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LITERATURE REVIEW

Exercise Programs for Children with Cerebral Palsy

A Systematic Review of the Literature

ABSTRACT

Verschuren O, Ketelaar M, Takken T, Helders PJM, Gorter JW: Exercise programs for children with cerebral palsy: a systematic review of the literature. Am J Phys Med Rehabil 2007;86:000–000.

The purpose of this literature review, regarding all types of exercise programs focusing 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 seems 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 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.

Key Words: Fitness, Exercise, Cerebral Palsy, Muscle Strength, Review

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] (The term *childhood* generally refers to the period of 2–12 yrs of age, and adolescence refers to the period of 13–21 yrs of age.[2] In this review, children and adolescents are referred to as *children*.) 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 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? and (3) what is the methodological quality of the studies?

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 (RCTs). 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 cardiovascular 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 September 2006: MEDLINE, PubMed, EMBASE, CINAHL, Sports Discus, Cochrane, and 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; and (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 (O.V.).

Data Extraction

Included papers were read in full by three (arbitrarily chosen out of a sample of five 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 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 cardiorespiratory component of fitness, typically performed for extended periods of time.[15] *Anaerobic (fitness) training* refers to exercises that require large bursts of energy over short ($<$30 secs) periods of time.[15] *Mixed (physical fitness) training* was, on the basis of the United States Department of Health and Human Services,[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 three 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 three 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 (zero points) or present (one 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) >85% follow-up for at least one key outcome, (9) intention-to-treat analysis, (10) between-group statistical analysis for at least one key outcome, and (11) point estimates of variability provided for at least one 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 = 0.54; 95% confidence interval (CI), 0.39–0.71).[18] To improve the reliability of this scale, any disagreement between the reviewers were resolved by discussion with an independent reviewer until consensus was reached.

Evidence Assessment

RCTs are the best method to ensure that any differences in outcome were attributable 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.

RESULTS

Search Results

The initial search of the electronic databases and the manual search of reference lists identified 581 citations. On the basis of 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 (Fig. 1): 11 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 to 20 yrs. Exercise interventions lasted for 6 wks in seven trials,[21–24,26,27,30] 8 wks in three trials,[9,25,29] and 9 mos 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 studies[22,23,25–30] reported on the activity level. In two 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 signifi-

TABLE 1 American Academy for Cerebral Palsy and Developmental Medicine (AACPDM) levels of evidence

Level	Nonempirical	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			

cant changes at all. Five trials reported significant improvements in tests of muscle strength after strength training programs lasting 6–8 wks.[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 long-term follow-up measurements, which varied from 4 wks up to 1 yr. 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.

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 to 20 yrs (except one subject in the study performed by Berg et al.[33] who was 25 yrs old.) Exercise interventions varied from 6 wks to 16 mos, with exercise frequencies varying from two to four times a week for 20–45 mins. 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 nonsignificant improvements on anaerobic capacity, muscle strength, and fat mass, was 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[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 (approximately 3 mos) 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 to 20 yrs, are shown. Exercise interventions varied from 4 wks to 6 mos. Exercise frequencies varied from two to three times a week and from 30 to 60 mins. 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

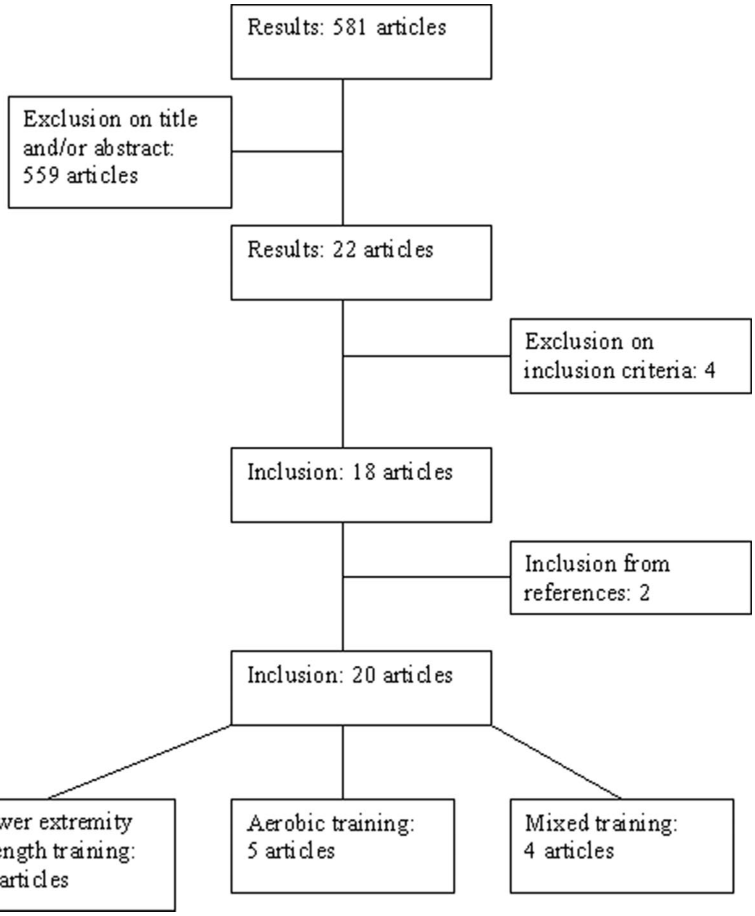

FIGURE 1 *Flow chart of included studies.*

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 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 8-wk follow-up.[39] The results found by Darrah et al. show that the significant changes in muscle strength were maintained 10 wks 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 handheld dynamometer,[21–23,27,35,36,39] the isokinetic dynamometer,[25] the Cybex,[31] the spring scale,[9] the Lateral Step-up Test,[39] the 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[27] were used in all included studies.

Fat Mass

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

Fitness Measures

The energy expenditure index,[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

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

	Subjects		Design			Intervention Program					Results According to the ICF Levels				
Study	Age	n	Number of Groups	Randomized	Time and Number of Measurements	Training Duration	Frequency of the Training	Ind/Gr	Training Program and Exercises	Sup	Body Function and Structure	Activity	Participation	PEDro	AACPDM
Dodd et al.[23]	8–18	21	2	Yes	1. Start 2. 6 wks 3. 18-wks follow-up	6 wks	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	Nonsignificant increase in muscle strength Significant increase in combined muscle strength	Nonsignificant increase in gross motor function, stair walking, and walking speed	—	7/10	I
McBurney et al.[30]	8–17	11	1	No	1. Posttraining	6 wks	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 wks 3. 6 wks	6 wks	3 times a week	Ind	Strength training 4 sets of 5 reps with each leg Load = 65% of max	ND	Significant increase in forceps muscle strength and nonsignificant change in hamstrings muscle strength	—	—	3/10	IV
MacPhail et al.[25]	12–20	17	1	No	1. Before 2. After. 3. 3-mos follow-up	8 wks	3 times a week for 45 mins	Ind	Strength training 3 sets of 5 max effort at 90% Knee flexors and extensors	ND	Significant increase for total muscle strength Nonsignificant change in spasticity and energy expenditure	Significant increase in gross motor function (9/17) Nonsignificant change in walking speed	—	3/10	IV
Damiano et al.[22]	6–12	11	1	No	1. Before 2. 2 wks 3. 4 wks 4. 6 wks	6 wks	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. Pretraining 2. Posttraining	6 wks	3 times a week for 40–60 mins	Gr	Strength training Load: 80% of 1RM 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
Dodd et al.[24]	8–16	17	2	Yes	1. Before 2. 6 wks 3. 18-wks follow-up	6 wks	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

(Continued)

TABLE 2 Continued

	Subjects		Design			Intervention Program					Results According to the ICF Levels				
Study	Age	*n*	Number of Groups	Randomized	Time and Number of Measurements	Training Duration	Frequency of the Training	Ind/Gr	Training Program and Exercises	Sup	Body Function and Structure	Activity	Participation	PEDro	AACPDM
Healy et al.[9]	8–16	5	1	No	1. 0 wks 2. 2 wks 3. 4 wks 4. 6 wks 5. 8 wks	8 wks	3 times a week	Ind	**Strength training** 2 programs: 1. Concentric 3 sets of 10 reps at half of 10 RM a. Half of 10 RM b. Three fourths of 10 RM c. 10 RM 2. Static 6 secs (two thirds of RM)	ND	Significant increase in muscle strength and range of motion No significant differences between gains when the two methods are compared	—	—	3/10	IV
Morton et al.[27]	6–12	8	1	No	1. Pretraining 2. Posttraining 3. Follow-up (4 wks)	6 wks	3 times a week	Ind	**Strength training** Progressive, free weight program for quadriceps and hamstrings; concentric and eccentric Load 65% of mean strength	ND	Significant increase in muscle strength and significant decrease in muscle tone	Nonsignificant increase in walking speed and step length Significant increase in self-selected cadence Significant (Dim E) and nonsignificant (Dim E) increase in gross motor function	—	3/10	IV
Patikas et al.[28]	6–16	39	2	Yes	1. Presurgery and pretraining (n = 39) 2. 1-yr postsurgery (n = 39) 3. Follow-up gait analysis (n = 22)	9 mos	3 times a week for 30–45 mins	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. Pretraining 2. Posttraining 3. Follow-up (4 wks)	8 wks	1–3 times a week for 40–60 mins	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

Ind/Gr, individual/group; sup, supervisor; PT, physical therapist; ND, not described; RA, research assistant.

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

	Subjects		Design			Intervention Program					Results According to the ICF Levels				
Study	Age	*n*	Number of Groups	Randomized	Time and Number of Measurements	Training Duration	Frequency of the Training	Ind/Gr	Training Program and Exercises	Sup	Body Function and Structure	Activity	Participation	PEDro	AACPDM
Van den Berg-Emons et al.[31]	7–13	20	2	Yes	1. Before trial 2. 2 mos 3. 9 mos 4. 12 mos	9 mos	4 times a week for 45 mins	Gr	**Aerobic training** Cycling, running, swimming, wheelchair driving, flying saucer, mat exercises	ND	Significant increase in aerobic capacity Nonsignificant 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 wks	2 times a week for 20 mins	Ind	**Aerobic training** Cycling or arm cranking at the AT point for 20 mins	ND	Significant increase in aerobic capacity for leg group, and nonsignificant 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. Posttraining 3. 3-mos follow-up	1.5–16 mos	3 times a week for 20 mins	Ind	**Aerobic training** 20 mins with various loads based on max cap cycling	PT	Nonsignificant increase for aerobic capacity	—	—	3/10	IV
Lundberg et al.[34]	15–20	14	1	No	1. Before 2. After	6 wks	2 times a week for 20 mins	Gr	**Aerobic training** Exercising large muscle groups for 1–2 mins(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→ 21 wks	3 times a week	Ind	**Aerobic training** Exercise on elliptical machine, treadmill, or recumber stepper between 40 and 70% HRmax	ND	Mixed results for energy expenditure Nonsignificant increase for muscle strength Nonsignificant increase in physical appearance (self-concept)	Nonsignificant increase in gross motor function	—	3/10	IV

Ind/gr, individual/group; sup, supervisor; PT, physical therapist; ND, not described.

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

	Subjects		Design			Intervention Program					Results According to the ICF Levels				
Study	Age	*n*	Number of Groups	Randomized	Time and Number of Measurements	Training Duration	Frequency of the Training	Ind/Gr	Training Program and Exercises	Sup.	Body Function and Structure	Activity	Participation	PEDro	AACPDM
Darrah et al.[36]	11–20	23	1	No	1. Before 2. Before 3. Before 4. 10 wks 5. 20 wks	10 wks	3 times a week	Gr	**Mixed training** Aerobic exercises Weight training 3 sets of 12 reps (upper and lower extremity) flexibility	PT students instruct	Significant increase in muscle strength. Nonsignificant change in heart rate and energy expenditure Non significant change in flexibility. Self-concept: significant increase for physical appearance and nonsignificant 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. Posttraining t5–t11	15 wks	2 times a week for 60 mins	Gr	**Mixed training** Balance 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. Pretraining 2. Posttraining	6 mos	3 times a week for 30 mins	2× Ind 1× 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. Posttest (6 wks) 4. Follow-up (8 wks)	4 wks	2 times a week for 60 mins	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

Ind/Gr, individual/group; sup, supervisor; PT, physical therapist; instruct, instructor; ND, not described.

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

	Outcome Measures According to the ICF Levels		
Study	Body Function and Structure	Activity	Participation
Strength training			
Dodd et al.[23]	HHD	GMFM (D&E) Timed stair test 10-m timed walking	—
McBurney et al.[30]	Self-constructed, semistructured interview	Self-constructed, semistructured interview	Self-constructed, semistructured 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-m timed walking 3-min 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-m timed walking GMFM D &E	—
Patikas et al.[28]	MAS EEI VO_2 measurement during two 5-min 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 (four 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-min walk 50-m sprint Balance 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-m timed walking 2-min walk test	—

HHD, handheld 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 Adolescents.

test. One study[28] measured the oxygen uptake (VO_2) during two 5-min 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 before and after training by using the sit and reach, the behind-the-back reach test, and the intermalleolar distance.

Self-Perception

McBurney et al.[30] used a semistructured 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, Self-Perception Profile for Adolescents, and the Self-Perception Profile for College Students 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 GMFM. Two studies[22,28] used the total GMFM score, and four studies[23,25,27,35] only used dimensions D (standing) and E (walking, running, jumping) to evaluate the effects of the intervention program.

Gait

The timed stair test,[23] the 10-m timed walking,[23,26,27,39] 3-min treadmill walking,[26] the computerized gait analysis,[22] and the 2-min 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

McBurney et al.[30] used a self-developed semistructured interview, containing a preliminary schedule of four questions about the program, to explore the changes in physical activity after a strengthening program.

Participation

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

Methodological Quality of Included Studies

Tables 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 (e.g., blinding of subjects is often difficult or impossible). Five of the 20 studies were RCTs.[23,24,28,29,31] The remaining 15 selected studies could not fulfill criteria related to RCTs (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 RCTs 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 nonphysically 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, and school- 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. On the basis of the strength training programs that were reviewed, it can be suggested that a training program for a minimum of 6 wks, 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 conclude 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 seven studies.[13]

To improve the aerobic capacity of children with CP, training sessions that vary from two to four times a week and that last at least 6 wks may be adequate. The mixed training programs that showed significant increases in muscle strength and stride length varied from 4 wks to 6 mos.

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 yrs of age) react different to the exercise programs compared with the older children (12 yrs of age and older). In general, aerobic capacity and muscle strength seem to be trainable in children of all ages.[44] Measures of anaerobic ability, such as peak and mean power and anaerobic capacity, seem 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 that almost all daily childhood activities are more of a short-term, high-intensity, than of a long-term activity character.[45,46] Because 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 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. On the basis of 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 posttraining. 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 programs 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 in 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 seems 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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