PHYSICAL FITNESS IN CHILDREN AND ADOLESCENTS WITH CEREBRAL PALSY

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PHYSICAL FITNESS IN CHILDREN AND ADOLESCENTS WITH CEREBRAL PALSY

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Dr. T. Takken
Dr. M. Ketelaar

Voor kinderen met cerebrale parese en hun ouders


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IN CHILDREN AND ADOLESCENTS
08.51.22
CEREBRAL PALSY
PHYSICAL FITNESS

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IN CHILDREN AND ADOLESCENTS
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INTRODUCTION

Cerebral palsy (CP) describes a group of permanent 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. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy and by secondary musculoskeletal problems.[1]

The Gross Motor Function Classification System (GMFCS)[2] is considered to be one of the most important scales for the measurement of function in CP. It was designed primarily to help clinicians prognosticate about the motor function of individuals with CP. The GMFCS consists of a 5-level scale with descriptors divided into four age bands: 0 to < 2 years, 2 to < 4 years, 4 to < 6 years, and 6 to 12 years. Children at Level I are relatively capable and by the time they reach 6 to 12 years old they can walk without limitations and can run and jump to some extent, with difficulties arising only with balance, speed, and coordination. At the other end of the scale, children at Level V will never achieve self-mobility unless they can learn to use a powered wheelchair with extensive adaptations. The GMFCS is based on self-initiated movements only, concentrating mostly on truncal control in sitting and functional mobility, taking into consideration everyday performance, rather than best capacity.

The 'Surveillance of Cerebral Palsy' in Europe (SCPE)[3] has classified CP into three main subtypes. These are *spastic*, *ataxic* and *dyskinesic* CP. Agreement has been reached on the clinical findings associated with each classification sub-group as follows:

Spastic CP is characterised by at least two of: 1) Abnormal pattern of posture and/or movement, 2) Increased tone (not necessarily constantly), 3) Pathological reflexes (hyper-reflexia or pyramidal signs e.g. Babinski response). It may be unilateral or bilateral.

Ataxic CP is characterised by both of 1) Abnormal pattern of posture and/or movement, 2) Loss of orderly muscular coordination, so that movements are performed with abnormal force, rhythm and accuracy.

Dyskinesic CP is characterised by both of 1) Abnormal pattern of posture and/or movement, 2) Involuntary, uncontrolled, recurring, occasionally stereotyped movements of affected body parts. Dyskinesic CP may be either a) Dystonic CP, dominated by both hypokinesia and hypertonus, b) Choreo-athetotic CP, dominated by both hyperkinesia and hypotonus.

As described above, the primary deficit in CP is an injury to the brain, not to the musculoskeletal or cardio-respiratory systems. However, the limitations in movement imposed by the brain lesion can have a marked secondary effect on these systems,[4] which may in turn become more debilitating for an individual with

CP than the direct effects of the initial injury. The secondary changes contribute to a vicious cycle whereby the disability leads to deconditioning that, in turn, worsens the level of disability.[5] Muscles need to be loaded adequately and frequently to maintain strength.[6] Similarly, in people with CP as in all people, the heart and lungs need to be exercised at moderately intense levels on a regular basis to maintain endurance and fitness. People with CP are already at a disadvantage with respect to achieving adequate levels of physical functioning because muscles and the cardio-respiratory system are not fully developed before the brain injury occurs and, therefore, are likely to have a lower starting point as well as slowed progress in developing these structures.[7]

It is becoming increasingly apparent that many of the adverse secondary changes may be preventable or reversible, although the extent to which this is possible has not been sufficiently explored or aggressively challenged. Management of motor disorder in CP is shifting gradually from the long-standing approach of alleviating impairments once they have occurred to a more proactive approach of promoting the type and degree of activity that may retard the development of muscle weakness. Alleviating the energy cost of performing typical daily activities is now a more frequently recognized and stated goal of motor interventions.[8-10]

Muscle strength and endurance training can be undertaken to improve performance of everyday activities. However, fitness implies a more generalized and greater degree of conditioning.

While health prevention and promotion efforts for persons with disabilities lag behind those for the non-disabled population, the potential benefits are enormous in terms of participation and health-related quality of life across the lifespan.

As stated in the revised 'Guide to Physical Therapist Practice' of the American Physical Therapy Association,[11] the role of the physical therapist is 'to restore, maintain, and promote not only optimal physical function, but optimal wellness and fitness and optimal quality of life as it relates to movement', which marks a significant expansion from its traditional mission. Strength, endurance, and fitness goals should be included in physical therapy treatment plans for the patients.

Importance of improving strength, endurance and fitness in CP

Historically, programs to promote physical fitness, including strengthening and cardio respiratory fitness exercise, were discouraged for patients with CP due to the concern that spasticity and abnormal movement patterns would worsen.[12] Therefore, traditional treatment of CP has focused primarily on attempting to improve abnormal motor patterns and maintain muscle length for daily activity and positioning. The historically used approaches failed to address adequately the ensuing muscle weakness, atrophy, and negative effects of diminished amounts and intensity of activity on the cardio-respiratory system. While these secondary

consequences have been recognized in CP for many decades, in the past, only a handful of proponents such as Winthrop Phelps[13] advocated physically intensive intervention. Scientific evidence has not supported the historically concern about the negative influence of physical fitness[14-16] and current research indicates that resistive exercise and endurance training is an effective intervention to improve strength, aerobic capacity and function in children and adolescents with CP.[6,17]
Documentation exists showing that individuals with CP have substantial generalized weakness.[18,19] Durstine and colleagues[5] described the circular process whereby persons with a chronic illness or disability experience less physical activity, which then leads to a cycle of deconditioning and further physical deterioration and reduction in activity (Figure 1). Many persons with CP lead a sedentary lifestyle with a decreased amount, variety, and intensity of physical activity.[4] Therefore, it is not surprising that also low levels of cardio-respiratory fitness have been reported.[20-24]

Improving the ability to walk or performing other functional activities are often the primary therapeutic goals for children with CP.[25] Because of existing impairments, many children and adolescents with CP have at least difficulty with activities such as walking independently, walking stairs, running or navigating safely over uneven terrain.[26] Additionally, children with CP have distinctly subnormal aerobic and anaerobic capacity in comparison with typically developing peers.[20,27,28]

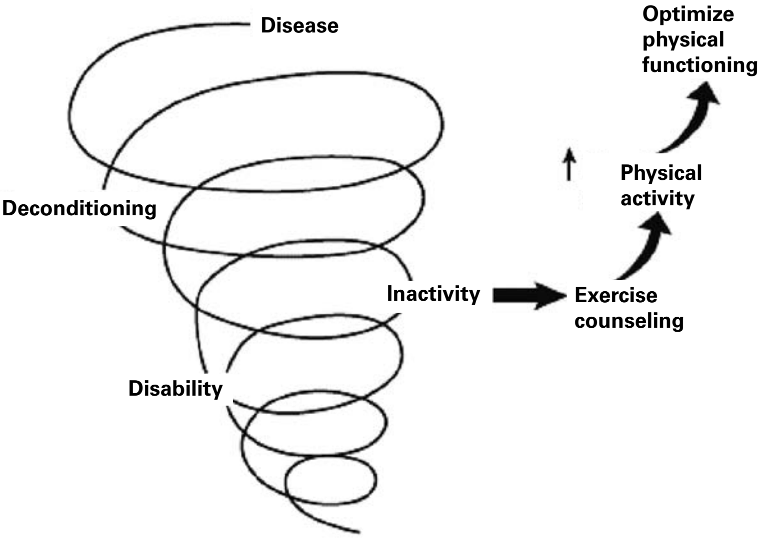

Figure 1. Cycle of deconditioning with physical inactivity. Disease can lead to inactivity and deconditioning. Deconditioning can lead to further inactivity and increase the potential for disability.

Also, muscle mass is low,[20] muscle strength is reduced[19,24,29] and energy cost of loco-motion is high.[30-32] Low levels on these fitness components may contribute to the difficulties in motor activities most children with CP encounter in daily life. More-over, evidence suggests that hypoactive children are more likely to become physically sedentary adults and that encouraging the development of physical activity habits in children will help us to establish activity patterns that continue into adulthood.[33]

The main question in this thesis is: “Do children with CP benefit from a fitness intervention program?” We focused on children that are classified at GMFCS level I and II. Due to the physical demands of the fitness tests and training exercises that were used throughout the study, recruitment solicited only children and adolescents who were classified at GMFCS level I (able to walk indoors and outdoors, and climb stairs without limitation) or level II (able to walk indoors and outdoors, and climb stairs holding onto a railing but experience limitations in walking on uneven surfaces and inclines, and walking in crowds or confined spaces).

The effects of this training program were considered by using the International Classification of Functioning, Disability and Health (ICF) framework for the description of health.[34] The ICF is applicable across cultures, age groups and sexes. In this framework, a person’s disability can be considered in terms of impairments, activity limitations, and participation restrictions. The ICF classification served as a framework for the present study.

Principles of fitness training

Training to improve a child’s performance obeys the three principles of training: specificity, overload and reversibility.[35,36]

Specificity

The application of the ‘Specific Adaptation to Imposed Demands (SAID)’ principle is essential in type of training.[37] Training programs are tailored to the exact demands of the activity and the individual needs of the child. This allows for specific gains to be made on the relative energy systems employed during training.

Specificity is an important principle in strength training, where the exercise must be specific to the type of strength required, and is therefore related to the particular demands of the event.

Overload

For training adaptations to occur the bodies systems must be overloaded beyond their normal levels. If these extra stresses are applied over a period of time the system will adapt and this becomes its new norm. During this adaptation-phase the bodies systems super or over compensate in order to cope with the next session. Training in this way along with sufficient recovery will allow for super compensation to occur thus resulting in an overall increase in fitness levels.

Reversibility or Detraining

During periods of exercise the human body makes adaptations to cope with the stresses placed on it. During periods of inactivity the human body will however reverse these adaptations in an attempt to return itself to a norm as this is the current level of stress placed upon it. Therefore gains that have been made will be lost.
Detraining is a de-conditioning process that affects performance due to the reduction in physiological capacity. During this period there is a loss of physiological adaptations associated with the training effect. Most training benefits are lost within a short period of stopping training. Most of the beneficial effects of training return to normal levels with 4-8 weeks dependent on the individual.

Measurements of fitness

Training effects are exercise mode specific.[38] Specificity of testing means that the modality of the testing tool needs to be similar to the type of activity the subjects train in. The outcome measures used in most studies in children with CP were not intervention-specific and often only focused on the International Classification of Function, Disability and Health (ICF) body function and activity level. 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. Since this study focuses on children that are able to walk independently (GMFCS I and II) we developed reliable and valid walking/running based exercise tests.

Age related physical activities

Recommended priorities for physical activities during childhood and adolescence relative to the development of skills and to behavioral, health, and fitness benefits are schematically illustrated in Figure 2. During the preschool and early school ages, general movement activities develop movement patterns and skills (dashed line in Figure 2). As these basic movements become established and skills improve, health, fitness, and behavioral components of physical activities increase in importance (solid line in Figure 2). Health related activities include those that emphasize cardiovascular and muscular endurance and muscular strength and those that involve weight bearing.

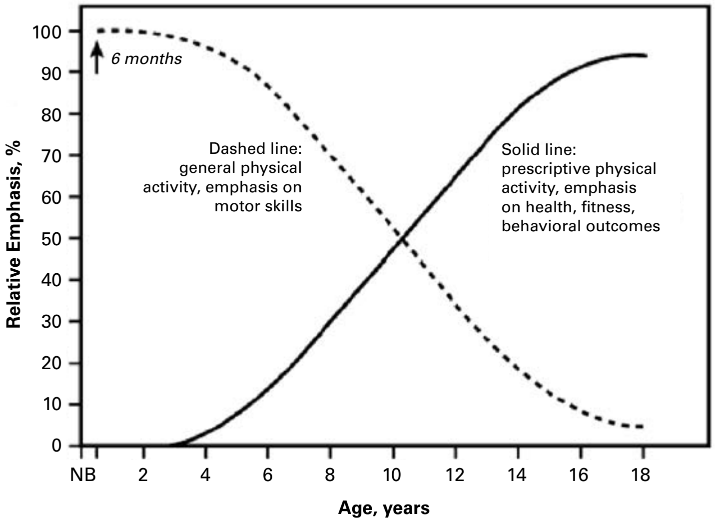

Figure 2. Changing emphasis of physical activity during childhood and adolescence.[39]

In this study we included children from 7 years of age and older, because we expected that children with CP of this age have established their motor patterns and skills, and are capable of increasing their fitness. Moreover, at this age children were expected to be able to understand and follow simple verbal commands.

Aim of this thesis

Based on the lack of exercise mode specific fitness-measures in children with CP, the first aims of this thesis are:

- Systematically review the literature regarding all types of exercise programs focusing on cardiovascular fitness (aerobic and anaerobic capacity) and/or lower extremity muscle strength in children with CP.
- To assess the psychometric qualities of measures for aerobic and anaerobic capacity that is specific for walking/running.
- To assess the psychometric qualities of a muscle strength measure that is exercise mode specific.

  Due to the scarce knowledge of the effects of a fitness intervention program in children and adolescents with CP, the second aim of this thesis is:
- To determine the effects of a functional fitness program in children and adolescents with CP not only on the domains of physical fitness, but also on gross motor function, self concept, participation and health-related quality of life.

Outline of this thesis

The first chapters address the assessment of fitness in children with CP. A review of the literature is presented in Chapter 2. In Chapter 3, a running based measure, the 10 m shuttle run test, is examined for its reliability and validity to measure aerobic capacity in children and adolescents with CP. In Chapter 4, two running based measures, the Muscle Power Sprint Test (MPST) and the 10x5 Meter Sprint Test, are being studied on its reliability and validity to measure respectively the mean and peak muscle power and agility. In Chapter 5, two ways to measure muscle strength are being studied. For Hand-Held Dynamometry the make-method and break-method are being compared regarding their reliability. Moreover, in this Chapter a new instrument (the 30-sec Repetition Maximum) to measure functional muscle strength is examined on its reliability.

The effects of a functional fitness program on aerobic and anaerobic capacity, agility, muscle strength, self-concept, gross motor function, participation and health-related quality of life are provided in Chapter 6. In Chapter 7, a summary and general discussion, focusing on the implications for research and clinical practice is presented.

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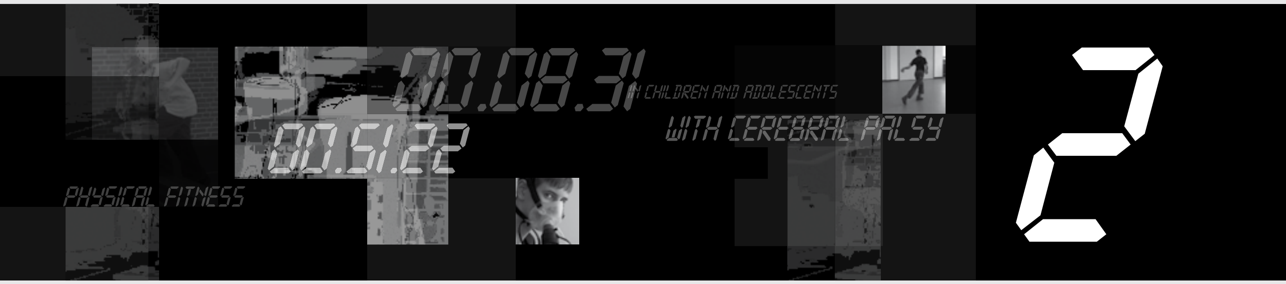

EXERCISE PROGRAMS FOR CHILDREN WITH CEREBRAL PALSY: A SYSTEMATIC REVIEW OF THE LITERATURE

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Paul J.M. Helders, Jan Willem Gorter

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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?

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* 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	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.[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	randomised	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	rando mised	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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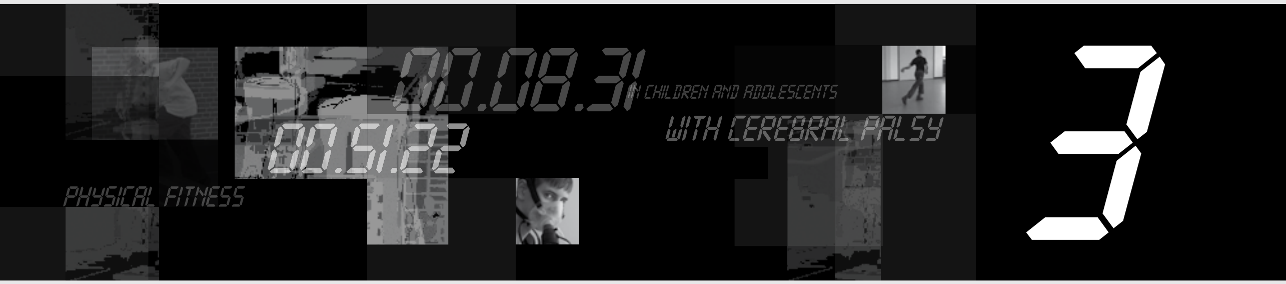

RELIABILITY AND VALIDITY OF DATA FOR 2 NEWLY DEVELOPED SHUTTLE RUN TESTS IN CHILDREN WITH CEREBRAL PALSY

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Abstract

Background and Purpose The purpose of this study was to examine the reliability and validity of data obtained with 2 newly developed shuttle run tests (SRT-I and SRT-II) to measure aerobic power in children with cerebral palsy (CP) who are classified at level I or II on the Gross Motor Function Classification System (GMFCS). The SRT-I was developed for children at GMFCS level I, and the SRT-II was developed for children at GMFCS level II.

Subjects Twenty-five children and adolescents with CP (10 female, 15 male; mean age=11.9 years, SD=2.9), classified at GMFCS level I (n=14) or level II (n=11), participated in the study.

Methods To assess test-retest reliability of data for the 10-m shuttle run tests, the subjects performed the same test within 2 weeks. To examine validity, the shuttle run tests were compared with a GMFCS-level based treadmill test designed to measure peak oxygen uptake.

Results Statistical analyses revealed test-retest reliability for exercise time (number of levels completed) (intraclass correlation coefficients of .97 for the SRT-I and .99 for the SRT-II) and reliability for peak heart rate attained during the final level (intraclass correlation coefficients of .87 for the SRT-I and .94 for the SRT-II). High correlations were found for the relationship between data for both shuttle run tests and data for the treadmill test (both r=.96).

Discussion and Conclusion The results suggest that both 10-m shuttle run tests yield reliable and valid data. Moreover, the shuttle run tests have advantages over a treadmill test for children with CP who are able to walk and run (GMFCS level I or II).

Introduction

Maximal aerobic power is an important component of physical fitness and often is used to measure the effect of physical exercise training. It also is used to assess cardiovascular capacity as well as the level of fitness in children and adolescents with cerebral palsy (CP).[1] Ascertaining maximal aerobic power is the most common way to assess the outcome of physical exercise training[2] in children with CP. Peak oxygen uptake (VO_{2peak}) determined at the limits of exhaustive treadmill or cycle exercise is generally viewed to be the best physiologic marker of aerobic fitness.[3]

Currently, tests for assessing aerobic power in children with CP that are clinically practical and easy to use and that yield reliable and valid data are absent. Most of the available literature has focused on assessments of VO_{2peak} using cycle ergometry[4] or arm ergometry.[5] Treadmill running engages a larger muscle mass than does cycling[6]; therefore, VO_{2peak} is typically 8% to 10% higher during treadmill running than during cycle ergometry.[7] A major disadvantage of cycle ergometry in children is that a high proportion of the total power output is generated by the quadriceps femoris muscle and the effort required to push the pedals during the later stages of a progressive test is high in relation to muscle strength (force-generating capacity).[8] Children with CP have weak quadriceps femoris muscles.[9] The maximal performance during a cycle ergometry test, therefore, can be limited by a peripheral factor. Still, many researchers reported VO_{2peak} of children with CP using the cycle ergometer.[4,10,11]

For children who are able to walk independently, the most functional way to assess their maximal aerobic power would be a walking- or running-based exercise test. The treadmill protocols that currently are often used in clinical practice (Bruce protocol and Balke protocol) are not appropriate for children with CP.[1] For most children with CP who have problems with movement coordination and an equinus position of the foot, the increasing speed and inclining floor are problematic and difficult. Research, however, has shown that it is possible for children with CP to perform a maximal exercise test on a treadmill,[12,13] although there is no published standardized protocol for administering VO_{2peak} tests to those children.

Although the "gold standard" assessment of exercise tolerance in children can be measured in a laboratory using a treadmill, the necessary equipment is expensive and may not be readily accessible. Thus, substantial logistical problems must be overcome before a child with CP can be assessed on the treadmill, and there is a clinical need to develop and validate field tests of exercise tolerance in children and adolescents with CP.

Shuttle run tests are field tests in which a participant walks or runs between 2 markers. These tests are potentially useful measures of exercise tolerance. Because there is no need for expensive equipment, shuttle run tests can easily be administered in a clinical setting. The shuttle run test commonly used is the

20-m shuttle run, which was developed and validated by Leger et al.[14] This test has been demonstrated to be an effective measure of the aerobic fitness in subjects who were healthy.[15,16] Validity has been determined in children who are developing typically,[14,17,18] in athletes,[19] and in a sedentary adult population.[20]

For most children and adolescents with CP, the 20-m shuttle test is not suitable, because the starting speed (8 km/h) and the increase (0.5 km/h) every minute are beyond their capabilities. Several experiments with the 20-m shuttle test showed that most of the children and adolescents with CP who were able to walk independently were not able to complete the first level of the test or reached exhaustion before 5 minutes. A continuous progressive exercise lasting 8 to 17 minutes is optimal for achieving a maximal myocardial oxygen demand for diagnostic and prognostic purposes.[21]

To facilitate assessment in children, the tests should be nonthreatening, inexpensive, and easy to administer.[22] For this reason, we developed 2 treadmill protocols and shuttle run tests for children with CP who are classified at level I or II on the Gross Motor Function Classification System (GMFCS).[23] To achieve a maximal exercise duration between 8 to 17 minutes, the GMFCS classification seems to be a useful system to distinguish between the 2 groups: children with a level I classification, who are able to run, and children with a level II classification, who encounter more difficulties while running. Therefore, we developed 2 different protocols with different starting speeds.

In this study, we examined the test-retest reliability and validity of data obtained with a GMFCS level I-specific 10-m shuttle run test (SRT-I) and a GMFCS level II-specific 10-m shuttle run test (SRT-II). We also evaluated the feasibility of both tests for use with children with CP.

Table 1. Subject Characteristics (n=25)

	GMFCS[a] Level I (n=14)				**GMFCS Level II (n=11)**			
Variable	**Mean**	**SD**	**Median**	**Range**	**Mean**	**SD**	**Median**	**Range**
Age (y)	11.5	2.8	11.7	7.5-16.1	12.5	3.0	12.1	7.2-17.0
Height (cm)	148.7	15.3	149.0	125.0-175.0	148.6	18.9	145.0	123.0-175.0
Body mass (kg)	40.3	12.4	35.1	23.8-60.8	38.6	12.1	32.7	24.0-59.7
Sum of 7 skinfold measurements (mm)	77.2	44.3	61.8	36.0-196.5	74.2	28.5	70.5	33.0-131.5

[a] GMFCS=Gross Motor Function Classification System.

Materials en Methods

Subjects

A convenience sample of 30 children and adolescents from a school for special education were invited to participate in the study. To be included, subjects were required to be within the age range of 7 to 20 years, had to be diagnosed with CP, and classified at level I or II on the GMFCS.[23] Cognitively, they had to be capable of following simple commands. Twenty-five subjects (15 male, 10 female) and their parents agreed to participate and signed an informed consent form. Group characteristics according to GMFCS level are described in Table 1.

Procedure

Prior to testing, each child was weighed on electronic scales (Seca, Hamburg, Germany). Height measurements were taken on the same visit while the child was standing against a wall. Body coposition was assessed using the sum of 7 skinfold measurements according to the method described by Pollack et al.[24] The skinfold measurements were taken at 7 sites on the right side of the body (triceps, biceps, subscapular, suprailiac, mid-abdominal, medial calf, and front thigh) by 2 of the investigators (OV and TT) in accordance with the American College of Sports Medicine guidelines.[25]

Study Design

To assess test-retest reliability of data for the 10-m shuttle run tests, each subject performed one shuttle run test (SRT-I or SRT-II) 2 times. To assess the validity of data for both shuttle run tests, the VO_{2peak} values obtained with gas analysis during the GMFCS level-based treadmill test and the shuttle run tests were compared. The treadmill test and the shuttle run tests were separated by a minimum of 2 days and a maximum of 7 days ($\overline{X}$=4.1, SD=1.4).

Treadmill testing on all subjects was performed using protocols that we developed. Subjects with a level I or II classification on the GMFCS performed different protocols. All subjects practiced walking on the treadmill before testing for a maximum of 3 minutes. The SRT-I was used for the subjects who were classified at level I on the GMFCS, and the SRT-II was used for the subjects who were classified at level II on the GMFCS.

To assess validity, we compared the VO_{2peak} achieved on the treadmill and the VO_{2peak} achieved on the 10-m shuttle run test. During the study, all subjects performed 1 treadmill test and 2 identical shuttle run tests within 2 weeks to assess the reproducibility of the measurements. One shuttle run test and the treadmill test were performed while the subjects were wearing a facemask for gas analysis. The subjects also completed one shuttle run test without a facemask and consequently no gas analysis. One child did not wear a facemask because the thought of breathing through the mouthpiece frightened her, and only the heart rate was monitored during all tests for that subject. The testing was done while the subjects were wearing regular sportswear and shoes, and orthoses if applicable.

In the tests followed by gas analysis, the subjects wore a firmly fitted facemask attached to a calibrated mobile gas analysis system (Cortex Metamax B3)[†] with an in-built gas analyzer, which allowed breath-by-breath gas analysis throughout the tests. Measurements of cardiopulmonary variables were collected. Before each trial started, the subjects rested until 1 minute had passed without an increase in heart rate. The subjects' heart rate was measured continuously, and the measurements were saved to a storage device during all tests using a reliable and accurate heart rate monitor.[‡][26] The Cortex Metamax is a valid and reliable system for measuring ventilatory parameters during exercise.[27-29]

The mobile gas analysis system consisted of a facemask, a transmitting unit (containing different oxygen and carbon dioxide gas analyzers), and a receiving unit. The transmitting unit with facemask with tubing (total weight=0.57 kg) was attached to the subjects with a harness, and the receiving unit was connected to a laptop computer located within 500 m of the transmitting unit. Metabolic stress test software (Metasoft, Version 2.6) was used to measure breath-by-breath minute ventilation, oxygen consumption (VO_2), carbon dioxide production (VCO_2), and heart rate and to calculate the respiratory exchange ratio ($RER=VCO_2/VO_2$). During all tests, the subjects were verbally encouraged to run as long as possible.

During the shuttle run tests without gas analysis, only the subjects' heart rate was monitored. The heart rate was read from the wrist monitor at the end of the test and recorded on the datasheet.

Measures and Treadmill Test and 10-m Shuttle Run Test Protocols

GMFCS. The GMFCS, translated into the Dutch language, was used by a pediatric physical therapist (OV), who was experienced in using the GMFCS, to classify the children and adolescents with CP into groups based on their functional ability. Level I represents the highest level of functional abilities, and level V represents the lowest level of functional abilities. Due to the physical demands of the tests, only children and adolescents who were classified at GMFCS level I (able to walk indoors and outdoors and climb stairs without limitation) or level II (able to walk indoors and outdoors and climb stairs holding on to a railing, but experience limitations in walking on uneven surfaces and inclines and in walking in crowds or confined spaces) were recruited. The original GMFCS has been reported to yield reliable and valid data for children aged 6 to 12 years.[23] Children over 12 years of age were classified using the same criteria as those used for 6- to 12-year-olds.

[†] Cortex Medical GmBh, Leipzig, Germany

[‡] Polar, Kempele, Finland

Treadmill tests. We could not adhere to "classic" treadmill protocols (Bruce protocol or Balke protocol) because they are not appropriate for children with CP due to the gait disturbance and accompanying problems. We performed a pilot study to develop the new test protocols. The starting speed and increase in speed were adjusted until mean total exercise time was between 8 and 17 minutes.[21] After this pilot study, we developed 2 treadmill protocols: 1 protocol for children who are classified at level I on the GMFCS and 1 protocol for children who are classified at level II on the GMFCS.

Our protocols started at 5 km/h for the GMFCS level I treadmill test and at 2 km/h for the GMFCS level II treadmill test. Speed was increased 0.25 km/h every minute by the assessor. Jones and Doust[30] showed that at the 2 lowest speeds, VO_2 during road running was not significantly different from treadmill running at low grades. Therefore, throughout the test we used a slope of 2% to compensate for the lack of air resistance, which results in a lower energy cost. With this incline on the treadmill, the results can be compared with those obtained during indoor running at the same speed.[30]

When walking, the subjects were permitted to support themselves with their fingers on the guardrails of the electromechanical treadmill (ENMil)[§] to maintain their balance. Subjects with a level II classification on the GMFCS performed their exercise tests on a wheelchair treadmill.[§] This treadmill is modified to accommodate the width of a wheelchair and is equipped with side guards. Children with a broad-based gait and other gait disturbances can stay more easily on this kind of treadmill. To ensure that the subjects were safe during the test, a therapist assisted them while walking behind or next to them. An emergency button was always within reach of the subjects and the therapist.

A maximal exercise test on the treadmill was done after a 3-minute treadmill practice session to familiarize the subjects with the equipment. The speed during this session was 2 km/h, and the practice session was followed by a 5-minute rest. The subjects were encouraged to push themselves to their limits, and the test was stopped when the subjects were unable or refused to continue the test despite encouragement.

All tests were performed under standardized conditions in a laboratory environment. The subjects maintained their normal diet before the day of testing. Only light physical activity was performed on the day before testing, and on the test day, subjects did not exercise before their test visit. During their visit, the subjects were given adequate explanation of the proposed protocol and its objectives.

[§] Eraf, Delft, The Netherlands

10-m shuttle run tests. Two new shuttle run tests for children with a GMFCS level I or II classification were developed. The starting speed and the increase in speed every minute are the same as for the treadmill protocol—5 km/h for the SRT-I and 2 km/h for the SRT-II—with the speed increased 0.25 km/h every minute.

Both shuttle run tests require children to walk or run between 2 markers delineating the respective course of 10 m, at a set incremental speed determined by a signal, which is played by a standard CD player. All subjects were accompanied by a physical therapist during the test to help them pace themselves with the audio signal. At the end of each level, the subjects were told to go a little faster. The test was finished when, on 2 consecutive paced signals, the subjects were more than 1.5 m away from the marker. Total exercise time was recorded and used for analysis.

The treadmill test and the shuttle run test with gas analysis were supervised by 2 experienced assessors (OV and TT). The shuttle run test without gas analysis was supervised by assessor (OV) and one pediatric physical therapist who was randomly chosen out of a sample of 6 therapists with no experience or formal training. One therapist was instructed to encourage the child, and 1 therapist (OV) accompanied the child during the test.

Feasibility

At the completion of the tests, subjects were asked a standardized question: “Which of the 2 tests, shuttle run or treadmill, did you prefer, and why?” All of the answers were recorded by the investigator for analysis.

Data Analysis

The data were analyzed using SPSS 12.0 and MS Excel 2003 for Windows. Intraclass correlation coefficients (ICC[2-way mixed]) for the number of levels completed were computed to assess test-retest reliability of data for both 10-m shuttle run tests. Acceptable reliability was considered to be an ICC value greater than .80.[31]

Limits of agreement also were calculated according to the procedure described by Bland and Altman.[32] A Bland-Altman plot is a graphic representation of the individual subject differences between the tests plotted against the respective individual means. Using this plot rather than the conventional test-retest scattergram, a rough indication of systematic bias and random error is provided by examining the direction and magnitude of the scatter around the zero line, respectively. Bland-Altman analysis describes the level of agreement between 2 measurements. In this analysis, the “precision” indicates how well the methods agree for an individual. By multiplying the precision by 1.96, the “limits of agreement” are calculated. This calculation represents the 95% likely range for the difference between a subject’s scores on 2 tests and is an indicator of absolute reliability. Typical error and total error were calculated as described by Hopkins.[33]Typical error was calculated as the standard deviation in each subject’s measurements between tests, after any shifts in the mean had been taken into account, and was expressed as a percentage of the subject's mean score to obtain a more easily interpretable percentage score. This percentage is also known as the "coefficient of variation." Total error was calculated as the average of all individual standard deviations for heart rate and time, based on the data of the 2 trials.[34] The level of statistical significance was set at $p = .05$.

In order to assess the amount of error associated with repeated measurements, the standard error of measurement (SEM) was calculated.[34] Standard errors of measurement between the 2 shuttle run test sessions were computed applying a 95% confidence interval. To determine whether there were significant differences for peak heart rate and exercise time between the treadmill tests and the shuttle run tests, the data were compared using paired *t* tests. Correlation coefficients (Pearson *r*) for VO_{2peak} achieved during treadmill tests and shuttle run tests were computed for validity. A linear regression analysis with backward elimination procedure also was performed to determine which measures could significantly predict VO_{2peak} in children and adolescents with CP.

Results

Peak RER was ≥1.0 and peak heart rate was >180 bpm in all subjects. These variables indicate that a maximal effort was reached during all tests.

Test-Retest Reliability for 10-m Shuttle Run Tests

The physiological variables measured during both shuttle run tests are described in Table 2. The test-retest reliability statistics of both exercise performances are shown in Table 3 and illustrated in Figures 1 and 2. Intraclass correlation coefficients (2-way mixed) for heart rate and exercise time were .87 or above for both shuttle run tests.

There were no significant differences in maximal heart rate and total exercise time between the 2 shuttle run tests. The results indicate that both 10-m shuttle run tests have good reproducibility.

The SEM values are shown in Table 2. The SEM values for exercise time ranged from 0.25 for the SRT-II to 0.42 for the SRT-I, and the SEM values for peak heart rate ranged from 1.52 for the SRT-II to 2.56 for the SRT-I.

Validity for 10-m Shuttle Run Tests and Treadmill Tests

Both 10-m shuttle run tests were compared with the treadmill tests for validity of VO_2 values. The peak heart rate values obtained during the GMFCS level I treadmill test ($\bar{X}$=192.9 bpm, SD=6.2) and the GMFCS level II treadmill test ($\bar{X}$=193.1 bpm,

Table 2. Reproducibility (Test-Retest) of Gross Motor Function Classification System (GMFCS) Level I and II Shuttle Run Test Measurements. (n=25)[a]

	Measurement 1		Measurement 2			
	Mean	**SD**	**Mean**	**SD**	**Change in Mean**	**SEM**
GMFCS level I						
HR_{peak} (bpm)	200.6	6.6	198.9	6.6	1.7	2.56
Exercise time (min)	8.2	2.3	8.6	2.0	0.4	0.42
GMFCS level II						
HR_{peak} (bpm)	197.9	6.6	197.3	5.2	0.6	1.52
Exercise time (min)	11.5	3.8	11.6	3.8	0.1	0.25

[a] Change in mean denotes the change between measurement 1 and measurement 2; SEM=standard error of measurement; HR_{peak}=peak heart rate.

Table 3. Reliability (Test-Retest) Statistics of the Gross Motor Function Classification System (GMFCS) Level I and II Shuttle Run Tests. (n=25)[a]

	ICC	**Typical Error**	**Total Error**	**LOA**	**Typical Error % (CV)**
GMFCS level I					
HR_{peak} (bpm)	.87	2.40	2.65	6.63	1.2
Time (min)	.97	0.36	0.43	1.01	4.8
GMFCS level II					
HR_{peak} (bpm)	.94	1.56	1.52	4.32	0.8
Time (min)	.99	0.31	0.30	0.86	2.7

[a] ICC=intraclass correlation coefficient, LOA=limits of agreement, CV=coefficient of variation, HR_{peak}=peak heart rate.

SD=6.1) were significantly lower ($p < .05$) from those obtained during the GMFCS level I and II shuttle run tests ($\overline{X}$=200.6 bpm, SD=6.7, and $\overline{X}$=199.4 bpm, SD=6.8, respectively). The subjects reached this higher peak heart rate in a significantly ($p < .05$) shorter time during the shuttle run tests compared with the treadmill tests (Tab. 4).

The physiological variables measured on both tests in which VO_{2peak} values were obtained are described in Table 4. Validity statistics for the shuttle run tests and the treadmill test in which VO_{2peak} values were obtained are shown in Table 5 and Figure 3. Pearson correlation coefficients for VO_{2peak} achieved during the shuttle run test with gas analysis and the treadmill test were .96 for subjects with a level I classification on the GMFCS and .96 for subjects with a level II classification on the GMFCS. The results indicate that both 10-m shuttle run tests are valid measures of aerobic capacity (VO_2) in children and adolescents with CP.

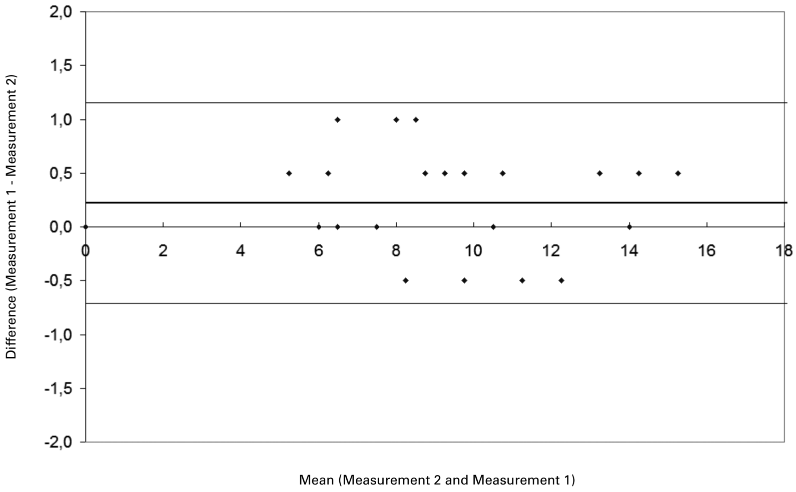

Figure 1. Bland-Altman plot of exercise time during measurement 1 and measurement 2 for Gross Motor Function Classification System (GMFCS) Levels I and II. The bold-type line shows the mean difference between the 2 measurements, and the 2 thin lines indicate ±2 standard deviations. On the X-axes, the average exercise time from both tests is displayed. On the Y-axes, the difference between the exercise times during measurement 1 and measurement 2 is displayed.

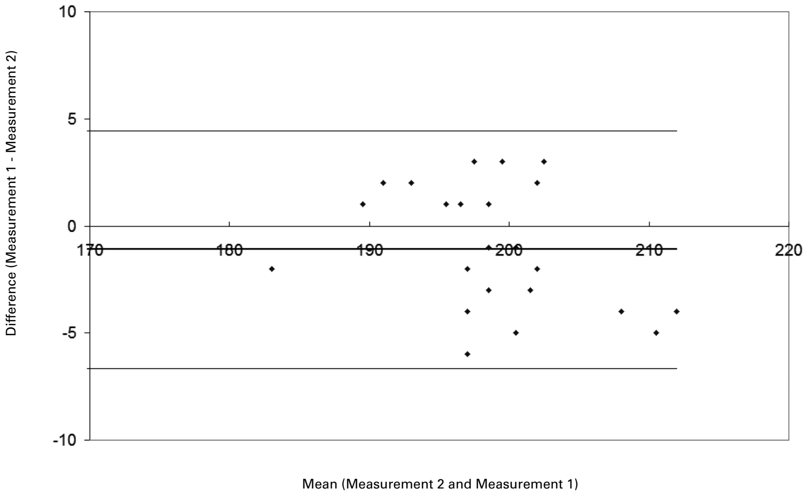

Figure 2. Bland-Altman plot of peak heart rate (HR_{peak}) during measurement 1 and measurement 2 for Gross Motor Function Classification System (GMFCS) Levels I and II. The bold-type line shows the mean difference between the 2 measurements, and the 2 thin lines indicate ±2 standard deviations. On the X-axes, the average HR_{peak} from both tests is displayed. On the Y-axes, the difference between the HR_{peak} during measurement 1 and the HR_{peak} during measurement 2 is displayed.

Table 4. Comparison of Gross Motor Function Classification System (GMFCS) Level I and II Shuttle Run Tests With Gas Analysis and Treadmill Tests. (n=24)[a]

	10-m SRT-STG		**Treadmill Tests**		
	Mean	**SD**	**Mean**	**SD**	**Change in Mean**
GMFCS level I					
VO_{2peak} (L/min)	1.7	0.5	1.7	0.5	0
RER	1.1	0.1	0.9	0.9	0.2
HR_{peak} (bpm)[b]	200.6	6.7	192.9	6.2	7.7
Exercise time (min)[b]	8.2	2.2	10.6	4.0	2.4
GMFCS level II					
VO_{2peak} (L/min)	1.7	0.6	1.6	0.6	0.1
RER	1.0	0.1	1.0	0.8	0
HR_{peak} (bpm)[b]	199.4	6.8	193.1	6.1	6.3
Exercise time (min)[b]	11.5	3.8	13.4	4.1	1.9

[a] SRT-STG=shuttle run tests with gas analysis, VO_{2peak}=peak oxygen uptake, RER=respiratory exchange ratio, HR_{peak}=peak heart rate. In one subject, VO_{2peak} and RER were not monitored, and therefore this subject's data were not included in the validity analysis.

[b] $p < .05$ for the comparison of the 10-m SRT and the treadmill test.

Table 5. Validity Statistics[a] of the Shuttle Run Tests With Gas Analysis and the Treadmill Tests (n=24)[b]

	r	**Typical Error**	**95% CI of Typical Error**	**Total Error**	**LOA**	**Typical Error % (CV)**
GMFCS level I						
VO_{2peak} (L/min)	.96	0.10	0.07-0.17	0.10	0.28	5.4
GMFCS level II						
VO_{2peak} (L/min)	.96	0.13	0.09-0.23	0.15	0.37	7.1

[a] Pearson product moment correlation (r).

[b] CI=confidence interval, LOA=limits of agreement, CV=coefficient of variation, VO_{2peak}=peak oxygen uptake. In one subject, VO_{2peak} and RER were not monitored, and therefore this subject's data were not included in the validity analysis.

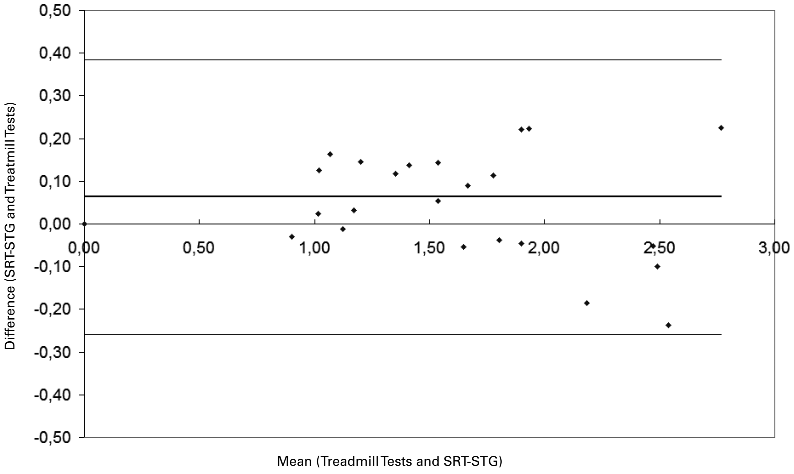

Figure 3. Bland-Altman plot of peak oxygen uptake (VO_{2peak}) during shuttle run tests with gas analysis STG) and treadmill tests for Gross Motor Function Classification System (GMFCS) Levels I and II. The bold-type line shows the mean difference between the 2 measurements, and the 2 thin lines indicate ±2 standard deviations. On the X-axes, the average VO_{2peak} from both tests is displayed. On the Y-axes, the difference between the VO_{2peak} during the treadmill tests and the VO_{2peak} during the STG is displayed.

Feasibility

The tests were generally well tolerated by the subjects. At the completion of tests, subjects were asked a standardized question: "Which of the 2 tests, shuttle run or treadmill, did you prefer, and why?" Of the 25 subjects, 23 preferred the shuttle run test over the treadmill test, 1 preferred the treadmill test over the shuttle run test, and 1 did not favor any type of testing. Predominant reasons for the shuttle run test preference were that the subjects felt they were in control of the test and did not need to acquire new skills. Several subjects added that they could "try harder" with the shuttle run test than in the treadmill test because they knew they "could stop at any time." Moreover, their gait disturbance was less of a problem during this field exercise test compared with the treadmill test.

A backward regression analysis indicated that VO_{2peak} or VO_{2peak}/kg could not be predicted from treadmill exercise time, maximal treadmill running speed, maximal shuttle run test running speed, or GMFCS level. The only significant predictors of VO_{2peak} and VO_{2peak}/kg were sex and body weight: $\overline{X}=0.29 \times \text{weight} - 0.474(\text{sex})[1=\text{male}, 2=\text{female}] + 1,159$; $r=.84$, $p < .0001$; standard estimate of error=0.31.

Discussions and conclusions

Field testing is widely used to estimate aerobic capacity in children and adolescents because it is impractical to obtain laboratory measurements for large groups of people.[14,35-38] Although it is generally agreed that cardiovascular field tests are good measures of cardiorespiratory fitness, it is sometimes difficult to compare individuals' performance on different field tests because scores are not often converted into VO_{2peak} values. Furthermore, to date, there are no validated field tests for children and adolescents with CP. The results of this study demonstrate that both recently developed 10-m shuttle run tests are reproducible, clinical applicable, and valid measures in children and adolescents with CP.

The shuttle run tests and treadmill tests were similar in that steady increments in heart rate with an increase in exercise intensity were apparent. The peak heart rate during the treadmill tests was different from that obtained during the shuttle run tests. The subjects reached a higher peak heart rate in a shorter time during the shuttle run tests. As with conventional maximal tests, there was a linear relationship between heart rate and VO_2 in the shuttle run and treadmill tests. Thus, while the newly developed shuttle run tests do not in any way challenge the use and significant benefits of conventional methods of exercise testing, our study suggests that both shuttle run tests have several characteristics that are similar to the accepted gold standard.

A limitation of this study is the fact that the treadmill VO_2 setup has never been validated in children and adolescents with CP. However, pilot testing showed that measurements of maximal VO_2 during an exercise test on the treadmill were reproducible. Together with the high maximal heart rates obtained in our subjects and the high RER values,[39] one might assume that treadmill testing yields valid measurements of maximal VO_2 in children and adolescents with CP. Therefore, we used the treadmill protocol as the gold standard, and we compared the results of the treadmill tests with the results of both shuttle run tests.

There are important differences between the shuttle run tests and the treadmill tests. First, during the shuttle run tests, a child is allowed to run faster than the set speed. The energy cost of changing direction (by 180° in the case of the shuttle run tests) and of decelerating and then accelerating is greater than continually running in a straight line on a treadmill. Children who are efficient in changing direction will be favored in this test compared with children who are poor in changing direction. However, in daily life, children require repeated changes of direction. Thus, the shuttle run tests may be more useful in predicting physical performance than a continuous gas analysis VO_{2peak} test on a treadmill. Second, when running on a treadmill, it is not possible to run faster or slower than the treadmill, because the running speed is externally paced. While performing the shuttle run tests, it is possible for a person to walk faster than the speed determined by the signal.

For some children, it is difficult to run at the audio signal that determines the running speed. Therefore, it is recommended that during the first stages of the test, a person should assist the child.[40] If the child understands the principle of the shuttle run tests, he or she can continue the test without assistance. If children have problems pacing themselves, they should be accompanied throughout the test. In these situations, an extra person to collect reliable and valid shuttle run test information is necessary.

In our study, there was no difference between the shuttle run tests for heart rate and exercise time. However, in both 10-m shuttle run tests, the exercise time was shorter than in the treadmill tests. This difference may have been due to the fact that a lot of energy was used to make a turn every 10 m. Some authors[10,41] have stated that the energy used for postural stabilization, high muscle tone, and involuntary movements causes an increase in VO_2 during submaximal exercise. During the treadmill tests in our study, the energy used for stabilization may have been low because the subjects were allowed to hold on to the guardrails with their fingers.

In the rehabilitation and fitness setting, VO_{2peak} is often estimated from the maximal speed and grade attained during a maximal treadmill exercise test.[28] For the exact determination of VO_{2peak} during exercise, a respiratory gas analysis system is necessary. However, this equipment is expensive and not always available. It would be of value if the VO_{2peak} could be predicted from surrogate measures in children with CP, because many clinicians do not have a respiratory gas analysis system. Unfortunately, in our study, neither maximal running speed nor treadmill exercise time could accurately predict VO_{2peak} in children with CP. Therefore, the shuttle run tests could be used only as an instrument to monitor changes in exercise capacity. The exercise time or level achieved is the most useful outcome measure. The heart rate during the shuttle run tests can be used to check whether a child has performed maximally (heart rate >180 bpm).

The calculated SEM can be used to determine the range in which a person's "true score" could be expected to lie, considering the amount of error associated with repeated measures. For example, we can be 95% confident that the "true score" for people performing the shuttle run tests lies within ±2 SEM. Thus, a change in person's performance of greater than 2 SEM most likely represents a real change that may not be attributed to measurement error. Based on the data in the "Results" section, total increases of >0.84 (2 × 0.42) minute for the SRT-I and 0.50 (2 × 0.25) minute for SRT-II could be attributed to real change with 95% confidence.

In this study, the subjects demonstrated a preference for the 10-m shuttle run tests over the treadmill tests. The shuttle run tests are nonthreatening and can easily be performed. A person can terminate the test at any point that he or she chooses. Moreover, as shuttle run tests require a child to either run or walk between 2 markers, the tests do not necessitate acquisition of new skills to participate. Shuttle run tests can be widely used in a mainstream school, schools for special education, or rehabilitation centers. None of the children or adolescents in our study required extensive instructions to participate. Both field tests seem to be useful for evaluating the aerobic performance of children and adolescents who are classified at level I or II on the GMFCS.

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IN CHILDREN AND ADOLESCENTS
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WITH CEREBRAL PALSY

4

RELIABILITY FOR RUNNING TESTS FOR MEASURING AGILITY AND ANAEROBIC MUSCLE POWER IN CHILDREN AND ADOLESCENTS WITH CEREBRAL PALSY

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Abstract

Purpose We investigated reliability, construct validity, and feasibility of two sprint tests for children with cerebral palsy (CP).

Methods Twenty-six children with CP participated (7-18 years of age; Gross Motor Function Classification System [GMFCS] level I or II). On different occasions, the 10 x 5 Meter Sprint Test and the Muscle Power Sprint Test were scored by different assessors.

Results Excellent interobserver reliability (intraclass correlation [ICC] = 1.0 and ICC ≥ 0.97) and test-retest reliability (ICC = 0.97 and ICC ≥ 0.97) were obtained. Scores differed significantly on both sprint tests for children classified at GMFCS level I and level II. Mean scores for feasibility ranged from 8.8 to 9.2 on a 10 cm. visual analog scale (10 = the best).

Conclusions Both exercise tests are reliable and have good feasibility for children and adolescents with CP (GMFCS level I or II). Construct validity is supported for both sprint tests between children classified at GMFCS level I and level II.

Introduction

Children with spastic cerebral palsy (CP) often have poor physical fitness[1,2] (muscle strength, anaerobic muscle power and aerobic capacity), which also may compromise their daily childhood functioning. Most of the current literature related to children with CP is focused on aerobic capacity[2-5] and muscle strength.[6-10] Less attention has been paid to high-intensity exercise lasting only a few seconds,[1,11,12] despite the fact that almost all daily childhood activities are more commonly of short-term high-intensity, than long-term activity.[13,14] In these short-term activities sufficient anaerobic muscle power and agility are extremely important for children with CP.

Anaerobic muscle power is the maximal anaerobic adenosine triphosphate (ATP) per second yield by a subject, during a specific type of short-duration, maximal exercise (e.g., 30 seconds).[15] Anaerobic muscle power generation is limited by the rate at which energy is supplied (ATP production) for the muscle contraction (ATP utilization). Anaerobic muscle power refers to the ability of the neuromuscular system to produce work in a short time period. In this report, peak power is defined as the highest power that can be generated during exercise of up to 30 seconds, while mean power is defined as the average power generated in 30 seconds.

Because many of a child's daily activities consists of short-term bursts of intense activity, anaerobic muscle power is thought to be an important measure of functional ability.[14] Bar-Or[12] stated that in children with a neurodevelopmental disease, anaerobic muscle power might be a better measure of functional ability than aerobic capacity.

For children with CP, peak power and mean power of the lower limbs have been reported to be distinctly subnormal.[1,12] Irrespective of the scaling method used (absolute or relative to body weight), when compared with control data derived from healthy children, children with CP scored between two and four standard deviations below the expected values.[1,11,16]

Anaerobic muscle power in children with CP has predominantly been measured using the Wingate Anaerobic cycling Test (WAnT).[17] The WAnT is a 30-second cycling test at all-out speed, against a constant braking force. The WAnT typically has been used in children developing typically as well as in children with neuromuscular diseases[1,11,18] and has been found to be a reliable and valid method. The WAnT, however, is more specifically geared to cycling, not to running. The necessary equipment is expensive, may require modification for the use in children with CP, and may not be readily accessible for most therapists. Moreover, impairment of the motor control system can reduce optimal performance during cycling. Specifically, maximizing anaerobic muscle power in cycle ergometry testing is complicated by difficulties with sustaining the circular motion of the pedals because of the necessary application of force on the pedal and the skill and co-ordination required for that task.[1,19]

The 10 x 5 Meter Sprint Test is currently used in clinical practice[20-22] and is an inexpensive measure that doesn't require special equipment and additionally is a measure of agility. Agility is the ability to change the direction of the body in an efficient and effective manner. To achieve this, a child needs a combination of balance, speed, muscle strength, and coordination. Children with CP often have difficulties changing direction of the body abruptly or shifting quickly the direction of movement without losing balance (agility).

To date, there is no reliable running-based exercise test to evaluate the effects of training programs that focused on agility and/or anaerobic muscle power in children with CP. For children and adolescents with CP, the test should be non-threatening, inexpensive, easy to administer in a nonresearch setting, and should be able to be administered in a short timeframe. Moreover, reliability is an important issue for clinical use, in follow-up as well as in clinical trials. The newly developed Muscle Power Sprint Test (MPST)[23] measures a different aspect of short term running performance: anaerobic muscle power.

In the present study, we evaluated the 10 x 5 Meter Sprint Test and the MPST with respect to reliability, construct validity, and feasibility in children with CP. To examine construct validity, results from both sprint tests were compared in children with different gross motor function as measured by the Gross Motor Function Classification Measure (GMFCS). We hypothesized a significant relationship between the time taken to perform both sprint test and the child's level on the GMFCS.[24]

Methods

Subjects

A convenience sample of thirty children and adolescents from a school for special education ('Ariane de Ranitz', Utrecht, The Netherlands) were invited to participate in the study. To be included, subjects had to be between seven and 20 years of age, diagnosed with spastic CP, and classified at level I or II on the GMFCS.[24] All children in the study were able to follow simple commands, had no contraindications or comorbidities, and were not ill or in pain. They were all receiving rehabilitation services at the time of the study.

In total 26 subjects (16 males, 10 females) and their parents agreed to participate and signed the informed consent form. Four subjects did not participate because they were participating in another study. Group characteristics according to GMFCS level are described in Table 1. The study was approved by the Institutional Review Board of the University Medical Center Utrecht.

Table 1. Subject characteristics. (n=26)

Number	**GMFCS I, n = 15 (10 male, 5 female)**				**GMFCS II, n = 11 (6 male, 5 female)**			
Variables	**Mean**	**SD**	**Median**	**Range**	**Mean**	**SD**	**Median**	**Range**
Age (y:m)	11.6	2.8	12.1	7.5-16.1	10.9	2.4	12.1	7.2-17.0
Height (cm)	148.7	15.3	149.0	125.0-175.0	148.6	18.9	145.0	123.0-175.0
Body mass (kg)	40.3	12.4	35.1	23.8-60.8	38.6	12.1	32.7	24.0-59.7
Body Mass Index (kg/m^2)	17.9	3.7	15.8	14.2-26.1	17.1	2.1	17.5	13.9-20.7

Legend: SD, standard deviation; GMFCS, Gross Motor Function Classification System

Procedures

Before testing, each child's body mass and height were measured. The participants' body mass was determined using an electronic scale (Seca, Hamburg, Germany). Height measurements were taken while the subject stood against a wall. The 10 x 5 Meter Sprint Test and the MPST were measured in a random order. Eight pediatric physical therapists performed all tests in randomly, changing pairs. Twelve children completed the MPST first, and 14 children performed the 10 x 5 Meter Sprint Test first.

All therapists were experienced pediatric physical therapists. Before data collection, the therapists were given written instructions in the application and scoring of the 10 x 5 Meter Sprint Test and the MPST, but had no formal training. To assess interobserver reliability for the 10 x 5 Meter Sprint Test and the MPST, two therapists assessed the subject at the same time. Both tests were performed on different days in the same week. So that the observers could assess test-retest reliability, the subject performed the same test at the same time and day in the following week. The same observers administered these tests. During the tests, the children were verbally encouraged to run as fast as they could. After each assessment, the child (if necessary with help of one therapist) and therapists filled out the feasibility questionnaire. The patients were tested and retested within two weeks.

Measures

GMFCS. The GMFCS,[24] translated into the Dutch language, was used by a pediatric physical therapist (OV), experienced in using the GMFCS, to classify the children and adolescents with CP into groups based on their functional ability. Level I is the highest level of functional abilities and level V the lowest. Because of the physical demands of the tests, we recruited only children and adolescents who were classified at GMFCS level I (i.e., able to walk indoors and outdoors, and climb stairs without limitation) or level II (i.e., able to walk indoors and outdoors, and climb stairs holding onto a railing but experience limitations in walking on uneven surfaces and inclines, and walking in crowds or confined spaces). Reliability and validity of the original GMFCS has been reported to be good to excellent for children age six to 12 years of age.[24] Children older than 12 years of age were classified using the same criteria as those used for six to 12-years of age.

10 x 5 Meter Sprint Test. Previous investigations into the 10 x 5 Meter Sprint Test with children developing normally have demonstrated good reliability.[25,26] The assessment of agility by measuring the time taken to execute the 10 x 5 Meter Sprint Test yields a good indication of a child's capacity in difficult tasks, for example, in transitions from running, turning, and resuming a run, one can assess if these are completed without falling, tripping, or deviating off course. However, the test does not provide information about short-term muscle power. The 10 x 5 Meter Sprint Test is a continuous sprint test. While performing this test, the child has to make nine fast turns after finishing every five meters. The child is not allowed to take a rest between each run. For children with CP who have problems with movement coordination, this is a problematic and/or difficult task, and the test measures agility rather than muscle power. The reliability and feasibility of this test has never been examined for children with CP.

The testing was performed in the gym while the children were wearing their usual clothing and shoes (and orthoses if applicable). Before the test was undertaken, there was a preparatory session in which the child performed the test at walking speed to make sure the subject understood how he/she had to perform the test. Normally, a three-minute recovery period is sufficient to repeat short running sprints without substantial fatigue.[27] Therefore, the subjects were given the cues "ready", "three", "two", "one" and "go" after a three-minute rest period. The subjects were instructed to complete 10 runs of five meters at a maximum pace. The distance (5 m) was marked by two taped lines on the floor and by cones. The subject had to run as fast as possible to each line, had to place at least one foot on each line, make a turn and run back as fast as possible. On the next line the subject had to make a similar turn, etc. There was no rest between the runs. At the end of the tenth run the subject had to cross the finish line. Two independent assessors assessed the time to a tenth of a second for the total 50 meters (10 x 5 meter) using a stopwatch and registered the time on a score form.

MPST. Because information about short-term muscle power can be of clinical importance for the clinician who wishes to measure objectively muscle function in children with CP, we developed a new running based exercise test: MPST.[23]

This test is similar to the WAnT in that it provides information about mean and peak power. The WAnT is more specific for cycling, and is a 30-second test at all-out speed,[17] while the MPST is a test that is specifically geared to running and consists of six separated sprints at maximum speed. There were several try-outs in which children and adolescents with CP (GMFCS-levels I or II) performed the MPST. Running-distances were modified until mean total test-time was around 30 seconds. In this new sprint test children have to complete six 15-meter runs at a maximum pace. The MPST is an intermittent sprint test, in which the child stops and starts at standardized intervals.

Before executing the test, the child performed the test at walking speed as a preparing warm-up session and to make sure the subject understood how to perform the test. After this practice session, the subjects were given a rest period of three minutes to recover.[27] For the MPST, they were instructed to complete six 15-meter runs at a maximum pace. The 15-meter distance was marked by two lines taped on the floor. Cones were placed at both ends of the lines. The subject had to run as fast as possible from one line to the other, and was instructed to cross the line. Between each run, the subject was allowed a timed 10-second rest before turning around and get prepared for the following sprint.

Testing was performed in a corridor in school. The children were wearing their usual clothing, shoes and orthoses if applicable. The subjects were given the cues "ready", "three", "two", "one" and "go" for the first run. For the second through the sixth run the assessors counted backwards from "ten" to "one" and then gave the cue "go". Two independent therapists recorded the time to a hundredth of a second for each 15-meter run using a stopwatch and recorded the time on a score form.

Power output for each sprint was calculated from the collected data using the following equations:[28]

- Velocity (m/sec) = 15 meter / Time
- Acceleration (m/sec^2) = Velocity / Time
- Force (kgm/sec^2) = Body mass x Acceleration
- Power (Watts) = Force x Velocity

For each of the six 15-meter runs the power was calculated and then the following parameters were determined. Peak power was defined as the highest calculated power output. This parameter provides information about the maximal sprint speed. Mean power was defined as average power output during the six runs. This parameter provides an indication of a child's ability to maintain power-output over time. The greater the average power output, the better the child's ability to maintain anaerobic performance. Mean power is considered the most important parameter during an anaerobic exercise test.[29]

Feasibility questionnaire

We developed a questionnaire (Appendix 1) that consists of five questions: three for the child and two for the assessors. Each question was answered using a 10-cm line Visual Analogue Scale (VAS). The child-form of the VAS had a picture of a green smiling face on the left side and red sad-looking face on the right side. The assessor-form had no pictures, but the words 'easy' and 'minimal' on the left side and 'difficult' and 'maximal' on the right side. In addition the assessors-form had a special box in which the assessors could comment on the application of the test.

Data Analysis

The data were analyzed using SPSS 12.0 (SPSS Institute, Chicago, IL) and MS Excel 2003 for Windows (Seatle, WA). Correlation coefficients, that is, Pearson's correlations (R) and intra-class correlations (ICC; two-way mixed), were computed for interobserver and the test-retest reliability. Acceptable reliability criteria for ICC values were values > 0.80.

Moreover, limits of agreement (LOA) were calculated to conform to the procedure described by Bland & Altman.[30] Bland-Altman analysis describes the level of agreement between two measurements. In this analysis, 'bias' is an estimate of how closely on average two measurements agree, and 'precision' indicates how well the methods agree for an individual. By multiplying the precision by 1.96, the LOAs are calculated. Typical error and total error were calculated according to Hopkins.[31] Typical error was calculated as the standard deviation in each subject's measurements between tests, after any shifts in the mean have been taken into account. Thereafter, the typical error was expressed as a percent of the subject's mean score to obtain an easier interpretable percentage score. This percentage is also known as the coefficient of variation. Total error was calculated as the mean of each subject's standard deviation between the trials.[32] The level of statistical significance was set at p = .05.

To assess the amount of error associated with repeated measurements, the standard error of measurement (SEM) was calculated.[32] SEMs between both tests were computed applying a 95% confidence interval.

To examine construct validity, we used an independent-samples t test to search for differences between GMFCS levels I and II for the mean and peak power derived from the MPST and for the results of the 10 x 5 Meter Sprint Test. The level of stati-stical significance was set at p = .05.

Results

All 26 subjects successfully performed both tests two times. The following results refer to both sexes and GMFCS levels combined. The physiological variables measured on both exercise tests can be found in Table 2.

Table 2. Reproducibility (test-retest) of the performance in the MPST and the 10 x 5 Meter Sprint Test. (n=26)

	Measure 1 Mean	SD	Measure 2 Mean	SD	Change in Mean	Range	P-value
MPST							
Peak Power (Watts)	101.1	84.5	94.0	75.6	7.1	10.7-350.4	NS (.17)
Mean Power (Watts)	78.5	66.6	76.0	60.8	2.5	9.4-272.3	NS (.24)
10 x 5 Meter Sprint Test							
Time (sec.)	32.1	9.4	32.1	9.7	0	19.3-57.8	NS (.91)

SD, standard deviation; NS, not significant. Change in mean denotes the change between measurement 1 and measurement 2. The mean score is the average of the scores from the 2 observers that administered the test.

Table 3. Reliability statistics (interobserver) of the MPST and the 10 x 5 Meter Sprint Test. (n=26)

	R	ICC	Typical Error	Total Error	LOA	Typical Error % (CV)
MPST						
Peak Power (Watts)	0.97	0.97	14.34	14.96	39.74	10.6
Mean Power (Watts)	0.99	0.98	8.78	8.94	24.33	7.1
10 x 5 Meter Sprint Test						
Time (sec.)	1.00	1.00	0.14	0.14	0.38	0.4

R: Pearson's product moment correlation, ICC: intra-class correlation, LOA: limits of agreement, CV: coefficient of variation.

Reliability

In Table 3, the interobserver reliability statistics of both the 10 x 5 Meter Sprint Test and the MPST can be found. R and ICC values for the 10 x 5 Meter Sprint Test of 1.00 were found. The total error (expressed as a percentage) of the 10 x 5 Meter Sprint Test showed a very low measurement error (0.4%). R values and ICC values for the MPST for mean power and peak power were found to be 0.97 and higher. The total error showed that the mean power was the variable with the lowest error of measurement.

In Table 4 test-retest reliability statistics for both the 10 x 5 Meter Sprint Test and the MPST are shown. R and ICC values for the 10 x 5 Meter Sprint Test were 0.97. The total error of the 10 x 5 Meter Sprint Test showed a low error of measurement (5.4%). R and ICC values for mean power and peak power were 0.97 or greater for the MPST.

Table 4. Reliability (test-retest) statistics of the MPST and the 10 x 5 Meter Sprint Test. (n=26)

	R	ICC	Typical Error	Total Error	LOA	Typical Error % (CV)	SEM
MPST							
Peak Power (Watts)	0.97	0.98	14.51	15.23	40.19	14.0	13.9
Mean Power (Watts)	0.99	0.99	7.22	7.27	20.01	10.3	9.0
10x5 meter sprint test							
Time (sec.)	0.97	0.97	1.59	1.58	4.41	5.4	1.6

Legend: R: Pearson's product moment correlation, ICC: intra-class correlation, LOA: limits of agreement, CV: coefficient of variation, SEM: standard error of measurement.

The test-retest data are based on the two assessors that tested the subject on both tests. As can be appreciated from the Bland-Altman plots (Figs. 1-3), there were some obvious outliers. These outliers are included in the calculations, and the reliability statistics are still good.

SEM values are shown in Table 4. These values ranged from 9.0 for mean power to 13.9 for peak power derived from the MPST. For the 10 x 5 Meter Sprint Tests the SEM value was 1.6.

Construct validity

An independent-samples t test was conducted to compare the scores for the mean and peak power for GMFCS levels I and II. There was a significant difference (p = .007) in peak power scores for children classified at level I on the GMFCS (M = 130.7, SD = 83.1) and children classified at level II on the GMFCS (M = 51.1, SD = 40.5). A significant difference (p = .006) also was found in mean power scores for children classified at level I on the GMFCS (M = 102.4, SD = 63.4) and children classified at level II on the GMFCS (M = 39.6, SD = 30.8;t[24] = 3.0).

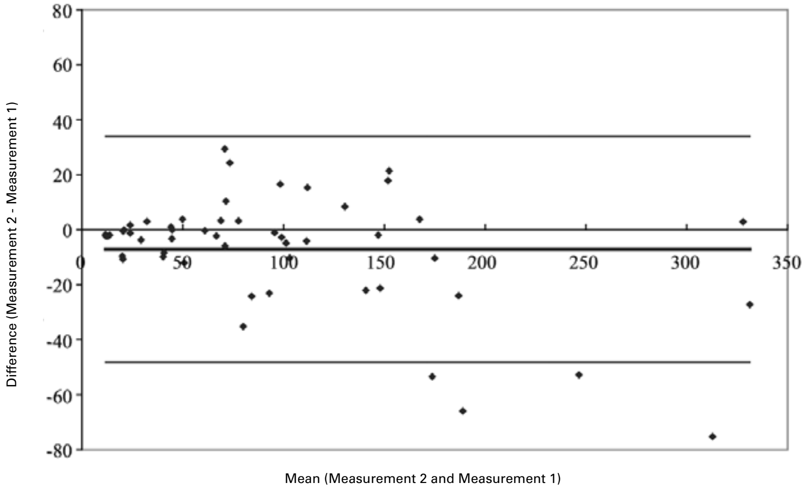

Figure 1. Bland–Altman plot of peak power during both test and retest on the MPST. The data that are used for this plot are the data from both assessors. The bold line shows the mean difference between the two measurements, the two thin lines indicate ± 2 standard deviations. On the X-axes the average peak power value from both tests is displayed. On the Y-axes the difference between the peak power during the test and the peak power during the retest is displayed.

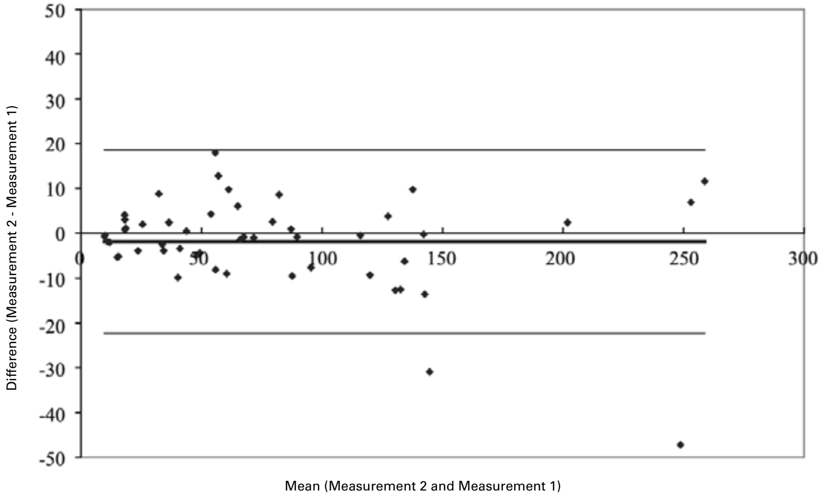

Figure 2. Bland–Altman plot of mean power during both test and retest on the MPST. The data that are used for this plot are the data from both assessors. The bold line shows the mean difference between the two measurements, the two thin lines indicate ± 2 standard deviations. On the X-axes the average mean power value from both tests is displayed. On the Y-axes the difference between the mean power during the test and the mean power during the retest is displayed.

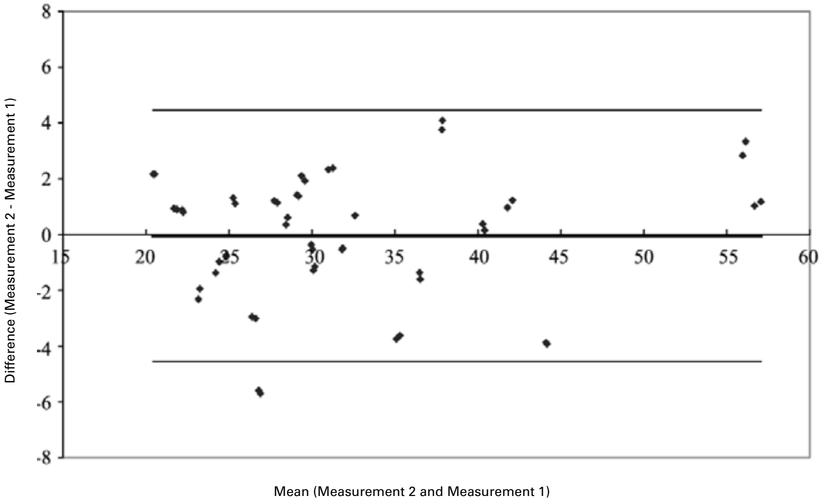

Figure 3. Bland–Altman plot during both test and retest on the 10 x 5 Meter Sprint Test. The data that are used for this plot are the data from both assessors. The bold line shows the mean difference between the two measurements, the two thin lines indicate ± 2 standard deviations. On the X-axes the time from both tests is displayed. On the Y-axes, the difference between the time during the test and the time during the retest is displayed.

For the 10 x 5 Meter Sprint Test, a significant difference (p = .002) was found in scores for children, classified at level I on the GMFCS (M = 27.5, SD = 5.6) and children classified at level II on the GMFCS (M = 38.5, SD = 9.9).

Feasibility

Table 5 presents results from the feasibility questionnaire. Mean scores ranged from 8.8 to 9.2 on the 10 cm VAS-scale for both tests, indicating a very high feasibility.

Discussion

In this study, we examined the feasibility, interobserver reliability and test-retest reliability and construct validity of a continuous and an intermittent sprint test in a group of children with CP, who were classified at GMFCS level I or II, using the 10 x 5 Meter Sprint Test and the MPST, respectively. The results demonstrate good feasibility, interobserver reliability, and test-retest reliability and construct validity for both tests. Subjects who performed both tests found the tests easy to perform. The observers also scored the tests as easy to perform and easy to administer.

Table 5. Feasibility Questionnaire Results for the MPST and the 10 x 5 Meter Sprint Test, reported by participating children (n=26, three questions) and by assessors (n=8, two questions).

	MPST			10 x 5 Meter Sprint Test		
	Mean	**SD**	**Range**	**Mean**	**SD**	**Range**
Children's questions						
Was the test easy (10) or difficult (0) to do?	9.0	1.0	5.5-10	8.8	1.0	7.0-10
Do you think the test was nice (10) or boring (0)?	8.9	0.9	7.0-10	9.0	0.9	6.5-10
Did you perform at minimal (0) or at maximal (10) level?	9.1	0.8	7.0-10	8.9	1.0	6.5-10
Assessor's questions						
Do you think the test was easy (10) or difficult (0) to administer?	9.0	0.9	7.0-10	9.2	0.5	8.0-10
Did the child, in your opinion, perform at minimal (0) or at maximal (10) level?	8.8	1.0	7.0-10	8.9	0.9	7.0-10

The large standard deviations for both tests suggest that there is a large inter-individual variability. This is likely the result of the large age-band and different classification levels of the subjects. For the 10 x 5 Meter Sprint Test the LOA were small. The LOAs of the MPST were large in the context of mean power and peak power that children generate. The coefficients of variation, which are particularly useful for comparing the reliability between performance tests, for the mean power and peak power test-retest reliability are also large (10% and 14%, respectively). In other studies that have assessed the reliability of the Wingate Anaerobic cycling Test in children with CP and myositis similar coefficients of variation were found. In a group of children with CP,[11] the coefficients of variation were 12.3 ± 12.1 for peak power and 14.2 ± 13.4 for mean power. Takken et al.[33] found coefficients of variation of 18.7 for peak power and 16.8 for mean power in a group of children with myositis. The coefficients of variation for the 10 x 5 Meter Sprint Test were small (5%). Therefore, the 10 x 5 Meter Sprint Test is the test that might be the most sensitive to change.

The total error (expressed as a percentage) showed that the mean power derived from the MPST was the variable with the lowest error of measurement. Therefore, the calculated mean power should be used as outcome measure. This is in accordance with the results of the WAnT, in which the mean power is considered the "gold standard."[29]

The calculated SEM can be used to determine the range in which a subject's "true score" could be expected to lie when the amount of error associated with repeated measures is considered. For example, we can be 95% confident that the "true score" for subjects performing the MPST lies within 2 SEM. Thus, a change in subject's performance of greater than 2 SEM most likely represents a real change that may not be attributed to measurement error. On the basis of the data in the results section, total increases of > 18.0 Watts (2 x 9.0) for mean power and a decrease of > 3.2 seconds (2 x 1.6) in exercise time for the 10 x 5 Meter Sprint Test could be attributed to real change with 95% confidence. Repeated periodically, the MPST and the 10 x 5 Meter Sprint Test can be used as a criterion for the effectiveness of rehabilitation treatment (e.g., physical therapy, fitness training) as well as the development of the activity level in this patient group.

For children with CP, anaerobic power is found to be distinctly subnormal (between two and four standard deviations below normal) on the WAnT.[1] It is of no additional value to compare the data from the 10 x 5 Meter Sprint Test and the MPST to data derived from children who are healthy, since therapy will not likely normalize their anaerobic exercise capacity. Optimizing the capacity of each individual patient must be the goal of the therapy.

Sprint tests are generally regarded to have face validity.[34] In this study, statistically significant differences in scores achieved on the 10 x 5 Meter Sprint Test and the mean power and peak power derived from the MPST within subgroups of children with CP (level I or level II on the GMFCS) are demonstrated. These findings support the construct validity of both instruments.

Ecological validity is the degree to which the behaviors observed and recorded in a study reflect the behaviors that actually occur in natural settings. Upper motor lesions have been demonstrated to cause atrophy of type II (fast) muscle fibers, resulting in a greater proportion of type I (slow) muscle fibers.[35] To measure muscle power, focused on type II muscle fibers, both sprint tests seem to be useful measures. In children with CP, activities predominantly are series of discrete jerky movements.[16] This supports the ecological validity of this kind of measurements.

The 10 x 5 Meter Sprint Test can be used to measure agility. It cannot be considered as a 'real' power test, according to Wilkie,[36] as the force component is not measured. The MPST can be considered as a muscle power test because the mean power and peak power are determined.

The 10x5 Meter Sprint Test and the MPST are inexpensive, nonthreatening, and easy to administer. Both tests do not need special equipment and training and are available for a variety of professionals working with children and adolescents with CP. The choice of the instrument depends on the goal of the intervention. The MPST measures the ability to exert muscular strength quickly. Therefore, when treatment is focused on muscle strength and high intensity exercises the MPST is the most appropriate outcome measure. When the intervention is focused on the ability to change the direction of the body abruptly or to shift quickly the direction of movement without losing balance the 10 x 5 Meter Sprint Test can be used to evaluate the training effects.

Our study, however, has some intrinsic methodological limitations. Just as for the WAnT,[33] both running tests are dependent on the individual's motivation. Currently there are no objective physiological criteria that can be used to establish a maximal performance of the patient. Moreover, maximal performance on the 10 x 5 Meter Sprint Test and the MPST may be influenced by other variables in children with CP, such as motor-planning, motor control, environmental factors, previous surgeries, and bracing. In the present study these variables were not studied.

The VAS provided us a subjective perception of the perceived exertion. We found motivation as well as encouragement of the child during the tests to be very important. As can be appreciated from the Bland-Altman plots (Figs. 1-3) seven obvious outliers were observed. Five of these subjects had the lowest scores on the performance-question (minimal/maximal) on the VAS scale. This means they did not perform at their maximal capacity during the exercise tests. These five outliers underline that exercise tests are influenced by the motivation of a patient to give a maximal effort and that a lack of motivation can influence the final test result.

The effects of the verbal encouragement of the different assessors on the child's performance were not studied. In the running-based anaerobic field tests and the WAnT, motivation and encouragement of the subjects play a very important role in the test performance. Therefore it is recommended that in future research this aspect will be studied as well.

Conclusions

This is the first study investigating field sprint tests in children with CP. This study found good feasibility and reliability for the 10 x 5 Meter Sprint Test and the MPST in children and adolescents with CP (GMFCS classification level I or II). The construct validity of both tests is supported by significant differences in scores between children classified at GMFCS level I and children at level II. To assess the muscle power during running performance in children with CP mean power derived from the MPST is the most appropriate outcome measure. To assess someone's running performance and coordination of speedy movements the 10 x 5 Meter Sprint Test is the most appropriate measure. In our opinion, the 10 x 5 Meter Sprint Test and the MPST can be incorporated in the exercise evaluation of the child with CP, classified at level I or II on the GMFCS.

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Appendix 1: Feasibility questionnaire

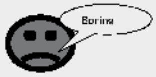
Children and adolescents were asked to answer the following questions:

1. Was the test easy or difficult to do?

I ---I

2. Do you think the test was nice or boring?

I ---I



3. Did you perform at minimal or at maximal level?

I ---I

The questions for the assessors were:

1. Do you think the test was easy or difficult to administer?

Easy I --- I Difficult

2. Did the child in your opinion perform at minimal or at maximal level?

Minimal I --- I Maximal


IN CHILDREN AND ADOLESCENTS
00.51.02
00.06.45
WITH CEREBRAL PALSY

5

RELIABILITY OF HAND-HELD DYNAMOMETRY AND FUNCTIONAL STRENGTH TESTS FOR THE LOWER EXTREMITY IN CHILDREN WITH CEREBRAL PALSY

Olaf Verschuren, Marjolijn Ketelaar, Tim Takken, Marco van Brussel,
Paul J. M. Helders, Jan Willem Gorter

Disability & Rehabilitation: accepted for publication

Abstract

Purpose To evaluate the intertester reliability of two methods for measuring lower-limb strength in children with cerebral palsy (CP).

Method Twenty-five subjects with CP (7-17 years of age) participated in this study. Lower-limb muscle strength was measured on 2 occasions using a Hand-held Dynamometer (HHD; break-method and make-method) and a 30-sec Repetition Maximum (RM) during three functional strength tests for the lower extremities. Reliability was measured using the intraclass correlation coefficients (ICCs), the standard error of measurement (SEM) and the coefficient of variation (CV).

Results The intertester reliability of strength measurement using a HHD was questionable with ICC values ranging from 0.42 to 0.73 for the break-method, and from 0.49 to 0.82 for the make-method. The SEM and CV(%) values ranged from 27.9 to 58.9 and 22.2% to 35.3% for the break-method and from 30.6 to 52.7 and 16.2% to 56.2% for make-method. The intertester reliability of strength measurement using the 30-sec RM was acceptable with ICC values ranging from 0.91 to 0.96, and SEM and CV(%) values ranging from 1.1 to 2.6 and 10.9% to 39.9% for the functional exercises.

Conclusion The intertester reliability of measuring muscle strength of the lower extremities using a hand-held dynamometer is questionable. The intertester reliability of the 30-sec RM for the lower extremity is acceptable.

Introduction

The assessment of muscle strength in children with cerebral palsy (CP) has become standard in clinical practice and research. Many children and adolescents with CP have a different distribution of muscle strength, and this can be associated with difficulties performing everyday functional activities.[1] Recent studies show that children with CP can benefit from strength training programs.[2-6] Therefore, it is important that clinicians and researchers who are interested in the efficacy of strength training programs have knowledge of the psychometric properties of their measurements of strength.

In general, there are two different muscle strength measurements that are used in children and adolescents with CP: isometric and isokinetic. An isometric-based test measures the ability of a muscle group to produce force without a change in overall muscle-tendon length. Isokinetic literally means 'same velocity' and refers to tests that are performed at a predetermined constant velocity.

In clinical practice, muscle strength is usually assessed by isometric resistance-based methods. Two methods to assess muscle strength are manual muscle testing (MMT) and the hand-held dynamometry (HHD). MMT uses a ordinal 6-point (0-5) Medical Research Council (MRC) scale.The HHD consists of a simple hand-held device equipped with a small internal load cell capable of measuring muscular force (in Newton).

Isokinetic dynamometry, most often used in a laboratory setting, uses computer-controlled equipment to measure the muscular force generated throughout a controlled movement. Isokinetic assessment has several advantages over isometric testing. Most activities of daily living involve phases of dynamic muscle action, and in this sense isokinetic testing may provide more specificity in terms of muscle action type than isometric testing.[7]

A maximal isometric contraction is only indicative of the capacity to produce force in that condition and at that particular muscle length, and can not necessarily be extrapolated to conditions where the muscle length is different or changing throughout the task.[8] Perhaps most important, however, is the inherent safety of isokinetic actions, afforded by the computer-controlled mechanism of accommodating resistance.

An alternative approach to measure muscle strength in a dynamical way is the use of the Repetition Maximum (RM) during functional exercises.[9] Since many lower extremity training and rehabilitation programs have been centred on functional exercises,[10,11] the RM during functional strength tests, based upon the specificity of training principle, may be better suited to detect actual improvements in lower extremity performance. Two ways in which this kind of functional strength tests may be used to assess lower extremity performance are by counting the number of repetitions performed at a specific activity over a specific period of time and by assessing the time necessary to complete a specified number of repetitions at a specific activity.[12,13] Because of the great diversity of functional abilities in young people with CP, counting the numbers of repetitions performed over a specific period of time seems the most appropriate. Using this way of testing all children can complete the test.

With respect to reliability of muscle strength measures it has been found that the reliability of MMT is fair when assessed for individual muscles.[14] Considerable training is required to achieve this modest reliability.[14] No data are available on the reliability of this method in children with CP. Moreover, this method is often not sufficiently sensitive to assess muscle strength of MRC grade 4 (active movement against moderate resistance) and grade 5 (normal strength; active movement against maximum resistance) or to detect small or moderate increases of strength over the course of rehabilitation.[15]

For the HHD, the topic of reliability is more complex. Many researchers use the HHD to measure muscle strength. However, in children and adolescents with CP there is no consensus about the way of testing that should be used. In published studies the break-method,[16,17] and the make-method[10,18] were used to measure muscle strength. In three studies that assessed intratester reliability in a group of children with CP, the used method (break or make) was different, and intraclass correlations were >0.79 for most muscle groups.[16,18,19] Damiano et al.[8] suggested that the peak value, achieved in a series of trials during the make-method, should be used to measure muscle strength in children with CP. Before using the HHD in general practice, training is generally recommended. This method might enhance reliability of measurement in this population. To date, there is no study in which the break and make method in a group of children and adolescents with CP has been compared.

Ayalon et al.[20] reported high intraclass correlation coefficients for test-retest reliability for isokinetic strength measurement in children with CP. Although these results are encouraging, many clinicians do not have access to expensive isokinetic dynamometry equipment.

Measuring muscle strength using a HHD, the most common way to assess muscle strength in studies involving children with CP, is difficult. The examiners need experience in hand-held dynamometry. HHD measures muscle strength in a single joint movement, whereas the functional strength tests are more related to the activity level and consist of multiple-joint movements. Moreover, the functional strength tests not only measure muscle strength, but also balance, coordination and some endurance and therfore correspond with the contemporary task-oriented approach.[21] The functional strength measures could be more appropriate for the use in everyday clinical practice. Testing is easily performed, functional and does not take a long time. Moreover, it seems that formal training is not required. To date, the reliability of the RM during functional exercises has not been studied in a group of children and adolescents with CP.

The consistency of measures of muscle strength is important for clinicians. In research and clinical practice it is preferable that a test can be reliably performed by different assessors. Therefore, we aimed to determine the intertester reliability of measuring lower-limb strength with the HHD (break vs. make) and the functional strength tests.

Methods

Subjects

A convenience sample of twenty-eight children and young adults from a school for special education were invited to participate in the study. To be included, subjects were required to be within the age range of 7 to 20 years, had to be diagnosed with Cerebral Palsy, and, due to the physical demands of the tests, classified as level I or II on the Gross Motor Function Classification System (GMFCS).[22] Cognitively, they had to be capable of understanding and following simple commands. Subjects had to have the degree of passive joint range of motion in the hips and knees that allowed them to assume the test positions (Table 1). The study was approved by the Institutional Review Board of the University Medical Center Utrecht.

Procedures

Prior to testing, each child was weighed on electronic scales (Seca, Hamburg, Germany). Height measurements were taken on the same visit while standing against a wall. Body composition was assessed using the sum of 7 skin folds method according to Pollack et al.[23] The measurements were taken at 7 sites (on the right side of the body: triceps, biceps, subscapular, suprailiac, mid-abdominal, medial calf, and front thigh) by the investigators (OV and TT) in accord with the American College of Sports Medicine guidelines.[24]

For the reliability study of the hand-held dynamometry, two examiners (OV and MvB), previously trained in hand-held dynamometry, performed hand-held dynamometry measurements on 5 muscle groups of the lower extremity (hip extensors, hip abductors, knee extensors, knee flexors, ankle plantar flexors), using break- and make- techniques. Normally, a three-minute recovery period is sufficient to repeat short running sprints without substantial fatigue.[25] Because of the number of measurements, the child was allowed a 5 minute rest between the break- and make-test, assessed by the first examiner in that order. Before the child performed the tests, which were assessed in the same order by the second examiner, he had a 10 minute rest-period.

For the reliability study of the functional strength tests, eight paediatric physical therapists performed the functional closed chain strength tests. For each child two therapists were randomly chosen out of a sample of eight to perform the tests.

All therapists were experienced paediatric physical therapists (4 to 20 years of experience). Prior to data collection, the therapists were given written instructions in the application and scoring of the functional strength measures, and had no formal training in how to perform functional strength measures. Since a decrease in muscle strength can be expected after five consecutive all-out functional tests the children were given a minimum and maximum time between the functional strength tests of respectively 24 and 72 hours.

All HHD and functional strength assessments were performed within one week. The minimum time between the HHD and the functional strength tests was respectively 24 and 96 hours. All tests were performed during school hours at the school of Special Education “Ariane de Ranitz, Utrecht, the Netherlands”.

Measures

GMFCS. The Dutch translation of the GMFCS[22] was used by a paediatric physical therapist (OV), who was experienced in using the GMFCS, to classify the children and adolescents with CP into groups based on their functional ability. Level I represents the highest level of functional abilities and level V the lowest. In this study, only children with GMFCS levels I or II were recruited (Level I: Children walk indoors and outdoors, and climb stairs without limitation, Level II: Children walk indoors and outdoors, and climb stairs holding onto a railing, but experience limitations in walking on uneven surfaces and inclines, and walking in crowds or confined spaces).

Hand-held dynamometry. Hand-held dynamometry was used to quantify subjects' wisometric muscle force production. Children were given two practice trials for each test until the investigators were confident that they understood the task. Each child performed three trials for each muscle group, and the peak force values from the dynamometer were recorded for break and make-method.[26] Clear standardized instructions regarding the testing procedure and strong verbal encouragement during the trials were used to produce maximal effort.

In the break test the examiner gradually applied force with the dynamometer for 1 second to allow the subject to adjust and to recruit the maximum number of muscle fibres. After 1 second the force of the examiner exceeds the force of the patient very slightly.[27,28]

The make test, operationally defined as a maximal effort exerted against a stationary HHD was used. The children were instructed to gradually “push as hard as possible” against the dynamometer (held rigidly by the examiner perpendicular to the child's limb segment) over a period of approximately five seconds until the examiner told them to relax.[27,28]

The HHD (type CT3001, Citec, C.I.T. Technics BV, Groningen, The Netherlands) that was used, could measure forces up to 500 Newton (N). The HHD was calibrated with weights before beginning data collection and was found to be accurate within ± 0.89 N. The HHD was equipped with a padded end piece provided by the manufacturer and was not modified for this study.

Test positions, except for the hip extensors, were standardized according to Berry et al.[18] and Wiley et al.[17] The position of the testers was not standardized. The following muscle groups were evaluated bilaterally: hip extensors and abductors, knee flexors and extensors and ankle plantar flexors. All muscle groups were assessed in the same order as shown in Table 1. The point on the extremity were resistance is applied was standardized. Absolute strength values in Newton were recorded and used in the analyses.

Table 1. Standardized Muscle Test Positions.

Muscle group	**Posture**	**Subject position**	**Dynamometer position**
Hip extensors	Supine	Hip in 90° flexion; pelvis stabilized	Anterior midthigh
Hip abductors*	Supine	Hips in neutral position and 0° abduction/adduction; pelvis stabilized to prevent sliding	Lateral midthigh
Knee extensors*	Sitting	Knee and hip flexed 90°; pelvis stabilized to prevent hip extension	Anteriorly 5 cm. proximal to lateral malleoli
Knee flexors*	Sitting	Knee and hip flexed 90°; thigh support to prevent hip flexion	Posteriorly 5 cm. proximal to lateral malleoli
Ankle plantar flexors¶	Supine	Knee extended and resting on a bench; lower leg stabilized	Dorsum of foot at level of metatarsal heads

* = Muscle test positions according to Berry et al.[18]; ¶ = Muscle test position according to Wiley et al.[17]

Functional strength tests. To assess functional performance in children with CP we chose functional exercises in which the large muscle groups that are important for standing and walking are being tested.[29] Three closed kinetic chain exercises were chosen:

1) The Lateral Step-up Test (on a 20 cm. bench). This step test is a closed kinetic chain test that has been utilized to assess lower extremity muscular performance.[30] For each test, the subjects were asked to stand with the extremity being tested on the step with their feet parallel and shoulder width apart. Appropriate lateral step-up technique was defined as achieving a position within 15° of knee extension for the tested extremity during the extension phase of the test. Repetitions were counted each time the heel or toes of the extremity not being tested touched the floor. Test-retest reliability in young healthy adult subjects was very good.[12] This protocol has never been examined before in subjects with CP.

2) Sit-to-Stand (from 90° flexion of the knee and hip to standing position). This test is a functional item in which the child has to attain stand without using the arms. The child was positioned on a small bench, and seated with feet flat on the floor and knees flexed at 90°. The child had to achieve standing, arms free, without any assistance from their arms on the bench or their body in the transition. Repetitions were counted each time the child's legs and hips were within 15° of the extended position.

3) Attain stand through half kneel, without using arms. This test is a functional item in which the child has to attain stand without using the arms. The child was positioned on a mat in high kneeling, arms free. This means that weight bearing is on one knee and the opposite foot, and that the alignment may vary as long as the buttocks are clear of the lower legs and/or the weight bearing surface. The child was instructed to assume standing without using any external support such as furniture or the floor. Repetitions were counted each time the child achieved a standing position, and both legs and hips are within 15° of the extended position.

Exercise 1 and 3 were assessed bilaterally. During exercise 2 both extremities were used to perform the task. Total scores for the left and right side were calculated from the repetition maximums for each side, so in total 5 scores were calculated; Lateral Step-up Test (scores for left and right side), Sit-to-Stand (score) and left and right Attain stand through half kneel. (scores for the left and right side). Following the command, "Ready, set, go," subjects started each test and timing began on a stopwatch accurate to .01 sec. Subjects were given 2 practice repetitions per extremity prior to formal testing. Subjects were given 30-sec rest following the practice session and 180-sec between tests. The exercises were assessed in the following order; 1) Lateral Step-up Test left, 2) Lateral Step-up Test right, 3) Sit-to Stand, 4) Attain stand through half kneel left, 5) Attain stand through half kneel right. The subjects were instructed to perform as many repetitions as possible in 30 seconds and they were verbally encouraged. The number of repetitions performed over 30 seconds were recorded and used in the analyses.

Data analysis

Data gathered from both tests and extremities were analysed using the statistical software package (SPSS, version 13.0; SPSS; Chicago, Ill) and MS Excel 2005 for Windows. Intra-class correlations (ICC; two way mixed) were computed for the intertester reliability of the HHD-measurements and the functional strength measures. The ICC gives a relative index of the ratio of variance between subjects to the variance between subjects plus error variance. Acceptable reliability criteria for ICC values were values > 0.80.[31]

In order to assess the amount of error associated with repeated measurements, the standard error of measurement (SEM) was calculated by means of the following equation: $SEM=SD\sqrt{1-ICC}$. Values were computed for each muscle group of each limb (HHD) and for each test of the functional strength tests.

Moreover, limits of agreement (LOA) were calculated to conform to the procedure described by Bland & Altman.[32] Bland-Altman analysis describes the level of agreement between two measurements. In this analysis, the 'bias' is an estimate of how closely on average the two measurements agree and the 'precision' indicates how well the methods agree for an individual. By multiplying the precision by 1.96, the 'limits of agreement' are calculated. The coefficient of variation (CV) was calculated to compare the reliability of the different measurement tools.[33]

The level of statistical significance was set at $p = .05$.

Results

In total 25 subjects (15 males, 10 females) and their parents agreed to participate and signed the informed consent form. Three subjects refused to participate because they were involved in another study. All included subjects were able to assume the test positions as described in Table 1. Group characteristics are described in Table 2. All 25 subjects successfully performed all four test-sessions. The following results refer to both sexes and GMFCS levels combined.

Table 2. Subject characteristics. (n=25)

	GMFCS I[a]				**GMFCS II[b]**			
Number of subjects	**14**				**11**			
Variables	Mean	SD	Median	Range	Mean	SD	Median	Range
Age (y.m)	11.11	2.8	12.6	7.8-16.8	10.11	2.7	12.2	7.5-17.4
Height (cm)	149.6	15.3	149.0	125.0-175.0	148.9	18.9	145.1	123.0-175.0
Body mass (kg)	40.3	12.4	35.1	23.8-60.8	38.6	12.1	32.7	24.0-59.7
Sum of seven skin folds (mm)	77.5	44.5	62.0	36.5-197	74.5	28.5	70.5	33.5-132

SD, standard deviation; GMFCS, Gross Motor Function Classification System

[a]:12 hemiplegia: 2 diplegia; [b]:1 hemiplegia; 10 diplegia

Hand-held dynamometry

Break test

The ICC and SEM for the break-test can be found in Table 3. ICC values ranged from 0.42 for left knee flexors to 0.73 for right hip abductors. The precision of our measures, represented as CV, ranged from 22.2% for the right hip abductors to 35.3% for the left hip abductors and left knee flexors. In most subjects the tested muscle groups could be measured bilaterally. For the ankle plantar flexors the strength could only be measured in 10 subjects for the left side and 8 (tester 1) and 5 (tester 2) subjects for the right side. In this study the break test has questionable reliability for measuring muscle strength using hand-held dynamometry for most muscle groups.

The SEM values, the standard error of the difference values, are listed in Table 3. SEM values ranged from 30.7 N to 58.9 N for hip extensors and hip abductors, knee extensors and knee flexors.

Make test

The ICC and SEM for the make-test can be found in Table 3. ICC values ranged from –0.04 for left ankle plantar flexors to 0.82 for right hip extensors. The precision of our measures, represented as CV, ranged from 16.2% for the right hip extensors to 56.2% for the left ankle plantar flexors.

Table 3. Mean scores, intertester reliability and standard error of measurements for Break- and Make-method of lower-limb strength, using a Hand-Held Dynamometer. (n=25)

	T1	T2	T1	T1	T2	T2				
	NT/T	**NT/T**	**Mean (N)**	**SD (N)**	**Mean (N)**	**SD (N)**	**ICC**	**SEM**	**CV (%)**	**LOA**
BREAK Method										
Hip extensors										
Left	3/22	2/23	209.6	81.7	187.5	57.4	0.53	55.5	28.5	134.6
Right	3/22	3/22	204.8	64.6	188.0	62.4	0.63	38.9	23.3	107.5
Hip abductors										
Left	1/24	1/24	176.4	66.1	189.2	67.3	0.49	47.2	35.3	131.7
Right	1/24	1/24	181.8	59.9	199.0	67.6	0.73	30.7	22.2	92.6
Knee extensors										
Left	1/24	1/24	206.9	91.8	168.8	77.0	0.60	57.5	32.7	148.8
Right	2/23	2/23	198.9	87.2	169.4	79.9	0.62	54.7	33.1	142.7
Knee flexors										
left	3/22	3/22	178.5	78.0	171.0	66.4	0.42	58.9	35.3	152.6
right	3/22	1/24	187.3	73.7	176.7	56.4	0.46	54.1	34.2	133.8
Ankle plantar flexors										
left	15/10	15/10	170.7	56.9	158.0	79.5	0.70	27.9	29.4	105.1
right	17/8	20/5	169.0	29.7	142.5	31.8	N/A	N/A	N/A	N/A
MAKE Method										
Hip extensors										
Left	0/25	0/25	236.2	70.3	158.8	70.4	0.67	40.4	22.4	111.2
Right	0/25	0/25	209.4	72.7	164.3	61.4	0.82	30.9	16.2	79.4
Hip abductors										
Left	0/25	0/25	136.0	55.5	127.0	53.0	0.69	30.6	27.4	83.2
Right	0/25	0/25	137.7	55.2	127.5	56.0	0.68	31.7	27.4	86.9
Knee extensors										
Left	0/25	0/25	188.7	73.8	143.9	62.0	0.70	40.4	21.6	103.3
Right	0/25	0/25	198.4	77.9	150.5	63.4	0.70	42.7	28.1	108.1
Knee flexors										
left	0/25	0/25	154.5	67.2	139.5	47.9	0.49	40.7	27.5	115.2
right	0/25	0/25	169.0	66.9	155.9	68.0	0.64	40.1	31.2	112.6
Ankle plantar flexors										
left	0/25	2/23	128.3	53.9	137.3	45.5	-0.04	52.7	56.2	141.0
right	1/24	3/22	127.3	47.2	136.7	51.2	0.48	35.6	28.8	98.3

NT/T= not testable/testable; ICC= intra class correlation; LOA=limits of agreement, CV=coefficient of variation; SEM= Standard Error of Measurement; reps = repetitions; N/A=not calculated because of the small sample size

Table 4. Mean scores, intertester reliability and standard error of measurements of functional lower-limb strength measurement. (n=25)

Test positions	NT/T	Mean	SD	ICC	SEM	CV (%)	LOA
Lateral step up test							
left	0/25	13.2 reps	10.5	0.94	2.4 reps	17.8	7.24
right	0/25	12.6 reps	10.4	0.94	2.6 reps	22.7	6.83
Sit-to stand	0/25	14.4 reps	5.0	0.91	1.5 reps	12.1	4.04
Half kneel to stand							
left	0/25	7.5 reps	5.5	0.96	1.1 reps	28.6	3.16
right	0/25	6.0 reps	5.3	0.93	1.4 reps	39.9	3.87
Total left	0/25	35.4 reps	18.9	0.96	3.7 reps	10.9	10.38
Total right	0/25	33.1 reps	18.2	0.94	4.5 reps	16.2	12.16

NT/T= not testable/testable; SD=standard deviaton; ICC= intra class correlation; SEM= Standard Error of Measurement; reps = repetitions, CV=coefficient of variation; LOA=limits of agreement.

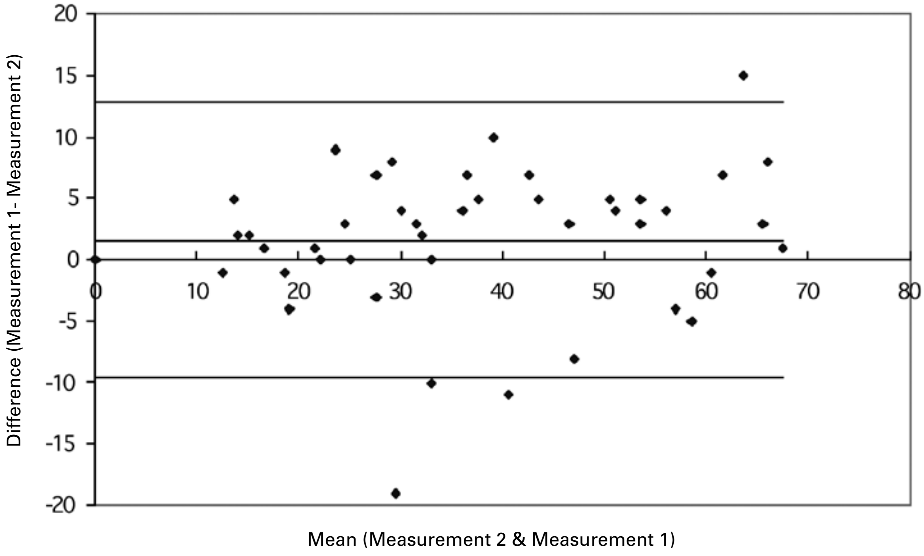


Figure 1. Bland-Altman plot for measurement 1 and measurement 2 for total repetitions for the left & right lower extremity. The bold-type line shows the mean difference between the 2 measurements, and the 2 thin lines indicate ±1.96 standard deviations. On the X-axes, the average repetitions from both tests is displayed. On the Y-axes, the difference between the total repetitions for the left & right lower extremity during measurement 1 and measurement 2 is displayed.

In all subjects the tested muscle groups could be measured bilaterally, except for the ankle plantar flexors in which for 25 subjects one right ankle plantar flexor by tester 1, two left and three right ankle plantar flexors could not be tested by tester 2. While specific muscle groups measured by hand-held dynamometry using the make test have questionable reliability, the tool also has questionable reliability for measuring the strength of knee flexors and ankle plantar flexors in this population. The SEM values, the standard error of the difference values, are also listed in Table 3. SEM values ranged from 30.6 N to 42.7 N for hip extensors and hip abductors, knee extensors and knee flexors.

Functional strength tests

The reliability statistics of all functional measures are shown in Table 4. ICC values ranged from 0.91 to 0.96, and SEM values ranged from 1.1 to 2.6 for the three functional exercises, and from 3.7 to 4.5 for the total scores on the right and left side. The test-retest data are based on the two assessors that tested the subject on both tests. The precision of our measures, represented as CV, for the total number of repetitions for the left and right lower extremity were respectively 10.9% and 16.2%. As can be appreciated from the Bland-Altman plot (Figure 1), there were some obvious outliers. Although these outliers are included in the calculations, the reliability statistics are still acceptable.

Discussion

In this study the intertester reliability of strength measurements in a group of children with CP was assessed. Functional tests showed acceptable reliability values (ICC ≥0.91; CV 10.9% - 39.9%) for all three functional strength measures (1.The Lateral Step-Up Test; 2. Sit-to-Stand; 3. Attain stand through half kneel) and variable reliability values (ICC = -0.04-0.82; CV 16.2%-56.2%) for HHD measurements between two assessors.

In the present study different statistics to examine reliability (i.e. ICC, SEM and CV) were used. The ICC indicates the proportion of the total variance in the measurements which is due to the true difference between subjects. The SEM and CV's are methods of analysing absolute reliability.[34] The SEM, that is unaffected by the range of measurements, is used if the goal is to determine the stability of a child's performance. The coefficients of variation (CV) are particularly useful for comparing the reliability between performance tests. Thus, when measuring the reliability, the ICC, SEM and CV yield useful, although quite different information. Previous reliability studies in children with CP have used the Pearson correlation as a measure of reliability.[35-37] However, the Pearson correlation is not very suited for the purpose since it cannot assess systematic bias and it depends greatly on the range of values in the sample.[38]

Measurements taken by an individual are typically more reliable than those taken by multiple examiners.[39] However, in research and clinical practice it is preferable that a test can be reliably performed by different assessors. Taking this into account, it is not surprising that in the studies, investigating the intratester reliability of HHD in subjects with CP,[16,18,19] the ICCs are higher than those found in the present study. The results of our study show that agreement among two observers with experience in the assessment of muscle strength using a HHD is low. It would be interesting to examine agreement in a group of observers with no or limited experience in HHD strength testing.

For rehabilitation practice, it is desirable that a measurement tool is reliable enough to be used on individuals. For example, a clinician may need to know whether an improvement in muscle performance following a rehabilitation program is real or merely due to measurement error. The results of the functional strength tests showed that they could be assessed in a highly reliable way between two assessors in children with CP. The calculated SEM can be used to determine the range in which a person's "true score" could be expected to lie, considering the amount of error associated with repeated measures. For example, we can be 95% confident that the "true score" for people performing the functional strength tests lies within ±1.96 SEM. Based on the data in table 4, total RM increases of 7.3 (1.96 x 3.7) and 8.8 (1.96 x 4.5) for the left and right side respectively can be attributed to real change with 95% confidence. Since the SEM values for each of the functional strength tests were small, it appears that these tests represent stable measures and may be used in the clinical evaluation of children and adolescents with CP.

The coefficients of variation (CV's) for the hip extensors and abductors, and the knee extensors and flexors HHD measures vary from 22.2% to 35.3%, and from 16.2% to 31.2% for respectively the break-method and make-method. The CV for the individual functional exercises vary from 12.1% to 39.9% for respectively the Sit-to-Stand and the Attain stand through half kneel (right) item. The CV's for the total score of the functional strength test for the left and right lower extremities are 10.9% and 16.2%, respectively. Since the CV's for the sit-to-stand test and the total scores for the lower extremities are smaller than for the other functional strength tests and HHD, these tests might be the most sensitive to change.

Poor selective control in some muscle groups may prevent an individual from being able to perform the task.[8] In the current study this was only found in the ankle plantar flexors during the make-method. This may be due to the fact that in children and adolescents classified as a level I or II on the GMFCS (the ones with the best motor abilities), motor control limitations are not likely to be a substantial factor in the ability to generate force. In the break-method the examiner attempts to exceed the force of the child. In this way of testing the examiner's strength can be inadequate to overpower the tested muscle. In this group of children the make-method has been shown to be more reliable than the break-method for the following muscle groups: hip extensors, hip abductors, knee extensors and knee flexors. We therefore recommend the use of the make method for measuring muscle strength in young people with CP.

In previous studies that assessed the intratester reliability for strength testing using HHD in children with CP, experienced examiners were used. In our study, that assessed the intertester reliability for HHD we also used experienced examiners. We found questionable reliability for the make and break method. As mentioned in the introduction it is preferable that a test can be reliably performed by different testers. However, it is clear from this and previous studies[40] that HHD suffers from questionable intertester reliability. Intratester reliability is high or moderate for most muscle groups and therefore it is recommended to use the same examiner if longitudinal measurements have to be done.

For the functional strength tests physical therapists, who had no formal training, were used to assess the intertester reliability. So, to perform the functional strength tests in a reliable way, a special training is not required. Therefore, this strength testing method is available and applicable for a variety of professionals working with children with CP. If a researcher or clinician is interested in the functional gain in task-related activities, the functional strength test is an appropriate and reliable measure. Moreover, the functional strength test is reliable when used by different testers.

There are some limitations in our study. In the present study the order in which all muscle groups and functional exercises were examined were fixed. Fatigue, concentration and distraction can play an important role in these kind of test protocols. The present study provides no information on the effect of order of testing and if these results apply to other testing orders or the testing of a single muscle or functional movement. As can be appreciated from the Bland-Altman plot (Figure 1) there were some outliers in muscle strength scores. Both muscle strength tests are influenced by the motivation or co-operation of a patient. Moreover, children with CP show a large variability in the generation of muscle strength.[41] Cognitive impairment has been cited as a possible reason for large within-participant variability when testing isokinetic strength.[35] Unfortunately, cognition of the children that participated in this study were not formally assessed.

In conclusion, the results of this study show that intertester reliability of HHD is low, and therefore it is advised to use the same examiner when HHD is the preferred muscle strength test method. When a task-related outcome is preferred the 30-second RM total scores for the left and right lower extremity can be used by different examiners to obtain highly reliable measures of lower extremity performance when used conform a standardized protocol.

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IN CHILDREN AND ADOLESCENTS
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EXERCISE TRAINING PROGRAM IN CHILDREN AND ADOLESCENTS WITH CEREBRAL PALSY: A RANDOMIZED CONTROLLED TRIAL

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Abstract

Objective To evaluate the effects of an 8-months training program with standardized exercises on aerobic and anaerobic capacity in children and adolescents with cerebral palsy.

Design Pragmatic randomized controlled clinical trial with blinded outcome evaluation.

Setting Participants were recruited from 4 schools for special education in the Netherlands.

Participants A total of 86 children with cerebral palsy (aged 7–18 years) classified at Gross Motor Function Classification System level I or II.

Intervention(s) Participants were randomly assigned to either the training group (n=32) or the control group (n=33). The training group met twice per week for 45 minutes to circuit train in a group format that focused on aerobic and anaerobic exercises.

Main Outcome Measure(s) Aerobic capacity was assessed by the 10-m shuttle run test, and anaerobic capacity was assessed by the Muscle Power Sprint Test. Secondary outcome measures included agility, muscle strength, self competence, gross motor function, participation level and health-related quality of life.

Results A significant training effect was found for aerobic ($p<0.001$) and anaerobic capacity ($p=0.004$). A significant effect was also found for agility ($p<0.001$), muscle strength ($p<0.001$) and athletic competence ($p=0.005$). The intensity of participation showed a similar effect for formal ($p<0.001$), overall ($p=0.002$), physical ($p=0.005$) and skilled-based activities ($p<0.001$). On the health-related quality of life measure, a significant improvement was found for the motor ($p=0.001$), autonomy ($p=.02$) and cognition ($p=.04$) domains.

Conclusions An exercise training program improves physical fitness, participation level, and quality of life in children with cerebral palsy when added to standard care.

Trial Registration http://www.ISRCTN.org number ISRCTN77274716

Introduction

Cerebral palsy (CP) describes a group of disorders that affect the development of movement and posture, causing activity limitation, and 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] Improving the ability to walk or perform other functional activities are often the primary therapeutic goals for children with CP.[2] Because of existing impairments, many children and adolescents with CP have difficulty with activities such as walking independently, negotiating stairs, running or navigating safely over uneven terrain.[3] Additionally, children with CP have distinctly subnormal aerobic and anaerobic capacity in comparison with typically developing peers.[4-6] Also, muscle mass is low,[4] and muscle strength is reduced.[7-9] Low levels on these fitness components may contribute to the difficulties in motor activities most children with CP encounter in daily life. Moreover, evidence suggests that hypoactive children are more likely to become physically sedentary adults and that encouraging the development of physical activity habits in children will help establish activity patterns that continue into adulthood.[10]

In general, aerobic capacity, anaerobic capacity and muscle strength can be trained in typically developing children of all ages.[11,12,13] Studies of the effects of lower limb exercises in children with CP have demonstrated an increase in muscle strength and function,[7,14-16] as well as in aerobic capacity,[17,18] while no negative effects on spasticity or abnormal movements have been reported.[14,15,19,20] To date, none of the training programs in children with CP have focused on anaerobic capacity, which is important, because almost all daily childhood activities are more short-term (<15 sec) and high-intensity than long-term.[21,22]

Activities of daily childhood life consist of well balanced aerobic, anaerobic and muscle strength components. The principle of specificity of learning states that learning is optimized by practice that approximates the target skill.[23] To date, we know of no study that trained the aerobic and anaerobic fitness component as well as muscle strength in task-specific, functional activities in children with CP. Therefore, we conducted a pragmatic randomized-controlled clinical trial to determine the effects of a standardized 8-months exercise program with an extra 4 months of follow-up after the training.

Accordingly, the primary objective of this study was to determine the specific (i.e. aerobic and anaerobic capacity) effects of an exercise program in addition to usual care at the end of the training period. Moreover, we investigated the effects of this program on all International Classification of Functioning, Disability and Health[24] levels and health-related quality of life (HRQOL) at the end of the training period and at 4 months' follow-up.

Methods

Participants

Children and adolescents with CP were recruited from 4 schools for special education in the Netherlands. To be included, participants had to be 7 to 20 years of age, diagnosed with spastic CP, and classified at level I or II on the Gross Motor Function Classification System (GMFCS).[25] Children older than 12 years of age were classified using the same criteria as those used for 6- to 12-year-olds. All children in the study were able to follow simple verbal commands. They were all receiving rehabilitation services at the time of the study. Participants were excluded if they had had orthopedic surgery or neurosurgery and/or botulinum toxin injection(s) within 6 months prior to study entry or cardiac or respiratory conditions that could negatively be affected by exercise.

Study design

A pragmatic randomized controlled clinical trial was conducted between June 2005 and October 2006. The Institutional Ethics Committee of the University Medical Center Utrecht approved the study. Participating schools for special education were informed about the study and the inclusion and exclusion criteria. Based on clinical examination, pediatric physiatrists working in these schools referred suitable participants.

The Dutch translation of the GMFCS[25] was used to classify the children with CP into 2 groups based on their functional ability. Participants were randomly assigned to 2 groups using a 4 block-randomization protocol. Each block represented all participants from 1 school. The groups within each block consisted of children classified at level I or II at the GMFCS.[25] From each block and group, every participant was randomly allocated to the training group or the control group. An independent off-site researcher not involved in the assessments used a concealed method for allocation. The children in the control group received their usual rehabilitation care. Because this might affect functional gains among participants, provided care was tracked from the medical progress records. Usual care ranged from no treatment to various therapeutic approaches. There was no difference in usual care between both groups.

Training program

We developed a functionally based exercise program that was easily implemented in clinical practice. This program consisted of 8 standardized aerobic exercises that lasted 3 to 6 minutes, and 8 standardized anaerobic exercises that lasted 20 to 30 seconds. All children, regardless of their age or GMFCS-level, performed the same exercises during the program. The task-specific exercises such as running and changing direction of the body abruptly, step-ups, and negotiating stairs, were repeated throughout the program and aimed to improve daily functioning. Each session lasted 45 minutes. The total program consisted of a 5-minute warm-up period; 25 to 35 minutes of functional aerobic, anaerobic and muscle strengthening exercises in circuit format; and a 5-minute cool down. During the first 4 months the main focus of the program was to improve aerobic capacity. After 4 months (after the second measurement), when the participants were expected to have improved their aerobic capacity, the focus shifted towards anaerobic capacity.

The children in the training group received the exercise program in addition to their usual care. The participants in the training group trained in age-related groups (7-12 and 13-18 years of age) that were formed after the randomization, and consisted of 4 to 6 children. Consequently, each school had 2 different age-related groups that participated in the training sessions. Standardized training sessions were led by 2 local pediatric physiotherapists during school hours at school. All therapists received the standardized fitness program training prior to the start of the fitness program. The training group trained 2 days per week for 8 months.

Outcome measures according to the international classification of functioning, disability, and health

Primary outcome measures were aerobic and anaerobic capacity measured at the end of the training period. Additional measurements included agility, muscle strength, body mass index, self-perception, gross motor function, participation and HRQOL.

Body function

Aerobic capacity was reflected by the achieved level on the 10-metre-shuttle run test.[26] This test requires children to walk or run between 2 markers delineating the respective course of 10 m, at a set incremental speed determined by a signal (every minute). The achieved level was recorded and used for analysis. Anaerobic capacity was measured using the mean power (measured in watts) derived from the Muscle Power Sprint Test.[27] For the Muscle Power Sprint Test the subjects were instructed to complete six 15-meter runs at a maximum pace. Between each run, the subject was allowed a timed 10 seconds rest. Power output (measured in watts) for each sprint was calculated. Agility was assessed by the 10 x 5 Meter Sprint Test.[27] Muscle strength of the lower extremity was measured with the 30-sec repetition maximum.[28]

The body mass index was calculated as weight in kilograms divided by height in meters squared. Participants' weight and height were measured using a standard protocol. Each child was weighed to the nearest 100 g. on electronic scales (Seca, Hamburg, Germany). Height measurements were taken to the nearest 0.5 cm. while the child was standing with his or her back against a wall.

We used the Self-Perception Profile for Children[29] to evaluate the self-concept of the children. This scale is designed to assess children's perceptions of themselves. We assessed the domains of athletic competence, physical appearance, and global perception of their worth or esteem as a person.

Activity

In this study, gross motor function was assessed using dimension D and E of the Gross Motor Function Measure (GMFM)[30] which measures activities in a standing position, and walking, running and jumping, respectively. These dimensions were chosen because they represent areas that many young people with CP, who are able to walk, have difficulty with.[7]

Participation

The Children's Assessment of Participation and Enjoyment (CAPE)[31] was used to document change in how children and youth participate in everyday activities outside mandated school activities. The CAPE provides 3 levels of scoring: (1) overall participation scores; (2) domain scores reflecting participation in formal and informal activities; and (3) scores reflecting participation in 5 types of activity (recreational, active physical, social, skill-based, and self-improvement activities). The intensity scores reflect the average amount of time that a child spends participating in different activities. The intensity scores of all types of activity were measured.

Health-Related Quality of Life

The TNO-AZL Questionnaire for Children's Health-Related Quality of Life (TACQOL) is a generic, multidimensional instrument.[32] It asks about health problems in the past few weeks using a 3-point Likert scale and about the emotional response to these problems on a 4-point scale. This instrument contains 7 scales: (1) pain and symptoms, (2) basic motor functioning, (3) autonomy, (4) cognitive functioning, (5) social functioning, (6) global positive emotional functioning and (7) global negative emotional functioning. We used the TACQOL Parent Form (TACQOL-PF) because we expected that a part of the participants were too young or too low functioning to complete the TACQOL themselves.

To reduce bias, 8 assessors who were not the treating therapist, who were blinded for the treatment modality, undertook the testing without review of previous scores. Prior to data collection, the assessors had formal training and were given written instructions in the application and scoring of all tests and measurements.

Assessments were performed at baseline, after 4 months and directly after the 8 months of the intervention period in both groups. There was a follow-up assessment with the same measures in both groups at 12 months after T0.

Statistical analysis

The sample size for the trial was determined by the most demanding hypothesis to detect effects of treatment on aerobic capacity. The sample size was calculated from data for the 10-metre shuttle run test,[26] with a significance level of .05, a *d* of 0.65, and a power of 80%. Therefore, 30 participants were required for each group. To compensate for drop-outs, we planned to enroll 35 patients per group.

All data analyses were carried out according to a pre-established analysis plan, and were performed according to the intention-to-treat principle. To assess the effects of training as compared with standard care, we used repeated-measures analysis of variance (group [2] x time [3]) with repeated aerobic and anaerobic capacity measures as dependent variables and treatment group as an independent variable. We used the same analysis to assess our secondary objective. In case of violation of the sphericity assumption, we used the Greenhouse–Geisser correction.

To assess the effects during and after the fitness program as compared with control treatment effects, repeated measures analysis of variance (group [2] x time [4]) with repeated contrasts was used. Violations of assumptions were checked in all cases.

The α-level was set at .05. Data were analyzed using SPSS, version 13.0 (SPSS Inc, Chicago, IL).

Results

Participant flow

Age-eligible participants were recruited from July 2005 to September 2005. Figure 1 shows the participant flow from initial recruitment to assessment after training and follow-up (October 2006). A total of 86 participants were assessed for eligibility for the study. One of the children did not meet the inclusion criteria, 9 of the eligible participants did not take part for logistical reasons (year of their graduation), and 8 children and adolescents refused to participate for unknown reasons. Sixty-eight patients and their parents gave written informed consent before entering the study, and were randomized to either the control group or training group. Baseline demographic and clinical characteristics of each group are listed in Table 1. At baseline, there were no significant differences between study groups. During baseline measurements, 3 children (boys aged 11.3, 15.1 and 16.1 years) discontinued the study because of personal reasons, such as lack of motivation, and were lost to follow-up. These children completed the fitness measures, but did not complete most of the other measures. Sixty-five participants completed the entire study. The median attendance in exercise training was 56 of 60 sessions (93%), with all children attending at least 85% of the training sessions. During a training session, 1 child fell, and fractured her radius; she missed 4 training sessions because she was wearing a cast.

Figure 1. Flow of participants through the trial. T0 indicates baseline; T1, after 4 months; T2, after 8 months.

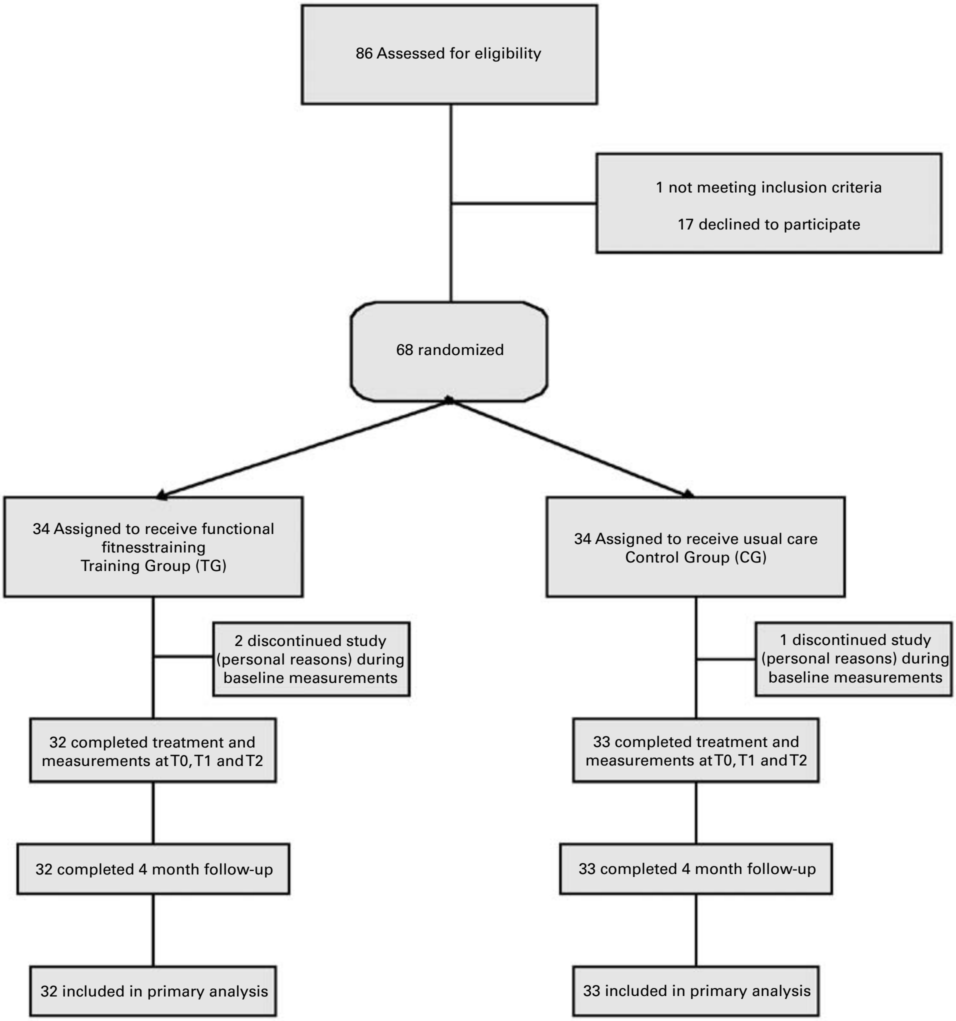


Table 1. Baseline characteristics of the participants.

	Training group (n=34)	Control group (n=34)
Demography		
Age (years)	11.6 (2.5)	12.7 (2.7)
Male/female	20/14	24/10
Anthropometry		
Body height (cm)	148.0 (17.5)	154.6 (18.4)
Body weight (kg)	44.7 (16.5)	48.4 (17.8)
BMI (kg/m^2)	19.8 (4.1)	19.6 (4.1)
Cerebral Palsy: classification and distribution*		
GMFCS level I	24 (71%)	23 (68%)
GMFCS level II	10 (29%)	11 (32%)
Unilateral	23 (68%)	22 (65%)
Bilateral	11 (32%)	12 (35%)
Aerobic capacity		
Level on Shuttle Run Test (minutes)	6.2 (3.1)	7.6 (4.8)
Anaerobic capacity		
Mean Power (Watts)	84.6 (78.6)	121.2 (88.1)

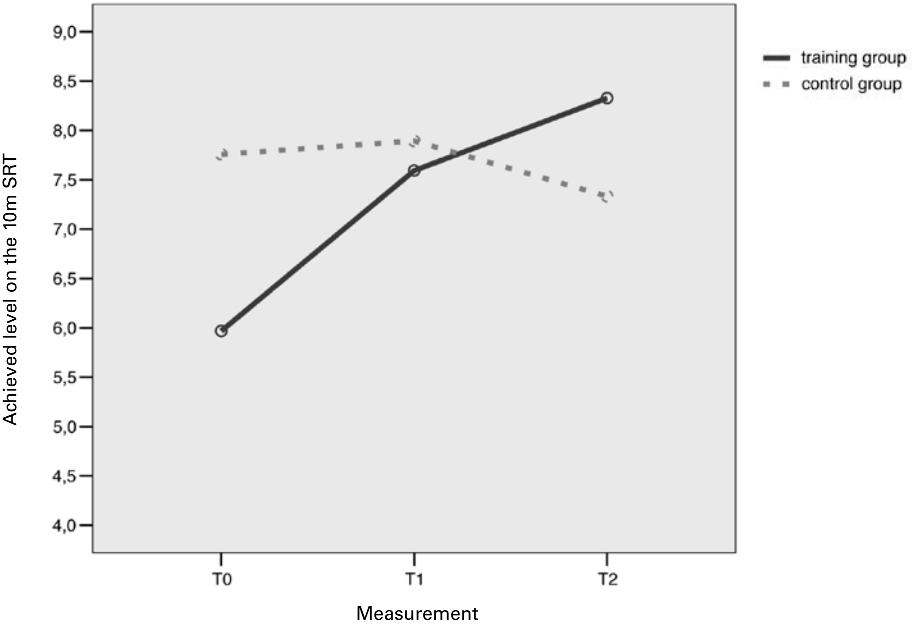
Legend: Values are means (SD) unless otherwise indicated; BMI=body mass index; GMFCS=gross motor function classification system; *Number of individuals

Effects of training

Body function

As shown in Figure 2, improvements on aerobic (+38%) and anaerobic capacity (+25%) were found for the training group. Moreover, as shown in Table 2, improvements were found on agility (+15%), muscle strength of the lower extremities (left and right side, +20% and +23%, respectively) and athletic competence on the Self-Perception Profile for Children (+11%).

Figure 2. Profile plots for aerobic capacity (p<0.001) and anaerobic capacity (p=0.004). P-values are for the repeated-measures analysis of variance (group [2] x time [3]). T0 indicates baseline; T1 after 4 months; T2 after 8 months.



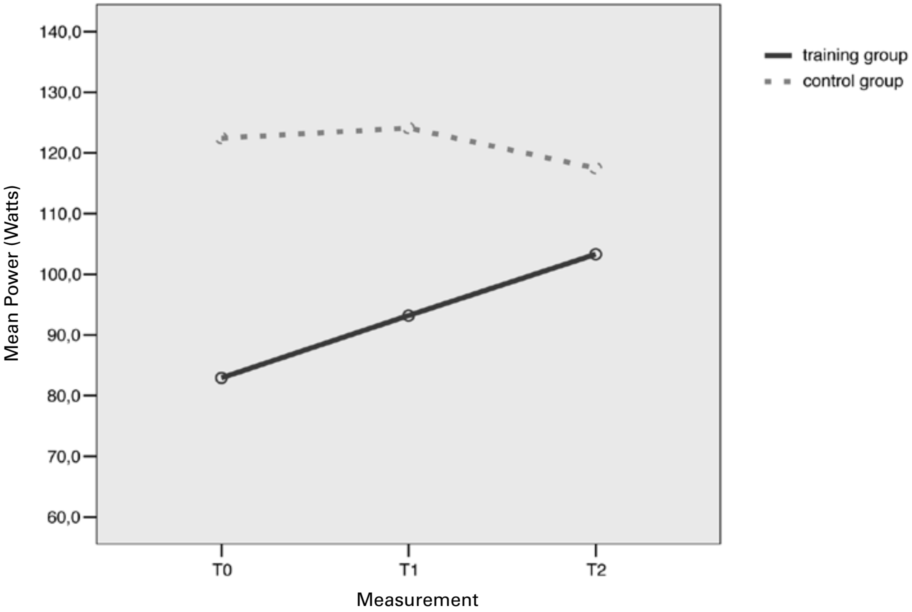


Table 2. Mean differences between T0 and T2 for the training group (TG) and the control group (CG) in outcome measures.

	Difference(SD) T0-T2 TG	Difference(SD) T0-T2 CG	P-value
Body function and structure			
Body Mass Index (BMI)	0.7 (2.1)	0.3 (1.1)	0.51
Aerobic capacity , level on the 10m Shuttle Run Test (minutes)	2.4 (1.9)	-0.4 (1.6)	<.001
Anaerobic capacity, Mean muscle power (Watts)	20.4 (38.0)	-4.8 (28.2)	.004
Agility (10x5 meter sprint test)	-4.5 (4.1)	0.2 (4.4)	<.001
Muscle strength left	6.9 (7.2)	-1.9 (8.7)	<.001
Muscle strength right	7.7 (9.0)	-1.9 (10.0)	<.001
Self concept (SPPC)			
Athletic competence	2.0 (4.2)	-1.3 (3.7)	.005
Physical appearance	0.03 (4.4)	0.2 (4.4)	.90
Global perception of their worth	0.5 (3.3)	-1.0 (4.0)	.20
Gross Motor Function (GMFM)			
Dimension D (standing)	2.6 (5.4)	-0.7 (5.1)	.03
Dimension E (walking, running and jumping)	1.5 (6.4)	-0.9 (3.5)	0.27
Participation (CAPE)			
Overall activities	0.0 (0.5)	-0.4 (0.6)	.002
Formal activities	0.2 (0.4)	-0.4 (0.8)	<.001
Informal activities	0.0 (0.7)	-0.4 (0.7)	.07
Recreational activities	-0.2 (1.0)	-0.4 (1.1)	.69
Physical activities	0.3 (0.8)	-0.3 (0.7)	.005
Social activities	-0.1 (0.8)	-0.4 (1.1)	.12
Skill-based activities	0.2 (0.5)	-0.6 (0.9)	<.001
Self-improvement activities	-0.1 (0.9)	-0.5 (0.8)	.10
Health-Related Quality of Life (TACQOL-PF)			
Pain and symptoms	-0.4 (3.6)	-1.0 (2.3)	.30
Basic motor functioning	2.1 (4.3)	-1.7 (4.3)	.001
Autonomy	0.5 (4.3)	-0.2 (3.1)	.02
Cognitive functioning	0.9 (4.7)	-0.2 (4.0)	.04
Social functioning	0.7 (4.0)	0.0 (3.9)	.13
Global positive emotions	0.3 (3.9)	-0.1 (1.9)	.25
Global negative emotions	0.7 (2.9)	0.0 (1.7)	.15

Legend: Values are means (SD). SPPC = Self-Perception Profile for Children; GMFM= Gross Motor Function Measure; CAPE = Children's Assessment of Participation and Enjoyment; TACQOL-PF = TNO-AZL Questionnaire for Children's Health-Related Quality of Life-Parent Form. P-values are for the repeated-measures analysis of variance (group [2] x time [3]).

Activity and Participation

A positive training effect was present for dimension D (standing) of the GMFM. For participation there were significant differences in favor of the training group on the overall, formal, physical and skill-based activities of the CAPE.

Health-Related quality of life

For HRQOL significant changes over time that differed by group were found for the motor, autonomy, and cognition domains of the TACQOL-PF (Table 2).

The contrasts between all measurements (at baseline, 4 months, 8 months) show that during the first 4 months of the training period (focused on aerobic capacity) there was a significant change in favor of the training group for aerobic capacity, agility, athletic competence, and GMFM dimension D (standing). Moreover, a similar change was also found for overall, formal, physical, skill-based and self-improvement activities as measured with the CAPE. On the TACQOL-PF, significant changes in favor of the training group were found for the motor, autonomy, cognition, and social domains during the first 4 months. No significant effects were observed for muscle power.

During the last 4 months of the training period (focused on anaerobic capacity) we found an improvement in the training group for aerobic capacity, anaerobic capacity, agility, muscle strength and GMFM dimension E (walking, running and jumping). There was no improvement on the CAPE. On the TACQOL-PF, the only significant change in favor of the training group was for the autonomy domain.

Follow-up effects at 12 months

At follow-up (4 months after the training period), we noted a significant difference in outcome measures, as measured with repeated contrasts between groups. The training group reached levels that were similar to the levels after 4 months of training; there was a decrease in aerobic capacity (-8.4%), anaerobic capacity (-8.5%), agility (-4.3%), muscle strength (left side, -4.4%; right side, -8.3%), and athletic competence (-9.8%). The control group remained stable on these measures.

Significant differences between groups were found at follow-up on dimension D (-1.1% for the training group) of the GMFM and on the skill-based activities of the CAPE (-20% for the training group). These measures remained stable in the control group.

For HRQOL there were no significant differences between both groups at follow-up.

Comment

To our knowledge, this is the first randomized multi-centre clinical trial studying the effects of an exercise program with emphasis on movements of daily childhood life, nowadays called "functional exercises," combining 3 fitness elements (aerobic and anaerobic capacity and muscle strength) in a large number of participants with CP. This study provides evidence that a group circuit-training program can be an effective and feasible strategy for increasing both aerobic capacity and anaerobic capacity in young people with CP.

To date, only 1 randomized controlled study investigating the efficacy of aerobic training in children with CP has been published.[18] After an aerobic training program of 9 months, observed improvements were 35% (4 sessions a week) and 21% (2 sessions a week) in aerobic capacity and there were no training-related improvements in anaerobic capacity. Our study showed improvements of 38% in aerobic capacity and significant improvements of 11% in anaerobic capacity in the second 4 months of the exercise program when anaerobic exercises were incorporated.

The main focus during the last 4 months of the fitness program was on anaerobic capacity. The significant interaction in favor of the training group during the last 4 months of training shows that a fitness training program with a predominantly anaerobic nature can improve the anaerobic capacity of children with CP. These improvements also led to significant changes in muscle strength (22%). Other studies focusing specifically on improving muscle strength, have reported similar results after training in children with CP.[7,14-16]

Children with CP often have difficulties changing direction of the body abruptly or quickly shifting the direction of movement without losing balance (agility). During both training periods, with different main foci (aerobic and anaerobic), there was a significant interaction between groups in favor of the training group for agility. The finding that training focus is not specifically related to improvement of agility could be explained by the fact that the children in the training group had more exercise and were training at higher velocities than in general.

Our study also showed a significant increase in perceived athletic competence but no significant changes over time for physical appearance or global perception of worth. One randomized controlled trial that studied the effects of an exercise program on the self-concept of children with CP found a change in self-concept.[33] The training group did not increase to the same extent in some aspects of self-concept as the children in the control group.[33] Another randomized study demonstrated an increase in self-concept of the children after training.[34] Possible explanations for these differences might be explained by differences in settings, the illness severity of the children included in either study, the length of the exercise program and the type of exercises. These and other possible factors need to be studied.

Participation in everyday childhood activities plays an important role in the development of a child's social relationships and skills and also influences long-term mental and physical health.[31,35,36] Participation enables children to explore their social, intellectual, emotional, communicative, and physical potential and is an important predictor of future life satisfaction.[37] Margalit[38] and Stevenson et al.[39] looked at children with CP and found significant limitations in participation. Law et al.[40] identified several child factors that affect participation, including that higher levels of physical function are associated with increased intensity of participation. Therefore, it could be expected that involvement in an exercise program might lead to more permanent changes in everyday physical activities that in turn might lead to maintenance of the fitness benefits. Based on the findings of this study, we conclude that having a higher physical function does lead to an increased intensity of participation. Overall, formal, physical, and skill-based activities were improved at the end of the training period. Unfortunately, most of these improvements were not maintained at follow-up. In future studies factors that may contribute to a positive long-term change in lifestyle habits should be taken into account.

Health related quality of life is an important outcome of treatment for chronic conditions such as CP.[41] This is, to our knowledge, the first study that assessed the effects of an exercise intervention program on the quality of life in children with CP. Improvements in HRQOL in favor of the training group were seen for 3 domains. The parents of the children with CP noticed improvements for the motor, autonomy and cognition domains.

The amount of time spent in physical activity has decreased markedly over the past decades.[42] Our study shows aerobic and anaerobic capacity, agility, and muscle strength decreased during the intervention period in the control group. Given this decline in fitness levels, children and adolescents with CP are at risk of a sedentary lifestyle that will continue throughout their lives.[43] The adverse effects of this decreased fitness may be reduced with involvement in a fitness program.

The findings of this study suggest that the benefits that children gained during training were only partially maintained at follow-up. A decrease in fitness after an exercise training program in children with CP was previously reported.[18] It seems very difficult for children with CP to maintain the gains of an exercise program. Consequently, it can be concluded that children with CP must continue their training to maintain their fitness levels. Limited exercise and sport opportunities in their neighborhood might impede transfer of the gains in fitness into extra participation in regular physical activities. Supervised training-programs at schools or in the community might provide fitness training for children and adolescents with CP. These possibilities need to be studied in future research.

Conclusions

An 8-month, standardized, functionally based exercise program significantly improved physical fitness, the intensity of activities, and HRQOL in children with CP when added to standard care.

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7

SUMMARY, GENERAL DISCUSSION AND CONCLUSION

Summary

Chapter 1 provides a brief introduction to the topic of the thesis. A review of the literature that focused on exercise training in children and adolescents with CP is presented in **Chapter 2**. We concluded that 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.

In **Chapter 3**, the newly developed 10-m shuttle run tests (SRT) for children who are classified at Gross Motor Function Classification System Level (GMFCS) I and II, are examined for their reliability and validity to measure aerobic capacity in children and adolescents with CP. The SRT-I was developed for children at GMFCS level I, and the SRT-II was developed for children at GMFCS level II. Twenty-five children and adolescents with CP (classified at GMFCS level I or II) participated in this study. To assess test-retest reliability the 10-m shuttle run tests were performed by the subjects within two weeks. To examine validity, the shuttle run tests were compared with a GMFCS level-based treadmill test designed to measure peak oxygen uptake. The results show that 10-m shuttle run tests yield reliable (intraclass correlation coefficients (ICC's) of 0.97 for SRT-I and 0.99 for the SRT-II) and valid (r=0.96 for both tests) data. Moreover, the shuttle run tests have advantages over a treadmill test for children with CP who are able to walk and run (GMFCS level I or II).

In **Chapter 4**, two running based measures, the Muscle Power Sprint Test (MPST) and the 10x5 Meter Sprint Test, are being studied on its reliability and validity to measure respectively the Mean and Peak Muscle Power and agility. Twenty-six children and adolescents with CP (classified at GMFCS level I or II) participated in this study. We found good feasibility and reliability for the 10 x 5 Meter Sprint Test and the MPST (ICC's of ≥ 0.97 for interobserver and test-retest reliability) in children and adolescents with CP (GMFCS classification level I or II). The validity of both tests is supported by significant differences in scores between children classified at GMFCS level I and children at level II. To assess the muscle power during running performance in children with CP the Mean Power derived from the MPST is the most appropriate outcome measure. To assess someone's running performance and coordination of speedy movements the 10 x 5 Meter Sprint Test is the most appropriate measure. In our opinion, the 10 x 5 Meter Sprint Test and the MPST can be incorporated in the exercise evaluation of the child with CP, classified as level I or II on the GMFCS.

In **Chapter 5**, two ways to measure muscle strength are being studied. For Hand-Held Dynamometry (HHD) the make- and break-method are being compared regarding their reliability. Moreover, in this Chapter a new instrument (the 30-sec Repetition Maximum(RM)) to measure functional muscle strength is examined on its reliability. To assess functional performance in children with CP we chose functional exercises in which the large muscle groups that are important for standing and walking are being tested. Three closed kinetic chain exercises were chosen: 1a) Lateral Step-upTest left, 1b) Lateral Step-upTest right, 2) Sit-to Stand, 3a) Attain stand through half kneel left, 3b) Attain stand through half kneel right. Twenty-five subjects with CP (GMFCS level I or II) participated in this study. The intertester reliability of strength measurement using a HHD was questionable with ICC values ranging from 0.42 to 0.73 for the break-method, and from 0.49 to 0.82 for the make-method. The Standard Error of Measurement (SEM) and Coefficient of Variation (CV(%)) values ranged from 27.9 to 58.9 and 22.2% to 35.3% for the break-method and from 30.6 to 52.7 and 16.2% to 56.2% for the make-method. The intertester reliability of strength measurement using the 30-sec RM was acceptable with ICC values ranging from 0.91 to 0.96, and SEM and CV(%) values ranging from 1.1 to 2.6 and 10.9% to 39.9% for the functional exercises. The results of this study demonstrate that the make-test is the preferred muscle strength test method when using HHD. Moreover, clinicians can use a 30-second RM as a functional strength test to obtain highly reliable measures of lower extremity performance when used conform a standardized protocol.

Chapter 6 describes the effects of a functionally based fitness program that is added to usual care for children with CP (GMFCS level I or II) on aerobic and anaerobic capacity, agility, muscle strength, self-concept, gross motor function, participation and health-related quality of life. To evaluate the effects of an eight-months training program with standardized exercises on aerobic and anaerobic capacity in children and adolescents with CP a pragmatic randomized controlled clinical trial was performed. A total of 86 children with CP (aged 7–18 years) classified at GMFCS-level I or II participated in this study. Sixty-eight participants agreed to participate and were randomly assigned to either the training group (TG; n=34) or the control group (CG; n=34). The TG met twice per week for 45 minutes circuit training in group format that focused on aerobic and anaerobic exercises.

Assessments were performed at baseline (T0), after 4 months (T1) and directly after the 8 months intervention period (T2) in both groups. There was a follow-up assessment with the same measures in both groups at twelve months after T0. Our primary outcome measures were aerobic and anaerobic capacity. They were assessed by respectively the 10-m shuttle run test and the Mean Power derived from the MPST. Secondary outcome measures included agility (10x5 Meter Sprint Test), muscle strength (30-sec RM), self competence (Self Perception Profile for Children), gross motor function (Gross Motor Function Measure), participation level

(Children's Assessment of Participation and Enjoyment) and health-related quality of life (HRQoL; TACQOL). A significant training effect was found for aerobic capacity (p<0.001) and anaerobic capacity (p=0.004). A significant effect was found for agility (p<0.001), muscle strength (p<0.001) and athletic competence (p=0.005) as well. The intensity of participation showed a similar effect for the formal (p<0.001), overall (p=0.002), physical (p=0.005) and skilled-based activities (p<0.001). On the HRQoL a significant improvement was found for the domains motor (p=0.001), autonomy (p=0.020) and cognition (p=0.042). Hence, an exercise training program improves physical fitness, the participation level and health-related quality of life in children with CP when added to standard care.

At follow-up we noted a significant difference in outcome measures. The training group reached levels that are similar to the levels at T1, after four months of training.

We concluded that an eight-month standardized exercise program consisting of functionally based exercises significantly improves physical fitness, the intensity of activities and health-related quality of life in children with CP when added to standard care.

General discussion

What we know

The RCT described in Chapter 6 showed that an eight-month standardized exercise program consisting of functionally based exercises significantly improves physical fitness, the intensity of activities (participation) and health-related quality of life in children with CP when added to standard care. However, at follow-up we noted a significant decrease in these outcome measures. The training group reached levels that are similar to the levels at T1; after four months of training.

Enhanced participation can be viewed as the ultimate outcome for pediatric rehabilitation.[1] Therefore, measurement of participation is necessary to evaluate the effectiveness of rehabilitation programs. King et al.[2] discussed that the Children's Assessment of Participation and Enjoyment (CAPE) could be useful in determining the effectiveness of clinical trials. The intensity-score of the CAPE showed its ability to detect change over time in our RCT. In a recent review by Sakzewski et al.[1] it was concluded that not one measure covered the full breadth of participation outlined by the ICF. This necessitates the use of multiple assessments to measure the broad perspective. The Children's Assessment of Participation and Enjoyment (CAPE)[3], the School Function Assessment (SFA)[4] and the Life Habits for Children (LIFE-H)[5] in combination cover participation in home, school, and community life.

Incorporation of participation measures in clinical trials will lead to a greater understanding of how interventions for children with CP have an impact on the broad context of participation.

Law et al.[6] provided a foundation from which to gain an improved understanding of the participation of children with physical disabilities in recreational and leisure activities. This information can assist families and service providers in planning activities that fit with their child's preferences and ensure active participation.

In using this knowledge it is important to consider that greater participation is not necessarily better, and lower participation does not imply personal failure.[7,8] A child could choose a variety of participation patterns, ranging from intense involvement in a few activities to participation in many activities. Participation in activities outside school is a choice that children and their families make to fit their needs, preferences, environment, culture, and lifestyle.[6]

As described in Chapter 1 (introduction) the human body makes adaptations to cope with the stresses placed on it during periods of exercise. For training adaptations to occur the bodies systems must be overloaded beyond their normal levels. If these extra stresses are applied over a period of time the system will adapt and this becomes its new norm. During periods of inactivity the human body will reverse these adaptations in an attempt to return itself to a norm as this is the current level of stress placed upon it. Therefore gains that have been made will be lost. Most training benefits are lost within a short period of stopping training. (Figure 1)

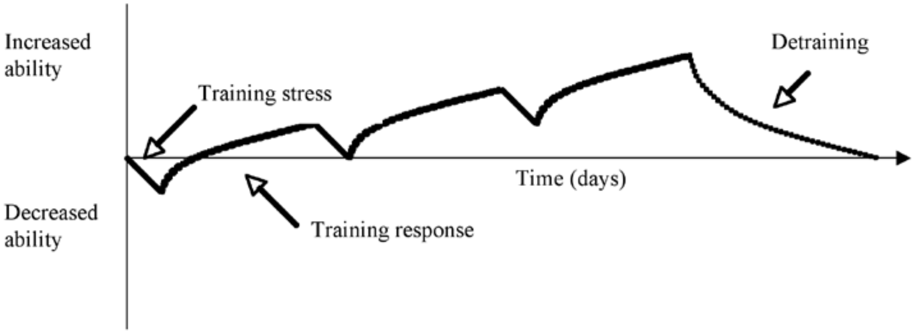

Figure 1. Schematic response of training and detraining.

The RCT described in Chapter 6 shows that changes seen in the school-based fitness intervention are reversed during a 4-month break. This observation illustrates the need to evaluate interventions for a sustained period. It is important to design interventions that will effectively improve childhood fitness. Developing and evaluating interventions to influence opportunities for healthful choices has been a focus of school-based health promotion research, including nutrition programs and physical education.[9] However, when interventions occur in a school-based setting, and are confined to the school year, an inherent question is one of sustainability.

In the RCT, that is described in Chapter 6 children were not specifically instructed to exercise during the four month summer break; in fact, no instructions were given about summer activity. Even during this relatively short break (four months) there was a loss of fitness benefits, resulting in a decline of fitness levels. These data show that in children with CP efforts to improve fitness should include exercise intervention in a sustained manner to improve fitness throughout the year, not just during the school-year. A more effective way to improve there general fitness could be by improving their participation in sports, play or active recreation outside the school-hours. Therefore, children with CP should be stimulated to participate in organized sports activities outside the rehabilitation center and schools for special education. However, at the moment, possibilities for handicapped children to be intensively active outside the rehabilitation center or school for special education are limited.

Moreover, it is unknown if barriers commonly observed in the general population (e.g. time, lack of interest, boredom) are similar or different in persons with CP. Therefore, barriers to exercise must be examined in persons with CP to have a higher chance of being successful.[10,11]

Effective assessment of outcomes in children with chronic health conditions is important to evaluate interventions.[12] The ICF provides a useful framework for both evaluation and intervention, considering aspects of body structure and function, activity, participation, and the influence of contextual factors on each domain. In order to conduct high-quality research into the effectiveness of rehabilitation and exercise programs, researchers need to be confident that their selected outcome will be sufficiently sensitive to change over time. The developed fitness tests in this thesis (10-m shuttle run test, Muscle Power Sprint Test, 10x5 meter Sprint Test and 30-sec Repetition Maximum) have not been investigated on its sensitivity to change.

The smallest detectable difference (SDD) is an indicator of sensitivity to change. Based on the Standard Error of Measurement (SEM), the SDD with 95% confidence is calculated as $1.96 \times \sqrt{2} \times SEM$. Only differences between two consecutive measurements greater than the SDD can be interpreted with 95% certainty as real change. There is an essential difference between 'clinically relevant change' and SDD. The SDD is a clinimetric property of a measurement instrument, while the 'clinically relevant change' is the change which clinicians and researchers minimally expect or judge as being an important change.[13,14] For the domains of functional outcome measured with the instruments in the present thesis, the 'clinically relevant change' is not known. However, in the RCT we found a 38% improvement for aerobic capacity using the 10-m shuttle run test, and a 25% improvement for anaerobic capacity using the Muscle Power Sprint Test after an 8 month exercise training program. Moreover, in the same study we found a 20 – 23% improvement in muscle strength using functional strength tests and a 15% improvement for agility using the 10x5 Meter Sprint Test. These values can be attributed with 95% certainty as real change since these values are greater than the calculated SDD. Moreover, these values are about the same magnitude as the measurement error of these outcomes, and show their sensitivity to change.

Therefore, it can be concluded that the newly developed outcome measures to assess aerobic capacity, anaerobic capacity and muscle strength are sufficiently sensitive to detect change over time.

As discussed in Chapter 2 (the literature review) the instruments used to measure the effects of fitness or strength training in the interventions were very diverse. Training effects are exercise mode specific.[15] Specificity of testing means that the modality of the testing tool needs to be similar to the type of activity the subjects train in. To find results that are more exercise-related, intervention-specific tests should be used in future research, since this may enhance the results of the studies and their interpretation. Therefore, efforts have to be made to develop core sets and standardize the exercise related outcome measures in CP. This will make study results interchangeable.

What we don't know

It is important to note the high prevalence of osteopenia and osteoporosis,[16] and poor nutrition[17] in people with CP, which are also very important health considerations that may affect and be affected by physical activity. Greater understanding of, and interventions directed at, these factors are also needed in order to optimize physical function and health-related quality of life in this population.[18,19] Information is lacking on the impact of exercise and fitness on specific health outcomes in people with CP, such as decreased body fat, lower blood lipids, and blood pressure; increased functional mobility; improved mood, self-efficacy, and life satisfaction. It is unknown if improvements in physical fitness will have similar benefits in improving various biomedical and psychological health outcomes observed in the general population.[20]

Bouchard et al.[20] have illustrated the relationship between physical activity, health-related physical fitness, and health outcomes (Figure 2). This indicates, that physical activity influences health directly as well as through its contribution to health-related physical fitness. Less clear is the impact of the amount or dosage of physical activity in this model. In children and adolescents with CP the dose-response relationship is unclear. Additional research is required to differentiate further the health benefits of daily physical activity versus exercise designed to enhance physical fitness.[21]

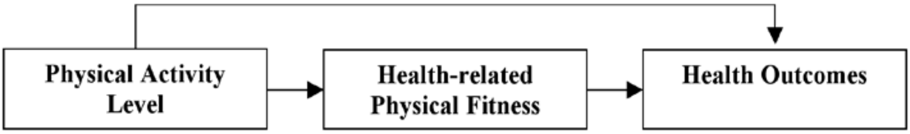

Figure 2. The relationship between physical activity, health-related physical fitness and health outcomes.[20]

Unwavering support from the individual's health care team for physical activity programming is essential. Their support is critical for assuring the child and their parents that physical activity is an important component in enhancing their overall health and well-being, and that exercise is safe for them.

A common problem among habitual exercisers is overtraining, which often results in musculoskeletal injuries.[22] Currently, there is no information on the prevalence of overuse injury in physically active persons with CP. Due to the higher incidence of inactivity and accommodating secondary changes (i.e. spasticity, contractures, joint pain), persons with CP may be more susceptible to overuse injuries than the general population.[23] Overuse injuries, which were not assessed in our randomized clinical trial, should be documented in future training studies to determine if certain exercises cause a higher or lower rate of injury.

In general, to be non-injurious to the patient and being effective for the purposes intended, physical activity must be prescribed according to age, gender, physical characteristics, related medical conditions, habitual physical activity and functional status. Special deficits or defects resulting from deformities and injury, and the needs and contra-indications imposed by these conditions must also be considered. The prescribed exercise dosage (i.e. intensity, frequency and duration) should be above the minimum level required to induce a "training effect", yet below the metabolic rate or intensity that evokes abnormal clinical signs, symptoms and/or injury.[24]

Evidence is increasing to suggest that fitness training is important for the pediatric CP population. However, little information is available on the clinical characteristics of the child that predict a response to such intervention. For the clinician it is important to have guidelines for selecting which children may benefit from an exercise program. Based on the data from the RCT we evaluated which baseline characteristics of the children and adolescents with CP could predict a positive functional response to the exercise program. The following baseline characteristics were used: GMFCS-level, sex, age, Body Mass Index (BMI), aerobic capacity, agility, muscle power, muscle strength (performance) and Gross Motor Function Measure (GMFM). In our RCT no baseline characteristics that could predict a positive functional response to the exercise program of the children that participated were found. In future studies possible characteristics, such as parent's attitude towards activity, cognition and motivation of the child and adolescent should be taken into account.

Because there are no specific exercise guidelines for individuals with CP, more innovative exercise research studies that develop and evaluate programs for accessibility, safety and effectiveness are needed. These types of studies will enable the exercise professional to develop an exercise program that is individually tailored to meet each patient's needs. Further, such studies will assist in developing guidelines for optimal exercise programming. As we gain experience, we will gradually be able to move beyond routine medical care issues to meeting the goals of optimizing physical functioning and overall health and well-being for children and adolescents with CP.

The general body of literature on physical fitness does not take into account the specific needs and concerns of persons with CP.[25] Public health recommendations often include guidelines for increasing physical activity that, in most circumstances, may be inappropriate for people with CP. For example, much of the exercise literature recommends walking is the safest and most convenient mode of exercise for improving cardio-respiratory fitness.[26] However, some persons with CP (e.g. GMFCS level III) have difficulty with walking, or may not be able to walk for a long enough duration or at a high enough intensity level to improve their physical fitness. Other persons with CP (e.g. GMFCS IV and V) may not be able to walk. Alternative exercises (i.e. (wheel)chair exercises) are usually not provided in public health reports that recommend physical activity to the general population, which makes it difficult for people with CP to develop their own personalized training program adapted to their specific needs.

Given these limitations, a skilled multidisciplinary assessment of the patient's abilities will help define a sports and activity program that is safe and tailored to the patient's needs. Safe activities can be suggested based on function and current activity levels. Each individual's limitations should be the focus of the exercise programming.

Function often decreases with age. Deficits from CP are not progressive. However, aging is often accompanied by a progressive spiral of inactivity leading to weight gain and decreased exercise tolerance.[25] The persistent underlying low fitness level of CP results in decreasing ability to ambulate and participate in physical activity. Few studies have addressed functional longitudinal outcomes in adults. By establishing a pattern of activity prior to adulthood, patients may avoid the health dangers of inactivity, be physically prepared to handle the aging process, and internalize the importance of activity and therapy. Additional research is needed on longitudinal effects of sports and physical activity through the lifespan.

Many children with CP tire of prolonged physical therapy from childhood into adolescence and adulthood. Therapy programs are simply abandoned due to boredom, expense, or lack of time.[27] Sports and sports therapy may offer an enjoyable alternative, providing children with CP skills for lifelong activity. They may experience normal sports enjoyment and a sense of mastery. Family life may be more normalized as the children are able to participate in enjoyable activities.[27] In most studies that focused on fitness-elements, the sports model of activity had a strong influence on the children's desire to participate and to continue participation during and after the studies.[27]

While many persons with disabilities can be encouraged to initiate a physical conditioning program, motivating them to continue is critical to promote favorable adaptation and improvement. Unfortunately, negative variables often outweigh the positive variable contributing to sustained participant interest and enthusiasm. Such imbalance leads to a decline in adherence while program effectiveness diminishes.

Research and empirical experience suggest that certain program modifications and motivational strategies may enhance participant interest and enjoyment.[28]

These include:

- Establishing short term goals
- Emphasizing variety and enjoyment
- Providing positive reinforcement through periodical testing
- Recruiting spouse support of the exercise program
- Including a modified recreational game to the conditioning program format that minimizes skill and competition and maximizes participant success
- Using progress charts to record exercise achievements
- Recognizing individual accomplishments.

In future studies and when developing an exercise program for children and adolescent with CP, these modifications and strategies should be taken into account. The chance of success and maintaining their fitness levels may be enhanced.

To conclude, we have learned from our study that children and adolescents with CP enjoyed being physically active and that this leads to increased fitness levels as well as improvement in the intensity of activities and health-related quality of life.

Implications for future research and clinical practice

- Evaluate exercise interventions for a sustained period
- Examine barriers (e.g. time, lack of interest, boredom) to exercise in persons with CP
- Develop core sets and standardize the exercise related outcome measures for all children and adolescents with CP
- Examine possible characteristics, such as parents attitude towards activity, cognition and motivation of the child and adolescent that could predict a positive response to the exercise training
- Examine if improvements in physical fitness will have benefits in improving various biomedical and psychological health outcomes
- Develop exercise programs based on each individuals limitations

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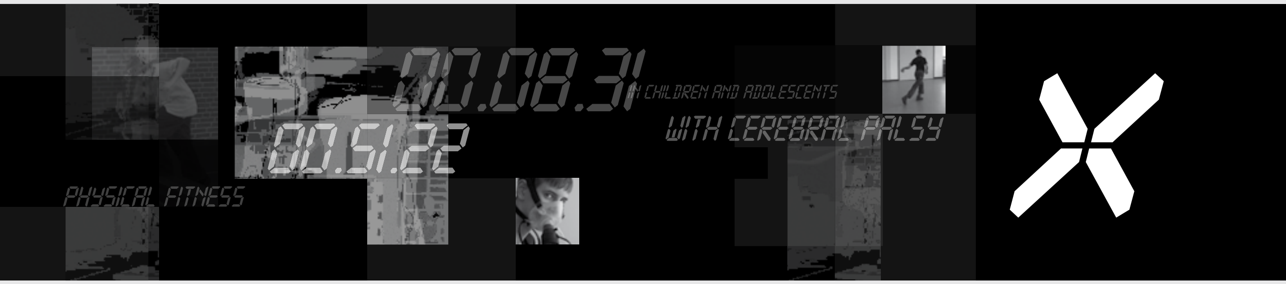


NEDERLANDSE SAMENVATTING

Dutch summary

Alhoewel cerebrale parese (CP) wordt gezien als een "non-progressieve" aandoening treden er wel degelijk secundaire complicaties op zoals afname van beweeglijkheid, spierkracht en cardiovasculair uithoudingsvermogen als kinderen ouder worden. Deze veranderingen kunnen een duidelijke invloed hebben op functionele onafhankelijkheid, vaardigheden en participatie van kinderen en adolescenten met cerebrale parese. Het onderzoek dat in dit proefschrift wordt beschreven is erop gericht deze secundaire complicaties indien mogelijk te stabiliseren of zelfs verminderen.

In **Hoofdstuk 1** wordt aangegeven dat het belangrijk is bij kinderen met cerebrale parese, die een laag fitnessniveau hebben, dit te verbeteren door middel van training. Ook worden drie belangrijke principes van training (specificiteit, overload en reversibiliteit) beschreven. Vanwege de aard van het trainingsprogramma zijn de kinderen die in dit proefschrift worden beschreven allen met de Gross Motor Function Classification System (GMFCS) geclassificeerd op niveau I en II. Bovendien zijn deze kinderen allen ouder dan 7 jaar. Uit de literatuur blijkt namelijk dat vanaf deze leeftijd bij kinderen de motorische patronen en vaardigheden zijn vastgesteld en dat ze in staat zijn om hun fitheid te verbeteren. Een overzicht van de studies die zich hebben gericht op fysieke training bij kinderen met CP en de effecten hiervan wordt gepresenteerd in **Hoofdstuk 2**. Uit dit literatuuroverzicht van verschillende effectstudies wordt geconcludeerd dat de methodologische kwaliteit van de geïncludeerde studies laag was. Toch lijkt het dat kinderen met CP kunnen profiteren van een trainingsprogramma dat zich richt op de spierkracht van de onderste extremiteit, cardiovasculaire fitness of een combinatie van beide. De meetinstrumenten die in de meeste studies werden gebruikt waren niet interventie-specifiek en richtten zich met name op het lichaamsfunctie- en activiteitenniveau van de ICF (International Classification of Functioning) van de World Health Organisation. Er wordt dan ook geconcludeerd dat er behoefte is om de effectiviteit van trainingsprogramma's die zich richten op verbetering van dagelijkse activiteiten en participatie van kinderen met CP alsmede een toename van de zelfwaargenomen competentie of kwaliteit van leven te bepalen.

In **Hoofdstuk 3** wordt de door ons nieuw ontwikkelde 10-m shuttle run test (SRT) voor kinderen met CP die geclassificeerd zijn op niveau I of II van de GMFCS geïntroduceerd. Deze test, die de aërobe capaciteit van kinderen met CP meet, wordt onderzocht op betrouwbaarheid en validiteit. De SRT-I en SRT-II zijn respectievelijk ontwikkeld voor kinderen met GMFCS niveau I en II.

Vijfentwintig kinderen en adolescenten met CP (geclassificeerd als GMFCS niveau I of II) hebben aan deze studie deelgenomen. Om de test-hertest betrouwbaarheid te onderzoeken werden de 10-m shuttle run tests binnen twee weken twee maal afgenomen bij ieder kind. Om de validiteit te onderzoeken werd een shuttle run test waarbij gasanalyse plaatsvond, vergeleken met een op het GMFCS-niveau gebaseerde loopbandtest waarbij ook de maximale zuurstofopname werd gemeten.

De resultaten laten zien dat de 10-m shuttle run tests betrouwbare (intraclass correlation coefficients (ICC's) van .97 voor SRT-I en .99 voor SRT-II) en valide (r=.96 voor beide tests) data geeft.

In **Hoofdstuk 4** worden twee tests die gebaseerd zijn op de vaardigheid lopen onderzocht op feasibility, betrouwbaarheid en validiteit. Deze twee testen zijn de Muscle Power Sprint Test (MPST) en de 10 x 5 Meter Sprint Test die respectievelijk de Peak en Mean Power en behendigheid tijdens snelle bewegingen meten. Zesentwintig kinderen en adolescenten met CP (geclassificeerd als GMFCS niveau I of II) hebben deelgenomen aan deze studie. We hebben goede feasibility en betrouwbaarheid gevonden voor de MPST en de 10 x 5 Meter Sprint Test (ICC's van ≥ .97 voor interobserver en test-hertest betrouwbaarheid). De validiteit van beide tests wordt ondersteund door significante verschillen in scores tussen kinderen die geclassificeerd zijn op GMFCS niveau I en II. Om de muscle power tijdens rennen bij kinderen met CP te onderzoeken is de Mean Power, afkomstig van de MPST de meest geschikte uitkomstmaat. Om de sprintprestatie en coördinatie tijdens snelle bewegingen te onderzoeken is de 10 x 5 Meter Sprint Test de meest geschikte test. Zowel de MPST als de 10 x 5 Meter Sprint Test kunnen een bijdrage leveren aan de trainingsevaluatie van kinderen en adolescenten met CP die geclassificeerd zijn op GMFCS niveau I of II.

In **Hoofdstuk 5** worden twee manieren om de spierkracht te testen bij kinderen met CP onderzocht. Bij de hand-held dynamometrie (HHD) worden de break- en makemethode vergeleken met betrekking tot hun intertester betrouwbaarheid. Daarnaast wordt er een nieuwe test (het 30-seconde Herhalings Maximum) om de functionele spierkracht te meten geïntroduceerd. Ook deze test wordt onderzocht op intertester betrouwbaarheid. Om de functionele spierkracht te meten bij kinderen met CP zijn functionele vaardigheden gekozen waarbij de grote spiergroepen van de onderste extremiteit die belangrijk zijn bij staan en lopen worden gebruikt. Dit heeft geleid tot drie gesloten keten oefeningen: 1a) Lateral Step-Up Test links, 1b) Lateral Step-Up Test rechts, 2) Sit-to Stand, 3a) Attain stand through half kneel links, 3b) Attain stand through half kneel rechts. Vijfentwintig kinderen met CP (GMFCS I of II) hebben deelgenomen aan dit onderzoek. De intertester betrouwbaarheid van de krachtmeting met een HHD was matig met ICC's die varieerden van .42 tot .73 voor de break-methode, en van .49 tot .82 voor de make-methode. De Standard Error of Measurement (SEM) en de Coefficients of Variation (CV(%)) varieerden van 27.9 tot 58.9 en van 22.2% tot 35.3% voor de break-methode en van 30.6 tot 52.7 en van 16.2% tot 56.2% voor de make-methode. De intertester betrouwbaarheid van de functionele spierkrachtmetingen met het 30-seconden Herhalings Maximum was goed met ICC's die varieerden van .91 tot .96. De waardes voor de SEM en CV(%) waren acceptabel. Deze varieerden van 1.1 tot 2.6 voor de SEM en van 10.9% tot 39.9% voor de CV(%). De resultaten van deze studie laten zien dat de makemethode geschikter is voor het meten van spierkracht bij kinderen met CP dan de break-methode als de HHD wordt gebruikt. Daarnaast is aangetoond dat het 30-seconden Herhalings Maximum voor de functionele spierkracht gebruikt kan worden om betrouwbare metingen van de onderste extremiteit te verkrijgen.

Hoofdstuk 6 beschrijft de effecten van een functioneel gebaseerd fitnessprogramma dat bestaat uit functionele oefeningen die erop gericht zijn de aërobe en anaërobe capaciteit bij kinderen en adolescenten met CP te verbeteren. Het programma bestond uit gestandaardiseerde aërobe en anaërobe oefeningen. Dit fitnessprogramma is additioneel aan de standaard zorg voor kinderen en adolescenten met CP (GMFCS niveau I of II) aangeboden. In een pragmatisch gerandomiseerde gecontroleerde trial werden de effecten van dit gestandaardiseerde fitnessprogramma onderzocht op aërobe en anaërobe capaciteit, behendigheid, spierkracht, zelfwaargenomen competentie, grof motorisch functioneren, participatie en gezondheidsgerelateerde kwaliteit van leven.

In totaal zijn 86 kinderen met CP (leeftijd 8-17 jaar) die geclassificeerd zijn op niveau I of II van de GMFCS benaderd om deel te nemen aan deze studie. Achtenzestig kinderen en adolescenten besloten mee te doen. Zij zijn vervolgens random verdeeld over een trainingsgroep (n=34) en een controle groep (n=34). De trainingsgroep trainde twee maal per week gedurende 45 minuten door middel van een circuit training bovenop het reguliere zorgprogramma. De controlegroep kreeg de reguliere zorg aangeboden.

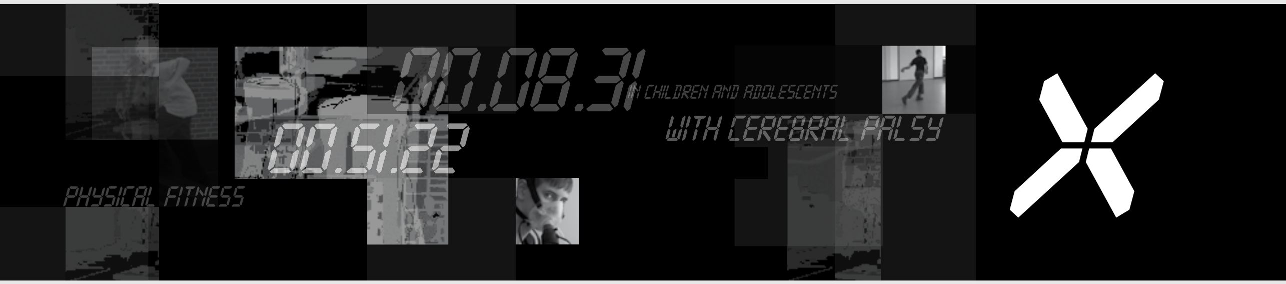
Metingen werden in beide groepen verricht bij aanvang van de training (T0), na 4 maanden (T1) en direct na de 8 maanden training (T2). Er was tevens een follow-up meting met dezelfde meetinstrumenten in beide groepen 12 maanden na T0. Primaire uitkomstmaten waren de aërobe en anaërobe capaciteit. Deze werden onderzocht met respectievelijk de 10-m shuttle run test en de Mean Power afkomstig van de MPST. Secundaire uitkomstmaten waren behendigheid (10 x 5 Meter Sprint Test), spierkracht (30-seconden Herhalings Maximum), zelfwaargenomen competentie (Competentie Belevings Schaal voor Kinderen), grof motorisch functioneren (Gross Motor Function Measure), participatieniveau (Children's Assessment of Participation and Enjoyment) en gezondheidsgerelateerde kwaliteit van leven (TACQOL-PF). Een significant trainingseffect ($p<0.05$) werd gevonden voor aërobe capaciteit en anaërobe capaciteit. Daarnaast werd er ook een significant effect gevonden voor behendigheid, spierkracht en sportvaardigheden. De intensiteit van de participatie liet een zelfde effect zien voor de formele, gezamenlijke en fysieke activiteiten en voor de activiteiten waar vaardigheden voor nodig zijn. Op de gezondheidsgerelateerde kwaliteit van leven werd een significante vooruitgang gevonden voor de domeinen motorisch, autonomie en cognitie.

Bij de follow-up bleek er een significant verschil te zijn in uitkomstmaten. De trainingsgroep ging terug naar het niveau dat gelijk was aan dat van T1, na 4 maanden training.

We concludeerden dan ook dat een 8 maanden durend fitnessprogramma dat bestaat uit functionele oefeningen de fysieke fitness, de intensiteit van de activiteiten en de gezondheidsgerelateerde kwaliteit van leven significant kan verbeteren wanneer het wordt toegevoegd aan de standaard zorg voor kinderen met CP.

In **Hoofdstuk 7** worden de implicaties van het onderzoek voor de praktijk beschreven en worden suggesties gedaan voor verder onderzoek.


IN CHILDREN AND ADOLESCENTS
00.06.45
WITH CEREBRAL PALSY
00.51.02


00.08.31
in children and adolescents
with cerebral palsy
00.51.22
physical fitness

DANKWOORD

Om een topsporter te laten pieken is keiharde training niet genoeg; geen enkele sport is tegenwoordig vrij van wetenschap en techniek. Of het nou gaat om video-analyse van de bewegingen, of om een aërodynamisch pak dat de luchtweerstand vermindert, iedereen heeft ermee te maken. En iedereen doet mee. Want wie wil er nou verliezen omdat hij als enige geen ribbels op zijn dijen heeft geplakt?
Het verhaal speelt zich telkens opnieuw af in allerlei sporten, maar ook in onderzoek. Een atleet of onderzoeker test een vernieuwend idee uit: een pak gebaseerd op haaienhuid, kromme schaatsen, nieuwe medicijnen of een nieuwe manier van therapie. Collega's twijfelen eerst aan het nut ervan, tot de pionier goede resultaten boekt. Ineens wil iedereen hetzelfde snufje hebben.
Tijdens iedere carrière, op sport en onderzoeksgebied, moet er op de juiste momenten worden gepiekt. Dat kan zijn tijdens een deelname aan de Olympische Spelen, een promotiewedstrijd naar een hogere klasse of het geaccepteerd krijgen van een eerste publicatie. Ook de voltooiing van mijn proefschrift zou kunnen worden gezien als pieken op het juiste moment. De promotie, die plaatsvindt op 18 december om 14.30 uur in het Academiegebouw te Utrecht, zou in de sportwereld dan ook niet misstaan. Het toewerken naar deze piek heb ik niet alleen gedaan. Ik wil dan ook iedereen bedanken die me heeft geholpen, bijgestaan en ondersteund bij het toewerken naar mijn piekmoment.

Allereerst wil ik de 68 pupillen en hun ouders bedanken voor hun onvoorwaardelijke steun en deelname aan het onderzoek. Dit had niet kunnen plaatsvinden zonder de enthousiaste deelname en medewerking bij het uitvoeren van alle testen en het invullen van diverse vragenlijsten die de afgelopen periode zijn langsgekomen. Ook dank voor alle leuke reacties die ik heb mogen ontvangen. Hierdoor wist ik, naast de resultaten uit de statistiek, dat het onderzoek waar jullie aan deelnamen ook waardevol is voor de dagelijkse fysiotherapiepraktijk. Daarnaast was het erg prettig om te horen dat het trainen als leuk werd ervaren.

Daarnaast wil ik de **TECHNISCHE STAF** bedanken bij de begeleiding tijdens de road to success.
Professor Dr. P.J.M. Helders, beste Paul. Jij was degene die ruim drie jaar geleden vertrouwen had in mij en in het onderwerp van dit proefschrift. Bedankt voor je overzicht, snelle reacties op manuscripten en strategische keuzes bij het insturen van de manuscripten. Daarnaast liet je me steeds weer versteld staan van je enorme kennis, niet alleen van je eigen vakgebied maar ook ver daarbuiten. Je bent een geweldig voorbeeld van een leidinggevende die werkt en laat werken "aan de grenzen van het weten." Ik heb veel van je geleerd.
Dr. M. Ketelaar, beste Marjolijn. Ik wil je bedanken voor de introductie in de methodologie en statistiek. Je kritische kijk op mijn analyses en artikelen waren steeds van grote waarde. De nauwgezette manier waarop jij ieder manuscript

van commentaar voorzag om ze vooral leesbaarder te maken en de opbouwende manier waarop jij altijd feedback gaf, zijn voor mij steeds een stimulans geweest om verder te gaan. Je deur stond altijd voor me open. Ik hoop dat we de komende jaren nog tijdens verschillende projecten zullen samenwerken.
Dr. T. Takken, beste Tim. Jouw expertise op het gebied van de pediatrische inspanningsfysiologie is ongekend en van onschatbare waarde voor de kindergeneeskunde. Jij hebt me de belangrijkste elementen bijgebracht van de inspanningsfysiologie. Een begin hiermee werd gemaakt tijdens een 'trainingskamp' op Papendal. Enorm bedankt voor je goede en snelle analyses bij de betrouwbaarheidsartikelen en je snelle reacties op verschillende versies van manuscripten. Je hebt mij keer op keer verrast door de enorme hoeveelheid energie die jij hebt. Jouw kennis van de literatuur op het gebied van de inspanningsfysiologie is onovertroffen. Ik hoop dat de samenwerking niet ophoudt bij het einde van dit promotietraject.
Dr. J.W. Gorter, beste Jan Willem. Jij hebt, samen met Marjolijn, mij vanaf het begin van mijn onderzoekscarrière begeleid en gecoacht. Samen hebben we, toen ik aangaf wat meer te willen in de kinderrevalidatie, een aantal kleinere onderzoeken uitgevoerd. Op deze manier kwam ik er achter dat onderzoek bij me paste en dat ik het bovendien leuk vond om te doen. Mede door jouw contacten in de revalidatiewereld en daarbuiten is het gelukt om dit proefschrift tot een goed einde te brengen. Je kennis en ervaring, je interesse in het verloop van het onderzoek maar vooral je passie en aanstekelijk enthousiasme hebben ervoor gezorgd dat ik steeds weer met volle moed en veel plezier met mijn werk doorging.

Door een viertal toegewijde en enthousiaste **SCOUTS** (revalidatieartsen Nanne Land uit Zwolle, Ruud Keijzer uit Goes, Agnes van Velzen uit Breda en Marie-Anne Kuijper uit Utrecht) zijn kinderen gevonden die hebben deelgenomen aan dit onderzoek. Deze kinderen zijn 8 maanden lang getraind door de beste **TRAINERS** van het land. Deze trainers werken op vier verschillende mytylscholen, die zijn gelegen in Goes, Breda, Utrecht en Zwolle. Bedankt Mischa, Ilse, Annelies, Yvonne en Joep (Utrecht), Anouschka, Caroline, Mirella en Joyce (Zwolle) Ruth, Elly, Cisca, Monique, Pim en Willy (Goes) en Koen, Astrid, Karin en Ruud (Breda) voor al die uren intensieve arbeid die jullie in de trainingen hebben gestopt. Zonder jullie inzet en doorzettingsvermogen was dit project niet zo soepel verlopen. Onderzoeksassistenten (Ilse, Chantal, Heidi, Radmer, Jose, Joep, Annelies, Elwin, Marianne, Anneleen); zonder jullie inzet en medewerking waren de metingen tijdens de interventiestudie niet zo goed verlopen als nu. Ik hoorde regelmatig dat jullie flexibiliteit, overredingskracht, doorzettingsvermogen en geduld op de proef werden gesteld. Vaak waren jullie de **MENTAL COACHES** van de kinderen. Ontzettend bedankt voor jullie hulp. Natuurlijk wil ik ook de zorgdivisie van revalidatiecentrum De Hoogstraat bedanken voor de flexibiliteit en medewerking bij de inzet van de onderzoeksassistenten.

Daarnaast wil ik mijn twee **PERSOONLIJKE ASSISTENTEN** Ger Scholtens en Ingrid van de Port bedanken voor al het plezier, vertrouwen, de hulp en adviezen in diverse periodes van mijn carrière. Ik vind het erg leuk dat jullie mijn paranimfen willen zijn. Ger, jij hebt mij vanaf het begin dat ik aangaf een ambitie richting onderzoek te hebben op alle mogelijke manieren gestimuleerd en gefaciliteerd. Zonder jouw inzet, enthousiasme en betrokkenheid zou mijn onderzoek niet zo voortvarend zijn begonnen en verlopen. Ingrid, mijn trainingsmaatje en sparringpartner. Jouw relaxte aanpak paste perfect bij mijn eigen ideeën. Jij hebt je promovenduscarrière vorig jaar afgesloten met een prachtige promotie, en bent nu senior onderzoeker. Je hebt nu een voet tussen de deur bij de "Lord of the Rings." We hebben heel wat gezellige uurtjes doorgebracht op kamer L2014. Jammer dat we niet meer naast elkaar zitten.

Gedurende mijn onderzoek heb ik goede professionele **BEGELEIDING** gehad, De fysiotherapiecollega's van mytylschool Utrecht (Ilse, Appie, Marianne, Marian, Yvonne, Heleen, Mischa, Lineke, Vera, Annelies, Manon en Liselotte) hebben zich allen volledig ingezet om van dit onderzoek iets moois te maken. Naast het meedenken over de uitvoering werd er zonder problemen in pauzetijden en andere vrije uurtjes met de stopwatch en pen en papier hard gewerkt. Bedankt voor alle leuke jaren dat ik met jullie heb mogen werken. Mijn carrière heeft een andere wending gekregen waardoor ik jullie wat minder vaak zal gaan zien. Hopelijk komen we elkaar toch nog regelmatig tegen. Ik zal jullie missen.

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In mijn laatste 'wedstrijd' als promovendus, die plaats zal vinden in het Academiegebouw te Utrecht, zal de beoordelingscommissie optreden als **SCHEIDSRECHTERS**.

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Jasper, jij bent zonder dat je het beseft mijn **MASCOTTE**. Het is na een werkdag en een rit naar huis prettig om thuis te komen. Je bent altijd opgewekt en vrolijk. Het is een genot je te zien opgroeien. Je vrolijke gezicht op mijn bureaublad heeft er de laatste maanden voor gezorgd dat ik op tijd een time-out nam.

Josine en Jasper, wat lief en gezellig dat jullie mij volgen naar Canada, om daar vier maanden lang een mooi vervolg te geven op mijn onderzoek en ambitie aan de University of Alberta. Gelukkig heeft Edmonton een enorm winkelcentrum. Jullie zullen je niet vervelen. Trust me, I am a doctor...

Hopelijk zal ik op mijn piek promoveren naar een hogere klasse; die van de doctors. In de sport wordt vaak gezegd dat men op het hoogtepunt wil (of had moeten) stoppen. Ik ben van plan toch nog even door te gaan, en wil nog vaker gaan pieken.


IN CHILDREN AND ADOLESCENTS
00.06.45
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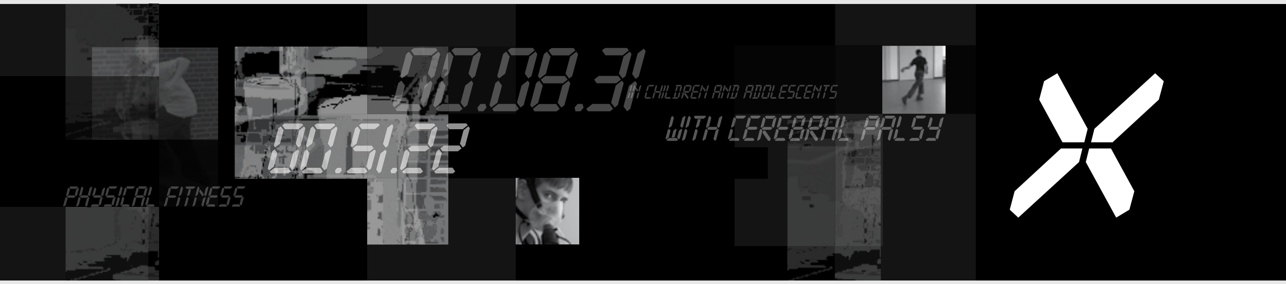

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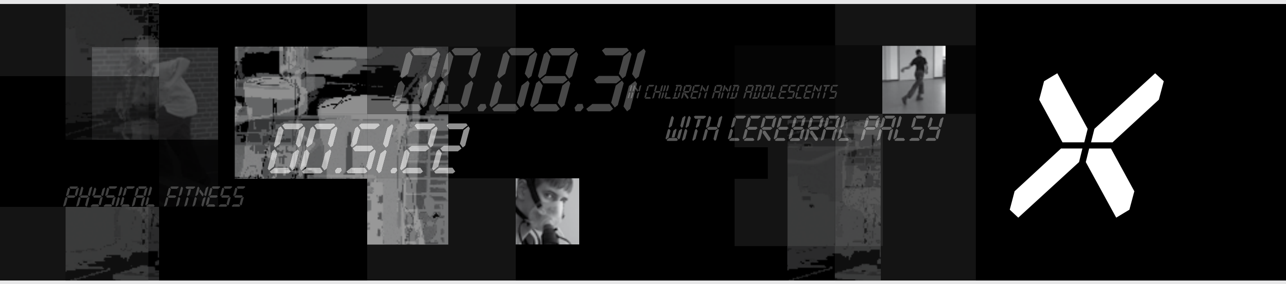
IN CHILDREN AND ADOLESCENTS
00.06.45
WITH CEREBRAL PALSY
00.51.02



CURRICULUM VITAE

De auteur is op 28 november 1973 te Hilvarenbeek geboren. Hij behaalde in 1993 zijn VWO-diploma aan het St. Odulphus lyceum te Tilburg. Van 1993 tot 1997 werd de opleiding fysiotherapie gevolgd aan de Hogeschool van Utrecht.

Van 1997 tot heden is hij werkzaam als (kinder)fysiotherapeut op Revalidatiecentrum De Hoogstraat te Utrecht. Aan het begin van deze periode (1998-2001) volgde hij een 3-jarige Post Academiale Scholing Kinderfysiotherapie aan de Hogeschool van Breda.

Specifieke wetenschappelijke scholing vond plaats via de basismodule ‘Statistiek 1’ van de Open Universiteit en ‘Methoden 1’ van de Katholieke Universiteit Nijmegen. Daarnaast werden bij het EMGO de cursussen ‘Epidemiologisch onderzoek; opzet en interpretatie’ en ‘Lineaire regressie en variantieanalyse’ gevolgd.