to obtain further details.
Lower extremity strength training was defined as prescribed exercises for the lower limbs, with the aim of improving strength and muscular endurance, that are typically carried out by making repeated muscle contractions resisted by body weight, elastic devices, masses, free weights, specialized machine weights, or

isokinetic devices.[15] *Aerobic (fitness) training* was defined as aiming to improve the cardio-respiratory component of fitness, typically performed for extended periods of time.[15] *Anaerobic (fitness) training* refers to exercises which require large bursts of energy over short (< 30 seconds) periods of time.[15] *Mixed (physical fitness) training* was, based on the United States Department of Health and Human Services (USDHHS),[15] defined as a planned, structured regimen of regular physical exercise deliberately performed to improve one or more components of physical fitness (i.e. muscle strength, aerobic and anaerobic capacity, flexibility and body composition).

Included trials were divided in four categories: lower extremity strength training, aerobic training, anaerobic training and mixed training. Because in some studies it can be difficult to distinguish between the different categories, any disagreements among the 3 reviewers were resolved by a discussion until a consensus was reached.

The outcome measures used in the studies were categorized by using the International Classification of Function, Disability and Health (ICF)[16] framework for the description of health. In this framework, a person's disability can be considered in terms of impairment on the body function or structure level, activity limitations and participation restrictions. In line with the ICF we consider a person's functioning as a dynamic interaction between the health condition (in this case, CP) and personal and contextual factors such as the environment.

Quality assessment

Obtained reports were assessed by the same 3 reviewers that performed the data extraction for each specific paper. Empirical studies that met inclusion criteria were rated for methodological quality with the PEDro Scale, based on the Delphi list described by Verhagen et al.[17] With the PEDro Scale, the following indicators of methodological rigor were scored independently as either absent (0 points) or present (1 point) by the reviewers: (1) specification of eligibility criteria, (2) random allocation, (3) concealed allocation, (4) prognostic similarity at baseline, (5) subject blinding, (6) therapist blinding, (7) assessor blinding, (8) greater than 85% follow-up for at least 1 key outcome, (9) intention-to-treat analysis, (10) between-group statistical analysis for at least 1 key outcome, (11) point estimates of variability provided for at least 1 key outcome. Points are only awarded when a criterion is clearly satisfied and reported in the trial report.

According to the PEDro guidelines, criteria 2 through 11 are used for scoring purposes so that a score from 0 to 10 can be obtained. The PEDro Scale has shown moderate levels of interrater reliability (intraclass correlation coefficient = .54; 95% confidence interval (CI), .39- .71).[18] To improve the reliability of this scale, any disagreement between the reviewers were resolved by discussion with an independent reviewes until consensus was reached.

Evidence assessment

Randomized Clinical Trials (RCTs) are the best method to ensure that any differences in outcome were due to the treatment and not other factors. They give one confidence in internal validity. So, the ideal method for determining efficacy of a treatment is through RCTs, but such trials are often difficult to pursue.[19] As a result, many studies employ less well-controlled research designs. The variety of research designs in the literature mandates use of a method to help evaluate diverse studies and give weight to their findings. To determine the degree of confidence that can be placed in the evidence available about an intervention, a grading system developed by the American Academy for Cerebral Palsy and Developmental Medicine (AACPDM) can be used.[20] For evidence levels see Table 1.

Table 1. AACPDM levels of evidence.

Level	Non-empirical	Group Research	Outcomes Research	Single Subject Research
I		- Randomized controlled trial - All or none case series		- N-of-1 randomized controlled trial
II		- Nonrandomized controlled trial - Prospective cohort study with concurrent control group	- Analytic survey	- ABABA design - Alternating treatments - Multiple baseline across subjects
III		- Case-control study - Cohort study with historical control group		- ABA design
IV		- Before and after case series without control group		- AB design
V	- Descriptive case series or case reports - Anecdote - Expert opinion - Theory based on physiology, bench, or animal research - Common sense/ first principles			

Results

Search results

The initial search of the electronic databases and the manual search of reference lists identified 581 citations. Based on title and abstract we excluded 559 studies that did not meet our inclusion criteria. Of the remaining 22 articles that were read full-text, 4 articles were excluded because the intervention did not meet the criteria. Screening of references of these studies led to another 2 studies being included. In total, 20 studies remained and were included in the present systematic review: Eleven studies on strength training interventions, 5 studies on aerobic training interventions and 4 studies on mixed training interventions. All information was obtained directly form the articles.

No article focused on anaerobic training; therefore the included trials were divided into three categories: lower extremity strength training, aerobic training and mixed training.

Intervention characteristics and effects

Lower extremity strength training

Table 2 shows the characteristics of the 11 included strength training interventions,[9,21-30] in children with CP, varying in age from 6-20 years. Exercise interventions lasted for 6 weeks in seven trials,[21-24,26,27,30] 8 weeks in three trials[9,25,29] and 9 months in one trial.[28] All exercise frequencies were three times a week. Nine programs were individually based,[9, 21-25, 27, 28, 30] and two programs were group programs.[26,29] In six studies[22-24,26,28,30] the supervisor was a physical therapist or parent/partner, in four studies[9,21,25,27] the supervisor was not described and in one study the supervisor was a research assistant.[29]

All studies reported outcome results on the ICF body structure and function level and eight[22,23,25-30] on the activity level. In two Randomized ControlledTrials (RCTs),[23,24] small improvements in performance on tests of muscle strength were found for the experimental group. In one RCT[29] only significant change in the perception of body image and a more upright posture were found. Another RCT[28] found no significant changes at all. Five trials reported significant improvements in tests of muscle strength following strength training programs lasting 6-8 weeks.[9,21,22,25,27] Dodd et al.,[23,24] Mac Phail et al.,[25] Morton et al.,[27] Unger et al.[29] and Patikas et al.[28] were the only studies that used a long-term follow-up measurement, that varied from four weeks up to one year. Only three studies concluded that the gained benefits on muscle strength,[23,25,27] gross motor function,[23,25,27] scholastic competence and social acceptance,[24] and muscle tone[27] of training were maintained.

Table 2. Strength training exercise studies for the lower extremity involving children with cerebral palsy.

Study	Subjects		Design			Intervention program					Results according to the ICF levels			PEDro	AAC PDM
	Age	N	Number of groups	Randomised	Time & number of measurements	Training duration	Freq. of the training	Ind /gr	Training program & exercises	Sup.	Body function and structure	Activity	Participation		
Dodd et al.[23]	8-18	21	2	yes	1. start 2. 6 weeks 3. 18 weeks (= follow up)	6 weeks	3 times a week	ind	**Strength training** 3 sets of 8-10 reps 3 muscle groups LE (ankle plant flex / knee ext / hipext)	PT parent	Non significant increase in muscle strength Significant increase in combined muscle strength	Non significant increase in gross motor function, stair walking and walking speed	-	7/10	I
McBurney et al.[30]	8-17	11	1	no	1. Post training	6 weeks	3 times a week	ind	**Strength training** 3 sets of 8-10 reps 3 muscle groups LE (ankle plant flex / knee ext / hipext)	PT parent	Improved perception of strength, flexibility, posture, walking and the ability to negotiate stairs. Increased well-being.	Improvement in mobility	Improvement in school, leisure, social and family events.	7/10	I
Damiano et al.[21]	6-14	14	1	no	1. before 2. 3 weeks 3. 6 weeks	6 weeks	3 times a week	ind	**Strength training** 4 sets of 5 reps with each leg Load=65% of max	ND	Significant increase in 4-ceps muscle strength and non significant change in hamstrings muscle strength	-	-	3/10	IV
MacPhail et al.[25]	12-20	17	1	no	1. before 2. after tr. 3. 3 months follow up	8 weeks	3 times a week for 45 minutes	ind	**Strength training** 3 sets of 5 max effort at 90%. Knee flexors and extensors	ND	Significant increase for total muscle strength Non significant change in spasticity and energy expenditure	Significant increase in gross motor function (9/17) Non significant change in walking speed	-	3/10	IV
Damiano et al.[22]	6-12	11	1	no	1. before 2. 2 weeks 3. 4 weeks 4. 6 weeks	6 weeks	3 times a week	ind	**Strength training** load = 65% of max. isom. strength 4 sets of 5 rep. for each muscle group lower extremity	PT parent	Significant increase in muscle strength No change in energy expenditure	Significant increase in gross motor function and walking velocity and cadence	-	3/10	IV
Eagleton et al.[26]	12-20	7	1	no	1. pre training 2. post training	6 weeks	3 times a week for 40-60 minutes	gr	**Strength training** Load: 80% of 1 RM Muscle groups: trunk and lower extremity	PT partner	Significant decrease in energy expenditure	Significant increase in walking speed, step length, cadence and distance	-	0/10	IV

N = number of participants, Ind/gr = individual/group, Sup. = supervisor, PT = Physical Therapist, ND = not described, RA = research assistant, wks = weeks

Study	Subjects		Design			Intervention program					Results according to the ICF levels			PEDro	AAC PDM
	Age	N	Number of groups	rando mised	Time & number of measurements	Training duration	Freq. of the training	Ind /gr	Training program & exercises	Sup.	Body function and structure	Activity	Participation		
Dodd et al.[24]	8-16	17	2	yes	1. before 2. 6 weeks 3. 18 weeks (follow up)	6 weeks	3 times a week	ind	**Strength training** 3 sets of 8-10 reps using 3 exercises for lower extremity	PT parent	Trend (borderline sign) in increase in muscle strength Significant decrease in self-concept for scholastic competence and social acceptance	-	-	6/10	I
Healy et al.[9]	8-16	5	1	no	1. 0 weeks 2. 2 weeks 3. 4 weeks 4. 6 weeks 5. 8 weeks	8 weeks	3 times a week	ind	**Strength training** 2 programs: 1. concentric 3 sets of 10 rep: a.½ of 10 RM b. ¾ of 10 RM c. 10RM 2. static 6 sec (2/3 of RM	ND	Significant increase in muscle strength and range of motion No sign. differences between gains when the 2 methods are compared.	-	-	3/10	IV
Morton et al.[27]	6-12	8	1	no	1. pre training 2. post training 3. follow-up (4 wks)	6 weeks	3 times a week	ind	**Strength training** Progressive, free weight programme for quadriceps and hamstrings; concentric and eccentric. Load 65% of mean strength.	ND	Significant increase in muscle strength and significant decrease in muscle tone	Non significant increase in walking speed and step length. Significant increase in self-selected cadence. Significant (Dim E) and non-significant (Dim E) increase in gross motor function.	-	3/10	IV
Patikas et al.[28]	6-16	39	2	yes	1. pre-surgery and pre-training (n=39) 2. 1 year post-surgery (n=39) 3. follow-up gait analysis (n=22)	9 months	3 times a week for 30-45 minutes	ind	**Strength training** Two sets of 5 repetitions. 7 exercises involving the following muscle groups: hip-, knee- and ankle extensors and flexors.	PT parent	No difference in spasticity	No significant difference in gross motor function	-	5/10	I
Unger et al.[29]	13-18	31	2	yes	1. pre training 2. post training 3. follow-up (4 wks)	8 weeks	1-3 times a week for 40-60 minutes	gr	**Strength training** 8-12 individually designed exercises selected from a 28 station circuit 1-3 sets of 12 repetitions	RA	Significant change in the perception of body image. No significant change in functional competence.	Significant change in a more upright posture. No significant change for stride length, velocity or cadence.	-	8./10	I

N = number of participants, Ind/gr = individual/group, Sup. = supervisor, PT = Physical Therapist, ND = not described, RA = research assistant, wks = weeks

Table 3. Aerobic training exercise studies for the lower extremity involving children with cerebral palsy.

Study	**Subjects**		**Design**			**Intervention program**					**Results according to the ICF levels**			**PEDro**	**AAC PDM**
	Age	N	Number of groups	rando mised	Time & number of measurements	Training duration	Freq. of the training	Ind /gr	Training program and exercises	Sup	Body function and structure	Activity	Participation		
Van den Berg-Emons et al.[31]	7-13	20	2	yes	1 before trial 2. 2 months 3. 9 months 4. 12 months	9 months	4 times a week for 45 minutes	gr	**Aerobic training** Cycling, running, swimming, wheelchair driving, flying saucer, mat exercises	ND	Significant increase in aerobic capacity. Non significant increase in anaerobic capacity. Trend to improve for muscle strength. Trend to improve for Physical activity Fat mass → CON > + EXP =	-	-	6/10	I
Shinohara et al.[32]	11.8 - 16.3	11	2	no	1.before 2. during 3. after	6-20 weeks	2 times a week for 20 minutes	ind	**Aerobic training** Cycling or arm cranking at the AT point for 20 minutes	ND	Significant increase in aerobic capacity for leg group and non significant increase for arm group. Increase for physical endurance for leg group.	-	-	3/10	IV
Berg et al.[33]	7-25	22	1	no	1. before 2. post training 3. 3 months follow-up	1.5-16 months	3 times a week for 20 minutes	ind	**Aerobic training** 20 minutes with various loads based on max cap cycling	PT	Non significant increase for aerobic capacity	-	-	3/10	IV
Lundberg et al.[34]	15-20	14	1	no	1. before 2. after	6 weeks	2 times a week for 20 minutes	gr	**Aerobic training** Exercising large muscle groups for 1-2 minutes (running and jumping)	PT	Significant increase for aerobic capacity	-	-	3/10	IV
Schlough et al.[35]	17-20	3	1	no	A1B1A2B2 design	Subject: 1→10 wks 2→20 wks 2→21wks	3 times a week	ind	**Aerobic training** Exercise on elliptical machine, treadmill or recumber stepper between 40 – 70% of HR-max.	ND	Mixed results for energy expenditure Non significant increase for muscle strength. Non significant increase in physical appearance (self concept)	Non significant increase in gross motor function	-	3/10	IV

N = number of participants, Ind/gr = individual/group, Sup. = supervisor, PT = Physical Therapist, ND = not described, wks = weeks

Aerobic training

Table 3 shows the results of the five studies[31-35] that focused the intervention on aerobic exercise in children with CP. They varied in age from 7-20 years (except one subject in the study performed by Berg et al.[33] who was 25 years old). Exercise interventions varied from 6 weeks to 16 months, with exercise frequencies varying from two to four times a week for 20 to 45 minutes. The intensity of the training programs varied from exercise at the anaerobic threshold point,[32] training at an intensity of ≥ 70% of the heart rate reserve[31] to various loads based on the maximal cycling capacity.[33] One study did not describe the intensity of the training.[34] Two programs[31,34] were group programs and three[32,33,35] were individually based programs. In two studies[33,34] the supervisor was a physical therapist, and in three studies[31,32,35] the supervisor was not described.

All included studies, of which one was an RCT,[31] reported results on the ICF level of body function. In the RCT performed by Van den Berg-Emons et al.[31] a significant increase in aerobic capacity, and non significant improvements on anaerobic capacity, muscle strength and fat mass were found. One study[35] investigated the activity level, measured with the Gross Motor Function Measure (GMFM) (dimension D: standing and E: walking, running, jumping) of the subjects. Three trials[31,32,34] reported statistically significant improvements of aerobic capacity.

Physical activity ratio,[31] fat mass,[31] anaerobic capacity[31] and the Energy Expenditure Index (EEI)[35] were studied as well. No statistically significant changes were found in the included studies.

In two studies follow-up measurement took place.[31,33] Both studies, including one RCT,[31] concluded that inactivity during summer vacation (approx. 3 months) significantly reduced the aerobic capacity.

Mixed training

In Table 4 the results of four studies that examined the effects of mixed training interventions[36-39] in children with CP, varying in age from 4-20 years are shown. Exercise interventions varied from 4 weeks to 6 months. Exercise frequencies varied from two to three times a week and from 30 to 60 minutes. All programs were group programs. However, one study[38] combined the group program with an individual swimming program. In three studies[36,37,39] the supervisor was a physical therapist, in one study[38] the supervisor was not described.

All included studies reported results on the level of body function. Two studies,[36,39] found a significant increase in muscle strength. One study[38,39] reported a significant increase in vital capacity, and another study[36] reported no significant change in heart rate and energy expenditure. The study performed by Darrah et al.[36] showed a significant increase for self-perception of physical appearance. Two studies investigated the effects on the level of activity.[38,39] Blundell et al.[39] reported a significant

Table 4. Mixed training exercise studies for the lower extremity involving children with cerebral palsy.

Study	Subjects		Design			Intervention program					Results according to the ICF levels			PEDro	AAC PDM
	Age	N	Number of groups	randomised	Time & number of measurements	Training duration	Freq. of the training	Ind /gr	Training program & exercises	Sup.	Body function and structure	Activity	Participation		
Darrah et al.[36]	11-20	23	1	no	1 .before 2. before 3. before 4. 10 weeks 5. 20 weeks	10 weeks	3 times a week	gr	**Mixed training** Aerobic exercises Weight training 3 sets of 12 rep (upper and lower extremity) flexibility	PT Students Instruct	Significant increase in muscle strength. Non significant change in heart rate and energy expenditure Non significant change in flexibility. Self-concept: significant increase for physical appearance and non significant changes for other subscales	Non significant change in walking speed	-	3/10	IV
Rintala et al.[37]	7-11	8	1	no	1. baseline t1-t4 2. post-training t5-t11	15 weeks	2 times a week for 60 minutes	gr	**Mixed training** Bal skills Balance coordination	PT Teacher	Non significant change for balance, grip strength, walking distance, sprint capacity and ball skills	-	-	2/10	IV
Hutzler et al.[38]	5-7	46	2	no	1. pre-training 2. post-training	6 months	3 times a week for 30 minutes	2x ind 1x gr	**Mixed training** Water orientation skills (group) Locomotion and ball handling (ind)	ND	Significant increase for vital capacity	Significant increase for water orientation.	-	5/10	II
Blundell et al.[39]	4-8	8	1	no	1. baseline 2. pretest 2 wks 3. post-test 6 wks 4. follow-up 8 wks	4 weeks	2 times a week for 60 minutes	gr	**Mixed training** Strength: circuit Aerobic training: treadmill	PT parent	Significant increase for muscle strength.	Significant increase in stride length and significant and non significant increases for walking speed	-	3/10	IV

N = number of participants, Ind/gr = individual/group, Sup. = supervisor, PT = Physical Therapist, Instruct = instructor, ND = not described, wks = weeks increase in stride length, and mixed results for walking speed. Darrah et al.[36] found a significant change in walking speed. There were two studies that used a follow-up measurement.[36,39] Blundell et al. concluded that all training improvements were maintained after eight weeks follow-up.[39] The results found by Darrah et al. showed that the significant changes in muscle strength were maintained 10 weeks after completion of the program.[36]

Outcome measures

The outcomes that were used in all included studies were categorized by using the ICF[16] framework for the description of health, and can be appreciated in Table 5.

Body function and structure

Muscle strength

To measure muscle strength the hand-held dynamometer,[21-23,27,35,36,39] the isokinenic dynamometer,[25] the Cybex,[31] the spring scale,[9] the Lateral Step-up Test,[39] Motor Assessment Scale (Sit-to-Stand),[39] a 10 repetition maximum[24] and the minimum chair height test[39] were used.

Spasticity and muscle tone

To measure spasticity and muscle tone the Modified Ashworth Scale of Spasticity[25,28] and the resistance to passive stretch (RPS)[27] were used in the included studies.

Fat mass

Fat mass was measured using skin fold measurement in one study.[31]

Fitness-measures

The Energy Expenditure Index (EEI),[22,25,26,28,35,36] which is defined as walking heart rate minus resting heart rate, divided by walking speed, expressed in beats per meter[40] was used to quantify the energy consumed during walking. To measure the aerobic capacity the cycle ergometer (arm and leg) was used in five studies.[31-34,36] One study[31] investigated the effects of an aerobic focused intervention on anaerobic performance using the Wingate test. One study[28] measured the oxygen uptake (VO_2) during two 5 minute walks.

Range of motion / flexibility

The goniometer was used to examine the range of motion of the lower extremity in one study.[9] Darrah et al.[36] examined the flexibility of the participants pre- and post training by using the sit-and-reach, the behind the back reach test and the inter-malleolar distance.

Table 5. Outcome measures used in exercise studies for the lower extremity involving children with cerebral palsy.

Study	Outcome measures according to the ICF-levels		
	Body function and structure	**activity**	**participation**
Strength training			
Dodd et al.[23]	HHD	GMFM (D&E) Timed stair test 10 meter timed walking	-
Mc Burney et al.[30]	self-constructed semi-structured interview	Self-constructed semi-structured interview	Self-constructed semi-structured interview
Damiano et al.[21]	HHD	-	-
MacPhail et al.[25]	isokinetic dynamometer Modified Ashworth Scale of Spasticity EEI	GMFM (D&E)	-
Damiano et al.[22]	HHD EEI	GMFM Gait analysis (comp.)	-
Eagleton et al.[26]	EEI	10 meter timed walking 3 minute treadmill walking	-
Dodd et al.[24]	10 repetition maximum SPPC	-	-
Healy et al.[9]	spring scale goniometer	-	-
Morton et al.[27]	HHD resistance to passive stretch (RPS)	10 meter timed walking GMFM D &E	-
Patikas et al.[28]	MAS EEI VO_2 measurement during two 5 minute walks	GMFM	
Unger et al.[29]	self-perception questionnaire	Six-camera video-based motion-capturing system: VICON 370 data station	
Aerobic training			
Van den Berg-Emons et al.[31]	cycle ergometer wingate cycling or arm cranking test Cybex Physical Activity-ratio skinfold measurement (4 sites)	-	-
Shinohara et al.[32]	cycle or arm ergometer physical endurance interview	-	-
Berg et al.[33]	cycle ergometer	-	-
Lundberg et al.[34]	cycle ergometer (and Douglas bag)	-	-
Schlough et al.[35]	EEI HHD SPPCS	GMFM D&E	-
Mixed training			
Darrah et al.[36]	EEI HHD cycle test sit-and-reach test behind the back reach test intermalleolar distance SPPC/SPPA	-	-
Rintala et al.[37]	balance test grip strength 9 minute walk 50 meter sprint bal skills	-	-
Hutzler et al.[38]	spirometer	Water orientation checklist	-
Blundell et al.[39]	HHD Lateral Step-up Test Motor Assessment Scale (Sit-to- Stand) Minimum chair height test	10 meter timed walking 2 minute walk test	-

HHD = Hand Held Dynamometer, EEI = Energy Expenditure Index, GMFM = Gross Motor Function Measure, SPPC = Self Perception Profile for Children, MAS = Modified Ashworth Scale, SPPCS = Self Perception Profile for College Students, SPPA = Self Perception Profile for Adolescent

Self perception

McBurney et al.[30] used a semi-structured interview to explore the changes in perception of strength, posture, walking and the ability to negotiate stairs, and one study[29] used a self developed self perception questionnaire. Four studies[24,29,35,36] investigated the effects of a training program on the self concept of the subjects using the Self Perception Profile for Children (SPPC), Self Perception Profile for Adolescents (SPPA) and the Self Perception Profile for College Students (SPPCS) and a short, self-administered self perception questionnaire.

Activity

Gross motor function

Six studies investigated the effects of an exercise program on the activity level by measuring changes in gross motor function using the Gross Motor Function Measure (GMFM).Two studies[22,28] used the total GMFM score, and four studies[23,25,27,35] only used dimension D (standing) & E (walking, running, jumping) to evaluate the effects of the intervention program.

Gait

The timed stair test,[23] the 10 meter timed walking,[23,26,27,39] 3 minute treadmill walking,[26] the computerized gait analysis[22] and the 2 minute walk test[39] were other instruments used to evaluate the effects on gait speed or stride length. Kinematic data were captured in the study performed by Unger et al.[29] using the VICON 370 data station.

Water orientation

The water orientation checklist[38] was used to evaluate the effects of a swimming program.

Physical activity

Mc Burney et al.[30] used a self-developed semi-structured interview, containing a preliminary schedule of four questions about the program, to explore the changes in physical activity following a strengthening program.

Participation

McBurney et al.[30] used the same semi-structured interview to evaluate the outcomes of a strength training program on the participation level.

Methodological quality of included studies

Table 2, 3 and 4 summarize the findings of the included publications. Initial inspection of the studies suggested that most were of a repeated-measures design without a control group.

The methodological quality was assessed with the PEDro scale. No article scored more than 8 (out of 10) on this scale, and the median score was 3. Not all the criteria on the PEDro scale can be satisfied in these studies (for example, blinding of subjects is often difficult or impossible). Five of the 20 studies were randomized controlled trials.[23,24,28,29,31] The remaining fifteen selected studies could not fulfill criteria related to randomized controlled trials (e.g. group allocation and blinding) as detailed in PEDro criteria 2 through 6. Most of the studies fulfilled criteria 8, 9 and 11, indicating that most subjects undertook the designated training program and that their outcome measures were reported.

To determine the degree of confidence the AACPDM levels of evidence were used. The five RCTs scored a level I on this assessment of degree of confidence placed on the evidence.[23,24,28,29,31] The median on the AACPDM levels of evidence scale was 4.

Discussion

There are only five randomized controlled studies investigating the efficacy of exercise training in children with CP and many of the extant studies have been poorly controlled. This is disappointing, because evidence suggests that non-physically active children are more likely to become physically inactive adults and that encouraging the development of physical activity habits in children helps establish patterns that continue into adulthood.[41] Prevention of this decline from childhood and adolescence to adulthood should emphasize increased physical activity.[42]

This systematic review examined the literature regarding exercise programs in children with CP, provides an overview of the intervention characteristics, and the outcome measures that are used in exercise programs in children with CP.

Intervention characteristics

The reviewed exercise studies involving children with CP vary in program design, population and evaluation. They include training programs conducted in a laboratory setting, the community, school-based and home-based settings. The supervisors in the studies varied from physical therapists to parents.

Thus far there is little evidence to identify the optimal mode, frequency, intensity, setting, supervision and duration of activity in exercise programs. Based on the strength training programs that were reviewed, it can be suggested that a training program with a minimum of 6 weeks with a frequency of three training sessions a week may be sufficient to improve the muscle performance of the lower extremity. This finding supports the findings of Dodd et al.[12] and Pippenger et al.[43] They concluded that there is evidence supporting the view that progressive

resistance exercise can increase the ability to generate muscle force in children with CP. This conclusion was supported by another systematic review of 7 studies.[13]
To improve the aerobic capacity of children with CP training sessions that vary from 2 to 4 times a week and last at least 6 weeks may be adequate. The mixed training programs, that showed significant increases in muscle strength and stride length, varied from 4 weeks to 6 months.
No study compared the training response in different age groups. In the studies that were reviewed there was no indication that young children (under 12 years of age) react different to the exercise programs compared to the older children (12 years of age and older). In general, aerobic capacity and muscle strength appear to be trainable in children of all ages.[44] Measures of anaerobic ability, such as peak and mean power and anaerobic capacity, appear also to be trainable in children, but there are apparently no reports in the literature examining the anaerobic trainability across different stages of maturation.[44]
None of the training programs focused on anaerobic capacity. This is surprising, considering the fact that almost all daily childhood activities are more of a short-term high-intensity, than of a long-term activity character.[45,46] Since many of the daily childhood activities consist of short-term bursts of intense activity, anaerobic fitness is thought to be an important measure of functional capacity.[45] In children with a neurodevelopmental disease, anaerobic power is considered to be a better measure of functional capacity than prolonged maximal aerobic power.[47]
The ability of the children with a diagnosis CP to maintain the gains achieved in the long term generally remains unknown because only a few trials have included a follow-up period. Based on the limited findings in this review it can be suggested that the benefits that children gained during strength training and mixed training were maintained at follow-up. However, aerobic capacity was significantly reduced at follow-up.
Activity patterns of youth vary considerably. Activities during the daily life of a child consist of aerobic, anaerobic and muscle strength components. To date, there is no study that trained all three fitness components combined. Exercise training, in which these three components are combined, may be more appropriate to improve the activity and participation level of children with CP. This needs to be investigated in future research.

Outcome measures

Instruments used to measure the effects of fitness training that were used in the included studies were diverse. To evaluate aerobic power five studies used cycle ergometers.[31-34,36] To assess the changes on the activity level no cycling-based test was used. There is a discrepancy between the instruments used on the body function and the activity level. Training effects are exercise mode specific.[48] Specificity of testing means that the modality of the testing tool needs to be similar to the type of activity the subjects train in. Because improvements in the fitness studies often used non-intervention-specific testing, to assess change, we suspect specificity was not an important factor in the ability to detect an improvement in cardiovascular fitness with the exercise programs. However, to find results that are more exercise-related, intervention-specific tests should be used in future research. This may enhance the results of the studies and their interpretation. However, intervention-specific measurement is often limited to the function level.

Only one study[30] reported examples of children who increased their participation in school, leisure, social and family events after undertaking an exercise program.It is surprising that only one study examined the effects on the participation level. Especially, because participation of children with CP in everyday activities is a goal shared by parents, service providers and organizations involved in children's rehabilitation.[49] Children with physical disabilities are at risk of limited participation.[49,50] In future research the effect of exercise programs on the participation level in children with CP needs to be studied.

There were two RCTs that studied the effects of an exercise program on the self-concept. Dodd et al.[24] reported a significant decrease in self-concept for scholastic and social competence, whereas the study performed by Darrah et al.[36] demonstrated an increase in the self-concept for physical appearance of the children post-training. A difference between both studies may be relevant. The study performed by Darrah et al.[36] was performed in a group environment, whereas the exercise program from Dodd et al.[24] was individually based. A group environment can be a motivating and socially stimulating therapy for children.[36] Within a group context, games, races, and cooperative activities can be used to enhance engagement of children with CP in exercise interventions.[51] Moreover, group treatment permits peer modeling, competition, and potentially, a wider range of activity which may benefit the child's overall participation in the prescribed exercises. However, Schlough et al.[35] reported an increase in self concept following a study that was individually based. Therefore, the underlying reasons for the discrepancy in findings are unclear. More research is needed to find out what kind of training, and what duration is the most beneficial for improvement in the self concept of children with CP.

Overall, only a few studies have measured the effects of an exercise program on activity in children with CP. In the studies that focused on muscle strength, only one study examined the effect of an exercise program on the societal participation of children with CP.[30] In the studies that focused on aerobic and mixed training the participation was not measured at all. These findings are similar to the results of the review that was performed by Dodd et al.[12] None of the studies they included in the review measured the effect of a strengthening program on participation limitation. The current review revealed the same result for other exercise program based studies.

Conclusion

In general, the methodological quality as well as the level of evidence of the included trials was low. Only five RCTs were included. However, from a critical evaluation of data currently available, it appears that children with CP may benefit from improved exercise programs that focus on lower extremity muscle strength, cardiovascular fitness or a combination. The outcome measures used in most studies were not intervention-specific and often focused on the ICF body function and structure and activity level. So, despite being able to increase muscle strength and aerobic capacity, more evidence is needed to determine whether training can make substantial or sustained improvements in daily activity, the participation level, self-competence or quality of life.

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