From exercise training to school-based sports

The effects on fitness and health in youth with physical disabilities

Brain Center Rudolf Magnus

Maremka Zwinkels

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From exercise training to school-based sports

The effects on fitness and health in youth with physical disabilities

Van trainingsprogramma naar (na)schoolse sport

De effecten op fitheid en gezondheid bij jongeren met een fysieke beperking

(met een samenvatting in het Nederlands)

Proefschrift

ter verkrijging van de graad van doctor aan de
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prof.dr. G.J. van der Zwaan, ingevolge het besluit van het
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Maremka Zwinkels

geboren op 4 september 1987
te Delft

Promotor Prof.dr. J.M.A. Visser-Meily

Copromotoren Dr. O. Verschuren
Dr. T. Takken

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1

General introduction

POPULATION

When a regular school is not able to support a child with physical disabilities, learning problems, or extra needs, a child can attend a school for special education. These schools have similar learning objectives as regular schools, but the children receive additional attention and support. Special education has been developed for both primary (4–12 year) and secondary (13–18 year) education. Children and adolescents following special education are divided into four different cluster divisions:

- Cluster 1: Children and adolescents who are visually impaired, have multiple disabilities with a visual impairment, or are blind. Most scholars go, with special supervision, to regular schools. The remainders attend special schools.
- Cluster 2: Children and adolescents who are deaf, have hearing impairments, speech or language difficulties, or communication problems.
- Cluster 3: Children and adolescents with physical, mental and/or multiple disabilities, and those with a chronic disease.
- Cluster 4: Children and adolescents with psychiatric disorders or severe behavioral problems.

Of all children and adolescents following primary or secondary education in the Netherlands, 4.7% go to a school for special education, and 1.9% are within cluster 3.[1,2] Due to their physical impairment, children and adolescents within cluster 3 often have difficulties with their mobility. Schools for cluster-3 education provide rehabilitation medicine within school hours. Dedicated pediatric physicians together with physical-, occupational- and speech pediatric therapists take care of therapies on a daily basis. In this thesis, we will focus on children and adolescents within cluster 3 with a chronic disease or physical disability, comprising a total of 26,500 children and adolescents in the Netherlands.[2]

The majority of children and adolescents within cluster-3 education have neuromuscular disorders, with cerebral palsy (CP) as the most common movement disorder with a prevalence well above two out of 1,000 live births in the Netherlands.[3] 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.[4] These children and adolescents can have poor coordination, spastic muscles, weak muscles and/or tremors. Signs and symptoms vary across individuals. The gross motor function of children with CP can be categorized into five different levels from walking independently on all surfaces (Gross Motor Function Classification System (GMFCS)-level I) to using an electric wheelchair with assistance (GMFCS V).[5] The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, behavior, by epilepsy and by secondary musculoskeletal problems.[4] Therefore, CP as a single diagnose, is a very heterogeneous disease and can manifest itself in different ways.

Another neuromuscular disorder frequently seen in cluster-3 education is spina bifida (SB). SB is a malformation of the spinal cord and often the brain, which can result in both motor and sensor impairment, incontinence for bowel and bladder, and cognitive impairment.[6] Depending on the height of the lesion level of the spinal cord, children and adolescents with SB experience difficulties with ambulation. A lot of research has been done in youth with CP who are ambulatory and to a lesser extent in those with SB. As a consequence, most literature for evidence-based practice within the cluster-3 population comes from children and adolescents with CP. Other neuromuscular disorders within cluster-3 education are psychomotor retardation without any specific diagnosis and acquired brain injury. Next to neuromuscular disorders, the population also includes children and adolescents with metabolic, musculoskeletal, cardiovascular and pulmonary disorders.

The cluster-3 population consists of a variety of diagnoses. Those children and adolescents go to school, play and exercise together, including children and adolescents who are ambulatory, those using walking aids or orthotic shoes for their locomotion, and those propelling a manual or use a powered wheelchair. In the current thesis, all children and adolescents with a chronic disease or physical disability, independent of their diagnosis or level of mobility with exception of those using a powered wheelchair, were included. In this way, our population is a reflection of daily-life at schools for cluster-3 education in the Netherlands. Because of the heterogeneity of our population, participants were divided based on their mobility level during physical education, as a representation of sports participation. The Functional Mobility Scale (FMS), originally developed for children with CP, is being used to classify children based on their level of mobility.[7] Participants were divided into three subgroups: (6) those who were able to run (i.e., runners), (5) those who could walk independently but were not able to run (i.e., walkers), and (1) those who propel a manual wheelchair (i.e., wheelchair users) (Figure 1.1).[7] Since no participants walked with assistive devices during physical education, subgroups 2–4 were redundant. In addition, subgroups would not be evenly distributed in a representative sample, since only 15% of Dutch individuals from 12 years of age with a motor disabilities use a wheelchair for their mobility.[8]

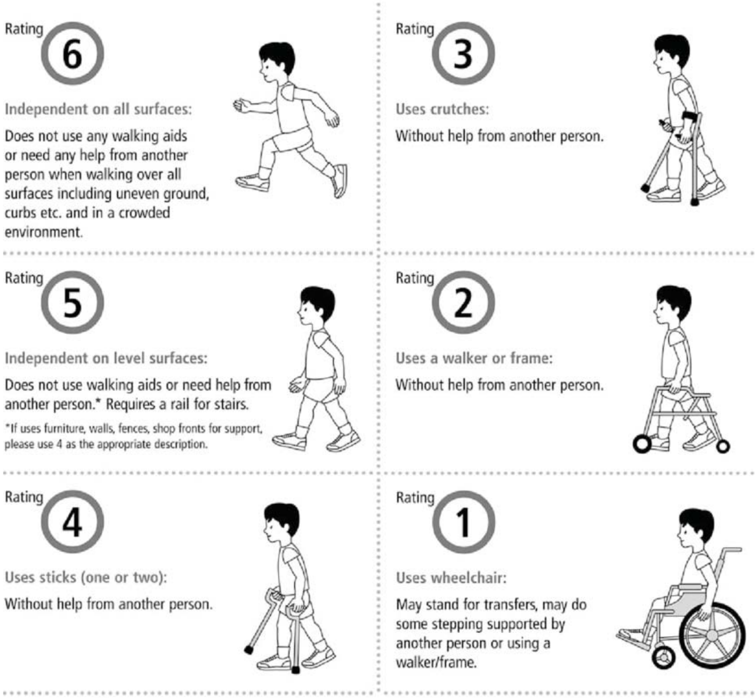

Figure 1.1: The functional mobility scale (FMS) to classify children's level of mobility.

PHYSICAL FITNESS

Physical fitness is a set of attributes that people have or achieve in relation to the ability to perform physical activities.[9] Different energy systems contribute to both short and long bouts of physical activity in daily life.[10] Short and vigorous bouts of activity rely on the anaerobic metabolism, where energy is quickly supplied with low reliance on aerobic combustion of carbohydrates and fat. These short intermittent bouts of intense activities (i.e., sprints) reflect children's daily activity pattern,[11] for example when playing tag or hide and seek. In addition to the anaerobic system, short bouts of activities in daily life are also attributed to coordination (i.e., sprint technique).[10] Therefore, the combination of the anaerobic system and coordinated exertion, as well as children's daily life activity pattern, resulted in our primary outcome measure: anaerobic performance. On the other hand, long and low-to-moderate intensity activities rely on the aerobic metabolism, involving the breakdown of fuels with the aid of oxygen. The maximum amount of oxygen consumed by the body during exercise (VO_2peak) reflects the aerobic capacity or cardiorespiratory fitness. VO_2peak is dependent on three systems: the respiratory system (i.e., lungs), the cardiovascular system (i.e., heart, blood and vessels), and the muscular system (i.e., muscles).[12] In youth with CP for example, a reduced VO_2peak can mainly be explained by the reduced oxygen extraction in the exercising muscles due to lower muscle mass.[13]

Many studies have reported that youth with a chronic disease or physical disability have decreased values of both anaerobic performance and VO_2peak compared to their typically developing peers.[14] Low physical fitness levels are highly associated with reduced physical activity, and increased cardiovascular diseases and overall mortality in healthy populations.[15–17] Cardiometabolic risk factors such as dyslipidemia, hypertension, hyperinsulinemia or insulin resistance, and obesity often already exist in children and adolescents who are typically developing.[18,19] Next to cardiometabolic health, physical fitness in youth with physical disabilities is also positively associated with psychosocial health, including self-perception and health-related quality of life.[20–22] Additionally, in typically developing youth physical fitness seems to be positively associated with attention as well,[23–25] where enhanced physical activity has been associated with improved attention.[26,27]

Since physical fitness has been associated with many factors, promoting physical activity and increasing physical fitness has been an important goal in clinical practice aiming for structural participation and optimal cardiometabolic health, psychosocial health and attention (i.e., exercise is medicine). In recent years, many efforts have been employed to implement exercise training in rehabilitation medicine of children and adolescents with a chronic disease or physical disability. Therefore, it is the question as to whether there is an upward secular trend in fitness levels among these children and adolescents. Therefore, we have investigated this question in a sample of children and adolescents with CP.

PHYSICAL ACTIVITY

Compared to their typically developing peers, children and adolescents with a chronic disease or physical disability are often restricted in their participation in physical activities and sports as a consequence of real or perceived limitations imposed by their condition.[28] The chronic condition itself often contributes to lower activity levels, which in turn leads to a deconditioning effect, lower performance levels and a downward spiral of deconditioning and lower activity level occurs.[29,30] To prevent health risks later in life, it is assumed that children and adolescents should be physically active on a structural basis. Adults with CP have significantly higher odds of chronic diseases including diabetes, asthma, hypertension, stroke, joint pain and arthritis compared to adults without CP.[31] Both level of mobility and physical activity were associated with these chronic diseases.[31]

In the Netherlands, only 21% of the cluster-3 children and adolescents meet the physical activity guidelines (NNGB: Dutch physical activity guideline) of at least 60 minutes of moderate-intensity every day (Figure 1.2).[32] Overall physical activity is divided into sports participation, active play and active transport. For the cluster-3 population, 26% participates in sports at least once a week, 30% plays outside for at least 60 minutes after school time, and 13% uses active transportation (i.e. walking or cycling) for at least three days a week. Compared to the typically developing youth, no differences in meeting the physical activity guidelines emerge; a minority of about a quarter is sufficiently active every day in both youth with and without physical disabilities. In contrast, these numbers differ largely for sports participation (71% vs 26%) and active transport (91% vs 13%) in youth who are typically developing and youth with a chronic disease or physical disability, respectively. Since there are many advantages of being physically active on a structural basis, the current thesis will focus on the transition from exercise training to sports participation.

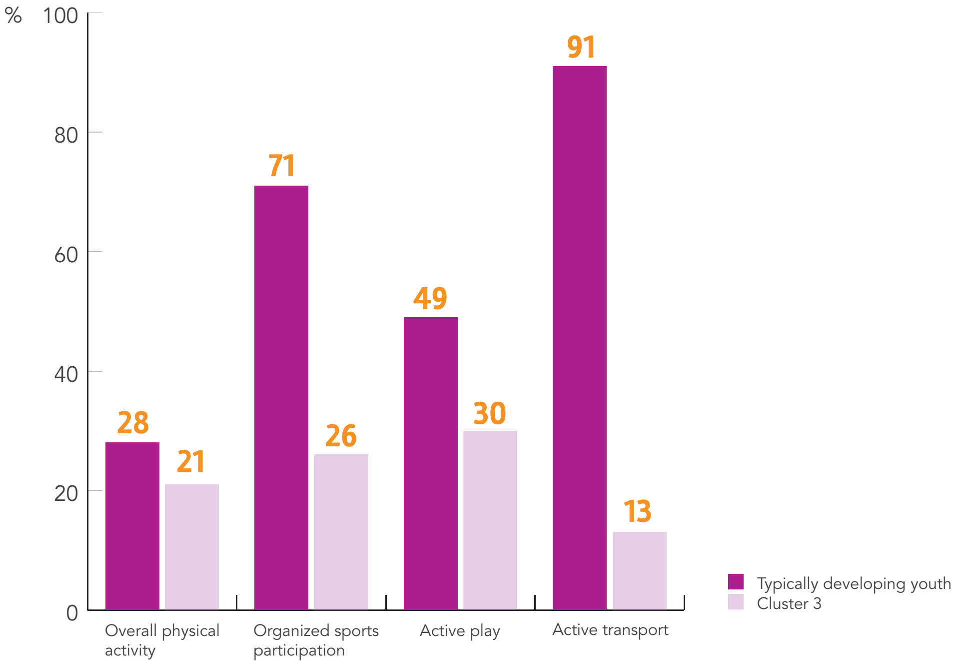

Figure 1.2: Overall physical activity of youth with physical disabilities compared to their typically developing peers described by Burghard et al.
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EXERCISE TRAINING

Exercise training is a planned, structured, and repetitive bodily movement done to improve or maintain one or more components of physical fitness.[9] It is generally acknowledged that children and adolescents with a chronic disease or physical disability can be trained according to general exercise principles.[33–35] For very deconditioned children and adolescents, it is recommended to start with 1–2 sessions a week.[34] Training interventions have shown that children and adolescents with various physical disabilities who are ambulatory can improve both health and exercise performance (i.e., performance-related fitness).[14,36–40] For example, an eight-months standardized twice a week exercise program in ambulatory children with CP consisting of functionally based exercises improved exercise time, anaerobic performance and agility.[41] However, the evidence of exercise training in children and adolescents using a manual wheelchair is lacking. It is interesting and of additional value to investigate whether wheelchair users can be trained following regular training principles and which training type is most beneficial for this population. In addition, field tests to measure their progress longitudinally are not available for this specific population.

It is often hypothesized that exercise training could break through the downward spiral of deconditioning and physical inactivity. However, the positive training effects after an exercise training program did not sustain in many studies (Figure 1.3),[14,40–42] nor did the positive training effects result in increasing physical activity levels.[43] Three to four months following an eight month training intervention, participants with CP scored 20–50% lower compared to their score immediately after intervention on both aerobic and anaerobic fitness.[40,41] In other words, for these children and adolescent the "use-it-or-lose-it" principle also applies. One should be physically active on a structural day to day basis to maintain his or her

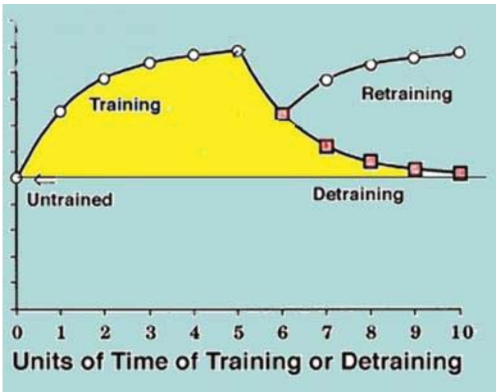

Figure 1.3: Schematic effects of training, detraining and retraining in time.

fitness level. In addition, it is assumed that regular physical activity during childhood, is a starting point for an active and healthy adulthood.[31,44] Both the abovementioned exercise interventions, and exercise training within physical therapy, are provided temporarily, but will not sustain into adulthood unless one adds physical activity structurally in leisure time. In this thesis, we will investigate whether participation in sports can increase the sustainability of the effects on physical fitness, cardiometabolic health, psychosocial health and attention of an exercise training.

HIGH-INTENSITY INTERVAL TRAINING

Before starting participation in sports, a short-term, high-intense, exercise training can familiarize children with exercise and provide a boost of their fitness level. Next to the more traditional forms of exercise training, high-intensity interval training (HIT) has shown to be a more effective and time-efficient form of training in adults with cardiometabolic diseases.[45] HIT involves alternating short bursts of high-intensity exercise followed by a period of low-intensity exercise or rest.[46] Despite large differences in training volume (i.e., 90% lower in HIT) and time commitment (i.e., 67% lower in HIT), HIT provides similar or greater benefits on fitness and health when compared to moderate intensity continuous training.[46–48] Next to the training time and volume benefits, children's daily physical activity pattern most often consists of brief, intermittent bouts of intense movement interspersed with varying intervals of low and moderate intensity.[11] As a result, the type of training also matches their daily activities where they are engaged in short intense movements.

SPORTS PARTICIPATION

For youth with a chronic disease or physical disability it seems more difficult to participate in sports and physical activities when compared to typically developing peers.[28] As mentioned before, only 26% of Dutch children and adolescents with a chronic disease or physical disability from cluster-3 education participate at least once a week in sports.[32] Reasons for this lower sport participation are: being physically active is more challenging because of their disability, lack of trained support personnel, transportation problems, lack of acceptance, and lack of sports clubs in the neighborhood.[49–51] When a sports program is provided at school, extracurricular, most of these barriers can possibly be eliminated, because this setting offers a familiar environment with supported trainers, acceptance, and no additional transportation except a postponed pick-up from school.

Providing a school-based sports program is an integrative approach targeting even the least active children.[52] Parents consider once a week sports participation to be feasible and sufficient,[53] since they need to prioritize the frequency with the demands of everyday life.[54] However, it is unknown whether once a week is beneficial in improving or maintaining fitness and health purposes in youth with physical disabilities. In addition, limited evidence exists for the effects on fitness and health in children and adolescents with a chronic disease or physical disability who participate in sports structurally. Because of the potential fitness and health effects of sports participation in children and adolescents with a physical disability or chronic disease, the Health in Adapted Youth Sports Study (HAYS) and Sport-2-Stay-Fit (S2SF) studies were initiated by the Shared Utrecht Pediatric Exercise Research Lab (SUPER-Lab) research group in Utrecht.

The aim of the HAYS study is to determine the effects of sports-related fitness and health outcomes in children and adolescents with a chronic disease or physical disability. The HAYS study cross-sectionally compared youth with physical disabilities, aged 10–19 years, who are actively participating in organized sports to their non-sporting peers. The results of the HAYS will be published separately.[55] The Sport-2-Stay-Fit study investigated whether a school-based sports program can increase the sustainability of a HIT program in children and adolescents with a chronic disease or physical disability. Both studies assessed similar outcome measures: physical fitness, physical activity, cardiometabolic health, psychosocial health and attention.

AIMS AND OUTLINE OF THIS THESIS

This thesis focusses on children and adolescents with a chronic disease or physical disability and consists of three main parts:

1. Exercise training types and performance tests for youth using a manual wheelchair (chapters 2 and 3)
2. Sport-2-Stay-Fit study where HIT is followed by a school-based once a week sports program for six months (chapters 4, 5, 6 and 7)
3. Ecological time trends over a decade in fitness levels of youth with spastic CP (chapter 8)

Chapter 2 presents a comprehensive review that summarizes the evidence of exercise training types on improving wheelchair propulsion capacity. **Chapter 3** introduces the reliability and validity of a newly developed anaerobic performance test in wheelchair using children and adolescents with CP. **Chapter 4** describes the design of the Sport-2-Stay-Fit study. **Chapter 5** summarizes the results of a HIT program of eight weeks on physical fitness and cardiometabolic health. **Chapter 6** addresses the results of a school-based once a week sports program for six months on physical fitness, physical activity and cardiometabolic health, where **chapter 7** shows the results on psychosocial health and attention. To place our results in perspective of the developments in rehabilitation medicine, **chapter 8** compares body mass index, performance-related fitness and cardiorespiratory fitness of children with CP measured in 2014 with a comparable sample from 2004. Finally, **chapter 9** discusses the general outcomes of this thesis, and how this may affect clinical practice.

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2

Exercise training programs to improve hand rim wheelchair propulsion capacity: a systematic review

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Tim Takken

Clinical Rehabilitation 2014; 28: 847–61

ABSTRACT

Objective: An adequate wheelchair propulsion capacity is required to perform daily life activities. Exercise training may be effective to gain or improve wheelchair propulsion capacity. This review investigates whether different types of exercise training programs are effective in improving wheelchair propulsion capacity.

Data sources: PubMed and EMBASE databases were searched from their respective inceptions in October 2013.

Review methods: Exercise training studies with at least one outcome measure regarding wheelchair propulsion capacity were included. In this study wheelchair propulsion capacity includes four parameters to reflect functional wheelchair propulsion: cardiorespiratory fitness (aerobic capacity), anaerobic capacity, muscular fitness and mechanical efficiency. Articles were not selected on diagnosis, training type or mode. Studies were divided into four training types: interval, endurance, strength, and mixed training. Methodological quality was rated with the PEDro scale, and the level of evidence was determined.

Results: The 21 included studies represented 249 individuals with spinal cord injury (50%), various diagnoses like spina bifida (4%), cerebral palsy (2%), traumatic injury (3%) and able-bodied participants (38%). All interval training studies found a significant improvement of 18–64% in wheelchair propulsion capacity. Three out of five endurance training studies reported significant effectiveness. Methodological quality was generally poor and there were only two randomized controlled trials.

Conclusion: Exercise training programs seem to be effective in improving wheelchair propulsion capacity. However, there is remarkably little research, particularly for individuals who do not have spinal cord injury.

Clinical messages

- Exercise training programs seems to be beneficial for improving wheelchair propulsion capacity, except for respiratory muscle training
- Interval training is the most promising training type
- There is an urgent need for research into the immediate and long-term benefits of exercise training in wheelchair using patients, particularly including people who do not have spinal cord injury

INTRODUCTION

Increasing physical activity level is associated with a lower risk of developing cardiovascular diseases.[1] Compared to able-bodied individuals, manual wheelchair users have reduced physical activity[2,3] and physical fitness levels.[4] This has a major impact on their daily life activities, social participation, and overall quality of life.[5-7] The intensity of wheelchair propulsion in daily life may not put sufficient stress on the cardiovascular system to induce positive health effects.[8] Hence, breaking through the vicious cycle of deconditioning in manual wheelchair users may require exercise training, i.e. a type of physical activity consisting of standardized, planned, structured, and repetitive bodily movement intended to improve or maintain one or more components of physical fitness.[9]

Improving physical fitness may also result in reduced relative effort in daily activities. Since manual wheelchair users' primary means of mobility is through hand rim wheelchair propulsion, exercise training should aim to improve their physical fitness levels in such a way that hand rim wheelchair propulsion has the greatest impact in daily life. The capacity for hand rim wheelchair propulsion is referred to as wheelchair propulsion capacity. Since both long and short bouts of activity are important in daily life,[10] wheelchair propulsion capacity is divided into endurance and sprint capacity.

Optimized wheelchair propulsion capacity requires cardiorespiratory fitness (i.e., aerobic capacity), anaerobic capacity and muscular fitness (Figure 2.1).[11] In addition, these physiological parameters need to be translated into functional wheelchair propulsion. Besides physical fitness, mechanical efficiency is also important for wheelchair propulsion capacity. Power output during hand rim wheelchair propulsion is regarded as the outcome measure most closely related to wheelchair propulsion capacity.[12] Other 'integrated' outcome measures for wheelchair propulsion capacity are the results of wheelchair propulsion tests in practical situations (i.e., field tests).

Valent et al.[13] reviewed studies on the effect of upper-body exercise and reported that no conclusions could be drawn because of the overall low quality of studies. They focused on persons with spinal cord injury only. Training of wheelchair propulsion capacity might be beneficial for more diagnostic groups, and knowledge on training effects in able-bodied subjects might provide more insight in the potential effects for persons with disabilities. In addition, exercise training should have its effect on daily life wheelchair propulsion. The focus of the present review will therefore be on integrated wheelchair propulsion outcome measures instead of specifically types of training or bodily related outcome measures. Moreover, several years have passed since the last review. Therefore, the present review

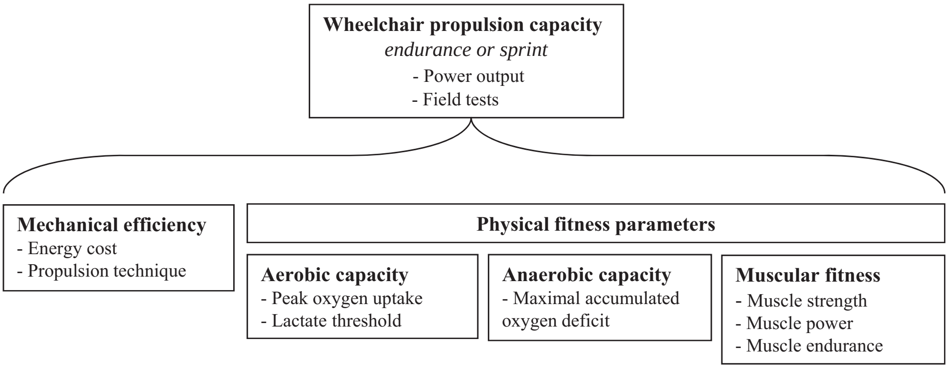

Figure 2.1: Schematic relationship of wheelchair propulsion capacity and underlying parameters.[17,57]

aims to systematically review the literature on the effectiveness of training programs in improving hand rim wheelchair propulsion capacity, including various groups and all types of training programs.

METHODS

The PubMed and EMBASE databases were searched from their respective inceptions until October 2013. Search terms included subject headings and text words based on the concepts of 'exercise' or 'physical' and 'wheelchair' with the following constraints: English language and human. Inclusion criteria were: standardized and clearly described exercise training, and at least one outcome measure regarding endurance or sprint wheelchair propulsion capacity. Articles were not selected on type or mode of exercise; training could be either with or without wheelchair. Titles and abstracts of the electronic searches were scrutinized by one author (MZ). References of included studies were screened to find articles that might have been missed. Included articles were read in full by two independent reviewers (MZ and OV), both trained in exercise physiology and rehabilitation. They recorded details of the study design, participants, intervention program, training type, outcome measures, results, and conclusions. Where key information was not reported, efforts were made to contact the authors to obtain further details.

Exercise training programs were divided into five predetermined categories: aerobic exercise training, subdivided into interval and endurance training; anaerobic exercise training; strength training; and mixed training. Aerobic exercise training was defined as training aimed at improving the function of the cardiorespiratory system.[14] There are roughly two ways of training the cardio-respiratory system: interval and endurance training. Interval training involves bursts of high-intensity work interspersed with periods of low-intensity exercise.[15] Endurance exercise training was defined as repetitive, aerobic exercise of large muscles at a constant intensity level for a prolonged period of time (>10 min).[14,16] Anaerobic exercise training refers to exercise that requires bursts of maximal intensity work over short periods of time (<30s).[14] Strength training was defined as a structured repetitive exercise, inducing an overload to increase strength, power, or muscular endurance.[14] Mixed training is a combination of interval, endurance, anaerobic, and/or strength training, or a training that does not fit the definition of a specific training type. All studies used different prescribed exercise intensities. Low intensity was defined as <40% heart rate reserve, <64% peak heart rate or maximal tolerated power, moderate intensity as 40–60% heart rate reserve, 64-76% peak heart rate or maximal tolerated power, and high intensity as >60% heart rate reserve, >76% peak heart rate or maximal tolerated power.[16]

Outcome measures were divided into endurance and sprint wheelchair propulsion capacity. Both are performance measures of wheelchair exercise including all parameters (Figure 2.1), recording for example power output, speed, or velocity. Endurance wheelchair propulsion capacity refers to the ability of the body to sustain prolonged wheelchair exercise, and reflects the cardiorespiratory system.[17] Sprint wheelchair propulsion capacity refers to the ability of the neuromuscular system to produce the greatest possible impulse over a given distance or time period up to 30 seconds.[18]

Methodological quality

The methodological quality of the studies was rated using the PEDro scale, based on the Delphi list developed by Verhagen et al.,[19] which consists of eight criteria for internal validity and two statistical criteria. Points were only awarded when a criterion was clearly satisfied and reported.

Evidence level

The ideal method to determine the efficacy of an intervention is through a randomized controlled trial, a design that ensures that any differences in outcome variables are indeed attributable to the exercise training. To evaluate how to interpret outcome variables of different studies, the levels of evidence were classified by means of a grading system developed by the Oxford Centre of Evidence-Based Medicine.[20] This classification system consists of five levels. Going from the highest to the lowest level of evidence, these are: 1) randomized controlled trial(s) (n>100), 2) randomized controlled trial, 3) cohort study, 4) clinical controlled trials or case series, and 5) case study.

RESULTS

Searching the PubMed and EMBASE databases in October 2013 initially resulted in 1158 articles. On the basis of title and abstract, we excluded 1114 studies. Another 23 studies turned out not to meet the inclusion criteria after the full text had been read. Reference screening of the included studies did not result in additional studies. A total of 21 studies were eventually included and divided in different training types (Figure 2.2). No studies were found on anaerobic training. The studies represented 249 individuals; 50% (n=126) diagnosed with spinal cord injury, 38% (n=94) were able-bodied participants and 12% (n=29) were patients diagnosed other than spinal cord injury. Other diagnoses included spina bifida (4%), cerebral palsy (2%), traumatic injury (3%), amputees (1%), polio (1%), and bilateral tarsal tunnel syndrome (1%).

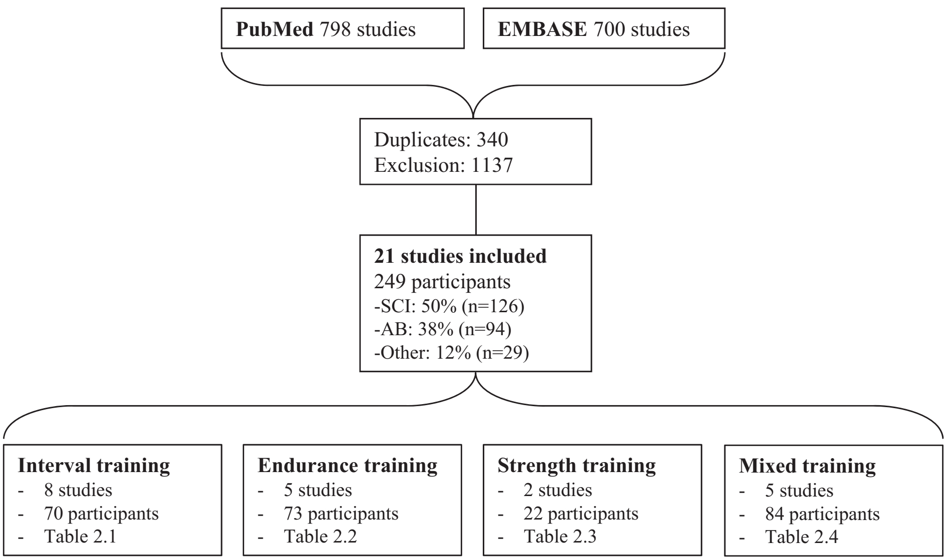

Figure 2.2: Flowchart for selection of eligible articles.
SCI, spinal cord injury; AB, able-bodied; other, participants having other diseases than spinal cord injury.

Interval training

Table 2.1 lists eight interval training studies,[21-28] of which three included a control group,[26-28] though none were randomized controlled trials. Four studies included participants with spinal cord injury,[21,22,24,25] and three included healthy ambulatory participants.[26-28] The number of interval sessions ranged from three to nine sets per training, high-intensity intervals ranged from high intensity to maximal, and low-intensity intervals from rest to moderate intensity. All training and test sessions were implemented on a wheelchair ergometer.

All studies measuring endurance wheelchair propulsion capacity found a significant improvement in the experimental group, ranging from 18–34% in participants with disabilities[21-25] and 49–66% in able-bodied participants.[26-28] No study reported sprint wheelchair propulsion capacity. All studies that measured mechanical efficiency[22,23,27] or peak oxygen uptake[21-23,25,26,28] reported significant improvements of 7–15% and 14–36%, respectively.

Endurance training

Table 2.2 lists the results of five studies examining the effects of endurance training.[29-33] Three studies included a control group,[29,32-34] one of them being a randomized controlled trial.[32] Three studies trained experienced wheelchair users, all diagnosed with spinal cord injury,[29-31] and four trained men only.[30-33] Training intensity ranged from 30 to 80% heart rate reserve or 75 to 85% peak heart rate, and exercise training was executed during arm crank exercise[30] or wheelchair propulsion on either a wheelchair treadmill,[32,33] wheelchair ergometer,[29] or indoor track.[31]

Three studies, of which 2 trained able-bodied participants, reported a significant positive effect on endurance outcome measures with an improvement of 30–78%.[30,32,33] Both studies training able-bodied participants found significant improvements in sprint capacity; 15% and 31% in 30-second power output.[32,33] Significant improvements in peak oxygen uptake of 94% and 10% were only found in two studies.[30,32] Mechanical efficiency was measured in four studies,[29,31-33] only one of which, with able-bodied participants, found a significant increase of 20%.[33]

Strength training

Two strength training studies were included (Table 2.3).[35,36] No randomized controlled trial was performed. The studies differed in training duration, frequency, strength exercises and participants: children with cerebral palsy and spina bifida[36] compared to male adults with and without spinal cord injury.[35]

Table 2.1: Interval training

	Study			Subjects				Intervention program				Results: Wheelchair propulsion capacity		Results: Other parameters
		Evidence level	PEDro score	n	Age (mean±SD)	M/F	Participants	Duration	Frequency	Modality	Training intensity	Endurance	Sprint	
Spinal cord injured	Bougenot et al. 2003[21]	IV Case series	2/10	7	35±13	7/0	SCI	6 weeks	3x p/w 45 min	WERG	9 sets: - 4 min at VT - 1 min at MTP	**+19.5% MTP (W)**[*,1] WERG	-	**+15.5% VO_2peak (ml/kg/min)**[*,1]
	Tordi et al. 200122	IV Case series	2/10	5	27±8	5/0	SCI	4 weeks	3x p/w 30 min	WERG	6 sets: - 4 min at 50% MTP - 1 min at 80% MTP	**+22.2% MTP (W)**[*,1] WERG	-	**+14.3% VO_2peak (ml/kg/min)**[*,1] **+9.3% ME *, #**
	Yim et al. 1993[24]	IV Case series	2/10	11	31±8	11/0	SCI	5 weeks	3x p/w 45 min	WERG	3 sets: - 10 min 80% HR_{peak} - 5 min rest	**-17.8% 100-meter time (s)**[*,1] WERG	-	**+39.5%, +24.4% total work of shoulder flexors**[*,1] **and extensors**[*,1] **(ft-lbs)** +15.5%, +14.5% total work of elbow flexors and extensors (ft-lbs)
	Le Foll-de Moro et al. 2005[25]	IV Case series	1/10	6	29±14	5/1	SCI	6 weeks	3x p/w 30 min	WERG	6 sets: - 4 min 50% MTP - 1 min 80% MTP	**+34.3% MTP (W)**[*,1] WERG	-	**+36.0% VO_2peak (l/min)**[*,1]

Other patients	Miles et al. 1982[23]	IV Case series	2/10	8	25±4	8/0	SCI, CP, polio, (traumatic) injury	6 weeks	3x p/w 45 min	WERG	2 sets: - 10 min 80% HRR - 5 min rest 4 sets: - 2 min 95% HRR - 2 min rest	**+30.7% MTP (W)*,1** WERG	-	**+26.0% VO_2peak (l/min)*,1** **+6.8% ME #,*,1**
Able-bodied subjects	Tordi et al. 1998[26]	IV CCT	5/10	5	27±11	5/0	AB	6 weeks	3x p/w 45 min	WERG	9 sets: - 4 min at VT - 1 min at MTP	**+66.0% MTP (W)**,2** WERG	-	**+35.0% VO_2peak (ml/min)**,2**
				5	22±3	5/0	AB	Control	Control		No training	0.0% MTP (W) WERG		0.0% VO_2peak (ml/min)
	Glaser et al. 1981[27]	IV CCT	3/10	7	21±3	0/7	AB	5 weeks	2x p/w 22 min	WERG	3 sets: - 4 min 80% HR_{peak} - 2 min relief period	**+48.5% training power output (kpm)*,1** WERG	-	+14.6% ME #,*,1
				6	22±2	0/6	AB	Control	Control		No training	-		+2.7% ME #
	Tordi et al. 2001[28]	IV CCT	5/10	5	23±7	5/0	AB	6 weeks	3x p/w 45 min	WERG	9 sets: - 4 min at VT - 1 min at MTP	**+63.6% MTP (W)**,1** WERG	-	**+29.3% VO_2peak (ml/kg/min)**,1**
				5	23±7	5/0	AB	Control	Control		No training	0.0% MTP (W) WERG	-	0.0% VO_2peak (ml/kg/min)

n, number of participants; SD, standard deviation; M/F, male/female; CCT, clinical controlled trial; SCI, spinal cord injury; CP, cerebral palsy; PP, post polio; AB, able-bodied; WERG, wheelchair ergometer; VT, ventilatory threshold; MTP, maximal tolerated power; HRR, heart rate reserve; HR, heart rate; VO_2, oxygen uptake; ME, mechanical efficiency. Bold, significant difference, *(*p*<.05), **(*p*<.01); [1] Significance level compared with baseline; [2] Significance level compared with control group; # ME is calculated with the formula: 'submaximal power output'/'submaximal oxygen uptake'. The calculated difference is the averaged relative improvement of ME.

Table 2.2: Endurance training

	Study			Subjects				Intervention program				Results: Wheelchair propulsion capacity		Results: Other parameters
		Evidence level	PEDro score	n	Age (mean±SD)	M/F	Partici-pants	Duration	Frequency	Modality	Training intensity	Endurance	Sprint	
Spinal cord injured	Hooker et al. 1989[29]	IV CCT	4/10	6	31±4	3/3	SCI	8 weeks	3x p/w 20 min	WERG	Moderate intensity 50–60% HRR	+24.0% MTP (W)[1] WERG	-	+10.3% VO_2peak (ml/kg/min)[1] +4.1% ME #,[1]
				5	30±5	3/2	SCI	8 weeks	3x p/w 20 min	WERG	High intensity 70–80% HRR	+13.3% MTP (W)[1] WERG	-	+12.0% VO_2peak (ml/kg/min)[1] +5.3% ME #
	DiCarlo et al. 1988[30]	IV case series	2/10	8	24±4	8/0	SCI	8 weeks	3x p/w 15–35 min	Arm cranking	Moderate intensity 50–60% HRR	**+78.0% 12-minute wheelchair test (km)*,[1]** FIELD	-	**+94.2% VO_2peak (ml/kg/min)*,[1]**
	Whiting et al. 1983[31]	V case study	1/10	2	21&33	2/0	SCI	8 weeks	3x p/w 20 min	indoor track	High intensity 75–85% HR_{peak}	+33.3% MTP (kg·m/min)[1] WERG	-	+8.6% VO_2peak (l/min)[1] +31.2% ME #,[1]

Able-bodied subjects	Van der Woude et al. 1999[32]	II RCT	5/10	9	23±3	9/0	AB	7 weeks	3x p/w 30 min	WT	Moderate intensity 50% HRR	**+29.8% MTP (W)****,[2] WERG	**+16.4% P30 (W)****,[2] WERG	+5.0% VO_2peak (l/min)[2] +13.4% ME[2] +11.9% $F_{iso\text{-}stroke}$ (N)[2]
				10	23±3	10/0	AB	7 weeks	3x p/w 30 min	WT	High intensity 70% HRR	**+42.2% MTP (W)****,[2] WERG	**+15.2% P30 (W)****,[2] WERG	**+9.7% VO_2peak (l/min)***,[2] +19.1% ME[2] +14.0% $F_{iso\text{-}stroke}$ (N)[2]
				8	22±2	8/0	AB	Control	Control		No training	-0.9% MTP (W) WERG	+5.3% P30 (W) WERG	-3.4% VO_2peak (l/min) +9.9% ME -0.3% $F_{iso\text{-}stroke}$ (N)
	Van den Berg et al. 2010[33]	IV CCT	4/10	10	23±2	10/0	AB	7 weeks	3x p/w 30 min	WT	Low intensity 30% HRR	**+34.0% MTP (W)****,[2] WERG	**+31.0% P30 (W)****,[2] WERG	-2.2% VO_2peak (ml//kg/min)[2] **+20.2% ME****,[2] +4.9% $F_{iso\text{-}stroke}$ (N) [2]
				15	23±2	15/0	AB	Control	Control		No training	+0.2% MTP (W) WERG	+7.9% P30 WERG	+6.3% VO_2peak (ml/kg/min) +0.6% ME +5.3% $F_{iso\text{-}stroke}$ (N)

n, number of participants; SD, standard deviation; M/F, male/female; CCT, clinical controlled trial; RCT, randomized controlled trial; SCI, spinal cord injury; AB, able-bodied; WERG, wheelchair ergometer; WT, wheelchair on treadmill; HRR, heart rate reserve; HR, heart rate; MTP, maximal tolerated power; VO_2, oxygen uptake; ME, mechanical efficiency; P30, peak anaerobic output (30 seconds).
Bold, significant difference, *(*p*<.05), **(*p*<.01); [1] Significance level compared with baseline; [2] Significance level compared with control group; # ME is calculated with the formula: 'submaximal power output'/'submaximal oxygen uptake'. The calculated difference is the averaged relative improvement of ME.

Table 2.3: Strength training

Study			Subjects				Intervention program				Results: Wheelchair propulsion capacity		Results: Other parameters
	Evidence level	PEDro score	n	Age (mean±SD)	M/F	Partici-pants	Duration	Frequency	Modality	Training intensity	Endurance	Sprint	
O'Connell et al. 1995[36]	IV case series	2/10	6	10±5	NS	CP, SB	9 weeks	3x p/w 30 min	Circuit training	3 sets - 8 upper-body exercises– 6-RM - 30 seconds rest	**+29.0% 12-minute wheelchair test (m)*,1** FIELD	-20.2% 50-meter sprint time (s)[1] FIELD	**+181.8% on average in all 8 exercises (kg)*,1**
Turbanski et al. 2010[35]	IV CCT	3/10	8	33±11	8/0	SCI	8 weeks	2x p/w	Bench press	5 sets - 10–12x 80% 1-RM - 3–5 minutes rest	-	-1.8% 10-meter sprint time (s)[1] FIELD	**+66.5% SE (reps)**,1** **+30.8% static F_{max} (N)**,1** **+39.3% dynamic 1RM (kg)**,1**
			8	25±2	8/0	AB	8 weeks	2x p/w	Bench press	5 sets - 10–12x 80% 1-RM - 3–5 minutes rest	-	-	**+56.5% SE (reps)**,1** **+12.8% static F_{max} (N)**,1** **+15.3%dynamic 1RM (kg)**,1**

n, number of participants; SD, standard deviation; M/F, male/female; CCT, clinical controlled trial; NS, not specified; CP, cerebral palsy; SB, spina bifida; SCI, spinal cord injury; AB, able-bodied; RM, repetition maximum; FIELD, field test; SE, strength endurance.
Bold, significant difference, *($p<.05$), **($p<.01$); [1] Significance level compared with baseline.

A significant improvement was found in endurance capacity during a field test.[36] Both studies reported sprint wheelchair propulsion capacity, but strength training did not improve sprint performance. However, a significant improvement was found in the strength exercises that were trained.[35,36]

Mixed training

Table 2.4 lists the characteristics of six mixed training studies.[37-42] Three studies included a control group,[39,41,42] one of them being a randomized controlled trial.[41] Participants had a variety of diagnoses: spinal cord injury, spina bifida, trauma, polio, and cerebral palsy. With one exception,[38] all studies trained experienced wheelchair users or even wheelchair athletes.[41,42] Three exercise programs consisted of a combination of strength and endurance training,[37-39] one study used wheelchair exercise to exhaustion,[40] and two studies investigated respiratory muscle training.[41,42] Exercise training programs lasted between six weeks and 10 months and involved one to 14 training sessions each week.

Endurance wheelchair propulsion capacity improved significantly, by 30–53% in terms of endurance time[39,40] and by 15% in terms of maximal tolerated power.[37] A case study reported an increase of 63% in 10-km time trial.[38] One study reported a significant improvement in sub-maximal endurance time, but found no increase in maximal tolerated power.[39] No difference was found in sprint capacity.[42]

Methodological quality

Tables 2.1–2.4 report the methodological quality as assessed with the PEDro scale. The median score was two out of 10, and no study scored more than six points, indicating generally poor methodological quality. Only two out of 21 studies were randomized controlled trials,[32,41] but these studies could not ensure concealed allocation and blinding of subjects, therapists or assessors. Most studies had positive scores on criteria eight and 11, indicating that more than 85% of the subjects completed the intervention and the effect of treatment was measured.

Evidence level

Two out of 21 studies obtained their results at the second highest level of evidence that of small sample size randomized controlled trial.[32,41] Another five studies were controlled clinical trials.[26-28,33,42] The majority of studies (57%, n=12) consisted of observational studies comparing post-training results with baseline values, and two articles reported on case studies.

Table 2.4: Mixed training

Study			Subjects				Intervention program				Results: Wheelchair propulsion capacity (WPC)		WPC parameters
	Evidence level	PEDro score	n	Age (mean±SD)	M/F	Partici-pants	Duration	Frequency	Modality	Training intensity	Endurance	Sprint	
Rodgers et al. 2001[37]	IV case series	2/10	19	44±11	16/3	SCI, SB, MT	6 weeks	3x p/w 60 min	Strength, stretch-ing, rowing	5 sets - 5 upper-body exercises - 75% max Rowing (30 min 60% HRR)	+14.6% MTP (W)*,[1] WERG	-	+7.6% VO_{2peak} (ml/min)[1] +6.7% ME[1] -2.9% $F_{dynamic\text{-}stroke}$ (N)[1] **+123.3% on average in all exercises (pounds)**,[1]**
Lakomy et al. 1996[38]	V case study	1/10	1	42	1/0	AB	10 months	6x p/w 1/2 a day	Strength, WT, swim-ming	Weight training, wheelchair propulsion, swimming	+63.0% 10 km time trial speed (m/s)[1] FIELD (road)	-	+12.9% VO_{2peak} (l/min)[1] +20.1% ME (VO_2 at constant speed)[1]
Keyser et al. 2003[39]	IV case series	2/10	20	43±10	15/4	SCI, SB, CP (no upper limb im-pairment)	12 weeks	3x p/w 25 min	Elastic bands	8–12x resisted stroke pushes 20 min wheelchair propulsion exercise with elastic bands	+0.0% MTP (W)[1] WERG **+30.0% endurance time of 60% MTP (min)*,[1]** WERG	-	-1.0% VO_{2peak} (ml/min)[1] +1.8% ME[1]
			7	36±7	5/2	(upper limb im-pairment)	12 weeks	3x p/w 25 min	Elastics bands	8–12x resisted stroke pushes 20 min wheelchair propulsion exercise with elastic bands	+14.7% MTP (W)[1] WERG +52.9% endurance time of 60% MTP (min)[1] WERG	-	+0.6% VO_{2peak} (ml/min)[1] -7.0% ME[1]

Gass et al. 1980[40]	IV case series	1/10	9	34±11	NS	SCI	7 weeks	5x p/w	WT	5 min 2.5 km/h every 5 min increase in slope to depletion	**+52.6% maximal exercise test time (s)*,**[1] WT	-	**+33.7% VO_{2peak} (ml/ /kg/min)**,**[1]
Mueller et al. 2008[41]	II RCT	6/10	6	29±11	4/2	athletes (SCI)	6 weeks	5x p/w 30 min	Respira-tory training	Respiratory muscle endurance test 65–75% MVV	-11.1% 10 km time trial (min)[2] WT	-	-2.2% VO_{2peak} (ml/ kg/min)[2] **+338.5% respiratory muscle endurance (min)*,**[2]
			6	28±10	5/1	athletes (SCI)	6 weeks	Control		No training	-0.4% 10 km time trial (min) WT	-	-2.6% VO_{2peak} (ml/ kg/min) +53.5% respiratory muscle endurance (min)
Goosey-Tolfrey et al. 2010[42]	IV case series	5/10	8	28±5	4/4	athletes (SCI, SB, polio)	6 weeks	2x a day	Respira-tory training	30 dynamic inspiratory efforts 50% MIP	-	-0.8% fifteen 20-meter sprint time (s)[2] FIELD	-
			8	30±11	4/4	athletes (SCI, SB, polio)	6 weeks	1x a day	Respira-tory training	60 slow breaths 15% MIP	-	+0.7% fifteen 20-meter sprint time (s) FIELD	-

n, number of participants; SD, standard deviation; M/F, male/female; RCT, randomized controlled trial; NS, not specified; SCI, spinal cord injury; SB, spina bifida; MT, multi trauma; AB, able-bodied; CP, cerebral palsy; HRR, heart rate reserve; VO_2, oxygen uptake; ME, mechanical efficiency; MTP, maximal tolerated power; WERG, wheelchair ergometer; FIELD, field test; WT, wheelchair on treadmill; MVV, maximal voluntary ventilation; MIP, maximum inspiratory pressure.
Bold, significant difference, *(p<.05), **(p<.01); [1] Significance level compared with baseline; [2] Significance level compared with control group; # ME is calculated with the formula: 'submaximal power output'/'submaximal oxygen uptake'. The calculated difference is the averaged relative improvement of ME.

DISCUSSION

Although the evidence base is weak, the present review does support exercise training programs as being beneficial for improving wheelchair propulsion capacity. The vast majority of participants involved had a spinal cord injury or were able-bodied individuals. It is surprising that there were only two proper randomized controlled trials.[32,41] Furthermore, only five out of 21 studies reported between-group differences.[26,32,33,41,42] The majority of studies were case series or even case studies, indicating changes over time that can be due to any factor, such as exposure to the test protocol, natural recovery, season or learning effects.[43] Studies without between-group differences do not provide sufficient evidence for the effectiveness of exercise training.

Arm cranking, an outcome measure used in many studies, requires a continuous pushing and pulling movement instead of the intermittent propelling pattern of wheelchair propulsion. Moreover, arm cranking exercise has a higher peak power output and mechanical efficiency.[44,45] Considering the concept of specificity of exercise, wheelchair propulsion exercise is more appropriate. It improves the capability in using a manual wheelchair, as shown in this review by improvements in mechanical efficiency. The focus in the current review was specifically on wheelchair propulsion, being the primary means of mobility in wheelchair users. So, studies that used arm cranking as an outcome measure were not included. Twenty studies included in the review by Valent et al.[13] were therefore not included in current review. The other focus of current review, including not only individuals with spinal cord injury, resulted in 12% of participants involved having another diagnosis and 38% able-bodied participants. This does not represent the wheelchair using population, and therefore more research is needed.

The intervention programs in the included studies varied in terms of duration, frequency, and intensity. The American College of Sports Medicine (ACSM) guidelines for wheelchair users with paraplegia recommend 3–5 exercise sessions a week lasting 20–60 minutes each, at moderate-to-vigorous exercise intensities, to increase aerobic capacity,[46] the most important physiological parameter. This suggests target heart rates between 50–80% of peak heart rate.[46] Therefore, strength training programs fall out of the scope in this comparison. Except for both respiratory muscle training programs,[41,42] all interventions met the ACSM guidelines for increasing aerobic capacity. The ACSM guidelines for wheelchair users only account for the physical fitness parameters. Therefore it seems reasonable to assume that respiratory muscle training was not effective in improving wheelchair propulsion capacity, since physiological parameters or mechanical efficiency during wheelchair propulsion were not trained.

Inclusion of all types and modes of training programs, as well as all diagnoses provided a nice overview, but did not result in potential training guidelines. However, despite our finding that no randomized controlled trial had been performed for interval training, this type of training seems to have the highest potential to improve endurance wheelchair propulsion capacity. All participants in seven intervention studies improved significantly in terms of endurance capacity and peak oxygen uptake, whereas the participants in endurance and mixed training programs showed improvements in endurance capacity and peak oxygen uptake in only two out of five and one out of five studies, respectively. This cautious conclusion supports the findings of Helgerud et al.[47] and Gibala et al.,[48] who found that high-intensity interval training is significantly more effective in improving peak oxygen uptake than performing the same (or more) total work during continuous aerobic exercise training. However, more research is needed to find out whether this also holds for wheelchair exercise.

Assuming that exercise intensity is the most important factor in improving aerobic capacity in healthy subjects, rather than exercise frequency and duration,[17] it is remarkable that studies training at moderate[29] or high[29,31] intensity found no significant improvements, while a study training at low intensity[33] did. The positive training effect was found in able-bodied participants. Despite that this group had similar peak power outputs compared to individuals with paraplegia, this suggest that training effects are achieved at lower intensities in able-bodied individuals.[49] Able-bodied participants gained the most effect at the start of the intervention in improving mechanical efficiency.[50] In contrast, despite of the similar training guidelines,[16] and baseline measurements[49] the effect of interval and endurance training in individuals with disabilities was lower compared to able-bodied participants. The limiting factor for improving peak oxygen uptake in individuals with a disability is generally a lack of active muscle mass.[51] Resistance training or electrical induced resistance training can increase muscle mass[52,53] and might have a positive effect on peak oxygen uptake.

Only six out of 21 studies investigated sprint wheelchair propulsion capacity. Significant improvements in sprint capacity in able-bodied participants may again represent an overestimation of the effects in experienced wheelchair users.[32,33] The improved strength in single- and multiple-joint exercises after strength training was not translated into an improvement in sprint capacity.[35,36] Although there is a correlation between muscle strength and the force imparted to the hand rim, increased force does not necessarily lead to more effective hand rim force or propulsion cadence.[54] This suggests that improving not only muscle strength, but also propulsion technique is important. Future research should use exercise programs incorporating both muscle strength exercises specifically designed to improve hand-rim propulsion and functional, effective propulsion technique training.

In wheelchair users with a spinal cord injury, aerobic capacity accounts for 69% of maximal tolerated power during wheelchair propulsion.[55] This makes peak oxygen uptake an important physical fitness parameter. All studies in which peak oxygen uptake improved significantly also found improved endurance capacity. The same pattern holds for mechanical efficiency; when mechanical efficiency improved significantly, endurance capacity improved as well. A highly significant relation between mechanical efficiency and maximal tolerated power supports this finding.[56] Both peak oxygen uptake and mechanical efficiency are good predictive physical fitness parameters for wheelchair propulsion capacity. In the study by DiCarlo et al.[30] peak oxygen uptake values improved by no less than 94.2%, from 12.1 to 23.5 mL O_2/kg/min. Baseline measurements indicated very low aerobic capacities, and the very large effect may be explained by the relative notation of peak oxygen uptake in combination with increased muscle mass and weight loss.

Our findings must be interpreted in light of certain limitations. Firstly, the search might have missed some studies. We have looked at outcome measures specifically performed during hand rim wheelchair propulsion and therefore training studies with outcome measures on endurance or sprint capacity during hand cycling have been missed. Secondly, there have been no studies on this topic with a high level of evidence and with good methodological quality. Without between-group analyses, changes over time can be due to any factor.[43] Moreover, in view of the low levels of evidence, low methodological quality and heterogeneity of both study sample and interventions, it was inappropriate to pool the data of the studies included. Thirdly, a plethora of different not equally divided diagnoses makes it difficult to interpret the results. Several studies were even conducted among healthy ambulatory subjects. Hence, further investigations are required to assess whether our results are generalizable to other diagnoses and medical conditions. Fourthly, the studies in our review predominantly included male participants. It is unclear whether the same effects would be observed in women. Fifthly, not all training types were equally represented. We could only include two studies on strength training, and no studies investigating the effects of anaerobic training. This is surprising, since for manual wheelchair users, most of the motor activities in daily life are of short duration[10] and produce a relatively high physical strain on the individual. Finally, there was only one training study that focused on children using a wheelchair. Although long-term effects of training are not known, one can only assume that an early introduction to wheelchair training would be beneficial for children's future health and function.

There is a need for randomized controlled trials involving manual wheelchair users, particularly including people who do not have spinal cord injury. Future research should also focus on the effects of interval training, since, based on the low-quality evidence, this seems to be the training type that has the greatest potential. In addition, there is a need for studies investigating the effects of exercise training in children using a wheelchair.

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3

Reliability and validity of short-term performance tests for wheelchair-using children and adolescents with cerebral palsy

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ABSTRACT

Aim: To investigate the test–retest reproducibility of the Muscle Power Sprint Test (MPST), the 10×5-m sprint test, and the arm-cranking Wingate Anaerobic Test (WAnT) in youth with cerebral palsy (CP). A secondary objective was to assess the construct validity of the MPST.

Method: Twenty-three participants with spastic CP (mean age 13.3, SD 3.6 years; 18 males, five females, two classified as having spastic unilateral CP, 21 as having spastic bilateral CP) using a manual wheelchair for at least part of the day were recruited and tested in different rehabilitation settings in The Netherlands. Participants were classified as in Gross Motor Function Classification System Expanded and Revised (GMFCS-E&R) level III and IV.

Results: Intra-class correlation coefficients (range, 0.93–0.99; range 95% CI, 0.82–1.0) for all variables indicated highly acceptable reproducibility. Limits of agreement analysis revealed satisfactory levels of agreement. The MPST variables demonstrated very strong significant positive correlations for peak power and mean power from both tests (peak power: r=0.91, p<.001; mean power: r=0.88, p<.001).

Interpretation: The MPST, the 10×5-m sprint test and the arm-cranking WAnT are reproducible tests for measuring anaerobic performance and agility in adolescents with spastic CP who self-propel a manual wheelchair. The MPST has been shown to be a valid test to measure anaerobic performance in this population.

What this paper adds

- Two inexpensive and easy to administer short-term performance wheelchair tests.
- A reproducible and valid anaerobic exercise test for youth with CP who self-propel a wheelchair.

INTRODUCTION

Anaerobic performance and agility are important physiological factors that play a critical role in the ability of a child or adolescent with cerebral palsy (CP) to participate in daily activities.[1] This is especially true for youth with CP who use a wheelchair for mobility, because most motor activities of daily living (ADL), including playing in the playground, moving around in the classroom, but also climbing a curb and ascending a ramp, are of short duration.[2] These short bouts of activity rely primarily on the energy produced by the ATP-PCr (or alactic) component of the anaerobic system and produce a relatively high physical stress on the individual.

Exercise testing over time can provide a quantitative assessment of the improvement or decline in the anaerobic performance of children and adolescents with CP and has the potential to be an important measurement tool in clinical practice as well as in research. For children with CP who are able to walk or run independently, running-based field tests, like the Muscle Power Sprint Test (MPST) and the 10×5-m sprint test, are currently used in clinical practice and are inexpensive measures that do not require special equipment or training.[3] The MPST has been developed with the specific goal of examining anaerobic performance,[3] while the 10×5-m sprint test is a marker of agility.[3] These tests, however, have not yet been examined in youth with CP who use wheelchairs.

Anaerobic performance in children with neuromuscular disorders such as CP is often assessed in the laboratory setting using the cycling Wingate Anaerobic Test (WAnT).[4] While much of the focus to date has revolved around lower-limb-based anaerobic performance, the upper-body or arm-cranking WAnT has been used to predict functional anaerobic performance in children with CP.[5] Tirosh et al.[6] reported that the arm-cranking WAnT was both feasible and reliable in a group of children with CP. This study, conducted over two decades ago, relied on the use of a mechanically braked ergometer with custom-made software calculating mechanical power based on the braking force applied as well as participant revolutions per minute, averaged over a 3- or 5-second interval. Today, the most commonly used and widely available ergometers are electro-magnetically braked, with the ability to calculate instantaneous mechanical power output with a higher frequency and precision. Given the potential utility of the arm-cranking WAnT in both clinical settings for patient follow-ups as well as outcome measure in clinical trials, an assessment of the reproducibility of this test in youth with CP using newly available and more commonly used technology seems warranted.

While the arm-cranking WAnT may be considered the criterion standard for assessing anaerobic performance, it must be noted that the results of the arm-cranking WAnT are not solely a reflection of the ATP-PCr (alactic) system (primarily responsible for supporting short bouts of activity) but also the anaerobic lactic and aerobic systems, each of which have been estimated to contribute to 28%, 60% and 11%, respectively, in an upper body WAnT.[7] Furthermore, perhaps the greatest limitation of the arm-cranking WAnT is the fact that its application requires the use of sophisticated and costly technical equipment and software, ultimately rendering it impractical for use in a field setting. It is, however, equally important to note that while a field test may be more practical, there is a shortage of options for field exercise testing to specifically examine agility and/or anaerobic performance in youth with CP who rely on a manually propelled wheelchair for locomotion.[8] Ideally, this test should be safe, inexpensive, easy to administer in a non-academic setting, and relatively quick, allowing patients to be assessed in a short time-frame. Moreover, much like laboratory-based tests, reproducibility and validity of field exercise tests are important issues for clinical outcomes. Since the 10×5-m sprint test measures agility and not anaerobic performance, validity of this test cannot be examined using the arm-cranking WAnT.

Therefore, the objective of the present study was to investigate the test–retest reproducibility of: (1) the MPST, (2) the 10×5-m sprint test, and (3) the arm-cranking WAnT in a sample of adolescents with CP who rely on a manually propelled wheelchair for locomotion. A secondary objective was to assess the construct validity of the MPST, where we hypothesised a significant positive correlation between MPST and arm-cranking WAnT power outcomes.

METHOD

Participants

This study focused on children with CP between the ages of 7 years and 18 years who were diagnosed with spastic CP and classified in Gross Motor Function Classification System Expanded and Revised (GMFCS-E&R)[9] level III and IV. All participants were further required to self-propel a manual wheelchair for at least a part of the day and be capable of following simple instructions.

All participants were receiving rehabilitation services in the Netherlands at the time of participation. Participant characteristics are provided in Table 3.1. A total of 23 children with CP and their participants in this study, which was approved by the Institutional Ethics Committee of the University Medical Center, Utrecht.

Table 3.1: Participant characteristics

	Mean	SD
Age (years)	13.3	3.6
Height (cm)	149.6	14.3
Height-for-age SDS	-1.48	1.07
Body mass (kg)	42.3	13.1
Body mass-for-age SDS	1.05	1.03
BMI (kg/m^2)	18.5	4.0
BMI-for-age SDS	-0.47	1.96
GMFCS		*n*
Level III		3
Level IV		20
Spastic unilateral		2
Spastic bilateral		21

Age, body height, body mass and BMI data were means ±SDs. BMI, Body Mass Index; GMFCS, Gross Motor Function Classification System; SDS, Standard Deviation Score.[25]

Procedures

All participants attended a total of three testing sessions. During the first two sessions, participants performed both the MPST and the 10×5-m sprint test. During a separate third session, participants performed the arm-cranking WAnT twice. In all sessions participants were given at least 15 minutes of rest between the two tests performed. Participants and assessors were blind to the child's performance on each of the tests. All tests were performed within a period of 2 weeks in four rehabilitation centres across the Netherlands.

Before testing, each participant's body mass was determined using an electronic scale (Stimag, Hoofddorp, the Netherlands), which was also used to determine the weight of the participant's wheelchair. Standing height was assessed, and arm span (fingertip to fingertip, with arms abducted 90° and elbow and wrists straight) was measured as a surrogate for standing height when the participant was unable to adopt a vertical position. Functional mobility was quantified using the Functional Mobility Scale (FMS).[10]

To assess the test–retest reliability of the MPST and the 10×5-m sprint test, the participant performed the same test at the same time of the day within 2 weeks, with both tests administered by the same two assessors. During each test, participants were verbally encouraged to propel the wheelchair as fast as they could.

Measures

GMFCS-E&R

A paediatric physical therapist (OV) experienced in using the GMFCS-E&R[9] used the translated Dutch version to classify the children and adolescents with CP according to their functional ability.

Functional Mobility Scale

The FMS, a reliable and valid instrument, is a 6-point ordinal scale that quantifies mobility according to the need for assistive devices at three specific distances: 5m, 50m and 500m.[11] These distances represent home, school and community environments, respectively. The FMS is administered by asking the child or parent a few questions to indicate if the child used a wheelchair (score 1) or was ambulatory with or without assistive devices (score 2–6) for every distance.

Muscle Power Sprint Test and 10×5-m sprint test

For both the MPST and 10×5-m sprint test, participants used their own wheelchair and back support, with no adjustments made to wheelchair configuration during the test period.

Muscle Power Sprint Test (anaerobic performance)

Peak power and Mean power (Watts) were calculated and used as markers of anaerobic performance in the MPST, which was performed as previously described.[3] 'Mean power' refers to the ability of the neuromuscular system to produce the greatest possible impulse in a given time period. 'Peak power' was defined as the highest mechanical power that can be delivered during exercise of up to 30 seconds' duration.[12]

Before executing the test, each participant performed the test at a slow speed, which served as both a warm-up, as well as a habituation for the participant to ensure that he/she understood how to perform the test. The warm-up was followed by a three-minute rest period. The participant was then asked to complete six 15m runs at maximum pace. The 15m distance was marked by two lines taped to the floor. Cones were placed at the end of each of the lines. The participant was instructed to propel the wheelchair as fast as possible from one line to the other, and to be sure to cross each line with all wheels of their chair. Between each run, the participant was given a 10-second period to turn around and prepare for the following sprint. Power output for each sprint was calculated using total mass (body mass and wheelchair weight) and propelling times, where:

$$\text{Power} = (\text{total mass} \times \text{distance}^2)/\text{time}^3.$$

Power was calculated for each of the six sprints. Peak power was defined as the highest calculated power, while mean power was defined as average power over the six sprints. The MPST was administered by two experienced researchers (OV and MZ).

The total exercise time in the arm-cranking WAnT is 30 seconds. We expected that total exercise time for the MPST would be greater than 30 seconds. After performing the MPST for all participants, the number of consecutive sprints that result in a mean total exercise time closest to 30 seconds was determined.

10×5-m sprint test (agility)

Previous investigations into the 10×5-m sprint test with typically developing children have demonstrated good reliability; however, it has yet to be examined in children with CP who self-propel a wheelchair.[13,14] The 10×5-m sprint test is a continuous sprint test whereby the participant is asked to perform nine fast turns upon completion of every 5m distance. The participant is not given the opportunity to rest between each turn. This may be a problematic and/or extremely difficult task for some children with CP who experience difficulty with movement coordination. Thus, this test is not designed to measure the muscle power; rather, the time taken to complete the 10×5-m sprints is a good indicator of participant agility.

All tests were performed in a gymnasium. Participants were provided with a practice session prior to test completion wherein they performed the test at a slow speed to ensure a proper understanding of the instructions. After a 3-minute rest period, participants were given the verbal cues of 'Ready? 3, 2, 1, go!' and were instructed to complete 10 runs of 5m at a maximum pace. The 5m distance was marked by two taped lines on the floor and by cones. The participant had to propel themselves as fast as possible to each line, had to place all wheels across each line, make a turn and sprint back as fast as possible, with no rest between the sprints. At the end of the 10th sprint, participants had to cross the finish line. The assessors recorded time to completion to a 10th of a second for the total 50m (10×5-m) using a hand-held stopwatch.

The arm-cranking Wingate Anaerobic Test (anaerobic performance)

The arm-cranking WAnT was performed on an electromagnetically braked cycle ergometer (Lode Angio, Procare BV, Groningen, the Netherlands). The ergometer was fixed to the floor to prevent any ergometer movement during arm-cranking. Participants sat in a chair (also fixed to the floor) and were asked to remain seated throughout the test. Seat height and backrest angle were adjusted such that the elbow joint was almost in full extension

(165–175°) and the shoulders were in line with the center of the ergometers shaft when the participant's hands were grasping the handles (synchronously) with the crank horizontally positioned away from the body. A braking force of 0.26 Nm/kg was used primarily based on our pilot work, as well as previously published literature.[10,15] The lowest braking force recommended for leg cycling is 0.53 Nm/kg. Because less active muscle mass is involved in the arm-cranking WAnT compared to the cycling WAnT, braking force was reduced, with our pilot testing suggesting a reduction of 50% as most appropriate for our age range. The arm-cranking WAnT was administered by two experienced assessors (OV and MZ).

The arm-cranking WanT protocol consisted of three parts: (1) warm-up – the child had to cycle at a comfortable pace for 2 minutes without a braking force; (2) arm-cranking WAnT – participants were given a 5-second countdown before the braking force was applied and were instructed to crank the handles as fast as possible over a 30-second period. All participants were verbally encouraged to maintain the highest possible cadence throughout the arm-cranking WAnT. The braking force was applied immediately at the start of the test (default: 0.26 × body weight in Nm); (3) recovery – once the arm-cranking WAnT was completed, participants were given a chance to cycle at their own pace for as long as they desired with a braking force of 20W.

There are two primary markers of performance on the arm-cranking WAnT: peak power and mean power. With the fully computerised Lode Ergometry Manager Software (LEM; Procare BV, Groningen, the Netherlands), instantaneous power values can be obtained. Specifically, peak power is defined as the highest mechanical power (Watts) achieved at any stage of the test; it represents the explosive characteristics muscle power and is closest to a person's 'real' maximal mechanical power. Mean power represents the average local muscle endurance over the entire 30 seconds of the arm-cranking WAnT.

Data analysis

The sample size for this study was determined by the most demanding hypothesis to examine the reproducibility for the MPST and the 10×5-m sprint test. Sample size was calculated based on data from children with CP in GMFCS level III who had performed the 7.5-m shuttle run test,[16] with a β of 0.9 and an α of 0.05. This resulted in a required sample size of 23 participants. Based on Donner and Eliasziw,[17] with 23 participants, we had a power value of >0.80 to detect an intraclass correlation coefficient (ICC) >0.80, with statistical significance of 0.05.

Reproducibility

Reproducibility encompasses both reliability and agreement.[18] For reliability the ICCs (two-way mixed) were used. An ICC >0.80 reflects excellent reliability, while ICCs from 0.70 to 0.79 reflect good reliability.[19] The recommended minimum for the lower bound of the 95% CI was 0.85.[20] The Bland–Altman procedure was used to check for heteroscedasticity of the test and retest of the MPST, the 10×5-m sprint test and the arm-cranking WAnT.[21] Furthermore, the consistency of measurements was verified graphically using the method of Bland and Altman.[21] This method plots differences between two measurements against the average of the two measurements. Size and range of differences, scoring distribution and possible measurement bias can be visually assessed. The Bland and Altman Limits of Agreement (LoA)[21] were used to evaluate the level of agreement between test and retest. The LoA define the limits within which 95% of the differences are expected to fall (mean 1.96 SD of the differences).

Construct validity

The association between the results of the MPST and the arm-cranking WAnT was tested using Pearson's correlation coefficients. An α of <0.05 (two-tailed) was considered as statistically significant.[19]

RESULTS

A total of 18 males and five females (mean age of 13.3 (3.6) years, range 7–18 years) completed all tests without complications. Twenty-one children were bimanually functional and two children used one arm to propel and one arm to steer their wheelchair using a steering wheel. The FMS data show that 11, 14 and 23 children used their wheelchair at home, school and community environments, respectively. Mean total exercise time for the MPST was 72.7 (40.5) seconds. The number of sprints completed within approximately 30 seconds was three (mean exercise time 36.4 (30.6) seconds). Therefore, the MPST for children who self-propel a wheelchair consists of three sprints of 15m.

There were no significant differences between both subgroups (males vs females, GMFCS levels and age). As such, the following results include both sexes and GMFCS levels combined. The physiological variables measured on both exercise tests are provided in Table 3.2.

Table 3.2: Results of the Muscle Power Sprint Test, arm-cranking Wingate Anaerobic Test and 10×5-m sprint test

Variable	Mean	SD	Range
Anaerobic performance			
MPST PP (Watts)	21.8	20.8	0.3–64.6
MPST MP (Watts)	19.7	19.4	0.3–62.0
WAnT PP (Watts)	85.6	56.9	15.2–206.2
WAnT MP (Watts)	43.8	36.8	3.3–115.7
Agility			
10×5-m sprint test (s)	74.0	38.1	39.8–186.5

MPST, Muscle Power Sprint Test; WAnT, arm-cranking Wingate Anaerobic Test; PP, Peak power; MP, Mean power.

Reproducibility

The test–retest reliability statistics of the MPST, the 10×5-m sprint test and the arm-cranking WAnT are presented in Table 3.3. As can be appreciated from the Bland–Altman plots (Figures 3.1–3.3), there were some obvious outliers. These outliers are included in the calculations. For the anaerobic performance tests, ICC values for mean power and peak power were 0.99 for both the MPST and arm-cranking WAnT. For the agility test (10×5-m sprint test) ICC values were 0.93.

Table 3.3: Test–retest reliability statistics

Variable	ICC	LOA	95% CI of ICC
Anaerobic performance			
MPST PP (Watts)	0.99	-39.9 to 59.7	0.98–1.0
MPST MP (Watts)	0.99	-7.1 to 6.4	0.98–1.0
WAnT PP (Watts)	0.99	-15.0 to 18.4	0.99–1.0
WAnT MP (Watts)	0.99	-11.3 to 10.7	0.99–1.0
Agility			
10×5-m sprint test (s)	0.93	-8.0 to 9.2	0.82–0.97

ICC, intra-class correlations; LOA, limits of agreement; CI, confidence interval; MPST, Muscle Power Sprint Test; WAnT, arm-cranking Wingate Anaerobic Test; PP, Peak power; MP, Mean power.

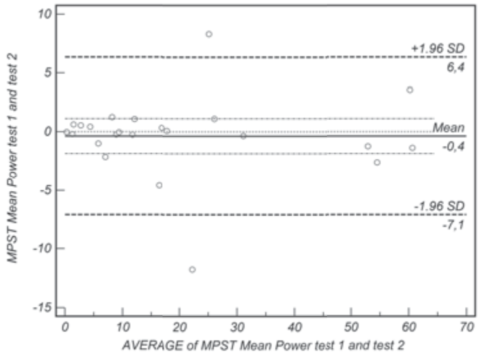

Figure 3.1: Bland–Altman plot of Mean power on test and retest on the Muscle Power Sprint Test.

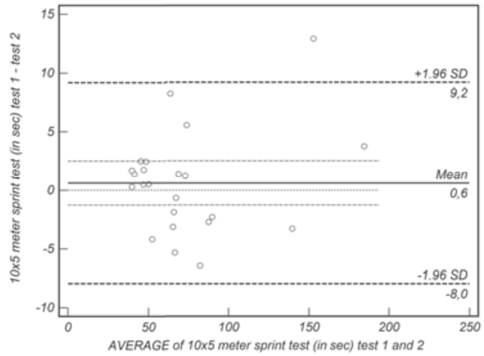

Figure 3.2: Bland–Altman plot of exercise time on test and retest on the 10×5-m sprint test.

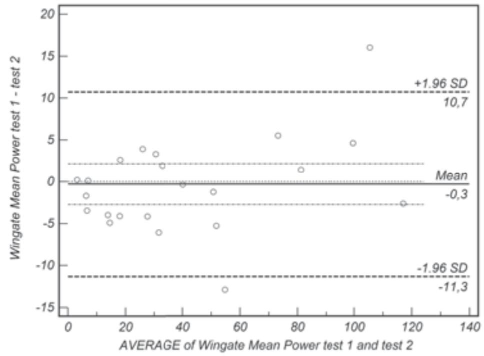

Figure 3.3: Bland–Altman plot of Mean power on test and retest on the arm-cranking Wingate Anaerobic Test.

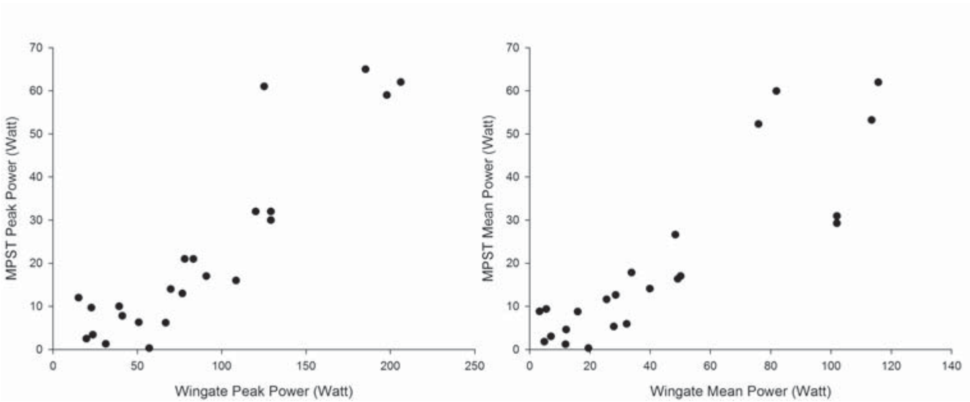

Figure 3.4: Scatterplot showing the relationship between Peak power and Mean power on arm-cranking Wingate Anaerobic Test and Muscle Power Sprint Test.

Construct validity of the Muscle Power Sprint Test

The MPST variables were significantly lower than the arm-cranking WAnT scores, but demonstrated very strong significant positive correlations for peak power and mean power from both tests (peak power: r=0.91, p<.05; mean power: r=0.88, p<.05, Figure 3.4).

DISCUSSION

The purpose of this study was to determine the two aspects of reproducibility (reliability and agreement) of the arm-cranking WAnT, MPST and 10×5-m sprint test in children and adolescents with CP who self-propel a manual wheelchair. Reliability of the arm-cranking WAnT and MPST can be considered excellent, with ICCs of 0.99 (with 95% CI between 0.98 and 1.0). Agreement was good, as reflected by the narrow LoA. The ICCs for the MPST and 10×5-m sprint test are similar to those previously reported for both tests in the literature, ranging from 0.97 to 0.99 for ambulatory children with CP and with ICCs for mean power and peak power on the MPST, of 0.98 for typically developing children.[3,22] Moreover, significant correlations between the performance on the arm-cranking WAnT and MPST were found, indicating that the MPST is a valid test for the assessment of anaerobic performance in children with CP who self-propel a manual wheelchair.

The MPST and the 10×5-m sprint test are inexpensive, safe, easy to administer, and are closely related to wheelchair-based ADL. Neither tests require any special equipment or training, making them available for use by 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 exercise, the MPST is probably the most appropriate outcome measure. When the intervention is more focused on functional outcomes, such as the ability to change direction of the wheelchair abruptly, the 10×5-m sprint test may be more suitable.

When power outputs (peak power and mean power) from both tests were compared, we found that MPST values were significantly lower than those of the arm-cranking WAnT. This difference may be explained by the fact that while both the MPST and arm-cranking WAnT measure power output of the upper limbs, the MPST uses intermittent propelling of the wheelchair instead of the continuous pushing and pulling used in arm cranking. Moreover, the outcomes of the MPST are based on averages over three 15m distances or roughly 30 seconds, while the arm-cranking WAnT outcomes are measured instantaneously (<1s). In spite of these differences, our results indicate that like the arm-cranking WAnT, the MPST can be used to measure the anaerobic performance of the arms. From a rehabilitation medicine perspective, this finding is very promising because of the minimal cost associated with performing the MPST, its easy application and its similarity and therefore relevance to children's ADL.

Field tests that rely on manual wheelchair propulsion performance are affected by many factors beyond motor limitations. Vanlandewijck et al.[23] have shown that the type of wheelchair (i.e. basketball/tennis wheelchair vs ADL wheelchair) affects field test performance. Also minimal changes to wheelchair configuration, such as the presence or absence of a caster wheel (a fifth wheel at the back of the wheelchair for stability), may also alter field test performance significantly. Therefore, field tests, such as the MPST and 10×5-m sprint test for wheelchair users should primarily be used to assess the individual progress of a child. Furthermore, the use of a standardised wheelchair should be discouraged due to the lack of individual adjustments that may impact, and potentially hamper, performance.[24]

Anaerobic testing has some intrinsic methodological limitations.[8] The arm-cranking WAnT and the MPST are largely dependent on participant motivation. Currently, there are no objective physiological criteria that can be used to establish a 'true' maximal anaerobic effort. Therefore, the researcher or the clinician must rely on the cooperation of the individual performing the exercise. Encouragements and a friendly environment are also important to ensure the participant performs the test to the best of their ability.

Our findings must be interpreted in light of certain limitations. First, this study included only youth with spastic CP. Whether our results are generalizable to other clinical types of CP and other medical conditions requires further investigation. Second, the participants in this study represented a convenience sample of children and adolescents with CP who were receiving physical therapy. This selection procedure may have led to some degree of selection bias, because it is unknown whether or not these participants differ from children and adolescents who are not receiving treatment in a rehabilitation centre or special education school. Third, we have selected a braking force of 0.26 Nm/kg for all children (and did not alter this for age or gender). This was primarily based on the fact that youths with CP may have different arm-ability that is not related to age or sex (e.g. spasticity, strength, mobility), but might influence the arm-cranking strength. The optimal braking force for males and females with CP of different age groups or between those with varying diagnoses remains to be investigated. Fourth, the large SDs in all tests suggest that there is large inter-individual variability in anaerobic performance in this group. This is probably the result of the large age-range and different classification levels of the participants. Fifth, our sample size was inadequate to compare the validity and reproducibility of specific subsets of the sample as grouped by age, sex or GMFCS level. Sixth, the degree to which the use of a static start in the MPST and a rolling start for the arm-cranking WAnT may have impacted our findings remains to be determined. Future studies should seek to assess these differences, or make use of only one of the two starting techniques.

CONCLUSION

The MPST, the arm-cranking WAnT and the 10×5-m sprint test are reproducible tests for measuring anaerobic performance and agility in children with CP who self-propel a manual wheelchair. Moreover, the MPST has shown to be a valid test to measure anaerobic performance. The MPST and 10×5-m sprint test are easy to administer and inexpensive. Clinicians using both tests do not need special equipment or training, which makes both tests available for a variety of professionals working with children and adolescents with CP.

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4

Sport-2-Stay-Fit study: Health effects of school-based sport participation in children and adolescents with a chronic disease or physical condition

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ABSTRACT

Background: Children and adolescents with a chronic disease or physical disability have lower fitness levels compared to their non-disabled peers. Low physical fitness is associated with reduced physical activity, increased cardiovascular diseases, and lower levels of both cognitive and psychosocial functioning. Moreover, children and adolescents with a chronic disease or physical disability participate less in both recreational and competitive sports. A variety of intervention studies have shown positive, but only temporary, effects of training programs. Next to issues related to the chronic condition itself, various personal and environmental factors play a key role in determining the extent to which they participate in sports or physical activities. Due to these barriers, sport participation in the immediate after-school hours seems to be a feasible solution to get these children and adolescents physical active structurally. To investigate if a school-based sports program can sustain the positive effects of an intervention, a standardized interval training will be given to improve physical fitness levels. High-intensity Interval Training (HIT) is superior to moderate-intensity continuous training in improving physical fitness in patients with chronic diseases. Therefore, the Sport-2-Stay-Fit study will investigate whether a school-based sports program can increase the sustainability of a HIT program in children and adolescents with a chronic disease or physical disability.

Methods: The Sport-2-Stay-Fit study is a clinical controlled trial. A total of 74 children and adolescents in the age of 6–19 years with a chronic disease or physical disability will be included. This could be either a cardiovascular, pulmonary, metabolic, musculoskeletal or neuromuscular disorder. Both children and adolescents who are ambulatory or propelling a manual wheelchair will be included. All participants will follow a HIT program of eight weeks to improve their physical fitness level. Thereafter, the intervention group will participate in school-based sports for six months, while the control group receives assessment only. Measurements will take place before the HIT, directly after, as well as, six months later. The primary objective is anaerobic performance. Secondary objectives are agility, aerobic fitness, strength, physical activity, cardiovascular health, cognitive functioning, and psychosocial functioning.

Discussion: If effective, school-based sports following a standardized interval training could be implemented on schools for special education to get children and adolescents with a chronic disease or physical disability active on a structural basis.

Trial registration: This trial is registered at the Dutch Trial Register #NTR4698.

INTRODUCTION

Children and adolescents with a chronic disease or physical disability have lower fitness levels compared to their non-disabled peers.[1-6] Low physical fitness is highly associated with reduced physical activity, increased cardiovascular diseases and overall mortality in healthy populations.[7-10] Additionally, cardiovascular risk factors such as dyslipidemia, hypertension, hyperinsulinemia or insulin resistance, and obesity often exist in children and young adults.[11,12] To be healthy later in life, it is assumed that children and adolescents should be physically active on a structural basis.

At the same time, children and adolescents who are healthy[13] or physical active[14,15] perform better at school. Moreover, childhood physical fitness is associated with higher levels of cognition.[15-17] Physical fitness level is a strong predictor in children and adolescents of school performance and level of cognition one year later.[18,19] School performance has improved in children and adolescents who are typically developing by intervention programs stimulating physical activity and sports.[20,21] However, this relationship between physical fitness and cognition level is still unknown in children and adolescents with a chronic disease or physical disability.

Another positive effect related to participation in sports is improving quality of life. This relation has been found in children and adolescents with cerebral palsy.[22,23] In addition, children with lower motor skills have a lower global self-perception than their well-coordinated peers.[24] Perceptions of physical competence are strong predictors of self-perception among children with motor coordination difficulties.[24,25] Successful performance experiences (i.e., sport participation) are the strongest means of changing self-perception.[26,27] Short-term sport interventions, however, did not significantly improve the self-perception of children with a physical disability although motor skills did improve.[28,29] The long-term effect of sport participation on self-perception in children and adolescents with a chronic disease or physical disability still has to be established.

It can be assumed that the benefits of participating in sports are universal for all children, including those with a chronic disease or physical disability. However, they participate less in competitive and recreational sports compared to their non-disabled peers.[30,31] A variety of interventions have shown that training programs improved physical fitness levels and participation in physical activities or sports. However, the positive effects following the training program did never sustain.[23,32,33] Therefore, more recently a shift in focus has moved from interventions to improve fitness to interventions to increase physical activity.[30,34]

For children and adolescents with a chronic disease or physical disability it is difficult to participate in sports or physical activities. Next to issues related to the chronic condition itself, various personal and environmental factors play a key role.[35-37] The most important barriers are: physical ability of the child, decreased motivation, lack of awareness of sports possibilities and lack of access to transportation. Most of these barriers can possibly be eliminated when an adapted sports program is provided at school. Interventions to promote sports in healthy young people conducted in the hours after school are effective in improving physical fitness, physical activities and health.[38,39] A few special schools in the Netherlands are experienced in organizing and facilitating school-based sports. Although they anecdotally notice a lot of progress in these children, they have never measured the effect accurately in a scientific and systematic way.

To investigate if school-based sports can sustain physical fitness level, health, and both cognitive and psychosocial functioning, a baseline fitness level is needed. Standardized exercise training with prescribed work-rest ratios, intensity, volume and time are more effective in improving physical fitness, especially short term.[23,32,33] In this regard, high-intensity Interval Training (HIT) is superior to moderate-intensity continuous training in improving physical fitness in patients with chronic diseases.[40-42] Furthermore, HIT is feasible for these children and adolescents.[23] Therefore, prior to school-based sports, a HIT program will be given to improve physical fitness level.

The current study, denoted as the Sport-2-Stay-Fit study, will investigate whether school-based sports can increase the sustainability of a HIT program in children and adolescents with a chronic disease or physical disability. In addition, sport participation in these children may have positive effects on cardiovascular health, and both cognitive and psychosocial functioning. While these positive relationships have been reported in children and adolescents who are typically developing, the benefits are not established in those with a chronic disease or physical disability.

METHODS

Design

The Sport-2-Stay-Fit study is a controlled clinical trial. All children and adolescents will participate in an eight week HIT program to improve their physical fitness level. Thereafter, the intervention group will participate in school-based sports for six months, while the control group will receive assessments only. Outcome measures will be assessed at baseline (T0), immediately after eight weeks of HIT (T1), and at completion of six months intervention (T2) (Figure 4.1). Similar outcome measures will be evaluated at T0, T1 and T2 except for physical activity at T1, because no effect or changed difference between groups was expected.[43,44] The current study is part of a larger project in which children and adolescents with a chronic disease or physical disability who are active in sports will be compared to their non-sporting peers.[45]

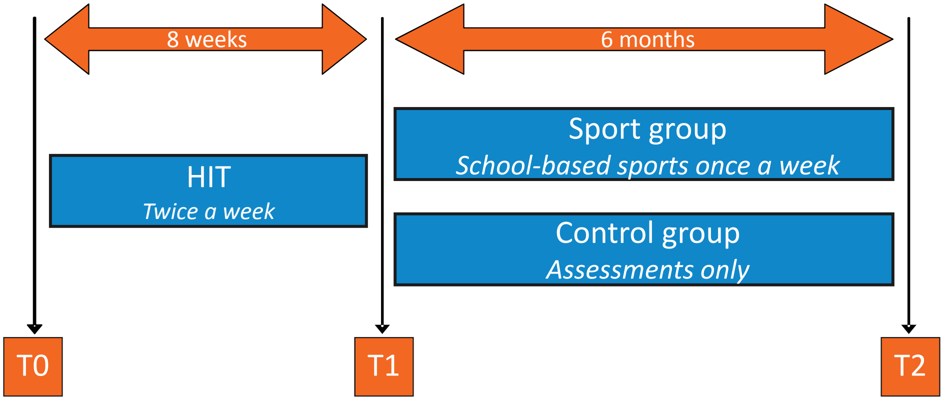

Figure 4.1: Schematic research design of the Sport-2-Stay-Fit study.
HIT: High-intensity interval training.

Setting

In total 74 participants will be recruited from four schools for special education in the Netherlands: Ariane de Ranitz | De Hoogstraat Rehabilitation in Utrecht, Lichtenbeek in Arnhem, Mariëndael in Arnhem, and Heliomare in Wijk aan Zee. Schools experienced in organizing and facilitating school-based sports, will be included in the intervention group. To prevent bias, the control group will only be included at schools where no school-based sports are provided. In this way, the research design is close to reality and participants are not unethically prohibited to sport at school. Both ethics approval and administrative site approvals were granted by the Medical Ethical Committee of UMC Utrecht in the Netherlands (#14-118). Additionally, all parents, and participants from 12 years of age, have to provide informed consent prior to study initiation.

Participants

Children and adolescents between the age of 6 and 19 with a chronic disease or physical disability will be included. This could be either cardiovascular, pulmonary, musculoskeletal, metabolic or neuromuscular disorders. Both children and adolescents who are ambulatory or propelling a manual wheelchair are asked to participate. They may participate in sports at most once a week during leisure time in the preceding three months or have a treatment goal on fitness level prescribed by their physician. Participants have to understand simple commands and are able to perform the physical fitness tests. Children and adolescents using powered wheelchairs or having progressive diseases will be excluded. In addition, during the length of the study, children and adolescents are not allowed to participate in other research projects which might influence current study results.

Intervention

All participants will follow a training program of eight weeks to increase their physical fitness level. A HIT program will be given at school by a physical educator and/or a physical therapist. Every training session will consist of a prescribed intensity, volume and time (Table 4.1). According to the review of Baquet et al.[46] the 30 seconds all-out approach for interval training is recommended in youth. An active recovery has been suggested in order to effectively aid the process of lactate removal.[47] Sitting will therefore be forbidden during the interval training and children will be encouraged to motivate and support the others while exercising. After eight weeks of HIT, the experimental group will participate in once a week school-based sports for six months. This is an extra sports lesson additional to the regular physical education schedule. During regular physical education half of the time is

Table 4.1: Schematic design of the HIT program

	Week 1	Week 2	Week 3	Week 4	Week 5	Week 6	Week 7	Week 8
Frequency (p/w)	2	2	2	2	2	2	2	2
Duration	30 sec	30 sec	30 sec	30 sec	30 sec	30 sec	30 sec	30 sec
Intensity	max	max	max	max	max	max	max	max
Work:rest ratio	1:4	1:4	1:4	1:4	1:3	1:3	1:3	1:3
Repetitions	4	4	5	5	6	6	6	6
Series	2	2	2	2	2	2	2	2
Total time	20 min	20 min	25 min	25 min	24 min	24 min	24 min	24 min

HIT, High-intensity interval training.

spend on skill practice, physical education knowledge and management in children with a physical disability.[48] In contrast, sport is about being physical active, playing the game, and being competitive.

Outcome measures

A general screening questionnaire will be used to inquire about possible factors influencing the outcomes: age, sex, physical activities, health status, injuries. The following outcome measures can be divided: physical fitness, cardiovascular health, physical activity, cognitive functioning, and psychosocial functioning (Table 4.2).

Table 4.2: Overview of outcome measures of the Sport-2-Stay-Fit study

Outcome measure	Parameter	Measurement
Physical fitness	Anaerobic performance	Muscle Power Sprint Test (MPST)
	Agility	10x5 meter sprint
	Aerobic fitness	Shuttle run/ride test (SRT)
	Strength	Grip strength Reverse curl Seated push up Standing broad jump/One stroke push
	Flexibility	Modified Apley test Modified Thomas test
Cardiovascular health	Morphologic	Length, weight, BMI Bioelectrical impedance analysis
	Metabolic	Sphygmomanometer Arteriograph Finger puncture
Physical activity	Modality	Activ8 Activity diary
Cognitive functioning	School performance	Type of education Educational achievement test
	Attention	Bourdon-Vos Cancellation task Capture task
Psychosocial functioning	Self-perception	Self-Perception Profile for Children (SPPC)
	Quality of life	Disabkids
	Self-efficacy	Exercise Self-efficacy Scale (ESES)

Primary outcomes

Anaerobic performance is the primary outcome measure in the current study, because daily physical activity in children most often consists of brief, intermittent bouts of intense movement.[49,50] Consequently, the anaerobic performance of healthy individuals involved in recreational sport programs is significantly better than that of those individuals who are not involved in any activities.[51] The Muscle Power Sprint Test (MPST) will be used to evaluate anaerobic performance. This test is suitable and validated for both children and adolescents who are typically developing, and with cerebral palsy who are ambulant or self-propel a manual wheelchair.[6,52-54] Subjects have to complete six 15-meter runs at a maximum pace. The MPST is an intermittent sprint test, in which the child starts and stops at standardized intervals. Power output will be calculated for each of the six sprints. Peak power will be defined as the highest calculated power, while mean power will be defined as average power over the six sprints.

Secondary outcomes

Agility

The 10x5 meter sprint test will be performed to assess agility, either while running or propelling a wheelchair. This is a reliable field test to measure agility in children with cerebral palsy.[52,53] During this test, the child has to sprint as fast as possible for 10 times in between two lines that are five meters apart. Since there is no resting period, the participant has to turn as fast as possible.

Aerobic fitness

Aerobic fitness, generally expressed as peak oxygen uptake (VO_2peak), is a good predictor of cardiovascular risk.[55,56] VO_2peak will be measured with a field test. Shuttle run/ride test (SRT) is a field test in which a participant runs, walks or rides between two markers. The SRT starts at 1.5 km/h, 2 km/h or 5 km/h and has shown to be a valid and reliable test in children with cerebral palsy measuring aerobic fitness.[57,58] The children have to adjust their running, walking of wheelchair propulsion pace to the beep-signals, until they fail to reach the line two times in a row, despite encouragements. Subsequently, a participants' mode of locomotion will be identified in order to apply a proper testing protocol (Table 4.3). Based on clinical observations, participants will be divided into four groups: subjects who are able to run, subjects who walk independently but are not able to run, subjects using walking devices, and wheelchair users. Running is defined in the ICF as moving with quick steps so that both feet may be simultaneously off the ground.[59]

Table 4.3: Overview of different SRT protocols

Participants	Protocol	Distance	Starting pace	Increasement per min
Able to run	SRT I	10-meter line	5.0 km/h	0.25 km/h
Walk independently (not able to run)	SRT II	10-meter line	2.0 km/h	0.25 km/h
Using walking devices	SRT II	7.5-meter square	1.5 km/h	0.19 km/h
Wheelchair users	SRT II	10-meter line	2.0 km/h	0.25 km/h

SRT, shuttle run/ride test.

Each test will last until volitional exhaustion and protocols will be chosen with the aim to achieve a total exercise time between 6 and 15 minutes.[60,61] The shuttle tests will be evaluated with the Cortex Metamax 3X (Samcon bvba, Melle, Belgium), a valid and reliable cardio-pulmonary exercise testing system.[62-64] The Cortex Metamax 3X consists of a facemask, transmitting unit attached to a harness containing oxygen and carbon dioxide analyzers, and a receiving unit. Metabolic stress test software (Metasoft Studio) will be used to measure minute ventilation, oxygen uptake (VO_2), carbon dioxide production (VCO_2), heart rate, and respiratory exchange ratio (RER $=VCO_2/VO_2$) every 10 seconds.

Strength

To assess explosive functional strength, the standing broad jump and one stroke push will be performed in children who are ambulatory and propel a manual wheelchair, respectively. Lower limbs strength will be measured by the distance jumped with two legs together from standing position,[65] and upper limbs strength by measuring the distance covered in a wheelchair by one push.[66] The following tests are chosen from the Brockport Physical Fitness Test, which was designed to test fitness of youths with various disabilities and is both health related and criterion referenced. Grip strength represents the general strength in healthy children.[67] The grip strength of the dominant hand will be tested through the use of a hand held dynamometer, as described by Beenakker et al.[68] The reverse curl is designed as a measure of hand, wrist, and arm strength.[69] In this test, the participant attempts to pick up a 0.5-kg dumbbell with the preferred arm while seated in a chair or wheelchair. The seated push up is designed to measure upper-body strength and endurance.[69] The participant attempts to perform a seated push-up and holds it for up to 20 seconds.

Flexibility

To test flexibility, the Modified Apley Test and the Modified Thomas Test were chosen from the Brockport Physical Fitness Test. To measure upper-body flexibility with the Modified

Apley test, participants attempt to reach back and touch the superior medial angle of the opposite scapula with one hand.[69] The Modified Thomas Test is designed to assess the length of participant's hip flexor muscles (i.e., iliopsoas and rectus femoris muscles).[69]

Cardiovascular health

BMI, body mass index, will be calculated as weight (kg)/length (m)^2. Height will be measured in standing position in case of ambulant subjects and supine in case of wheelchair bound persons. When a person has spastic legs, arm span width will be measured and corrected as described by Dosa et al.[70] Fat free mass will be determined with bioelectrical impedance analysis (BIA), using the Bodystat Quadscan 4000 (Euromedix, Leuven, Belgium). BIA is a non-invasive test comparing conductivity and resistance in the body to distinguish lean body mass and fat.[71]

Increased arterial stiffness is associated with a higher likelihood of cardiovascular disease.[72] Arterial stiffness exists of two independent values. The Pulse Wave Velocity (PWV) is measured as the speed at which an aortic pulse travels; increased speed will indicate stiffer arteries. The Augmentation Index (AIX) provides information on the peripheral resistance of the endothelial vessels. Blood pressure and arterial stiffness measurements will be performed using arteriography. The arteriograph (Litra, Amsterdam, the Netherlands) will measure blood pressure, as well as arterial stiffness, using oscillometric tonometry. This method is valid compared with the golden standard: invasive measurement during cardiac catheterization.[73] Each subject will rest for at least 10 min prior to the recording. The measurement will be executed using an inflatable cuff at the right upper arm. The participants may not move or talk during the measurement.

To determine metabolic parameters, a finger puncture will be performed to measure cholesterol, low density lipoprotein, high density lipoprotein, glucose and triglyceride. The finger puncture will be performed using a Cholestech LDX analyzer (Mediphos Medical Supplies BV, Renkum, the Netherlands). Participant may not eat or drink for three hours prior to this procedure.

Physical activity

An Activ8 (2M Engineering, Valkenswaard, the Netherlands) accelerometer will be used to measure the type, duration, frequency and intensity of physical activity in daily life. The system is valid and reliable to detect the type, duration, frequency and intensity of physical activity in persons able to walk.[74] Both children who are ambulatory and wheelchair dependent wear the Activ8 on their upper leg for five consecutive days: three weekdays and two weekend days. Wearing the activity monitor will be no burden for the participants; all activities in daily life and during sport activities can be performed. The general activity pattern, incidence and type of injury during the last three months will be measured by a questionnaire.

Cognitive functioning

Childhood physical fitness is associated with higher levels of cognition; attention and school performance.[15,16,75] For quantifying school performance, a Dutch educational achievement test will be used, which should be administered at school annually. All scores can be converted to a didactic age equivalent. In addition, a more sensitive outcome measure has been chosen to detect changes longitudinally. Attention was operationalized into three components: sustained attention, search efficiency and distractibility. All tasks will be performed on a tablet (Asus Eee Slate Tablet, with a 12.1 inch display and clock speed of 1.33 GHz) overcoming coordination problems and guaranteeing detection of subtle improvements in performance. Sustained attention will be measured using an adapted digitalized version of the Bourdon-Vos task.[76,77] The Bourdon-Vos task is a cancellation test requiring high-speed visual selectivity and a repetitive motor response.[76] Given the length and duration of the test, sustained attention will be assessed.[78,79] Children are instructed to cross out the target items, i.e. the dot patterns with four dots on a tablet covered in three, four, and five dot patterns. Dependent variables are performance and accuracy over time indicating fluctuations in attention. To assess search efficiency an object cancellation task was used. The presented screen contained 130 stimuli: red and green apples, green and brown pears, letters, and strings of letters. Children had to cancel all apples, while ignoring all other stimuli. Search efficiency will be measured including consistency of search direction, distance between two clicks, and number of intersections. The original capture task used in Van Der Stigchel & Nijboer[80] was adapted to measure distractibility. Each trial start with the presentation of central fixation cross. When the cross disappeared, the target, represented as an apple, appeared in one of the corners. In 50% of the trials, a distractor will appear as well. Reaction time will be measured for both conditions to calculate distractibility.

Psychosocial functioning

In children who are typical developing several studies proved a positive correlation between sports participation and self-perception.[81] To evaluate self-perception, the self-perception profile for children will be used.[82] This is a self-report scale that measures five specific domains of self-concept and the sense of general self-worth.

Children with physical disabilities who participate in sports had significantly higher health related quality of life satisfaction scores compared to their non-sporting peers.[83] The

Disabkids questionnaire will be used to assess health-related quality of life of children with chronic health conditions.[84] The questionnaire consists of 37 Likert-scaled items and is associated with three domains: mental, social and physical. Additionally, the three domains can be subdivided to six dimensions: independence, emotion, social inclusion, social exclusion, limitation, and treatment. The Disabkids has been developed and validated for children of age 4–16 years.[85-87] Older kids probably have not been included before, because some questions are related to school situations. Since all participants are still attending school, the questionnaire is considered to be applicable for all participants.

To establish the extent of participation in and adherence to a program of regular exercise or physical activity, the Exercise Self-Efficacy questionnaire scale (ESES) will be conducted.[88] A persons self-efficacy is largely influenced by past performances and accomplishments, or mastery experiences.

Sample size

According to a study of Verschuren et al.[23] an average decline of 10±27% in anaerobic performance was calculated 3–4 months following a training program. It has been hypothesized that participants following the sports program will show a difference of 20% compared to the control group. The effect of 20% seems realistic, since we previously found a 24% increase in anaerobic performance in children with cerebral palsy after a training program.[23] With an alpha of 0.05 and beta of 0.20 (power of 0.80) a sample size of 32 subjects per group will be required. When taking a failure rate of 15% into account, 74 subjects should be included in total in the Sport-2-Stay-Fit study.

Statistical analyses

The effect of the interval training intervention and the six month sports program will be analyzed using a multivariate repeated measures ANOVA. The possible differences between and within T0, T1 and T2 for the intervention group and control group will be calculated with a statistical significance level of p=.01. If there is a significant difference, a post-hoc test (a Bonferroni or LSD) will be executed to further investigate group differences. Quantitative descriptive statistics will be used to present demographics of the primary and secondary study parameters. All statistical analyses will be performed using SPSS for Windows (version 21.0, SPSS Inc, Chicago, Ill.) with a statistical significance level of p=.05.

DISCUSSION

The Sport-2-Stay-Fit study will provide insight in the effectiveness of an eight weeks HIT program in children and adolescents with a chronic disease or physical disability. Furthermore, the current study gives answer to the important question whether school-based sports can increase the sustainability of the effectiveness of a HIT program. To our knowledge, this is the first study investigating the sustainability of a standardized training with a sport program in a group of children and adolescents with a chronic disease or physical disability.

In addition, sport participation in these children may have positive effects on cardiovascular health, injuries, and both cognitive and psychosocial functioning. While these positive relationships have been reported in healthy children, the benefits are not yet established in children with a chronic disease or physical disability.

The Sport-2-Stay-Fit study provide insight and understanding in both the effectiveness and feasibility of school-based sports following a standardized interval training. Changing physical activity during leisure time is complicated. Especially in children and adolescents with a chronic disease or physical disability who experience a lot of barriers in joining sports clubs. In this perspective, after-school sport participation may be a feasible solution to get those children and adolescents structurally active. The results are expected to be available in 2017.

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Effects of high-intensity interval training on fitness and health in youth with physical disabilities

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Submitted

ABSTRACT

Purpose: To investigate the effects of high-intensity interval training (HIT) on physical fitness and cardiometabolic health in youth with physical disabilities.

Methods: For this quasi-experimental study 70 participants were recruited (mean age 13.4 (2.9) years, range 8–19, 56% boys) from schools for special education. HIT was performed for eight weeks, twice a week, containing 8–12 intervals of 30 seconds all-out exercises.

Results: Exercise adherence was 84.5%. Following HIT, improvements were found in anaerobic performance (MD=19.2W, p=.002, d=0.40), agility (MD=-1.9s, p<.001, d=0.54), aerobic performance (MD=1.4 shuttles, p<.001, d=0.94), and both systolic- (MD=-2.9 mmHg, p=.008, d=0.34) and diastolic blood pressure (MD=-2.4 mmHg, p=.022, d=0.29). No changes were found for VO_2peak, arterial stiffness, body composition and the metabolic profile.

Conclusions: Both anaerobic and aerobic performance improved after HIT, with no changes in VO_2peak. No effects were found for cardiometabolic health, except for a decrease in blood pressure.

INTRODUCTION

Given the fact that youth with physical disabilities have lower physical fitness levels compared to their non-disabled peers,[1] they are thought to be at increased risk for cardiometabolic diseases.[2] While several studies have reported positive effects of more traditional forms of exercise training on physical fitness in youth with cerebral palsy (CP) and spina bifida,[3,4] high-intensity interval training (HIT) has shown to be a more effective and time-efficient form of training in adults with cardiometabolic diseases.[5] HIT involves alternating short bursts of high-intensity exercise followed by a period of low-intensity exercise or rest.[6] Despite large differences in training volume (i.e., 90% lower in HIT) and time commitment (i.e., 67% lower in HIT), HIT provides similar or greater benefits in improving physical fitness and reducing cardiometabolic risk factors when compared to moderate intensity continuous training (MICT) in both healthy adults and those with cardiometabolic diseases.[5,7]

Following HIT, positive effects on exercise performance, such as exercise time, sprint performance or agility, can be expected.[6,7] In addition, cardiorespiratory fitness, generally expressed as peak oxygen uptake (VO_2peak), almost doubled after HIT as compared to MICT in a meta-analysis among adults with lifestyle-induced cardiometabolic disease.[5] In addition to the physical benefits, HIT also reduce cardiovascular (i.e., blood pressure and arterial stiffness) and metabolic risk factors (i.e., body composition, lipid profile and fasting glucose) in both healthy and diseased adults.[5,8,9]

Daily physical activity in youth most often consists of brief, intermittent bouts of intense movement, similar to HIT.[10] Since the physiological adaptations after training are task specific, HIT might be more relevant to the activity patterns during childhood and adolescence as compared to MICT. This holds true for children and adolescents without motor problems. However, it is unknown whether intermittent bouts of intense exercise have the same effect on youth with physical disabilities. In addition, since performance-related fitness is dependent on motor function, it is unknown how HIT affects children and adolescents with different levels of mobility.[11]

The higher intensities of HIT causes greater disturbances of the physiological system, resulting in enhanced metabolism greater or similar to MICT.[6] Several studies have shown short-term (i.e., ≤12 weeks) effects of HIT in physical fitness and cardiometabolic health in children and adolescents who are overweight.[12,13] A recent study showed that VO_2peak increased and body composition remained similar after 24 HIT-sessions in children with CP.[14] However, little is known about the effect of HIT on fitness and health in children and adolescents with various disabilities. Since these children and adolescents exercise together at school, this study aimed to include a reflection of health conditions and mobility levels. The purpose of current study is to investigate the short-term effects of HIT on physical fitness and cardiometabolic health in youth with physical disabilities including different levels of mobility.

METHODS

Design

This study was a quasi-experimental study with pre and post measurements. All children and adolescents had to perform HIT for eight weeks, twice a week, for 30 minutes. Outcome measures were assessed at baseline (T0) and immediately after HIT (T1) by the same trained researcher across all schools. The current study is part of the Sport-2-Stay-Fit study (Dutch Trial Register #NTR4698), where HIT is followed by a school-based sports program. The complete protocol is described elsewhere.[15] Both ethics approval and administrative site approvals were granted by the Medical Ethical Committee of UMC Utrecht in the Netherlands (#14-118).

Participants

Children and adolescents, both ambulatory and those propelling a manual wheelchair, were recruited from four schools for special education in the Netherlands. These schools have similar learning objectives as regular schools, but the children receive additional attention and support. Rehabilitation medicine is provided within school hours by rehabilitation professionals. In the Netherlands 1.9% (26,500) of the children and adolescents following primary or secondary education follow these schools dedicated to youth with special education.[16] Children and adolescents were screened by a physical therapist, physical educator or physician for eligibility. Inclusion criteria were: (1) aged between 6 and 19 years, (2) having a chronic disease or physical disability including a cardiovascular-, musculoskeletal-, metabolic- or neuromuscular disorders, (3) participated in sports less than twice a week during leisure time in the preceding three months or were advised to participate by their physical therapist or physician (4) understood simple commands, and (5) were expected to be able to perform the physical fitness tests. Children and adolescents using an electric wheelchair for sport purposes or those with progressive diseases were excluded. In addition, during the length of the study, children and adolescents were not allowed to participate in other studies that could possibly influence results in the current study. Parents and children were invited by letter for participation in the Sport-2-Stay-Fit study.[15] Additionally, all parents and participants from 12 years of age provided written informed consent prior to study initiation.

Intervention

The HIT program was provided by an experienced physical educator and/or a physical therapist at school. Every training session consisted of a prescribed intensity, volume and time (Table 5.1). The 30 seconds all-out approach, as recommended for interval training in youth, was used.[6,10,18] The all-out approach means participants were extensively encouraged to sprint with maximal effort without need for extensive heart rate monitoring.[9] Easily executable sprint exercise, like transferring bean bags and sprinting between cones, were chosen to provoke the cardiorespiratory system as much as possible within 30 seconds. For motivation purposes, participants were instructed to count their repetitions and report it to the trainers. In between intervals participants performed lower intensive activity, for example gathering bean bags, as active recovery has been suggested to effectively aid the recovery process. The presence of the participants was documented every training session by the physical educator or physical therapist.

Table 5.1: Schematic design of the high-intensity interval training (HIT) twice a week

	Week 1	Week 2	Week 3	Week 4	Week 5	Week 6	Week 7	Week 8
Series	8	8	10	10	12	12	12	12
Duration	30 sec	30 sec	30 sec	30 sec	30 sec	30 sec	30 sec	30 sec
Intensity	max	max	max	max	max	max	max	max
Work:rest ratio	1:4	1:4	1:4	1:4	1:3	1:3	1:3	1:3
Total time	20 min	20 min	25 min	25 min	24 min	24 min	24 min	24 min

Outcome measures

Anaerobic performance was the primary outcome, since daily physical activity in youth most often consists of intermittent bouts of intense exercise.[10] Outcome measures were tested during school hours subdivided into four different occasions within two weeks: a) height, weight, anaerobic fitness and strength, b) aerobic fitness, c) blood pressure, arterial stiffness, body composition, and d) metabolic profile. For analyses, participants were divided into three subgroups based on the Functional Mobility Scale (FMS): those who were able to run (i.e., runners), those who could walk independently but were not able to run (i.e., walkers), and those who propel a manual wheelchair (i.e., wheelchair users).[19] A participant's level of mobility was rated by their physical therapist or physical educator during free play or physical education.

Physical fitness

Anaerobic fitness

Both anaerobic performance and agility were assessed. Anaerobic performance was assessed by the Muscle Power Sprint Test (MPST).[11,20] In this test children and adolescents had to complete 15-meter sprints with a standardized rest of 10 seconds between sprints. Both mean and peak power were calculated from the sprints.[11] To assess agility, the 10x5 meter sprint test was performed, either while running, walking or propelling a wheelchair.[11,20] During this test, the child has to sprint 10 times as fast as possible between two lines five meters apart, while time was measured.

Aerobic fitness

A rrun/ride test (SRT) was performed to assess both aerobic performance (achieved shuttles) and VO_2peak. During this field test, a participant runs, walks or rides between two markers which were 10 meters apart, as described extensively elsewhere.[21,22] Participants have to adjust their running, walking or wheelchair propulsion pace to the beep-signals, until they fail to reach the line twice in a row, despite strong verbal encouragements. During the SRT a calibrated Cortex Metamax 3X (Samcon bvba, Melle, Belgium) measured oxygen uptake (VO_2), carbon dioxide production (VCO_2), peak heart rate, and respiratory exchange ratio ($RER=VCO_2/VO_2$) every 10 seconds, using metabolic stress test software (Metasoft Studio). To determine whether a subject reached their maximal effort during the test, two out of the following three criteria had to be achieved as reported previously in children and adolescents with CP: heart rate ≥180 bpm, RER ≥1.00 at peak exercise, or subjective signs of intense effort, such as sweating, facial flushing, or a clear unwillingness to continue.[23]

Strength

To assess explosive strength, the standing broad jump or one stroke push was performed in youth that were ambulatory and those that propelled a manual wheelchair, respectively. The standing broad jump was assessed as the distance in centimetres jumped with two legs. The one stroke push was assessed as the distance in meters covered in their own wheelchair by one push. Grip strength was measured using a hand held dynamometer (CITEC, CIT Technics, Haren, the Netherlands). Participants sat on a chair or in their wheelchair with their elbow situated on the armrest. After a practice session, grip strength was performed three times with the preferred hand. Mean grip strength (N) was calculated by averaging the three attempts.

Cardiovascular health

Blood pressure

Both resting blood pressure and arterial stiffness were simultaneously and noninvasively measured with the Arteriograph (Litra, Amsterdam, the Netherlands). The measurement was performed in a supine position, after a 10-minute rest, using an inflatable cuff on the right upper arm. Participants were asked not to move or talk during the measurement.

Arterial stiffness

Arterial stiffness was measured using two independent and validated values.[24,25] The Pulse Wave Velocity (PWV) was measured as the speed at which an aortic pulse travels; increased speed indicates stiffer arteries. The Augmentation Index (AIX) provides information on the peripheral resistance of the endothelial vessels; increased index indicated higher peripheral resistance. To control for differences in sex and age, Z-scores of AIX were calculated according to reference values of Hidvégi et al.[24]

Metabolic health

Body Mass Index

Body mass index (BMI) was calculated as weight (kg) / height (m)^2. A detailed description was published previously.[15] To control for differences in age, Z-scores of BMI for age were calculated according to Dutch reference values.[26]

Body composition

Fat mass was measured in supine position using the Bodystat Quadscan 4000 (Euromedix, Leuven, Belgium) and determined with Bioelectrical Impedance Analysis (BIA). For waist and hip circumference, a horizontal measure was taken at the umbilicus and spina iliaca anterior superior, respectively.

Metabolic profile

To determine the metabolic profile, a finger puncture was performed from which total cholesterol, low density lipoprotein (LDL), high density lipoprotein (HDL), ratio (total cholesterol/HDL), triglyceride and fasting glucose were determined. This was an optional measurement and consent was asked separately. The finger puncture was performed using a Cholestech LDX analyzer (Mediphos Medical Supplies BV, Renkum, the Netherlands). All participants were instructed to refrain from eating and drinking three hours prior to this procedure. Before the finger puncture, participants were asked about the fasting period and if possible the measurement was postponed. Otherwise, only total cholesterol, LDL, HDL, and triglyceride data were used for analysis.[27]

Statistical analysis

The sample size calculation for anaerobic performance was based on a previous study in children with CP.[3] Using a mean (SD) training effect of 20.4 (38.0), with an alpha of 0.05 and power of 0.80, a sample size of 30 participants is required. However, for the Sport-2-Stay-Fit study, where the current study is a part of a sample size of 74 participants was calculated, taking a drop-out rate of 15% into account.[15] Therefore, we aimed to include a total of 74 instead of the required 30 participants, and performed secondary analyses per subgroup.

All data analyses were performed according to the intention-to-treat principle. We checked for normality of data, normality of residuals and outliers. When residuals were not normally distributed, a log transformation was performed after which normal distribution was checked again. Finally, all outcomes were normally distributed. For comparison of outcomes pre- and post-training, a paired-sample t test was used to determine the effect of training. Outcomes measures were reported as means with their standard deviation (SD). Effect sizes were calculated using Cohen *d* for significant differences. Values of Cohen *d* less than 0.20 were considered small, between 0.50 and 0.80 were considered medium, and effect sizes greater than 0.80 were considered large. All statistical analyses were performed using SPSS for Windows (version 21.0, SPSS Inc, Chicago, Ill.) with a statistical significance level of p=.05.

RESULTS

A total of 70 participants completed the study (Figure 5.1). Participant characteristics are presented in Table 5.2. The number of planned HIT sessions ranged from 9 to 16 sessions in eight weeks, due to the school schedule incorporating holidays, days off and school activities with priority. The adherence was 84.5% containing an average number of 11.4

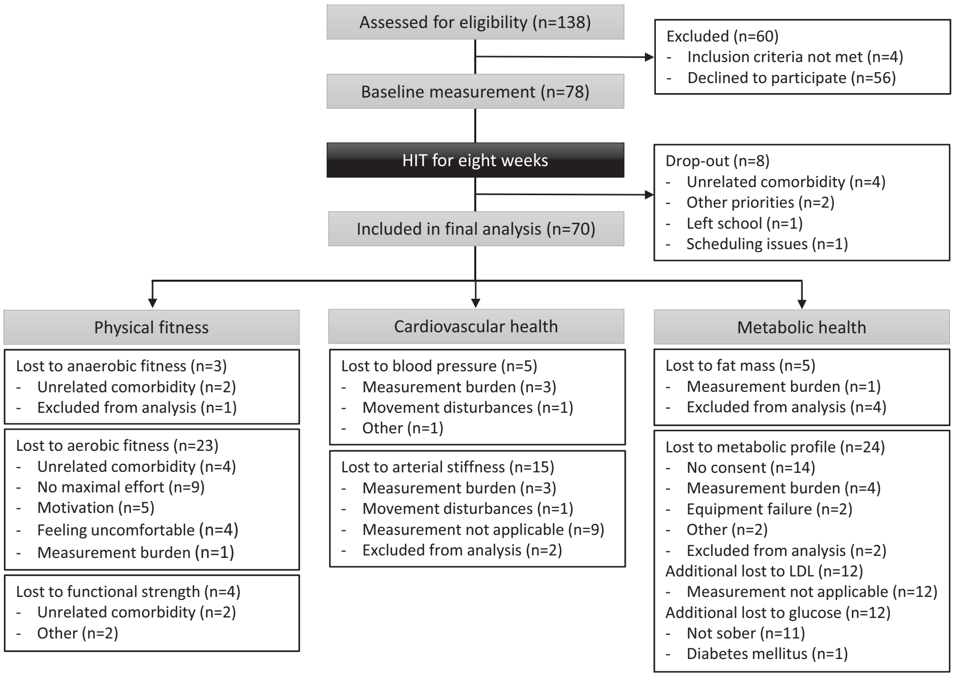

Figure 5.1: Flow-chart from initial inclusion to intervention and reasons for detraction from data analysis per outcome measure.

(3.0) training sessions. This was similar for all subgroups. No adverse events were reported during the study, suggesting HIT can be considered a safe and feasible intervention for this population.

Physical fitness

Anaerobic fitness

As shown in Table 5.3, for the total group both mean (p=.002) and peak (p=.009) power improved on the MPST with a mean difference (MD) of 19.2 W and 22.6 W, respectively. Secondary analysis showed similar improvements for the runners subgroup in mean (MD=31.9 W, p=.011) and peak (MD=40.3 W, p=.017) power. The walkers subgroup improved only with regards to mean power (MD=9.1 W, p=.020), while anaerobic performance in wheelchair users did not change (Figure 5.2a). For agility, the total group (MD=-1.9 s, p<.001) performed better on the 10x5 meter sprint test after training (Figure 5.2b). Effect sizes showed medium effects (0.33–0.51) for anaerobic performance and medium to large effects (0.49–0.92) for agility.

Table 5.2: Baseline characteristics of participants

	Total n=70	Runners n=36	Walkers n=25	Wheelchair users n=9
Age (y), mean (SD)	13.4 (2.9)	13.5 (2.7)	13.2 (3.1)	13.8 (3.0)
Boys, n (%)	38 (54)	22 (61)	9 (36)	7 (78)
Chronic diseases, n (%)				
Neuromuscular	56 (80)	29 (80)	21 (84)	6 (67)
Cerebral palsy	25 (36)	10 (28)	13 (52)	2 (22)
Psychomotor retardation	9 (13)	5 (14)	4 (16)	-
Spina bifida	5 (7)	-	1 (4)	4 (45)
Acquired brain injury	3 (4)	2 (6)	1 (4)	-
Other	14 (20)	12 (33)	2 (80	-
Metabolic	6 (9)	4 (11)	1 (4)	1 (11)
Musculoskeletal	5 (7)	2 (6)	2 (8)	1 (11)
Cardiovascular	3 (4)	1 (3)	1 (4)	1 (11)

Aerobic fitness

A total of 47 children and adolescents (67%), performed at their maximal effort both pre- and post-test (Figure 5.1). No significant differences were found between pre- and post-test measurements regarding peak heart rate and RER. Baseline values of respectively peak heart rate and RER, were for the runners 192 (11) bpm and 1.07 (.07), for the walkers 191 (13) bpm and 1.12 (.06), and for the wheelchair users 167 (26) bpm and 1.12 (.07). Aerobic performance on the SRT improved (MD=1.4 shuttles, p<.001) (Figure 5.2c) Effect sizes showed large effects (0.78–1.57) for aerobic performance in all subgroups. However, both absolute VO_2peak (l/min) and relative VO_2peak (ml/kg/min) did not increase after HIT (Figure 5.2d).

Strength

The standing broad jump test improved only in the runners subgroup (MD=4.6 cm, p=.042). One stroke push and mean grip strength did not change over time in all subgroups.

Cardiovascular health

A positive change was found in the total group for resting systolic (MD=-2.9 mmHg, p=.008) and diastolic (MD=-2.4 mmHg, p=.022) blood pressure (Figure 5.2e and 5.2f). Arterial stiffness showed no differences after HIT in AIX and PWV, with exception of both AIX (MD=-8.7%, p=.044) and AIX Z-score (MD=-1.26, p=.050) in wheelchair users (Figure 5.2g).

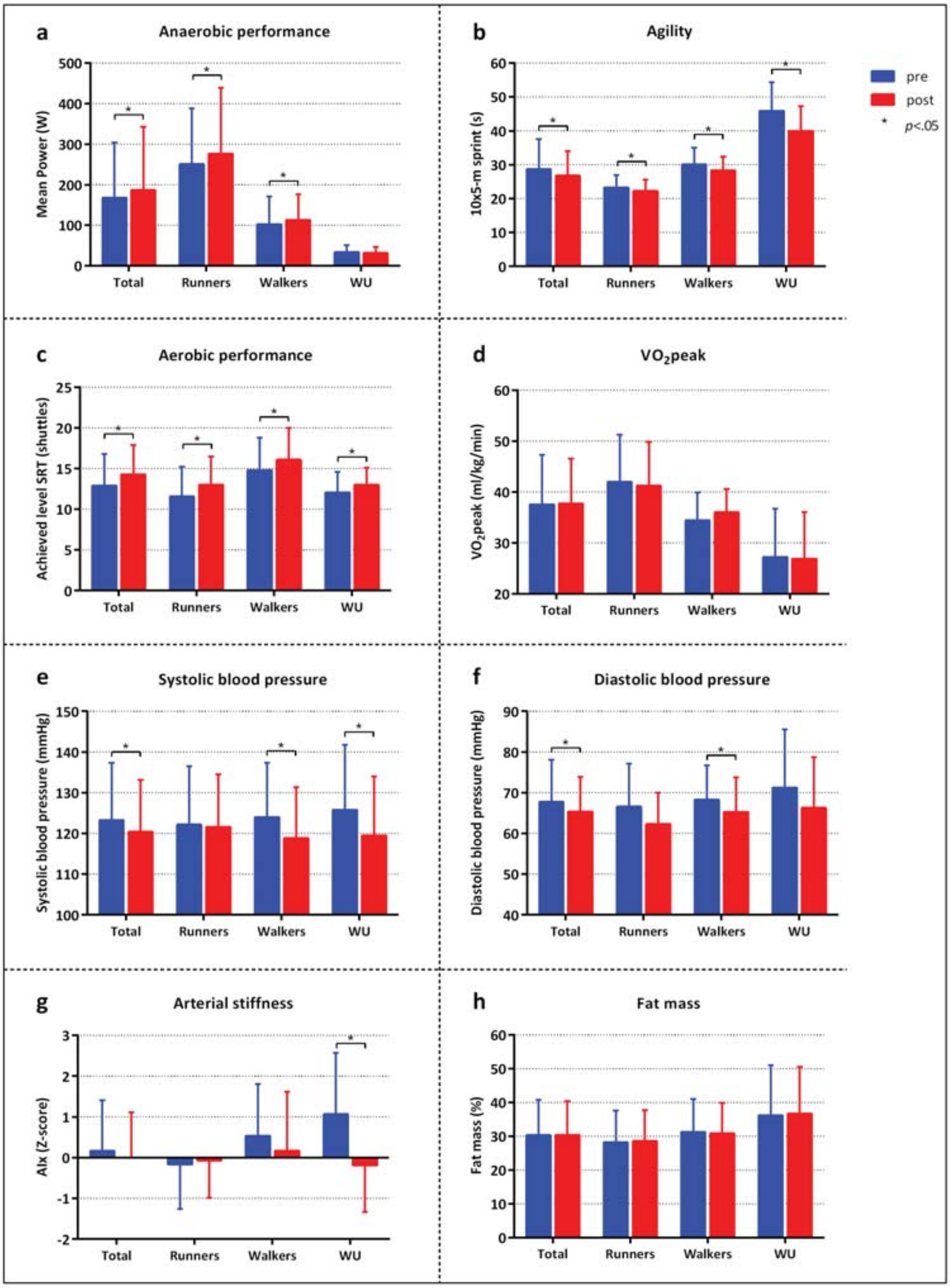

Figure 5.2: Effects of HIT on different outcome measures.
Mean (SD) values pre and post HIT for the total group and subdivided for all subgroups on a) anaerobic performance, b) agility, c) aerobic performance, d) VO_2peak, e) systolic blood pressure, f) diastolic blood pressure, g) arterial stiffness, and h) fat mass. WU, wheelchair user; SRT, shuttle run/ride test; VO_2peak, peak oxygen uptake; AIx, augmentation index.
* Significant difference (p<.05) between pre and post measurement.

Table 5.3: Mean (SD) of physical fitness and cardiometabolic health pre and post HIT

	Total					Runners					Walkers					Wheelchair users				
	n	Pre	Post	*p*	*d*	n	Pre	Post	*p*	*d*	n	Pre	Post	*p*	*d*	n	Pre	Post	*p*	*d*
Height (cm)	70	156 (13.6)	156 (13.6)	<.001*	0.54	36	159 (13.1)	160 (13.2)	.001*	0.58	25	153 (12.9)	154 (12.7)	.001*	0.72	9	149 (15.0)	150 (15.0)	.427	
Weight (kg)	70	54.5 (17.9)	54.9 (18.0)	.062	0.18	36	58.5 (19.1)	58.7 (19.2)	.660		25	51.5 (15.8)	52.2 (16.2)	.005*	0.54	9	46.5 (15.7)	47.2 (15.7)	.085	
PHYSICAL FITNESS																				
Anaerobic performance – MP (W)	67	168 (136)	187 (156)	.002*	0.40	34	249 (139)	280 (163)	.011*	0.46	24	104 (67.8)	113 (64.3)	.020*	0.51	9	36.2 (20.6)	33.2 (15.6)	.463	
Anaerobic performance – PP (W)	67	199 (161)	222 (188)	.009*	0.33	34	295 (165)	335 (197)	.017*	0.43	24	123 (75.8)	131 (69.3)	.060		9	39.7 (23.1)	35.4 (16.7)	.268	
Agility - 10x5 (s)	68	28.7 (8.9)	26.8 (7.2)	<.001*	0.54	35	23.3 (3.7)	22.3 (3.3)	.001*	0.62	24	30.1 (4.9)	28.4 (4.0)	.024*	0.49	9	45.9 (8.4)	40.0 (7.3)	.024*	0.92
Aerobic performance SRT (shuttles)	47	12.9 (3.9)	14.3 (3.6)	<.001*	0.94	24	11.8 (3.6)	13.3 (3.3)	<.001*	1.06	17	14.8 (4.0)	16.1 (3.9)	.006*	0.78	6	12.1 (2.5)	13.0 (2.1)	.012*	1.57
VO_2peak (l/min)	46	2.02 (.77)	2.03 (.71)	.400		24	2.39 (.82)	2.34 (.72)	.541		16	1.75 (.35)	1.87 (.48)	.078		6	1.23 (.47)	1.22 (.36)	.903	
VO_2peak (ml/kg/min)	46	37.6 (9.7)	37.7 (8.8)	.823		24	42.3 (9.4)	41.6 (8.6)	.497		16	34.4 (5.5)	36.0 (4.6)	.141		6	27.2 (9.5)	26.9 (9.1)	.874	
Grip strength (N)	66	151 (76.3)	150 (72.3)	.907		33	172 (86.0)	172 (78.6)	.611		24	135 (55.6)	133 (54.7)	.700		9	114 (69.2)	112 (68.3)	.672	
Standing broad jump (cm)	56	87.4 (35.6)	91.7 (38.7)	.016*	0.33	34	107 (26.6)	112 (33.3)	.042*	0.36	22	56.9 (24.5)	60.8 (23.1)	.204		-	-	-	-	
One stroke push (m)	8	6.52 (.73)	6.48 (1.11)	.897		-	-	-	-		-	-	-	-		8	6.52 (.73)	6.48 (1.11)	.897	
CARDIOVASCULAR HEALTH																				
Systolic blood pressure (mmHg)	65	123 (14.0)	120 (12.8)	.008*	0.34	33	122 (14.3)	122 (12.9)	.714		24	124 (13.4)	119 (12.5)	.006*	0.62	8	126 (16.0)	120 (14.5)	.026*	0.99
Diastolic blood pressure (mmHg)	65	67.8 (10.3)	65.4 (8.5)	.022*	0.29	33	66.6 (10.6)	65.3 (7.7)	.440		24	68.3 (8.4)	65.3 (8.5)	.010*	0.58	8	71.3 (14.2)	66.3 (12.4)	.256	
AIX (%)	55	10.9 (9.3)	9.3 (8.1)	.074		32	8.2 (7.2)	8.3 (6.5)	.900		18	14.0 (10.9)	11.3 (10.5)	.065		5	17.3 (11.0)	8.6 (7.5)	.044*	1.30
AIX (Z-score)	55	0.17 (1.24)	-0.00 (1.12)	.176		32	-0.17 (1.09)	-0.07 (0.92)	.461		18	0.54 (1.27)	0.17 (1.45)	.057		5	1.07 (1.50)	-0.19 (1.14)	.050*	1.24
PWV (m/s)	55	5.8 (0.89)	5.8 (0.82)	.575		32	5.8 (0.90)	5.8 (0.75)	.837		18	6.0 (0.81)	5.8 (0.85)	.150		5	5.4 (1.1)	5.4 (1.2)	.898	

METABOLIC HEALTH																			
BMI (kg/m^2)	70	22.2 (4.8)	22.3 (5.1)	.647	36	22.7 (5.1)	22.6 (5.2)	.576		25	21.6 (4.7)	21.6 (4.7)	.394	9	22.4 (4.7)	23.3 (5.8)	.354
BMI (Z-score)	70	1.31 (1.40)	1.29 (1.40)	.526	36	1.42 (1.43)	1.34 (1.44)	.152		25	1.14 (1.44)	1.13 (1.41)	.683	9	1.38 (1.27)	1.52 (1.35)	.531
Waist circumference (cm)	69	79.2 (14.5)	79.4 (14.1)	.665	35	79.3 (14.8)	80.3 (15.0)	.257		25	80.4 (15.1)	79.4 (13.4)	.117	9	75.6 (12.3)	76.2 (13.4)	.700
Waist-Hip ratio	69	0.94 (0.08)	0.95 (0.07)	.098	35	0.92 (0.06)	0.94 (0.06)	.037*	0.37	25	0.97 (0.09)	0.99 (0.07)	.246	9	0.95 (0.10)	0.92 (0.09)	.234
Fat mass (%)	65	30.4 (10.4)	30.4 (10.0)	.986	33	28.3 (9.4)	28.6 (9.2)	.502		24	31.4 (9.6)	30.9 (9.0)	.288	8	36.3 (14.8)	36.7 (13.9)	.581
Total cholesterol (mmol/l)	46	3.80 (0.67)	3.81 (0.68)	.943	24	3.78 (0.75)	3.89 (0.76)	.354		16	3.82 (0.66)	3.71 (0.66)	.377	6	3.85 (0.37)	3.74 (0.32)	.453
HDL (mmol/l)	46	1.23 (0.36)	1.25 (0.36)	.342	24	1.30 (.43)	1.35 (0.43)	.207		16	1.17 (0.28)	1.16 (0.17)	.970	6	1.07 (0.17)	1.09 (0.32)	.977
LDL (mmol/l)	34	2.25 (0.56)	2.20 (0.51)	.498	17	2.19 (0.58)	2.15 (0.49)	.744		12	2.29 (0.62)	2.25 (0.63)	.794	5	2.39 (0.32)	2.26 (0.27)	.452
Total cholesterol/HDL	46	3.36 (1.15)	3.24 (0.89)	.166	24	3.24 (1.34)	3.13 (1.02)	.395		16	3.41 (0.93)	3.27 (0.73)	.280	6	3.70 (0.84)	3.59 (0.69)	.705
Triglyceride (mmol/l)	46	1.01 (0.59)	1.11 (0.68)	.259	24	1.10 (0.69)	1.25 (0.81)	.108		16	0.93 (0.47)	0.98 (0.54)	.887	6	0.87 (0.40)	0.89 (0.32)	.917
Glucose (mmol/l)	34	4.68 (0.61)	4.80 (0.58)	.268	16	4.88 (0.69)	5.11 (0.54)	.252		14	4.44 (0.51)	4.57 (0.51)	.377	4	4.69 (0.29)	4.40 (0.19)	.304

d, effect size; MP, mean power; PP, peak power; SRT, shuttle run/ride rest; VO_2peak, peak oxygen uptake; AIX, augmentation index; PWV, pulse wave velocity; BMI, body mass index; HDL, high density lipoprotein; LDL, low density lipoprotein.
*p<.05.

5

Metabolic health

In the total group, no changes were found in BMI, waist circumference, waist-hip ratio and fat mass (Figure 5.2h), nor in any of the subgroups, except from an increase in waist-hip ratio in the runners subgroup (MD=0.02, *p*=.037). In addition, no differences were found on the metabolic profile; total cholesterol, HDL, LDL, triglyceride, and glucose.

DISCUSSION

In the current study, we investigated the short-term effects of HIT on physical fitness and cardiometabolic health in youth with physical disabilities including different levels of mobility. Anaerobic performance improved in the total group as well as in the runners and walkers subgroups, except for peak power in the walkers subgroup. Effect sizes showed medium effects for anaerobic performance. Agility improved with medium effect sizes across all groups. For aerobic performance, large effect sizes were found in all subgroups, while VO_2peak did not change. Furthermore, no changes were found on cardiometabolic health for the total group, except for a decrease in resting systolic and diastolic blood pressure.

Compared to the current study, similar results in anaerobic performance were found following a 30 seconds all-out protocol on a cycle ergometer in healthy young adults.[7] In the latter study, improvements of 7% and 17% for mean and peak power were found compared to 9% and 10% in the current study respectively. Another HIT program in overweight children consisted of 16 intervals of 15 seconds at 100% of maximal running speed followed by 15 seconds of active recovery.[13] They found similar effects after 6 weeks; 11% improvement in aerobic performance on a SRT, compared to 11% in this study. In contrast, an eight months training intervention comprised of both aerobic and anaerobic exercise resulted in improvements of 37% in aerobic performance on a SRT in children and adolescents with CP.[3] Both the length of the program and lower anaerobic performance at baseline may declare the discrepancy.[28]

For VO_2peak, a recent study in adolescents with cerebral palsy reported a significant improvement of 10% in VO_2peak (ml/kg/min) after 24 HIT sessions on a treadmill.[14] They used an individualized protocol of 1.5–4 minutes interval resulting in 16 minutes at >85% of peak heart rate in total. However, these results were based on only eight children with CP. Since literature on 30 seconds all-out protocols on VO_2peak in youth with physical disabilities is lacking, the effects on VO_2peak remain unclear. For improving VO_2peak with short-term HIT in adults who are healthy, a recent meta-analysis suggests longer intervals (i.e., 3–5 min) and a training frequency of three times a week.[29] To improve VO_2peak within eight weeks in this population, we suggest for future research to increase training frequency to three times a week and/or increase time per interval (i.e., training volume).

In the current study, baseline levels of resting systolic blood pressure were slightly elevated compared to non-overweight reference values in typically developing youth.[25] For children 13 years of age reference values of systolic blood pressure for boys are 117 mmHg and for girls 115 mmHg compared to an average of 123 mmHg in the current study. Although both systolic (MD=-2.9 mmHg) and diastolic (MD=-2.4 mmHg) blood pressure decreased after HIT, the improvement is relatively small and the clinical relevance can be argued. Furthermore, the decrease in blood pressure did not hold true for all subgroups. These findings are in line with a recent review showing inconsistent effects of HIT on blood pressure in adults with common metabolic diseases.[9]

To our knowledge, this is the first study that measured the effect of HIT on arterial stiffness in youth with physical disabilities. Arterial stiffness, which is regarded to be the first sign of arteriosclerosis, was for the total group between normal ranges at baseline.[24,25] This presumably explains why there was no reduction after training, similar to previous findings in other populations.[30] However, in wheelchair users, AIX Z-scores were slightly elevated at baseline. In this subgroup a significant improvement can be seen in AIX, although interpretation must be done carefully with the low number of participants (n=5). Probably, especially children and adolescents at risk at baseline seem to benefit from HIT regarding arterial stiffness.

Recent studies, published after our design paper,[15] have shown that positive effects of HIT on body composition occur either when HIT is performed long-term (>12 weeks), and/or when participants are overweight at baseline.[8,12,13] This may explain why we found no effects on body composition. Similar results were found after HIT in children with CP; no effects on BMI and fat mass were shown.[14] The negative effect on waist-hip ratio in the runners subgroup could probably be explained by a measurement error, since the hip circumference decreased unexpectedly. Despite the fact that some studies found improvements in the metabolic profile after HIT, especially on HDL, triglycerides and fasting glucose, the evidence is inconsistent.[8] A recent study with meta-analysis showed a positive, but small, effect on fasting glucose in overweight/obese adults, but not in the normal population.[8] The results we present are in accordance with recent findings showing that there is no evidence for improvement of the metabolic profile after an short-term HIT program, in case baseline values are within normal ranges.[8]

Several limitations should be taken into account. Firstly, due to a small sample size (n=9), results of the wheelchair users subgroup should be considered exploratory. Secondly, since a control group with random assignment was lacking, current results do not account for natural development or other possible confounders. Thirdly, the study population comprised a large age range, a variety of diagnoses and mobility levels with consequently different methodologies between subgroups. This was however a deliberate choice, aimed to increase our sample size, and in the knowledge that many disabilities reported in literature, such as CP, are often heterogeneous. Furthermore these heterogeneity actually reflects real-life as they probably exercise together at the schools they are recruited. In addition, the total number of sessions conducted within eight weeks of HIT might not have been high enough to elicit improvements, but is again a reflection of daily life. A fourth limitation of this study is the drop-out rate of 33% in aerobic fitness. On the other hand, in a similar population higher drop-out rates were found before. A study in children with CP reported a drop-out rate of 44% in two maximal exercise tests using similar criteria for maximal effort.[23]

CONCLUSIONS

Following eight weeks of HIT, youth with physical disabilities improved anaerobic performance, agility and aerobic performance, with no changes in VO_2peak. No effects were found for cardiometabolic health, except for a decrease in blood pressure. For clinical practice, 30 seconds all-out is feasible and safe in youth with physical disabilities and is useful in improving exercise performance short-term. Future research should focus on wheelchair users specifically. To improve VO_2peak short-term, we suggest for future research to increase training frequency to three times a week and/or increase time per interval (i.e., training volume). Regarding health, research should focus on children and adolescents at risk at baseline.

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6

Effects of a school-based sports program on physical fitness, physical activity and cardiometabolic health in youth with physical disabilities: data from the Sport-2-Stay-Fit study

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ABSTRACT

Objective: To investigate the effects of a school-based once a week sports program on physical fitness, physical activity, and cardiometabolic health in children and adolescents with a physical disability.

Methods: This controlled clinical trial included 71 children and adolescents from four schools for special education (mean age 13.7 (2.9) years, range 8–19, 55% boys). Participants had various chronic health conditions including cerebral palsy (37%), other neuromuscular (44%), metabolic (8%), musculoskeletal (7%), and cardiovascular (4%) disorders. Before recruitment and based on the presence of school-based sports, schools were assigned as sport or control group. School-based sports was initiated and provided by motivated experienced physical educators. The sport group (n=31) participated in a once a week school-based sports program for six months, which included team sports. The control group (n=40) followed the regular curriculum. Anaerobic performance was assessed by the Muscle Power Sprint Test. Secondary outcome measures included aerobic performance, VO_2peak, strength, physical activity, blood pressure, arterial stiffness, body composition and the metabolic profile.

Results: A significant improvement of 16% in favour of the sport group was found for anaerobic performance (p=.003). In addition, the sport group lost 2.8% more fat mass compared to the control group (p=.007). No changes were found for aerobic performance, VO_2peak, physical activity, blood pressure, arterial stiffness, and the metabolic profile.

Conclusions: Anaerobic performance and fat mass improved following a school-based sports program. These effects are promising for long-term fitness and health promotion, because sports sessions at school eliminate certain barriers for sports participation and adding a once a week sports session showed already positive effects over six months.

INTRODUCTION

Daily physical activity is beneficial for all children and adolescents. For those with physical disabilities similar physical activity recommendations account.[1] In addition, exercise interventions have shown that youth with physical disabilities can improve physical fitness levels[2–7] and decrease cardiometabolic risk factors.[4–7] Moreover, they have lower levels of physical fitness and participate less in competitive and recreational sports compared to peers who develop typically.[3,8] Since limited physical ability can interfere with being physically active in daily life and consequently affect their health later in life, maintaining sports participation and adequate performance-related fitness levels is especially important in this population.[9,10]

For youth with a physical disability it seems more difficult to participate in sports and physical activities when compared to typically developing peers.[11] In 2011, only 26% of Dutch children and adolescents with a physical disability from schools for special education participate in sports at least once a week compared to 71% in youth without physical disabilities.[12] Reasons for this lower sports participation are: being physically active is more challenging because of their disability, lack of trained support personnel, transportation problems, lack of acceptance, and no sports clubs in the neighborhood.[13–15] Most of these barriers to be physical active, can possibly be eliminated when a sports program is provided at school in the immediate after-school hours. This setting offers a familiar environment with supported trainers, acceptance, and no additional transportation except a postponed pick-up from school.

Additionally, a recent study has proposed the need for school-based initiatives, since their integrative approach is effective and even target the least active children.[16] Recent work in the typically developing population showed positive but inconsistent effects in increasing physical activity with after-school interventions.[17,18] Interventions which lasted more than 12 weeks and that focused solely on increasing physical activity by providing a sports program were most effective.[18] According to the exercise principles, a training frequency of at least three sessions a week is recommended to improve cardiorespiratory fitness in youth who are typically developing.[19,20] In contrast, for children and adolescents with cerebral palsy (CP) who are very deconditioned, two sessions a week is also possible to induce or maintain effects.[1] On the other hand, from a parents' perspective, they consider sports participation of once a week to be sufficient.[21] Parents need to prioritise the frequency of sports participation with the demands of everyday life.[22] In a recent qualitative research, all parents indicated that the intensity or frequency of a sports program to improve fitness levels was not of importance for them. They emphasized the importance of sports as "being active", having fun, and socialization.[21]

For improving fitness levels, there is a discrepancy between exercise recommendations and feasibility in daily life for youth with disabilities and their parents. Providing a once a week school-based sports program can relatively easy be implemented and increase children's level of sports participation. However, it is unknown whether once a week is beneficial for fitness and health purposes in this population. The current study, denoted as the Sport-2-Stay-Fit study, will investigate the effects of a school-based once a week sports program on physical fitness, physical activity, and cardiometabolic health in children and adolescents with a physical disability.

METHODS

Design

The Sport-2-Stay-Fit study is a controlled clinical trial. The study was conducted at four schools for special education in the Netherlands between September 2014 and July 2016. In a previous publication we described the study design extensively.[23] The results of the school-based sports program will be published in two separate papers where this paper focuses on the fitness and health aspects. Both ethics approval and administrative site approvals were granted by the Medical Ethical Committee of UMC Utrecht in the Netherlands (#14-118). This trial was registered with the Dutch Trial Registry (NTR4698).

Participants

Children and adolescents with a physical disability were recruited via four schools for special education in the Netherlands. These schools, dedicated to youth with physical disabilities, have similar learning objectives as regular schools, but the children receive additional attention and support. Children and adolescents were screened by a physical therapist, physical educator or physician for eligibility. Inclusion criteria were: (1) a chronic disease or physical disability; neuromuscular, musculoskeletal, metabolic or cardiovascular disorder, (2) aged between 6 and 19 years, (3) participation in sports less than twice a week during leisure time in the preceding three months or advised to participate in sports by their physical therapist or physician, (4) understands simple instructions, and (5) were expected to be able to perform the physical fitness tests. Exclusion criteria were (1) having a progressive disease, (2) using a powered wheelchair for sport purposes, (3) participation in other research that could possibly influence current results. Additionally, all parents, and participants from 12 years of age, provided informed consent prior to study initiation.

Procedure

Before recruitment, schools (n=4) were assigned as sport or control site. This was directed by current presence of school-based sports initiated by motivated physical educators. Schools who already provided (n=1) or intended to provide (n=1) school-based sports in addition to the regular curriculum, were assigned sport sites. Otherwise, schools were assigned as control sites (n=2). In case school-based sports had been provided before the start of the study, children and adolescents were only included if they had not participated during the preceding three months. Regardless of the group enrolled, all participants followed a high-intensity interval training (HIT) for eight weeks as an initial start-up for their fitness level and to get familiarized with exercise.[23] In this way, all participants knew in what group they would be enrolled: HIT and school-based sports (i.e., sport group) or HIT and control (i.e., control group). The focus of the current study is the school-based sports program, results of the HIT are described elsewhere. [24] Outcome measures were assessed at baseline (T0), after eight weeks of HIT (T1) and at completion of six months intervention (T2). Similar outcome measures were evaluated at T1 and T2 except for physical activity. Because no short-term effect was expected on physical activity following HIT (T1),[25,26] baseline physical activity (T0) was used to analyse the effectiveness. All outcome measures across all schools were assessed by the same trained researcher (MZ) together with research assistants. The assessors were not blinded for group allocation.

Intervention

All participants performed HIT for eight weeks, twice a week for 30 minutes, containing 8–12 series of 30 seconds all-out exercises. Detailed information about the training schedule is described elsewhere.[23] After eight weeks, the sport group commenced with the school-based sports program. The program was provided once a week for 45 minutes by an experienced physical educator at school additional to the regular physical education schedule. In contrast to regular physical education where half of the time is spend on skill practice, cooperation and management,[27] sport is about moving more intensively. No instructions were given on exercise intensities, but children and adolescents were encouraged to be physically active, play the game and have fun.[21] The content of the lesson was adapted by the physical educator based on the children's skills- and cognitive level. The sports program included, but was not restricted to, soccer, (wheelchair) basketball, (wheelchair) hockey, and/or easy administered games like playing tag. The presence of the participants was documented every session.

Outcome measures

Since intermittent bouts of intense exercise reflects children's daily activity pattern, the primary outcome was anaerobic performance.[28] Outcome measures were tested during school hours, except from physical activity, and were subdivided in four different occasions within two weeks: a) height, weight, anaerobic fitness and strength, b) aerobic fitness, c) blood pressure, arterial stiffness and body composition, and d) metabolic profile.

Physical fitness

Anaerobic fitness

Anaerobic performance was measured with the Muscle Power Sprint Test (MPST) either while running, walking or propelling a manual wheelchair as described previously.[29,30] This is an intermittent sprint test consisting of three or six 15-meter sprints with a standardized rest of 10 seconds between sprints. Participants who were ambulatory had to complete six 15-meter runs, while wheelchair users completed three 15-meter sprints at maximal pace. Both peak and mean power were calculated from the results of the sprints. To assess agility, time was recorded during a 10x5 meter sprint test where children or adolescents had to sprint 10 times as fast as possible between two lines five meter apart without rest.[29]

Aerobic fitness

For aerobic fitness, both performance (achieved shuttles) and VO_2peak were measured during a 10-m shuttle run/ride test (SRT).[31,32] The SRT is an incremental exercise test where participants had to adjust their running, walking or wheelchair propulsion pace to the beep-signals until they failed to reach the line twice within one level. The test protocol was selected based on their level of mobility as described previously.[23] During the SRT a calibrated mixing chamber Cortex Metamax 3X (Samcon bvba, Melle, Belgium) was used to measure VO_2peak. Metabolic stress test software (Metasoft Studio) was used to measure oxygen uptake (VO_2), carbon dioxide production (VCO_2), peak heart rate, and respiratory exchange ratio ($RER=VCO_2/VO_2$). Each test lasted until exhaustion. To determine whether a subject reached their maximal effort, two out of the following three criteria had to be achieved: heart rate ≥180 bpm, RER ≥1.00 at peak exercise, or subjective signs of intense effort, such as sweating, facial flushing or a clear unwillingness to continue.

Strength

Grip strength was measured using a hand held dynamometer (CITEC, CIT Technics, Haren, the Netherlands). Mean grip strength was calculated out of three attempts with the preferred hand. To assess explosive strength either the standing broad jump or one stroke push was performed in those who were ambulatory and propelling a wheelchair, respectively.[33] The standing broad jump referred to the distance jumped with two legs together, while one stroke push referred to the distance covered in their wheelchair by one push.

Physical activity

Total physical activity was measured objectively using the Activ8 activity monitor (2M Engineering, Valkenswaard, the Netherlands). The system measures acceleration in three planes and is valid to detect six types of activities in persons who are ambulatory: lying, sitting, standing, walking, cycling and running.[34] Participants wore the Activ8 for seven consecutive days. The device was fixed with Tegaderm™ waterproof skin tape on the ventral side of the upper leg allowing participants to take a shower or swim. At least two schooldays with a minimum of 600 minutes wear time was needed for a representative weekday. For weekend days at least one day of 600 minutes wear time was required.[35] Time spent lying and sitting (sedentary time), and time spent standing, walking, cycling and running (active time) was calculated in minutes. Children and adolescents who were manual wheelchair-using (n=9) wore the device as well. However, we omitted the data from the analyses since this device has not yet been validated for wheelchair users.

Cardiometabolic health

Cardiovascular

Both resting blood pressure and arterial stiffness were noninvasively measured with the Arteriograph (Litra BV, Amsterdam, the Netherlands). The measurement was performed in a supine position using an inflatable cuff on the right upper arm. Participants rested supine for 10 minutes prior to the recording, and they were asked not to move or talk during the test. Arterial stiffness contained two independent values: pulse wave velocity (PWV), and the augmentation index (AIx). The PWV was measured as the speed at which an aortic pulse travels; increased speed indicates stiffer arteries. The AIx provides information on the peripheral resistance of the endothelial vessels; increased index indicates higher peripheral resistance. To control for differences in sex and age, Z-scores of AIx were calculated according to reference values of Hidvégi et al.[36]

Metabolic

Height and weight were measured to determine body mass index (BMI). A detailed description has been described previously.[23] To control for differences in age, Z-scores of BMI were calculated according to Dutch reference values.[37] For waist and hip circumference, a horizontal measure was taken at the umbilicus and trochanter major, respectively. Fat mass was measured in supine position with bioelectrical impedance analysis, using the Bodystat Quadscan 4000 (Euromedix, Leuven, Belgium). To determine the metabolic profile, a finger puncture was performed. This was an optional measurement and consent was asked separately to parents and participants from 12 years of age. During this procedure blood was drawn through a finger puncture from which total cholesterol, low density lipoprotein (LDL), high density lipoprotein (HDL), fasting glucose and triglyceride were analysed. The analyses were performed using a Cholestech LDX analyzer (Mediphos Medical Supplies BV, Renkum, the Netherlands). All participants were instructed not to eat or drink for three hours prior to this procedure. Before the finger puncture, participants were asked about the fasting period and if possible the measurement was postponed. Otherwise only total cholesterol, LDL, HDL, and triglyceride data were used for analysis.[38]

Data analysis

A sample size calculation showed that 32 participants per subgroup was needed to detect a 20% difference between groups in anaerobic performance.[23] Statistical analysis was performed using SPSS for Windows (version 21.0, SPSS Inc, Chicago, Ill) with a statistical significance level of p=.01 to correct for testing multiple hypotheses. Descriptive statistics were presented as means and standard deviation (SD). To determine the intervention effect, linear regression analyses were performed according to the intention to treat principle. In the linear regression analyses, outcome measures at T2 were the dependent variables, with group allocation and the measured outcome at T1 as independent variables. Since participants were not randomly allocated, outcome measures at T1 were included in the analyses to correct for potential baseline differences between groups. In addition, baseline differences in subject characteristics were checked. Subgroup response patterns on age, sex and mobility level were analysed and included as confounders in the analyses when they changed the intervention effect. Besides, response patterns of the different schools were analysed and included as cluster variable if they changed the intervention effect. Data were graphically checked for normal distribution using residual plots. Variables with non-normally distributed residuals were logarithmically transformed prior to linear regression, after which the results were transformed back, providing a between-group regression coefficient which has to be interpreted as a ratio. The residuals of all variables were normally distributed after logarithmic transformation. Since the dataset was expected to contain incomplete data for some variables, we used multiple imputation to create and analyse 10 imputed datasets. The imputation model included the outcome measures and sex, height, age, weight and mobility level in the regression model. Regression coefficients (β), and the 95% confidence interval (CI) were reported for the regression model. For clinical purposes, the estimated marginal means (EMM) of both groups, the mean difference (MD) and relative effect (%) were calculated. Linear regression analyses were both performed on the original data and multiple imputation data. Since both models resulted in similar effects, only the multiple imputation model is shown here.

RESULTS

A total of 138 participants were invited to participate between September 2014 and November 2015 of whom 78 decided to participate. Seventy-one children and adolescents participated in the current study (Table 6.1, Figure 6.1). Due to practical reasons, one participant of the sport group was not able to attend the sports program and was therefore

Table 6.1: Characteristics of participants

	Sport (n=31)	Control (n=40)
Age (y), mean (SD)	13.4 (3.0)	14.0 (2.8)
Sex, n male (%)	23 (74)	16 (40)
Height (cm), mean (SD)	156.4 (17.5)	157.1 (10.4)
Weight (kg), mean (SD)	54.4 (20.8)	56.9 (18.6)
Level of mobility, n (%)		
Able to run	22 (71)	15 (38)
Able to walk	6 (19)	19 (47)
Wheelchair user	3 (10)	6 (15)
Diagnoses, n (%)		
Neuromuscular	22 (71)	35 (87)
Cerebral palsy	10 (32)	16 (40)
Spina bifida	-	5 (12)
Psychomotor retardation	4 (13)	5 (12)
Acquired brain injury	2 (97)	1 (3)
Other	6 (19)	8 (20)
Cardiovascular	2 (6)	1 (3)
Metabolic	4 (13)	2 (5)
Musculoskeletal	3 (10)	2 (5)

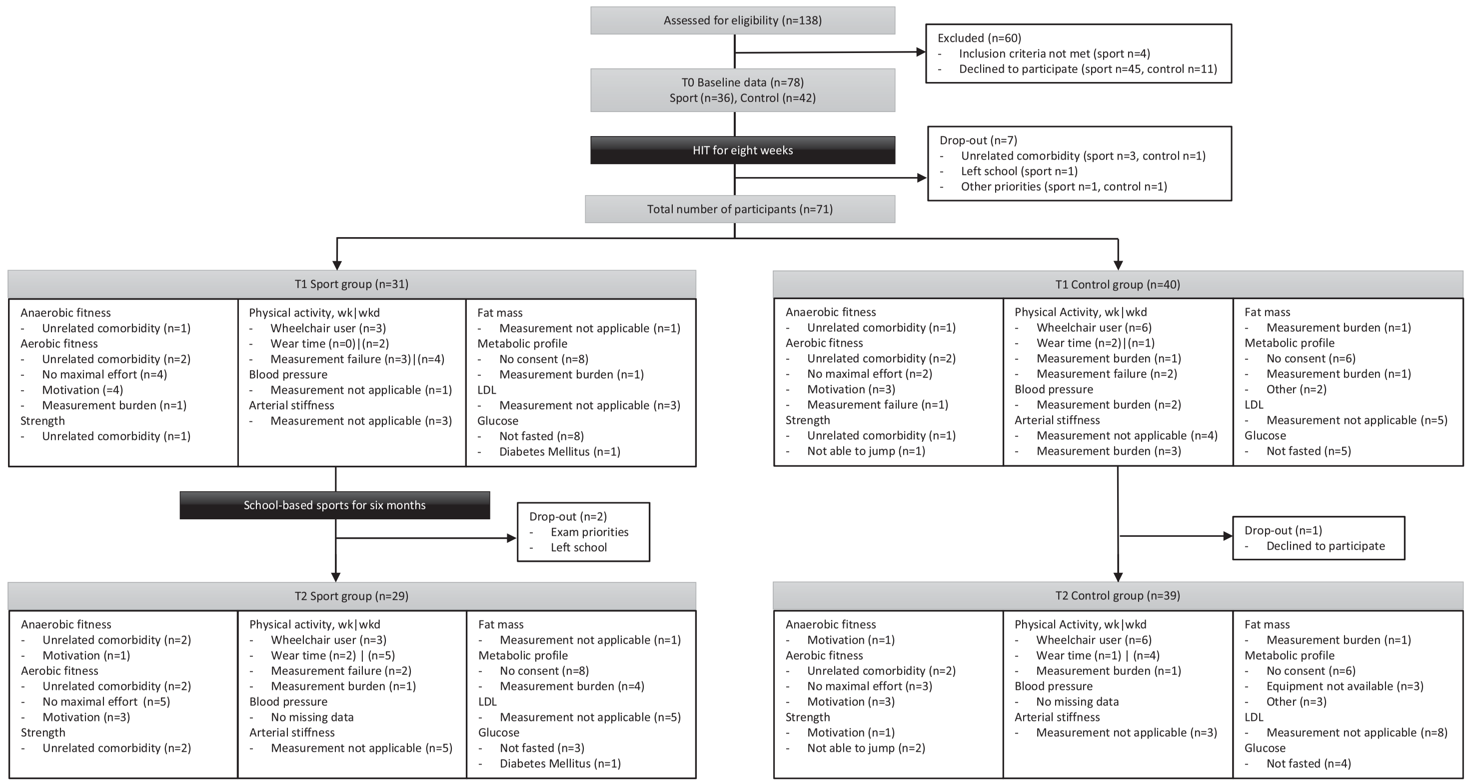

Figure 6.1: Flowchart from initial inclusion to intervention and reasons for missing data per outcome measure.

assigned to the control group. Prior to the start of the study, sports participation at T0 did not differ between the sport (1.1 (1.1) times a week) and control group (0.8 (0.8) times a week) (*p*=.285), whereas at T2 sports participation differed significantly between the sport (2.1 (1.0) times a week) and control group (0.9 (0.8) times a week) (*p*<.001).

Following the school-based sports program, three participants dropped out and some participants did not complete all assessments as illustrated in Figure 6.1. According to the finger puncture, 74% and 85% of the participants provided consent for the sport and control group, respectively. All measured data at T1 and T2 is shown for physical fitness and physical activity in Table 6.2 and for cardiometabolic health in Table 6.3.

Adherence to the sports program was on average 86%, with children and adolescents attending on average 14.4 (4.1) with a range of 5 to 20 sport sessions. To illustrate, five adolescents attended less than 75% of the program, due to surgery (n=1), other priorities (n=1) and truancy (n=3). No adverse events related to the sports program were reported. Time between measurements was 6.6 (1.3) months with a range of 4.6–10.6 months. The sport group was measured within two weeks after finishing the sports program. The huge range between measurement is due to participants of the control group who left school during the study period and agreed to return to finish the assessments on a different occasion.

Effect of intervention

Physical fitness

As shown in Figure 6.2a and Table 6.4, a significant effect in favour of the sport group was found for anaerobic performance on mean power (β=1.16, IC=1.05–1.28[1]) and peak power (β=1.15, IC=1.04–1.27[1]). The between-group difference was 23 Watt (16%) and 25 Watt (15%) for mean and peak power respectively. No significant effect was observed for agility (Table 6.4). In addition, no intervention effect was demonstrated for aerobic performance (Figure 6.2b), VO_2peak and strength (Table 6.4).

Physical activity

At baseline no seasonal differences were found among participants measured in the autumn, winter and spring. Sports participation was increased during the school-based sports program in the sport group with 1.2 (0.9) times a week compared to 0.1 (0.9) times a week in the control group (*p*<.001). Between group differences showed no intervention

1 Regression coefficients should be interpreted as a ratio as it was logarithmically transformed.

Table 6.2: Mean (SD) of physical fitness and physical activity before (T1) and after (T2) the school-based sports program

		T1				T2			
		n	Sport Mean (SD)	n	Control Mean (SD)	n	Sport Mean (SD)	n	Control Mean (SD)
Anaerobic fitness	Anaerobic performance – MP (W)	30	247.9 (188.7)	39	144.0 (107.2)	26	279.2 (208.7)	38	146.3 (109.5)
	Anaerobic performance – PP (W)	30	300.0 (226.5)	39	167.3 (128.3)	26	326.7 (227.3)	38	168.7 (124.7)
	Agility (s)	30	24.6 (6.9)	39	28.4 (7.0)	26	25.2 (8,4)	38	28.2 (6.9)
Aerobic fitness [a]	Aerobic performance (shuttles)	21	13.5 (3.7)	33	14.0 (4.2)	19	12.6 (3.4)	31	13.6 (4.6)
	VO_2peak (ml/kg/min)	20	42.6 (8.8)	32	34.6 (7.0)	19	37.9 (7.2)	31	34.3 (8.0)
	VO_2peak (ml/fat free mass/min)	20	58.2 (9.7)	32	49.9 (9.4)	19	51.9 (6.1)	31	50.0 (9.9)
	HRpeak (bpm)	20	194 (11)	32	184 (16)	19	192 (11)	31	185 (14)
	RER	20	1.11 (0.11)	32	1.10 (0.09)	19	1.11 (0.07)	31	1.11 (0.10)
	VEpeak (l/min)	20	79.6 (35.6)	32	69.5 (28.0)	19	72.9 (33.7)	31	70.2 (27.2)
Strength	Grip strength (N)	30	153.1 (88)	38	144.7 (57.6)	27	147.8 (84.8)	38	149.2 (56.2)
	Standing broad jump (cm) [b]	27	110.9 (40.6)	31	71.9 (28.5)	24	110.0 (40.3)	30	75.8 (23.5)
	One stroke push (m) [c]	3	7.65 (0.83)	6	6.78 (2.50)	3	6.00 (0.97)	6	6.21 (2.09)
Physical activity [b]	Sedentary time – week (min)	25	588.8 (90.4)	29	554.1 (101.9)	21	551.1 (79.2)	31	562.0 (85.1)
	Sedentary time – weekend (min)	22	518.3 (112.0)	28	540.1 (85.1)	18	494.9 (151.8)	28	544.5 (106.1)
	Active time – week (min)	25	271.7 (63.3)	29	259.2 (104.4)	21	302.1 (69.8)	31	282.1 (83.6)
	Active time – weekend (min)	22	281.7 (86.3)	28	229.1 (80.9)	18	301.7 (117.1)	28	245.9 (90.3)

MP, mean power; PP, peak power; HR, heart rate; RER, respiratory exchange ratio; VE, ventilation.
[a] Included only participants who reached maximal effort.
[b] Included only participants who were ambulatory.
[c] Included only wheelchair users.

Table 6.3: Mean (SD) of cardiometabolic health before (T1) and after (T2) the school-based sports program

			T1				T2			
		Ref[a]	n	Sport Mean (SD)	n	Control Mean (SD)	n	Sport Mean (SD)	n	Control Mean (SD)
Cardiovascular	Systolic blood pressure (mmHg)	<130	30	119.6 (15.3)	38	121.9 (11.4)	29	120.8 (14.6)	39	123.7 (1323)
	Diastolic blood pressure (mmHg)	<80	30	64.1 (8.4)	38	67.1 (8.4)	29	65.8 (11.3)	39	71.0 (10.3)
	AIx (%)	<15	28	9.82 (9.93)	33	9.58 (8.45)	24	9.50 (10.1)	36	12.0 (8.34)
	AIx (Z-score)	NA	28	0.06 (1.25)	33	0.03 (1.22)	24	0.01 (1.16)	36	0.49 (1.15)
	PWV (m/s)	<7	28	5.74 (0.89)	33	6.02 (0.94)	25	5.95 (0.94)	36	6.03 (0.81)
Metabolic	BMI (kg/m^2)	<23	31	21.7 (5.4)	40	23.2 (5.2)	29	22.2 (6.1)	39	23.5 (5.1)
	BMI (Z-score)	NA	31	1.15 (1.54)	40	1.45 (1.33)	29	1.16 (1.64)	39	1.39 (1.39)
	Waist-circumference (cm)	<85	31	77.3 (14.8)	39	82.4 (15.3)	29	78.4 (16.4)	39	84.3 (16.1)
	Waist-hip ratio	<0.95	31	0.96 (0.05)	39	0.95 (0.08)	29	0.97 (0.06)	39	0.99 (0.07)
	Fat mass (%)	<25	30	26.7 (9.8)	39	32.4 (10.0)	28	25.9 (9.5)	38	32.7 (8.93)
	Total cholesterol (mmol/l)	3.0–5.0	22	3.88 (0.84)	31	3.85 (0.57)	17	3.78 (0.80)	27	3.99 (0.58)
	HDL (mmol/l)	>1.0	22	1.37 (0.48)	31	1.18 (0.29)	17	1.31 (0.46)	27	1.23 (0.30)
	LDL (mmol/l)	<3.2	19	1.93 (0.55)	26	2.32 (0.46)	12	2.07 (0.50)	19	2.28 (0.52)
	Total cholesterol/HDL	<5.0	22	3.07 (1.00)	31	3.43 (0.87)	17	3.14 (1.07)	27	3.43 (0.96)
	Triglyceride (mmol/l)	0.6–2.2	22	1.56 (1.00)	31	0.96 (0.52)	17	1.10 (0.72)	27	1.28 (0.94)
	Glucose (mmol/l) [b]	3.5–5.6	13	5.05 (0.70)	26	4.70 (0.46)	13	4.80 (0.62)	23	5.00 (0.55)

AIx, augmentation index; PWV, pulse wave velocity; BMI, body mass index; HDL, high density lipoprotein; LDL, low density protein.

[a] Cutoff references values of Hidvegi et al. 2015; Talma et al. 2010; Bodystat Quadscan 4000 and Cholestech LDX software.

[b] Included only participants who were fasted.

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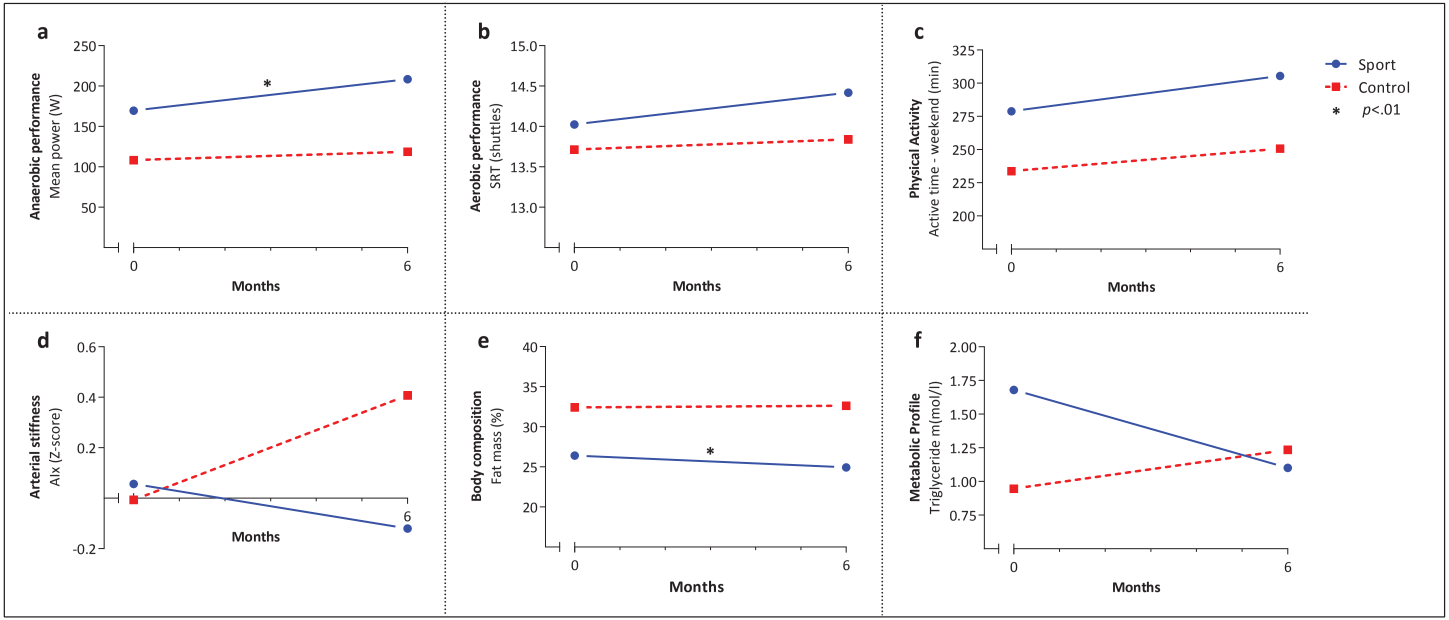

Figure 6.2: Effects of the school-based once a week sports program.
Mean values for the sport (solid blue line) and control (dashed red line) group before (T1) and after (T2) the school-based sports program of a) anaerobic performance, b) aerobic performance, c) physical activity, d) arterial stiffness, e) body composition, f) metabolic profile on the multiple imputed model. Linear regression analyses were done on the multiple imputed model adjusted for T1. * Significant (*p*<.01) effect in favor of the sport group.

Table 6.4: Results from the linear regression analyses assessing the intervention effect on physical fitness and physical activity

		Linear regression[a]		Estimated marginal means Differences between groups			
		β	95% CI	Sport	Control	MD	%
Anaerobic fitness	Anaerobic performance – MP (W) [b]	1.16	1.05–1.28*	164.8	142.2	22.6	16
	Anaerobic performance – PP (W) [b]	1.15	1.04–1.27*	192.3	167.1	25.2	15
	Agility (s) [b]	1.02	0.96–1.07	25.8	25.4	0.41	1.6
Aerobic fitness	Aerobic performance (shuttles)	0.31	-1.19–1.81	14.3	14.0	0.31	2.2
	VO_2peak (ml/kg/min)	-1.55	-4.66–1.56	35.5	37.0	-1.55	-4.2
	VO_2peak (ml/fat free mass/min)	-3.00	-7.92–1.92	49.5	52.5	-3.00	-5.7
Functional strength	Grip strength (N)	-7.72	-17.97–2.53	147.6	155.3	-7.72	-5.0
	Standing broad jump (cm) [c]	-0.12	-7.31–7.07	96.8	94.0	-0.12	-0.1
	One stroke push (m) [d]	-0.80	-2.86–1.25	5.61	6.41	-0.80	-13
Physical activity [c]	Sedentary time – week (min)	-26.7	-66.8–13.4	544.8	571.6	-26.7	-4.7
	Sedentary time – weekend (min)	-40.6	-119.0–37.8	497.8	538.4	-40.6	-7.5
	Active time – week (min)	8.0	-31.4–47.4	294.7	286.6	8.0	2.8
	Active time – weekend (min)	33.7	-40.1–107.4	291.5	257.8	33.7	13

*p<.01; CI, confidence interval; MD, mean difference; MP, mean power; PP, peak power; VO_2, oxygen uptake.
[a] Multiple imputation model adjusted for baseline values. The intervention effect was not substantially confounded by age, sex, level of mobility or school.
[b] Regression coefficients should be interpreted as a ratio as it was logarithmically transformed.
[c] Included only participants who are ambulatory.
[d] Included only wheelchair users.

Table 6.5: Results from the linear regression analyses assessing the intervention effect on cardiometabolic health

		Linear regression[a]		Estimated marginal means Differences between groups			
		β	95% CI	Sport	Control	MD	%
Cardiovascular	Systolic blood pressure (mmHg)	-0.46	-5.50–4.59	122.5	123.0	-0.46	-0.4
	Diastolic blood pressure (mmHg)	-2.57	-7.24–2.10	67.2	70.0	-2.57	-3.7
	AIx (%)	-3.02	-7.35–1.31	8.93	11.95	-3.02	-25
	AIx (Z-score)	-0.53	-1.10–0.04	-0.08	0.45	-0.53	-117
	PWV (m/s)	0.20	-0.15–0.54	6.18	5.98	0.20	3.3
Metabolic	BMI (kg/m^2)	0.05	-0.64–0.90	22.8	22.8	-0.05	-0.2
	BMI (Z-score)	-0.06	-0.25–0.14	1.23	1.28	-0.06	-4.3
	Waist-circumference (cm)	-1.13	-3.41–1.15	80.8	82.0	-1.13	-1.4
	Waist-hip ratio	-0.02	-0.04–0.00	0.97	0.99	-0.02	-1.9
	Fat mass (%)	-2.78	-4.78–-0.78*	27.7	30.5	-2.78	-9.1
	Total cholesterol (mmol/l)	-0.35	-0.80–0.10	3.67	4.02	-0.35	-8.8
	HDL (mmol/l)	-0.07	-0.29–0.15	1.21	1.28	-0.07	-5.6
	LDL (mmol/l)	-0.05	-0.37–0.28	2.08	2.13	-0.05	-2.2
	Total cholesterol/HDL	0.02	-0.44–0.47	3.31	3.29	0.02	0.5
	Triglyceride (mmol/l) [b]	0.64	0.43–0.94	0.78	1.21	-0.44	-36
	Glucose (mmol/l) [c]	-0.31	-0.68–0.07	4.80	5.11	-0.31	-6.0

*p<.01; CI, confidence interval; MD, mean difference; AIx, augmentation index; PWV, pulse wave velocity; BMI, body mass index; HDL, high density lipoprotein; LDL, low density protein.

[a] Multiple imputation model adjusted for baseline values. The intervention effect was not substantially confounded by age, sex, level of mobility or school.

[b] Regression coefficients should be interpreted as a ratio as it was logarithmically transformed.

[c] Included only participants who were fasted.

effect on physical activity. Sedentary and active time during both week and weekend days showed no between group difference following the school-based sports program (Table 6.4, Figure 6.2c).

Cardiometabolic health

No effects were found for blood pressure and arterial stiffness (Table 6.5). Although not statistically significant, a change of 0.53 Z-score on AIx might be clinically relevant (Figure 6.2d). A significant effect in favour of the sport group was demonstrated for fat mass (β=-2.78, IC=-4.78–-0.78). The sport group lost 2.8% more fat mass compared to the control group (Figure 6.2e). No effects were observed for BMI, waist-hip ratio, and metabolic profile. For triglyceride a small but non-significant effect was found (β=0.64, IC=0.43–0.94[1]) with a between-group change ratio of -0.44 mmol/l in favour of the sport group (Figure 6.2f).

DISCUSSION

The aim of current study was to evaluate the effects of a school-based once a week sports program on physical fitness, physical activity and cardiometabolic health in youth with physical disabilities. For all participants, both able to walk/run or propel a manual wheelchair, a school-based sports program is feasible and can be performed safely. Despite the heterogeneity of the group, increasing the level of sports participation once a week with 45 minutes showed already positive effects after six months. We found effects in favour of the sport group in both anaerobic performance and fat mass.

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The school-based sports program resulted in a positive within group difference of 23 Watt (16%) in anaerobic performance. This absolute increase is comparable with the 20 Watt (38%) improvement after eight months of exercise training in children and adolescents with CP.[39] The higher relative difference, 16% in the current study versus 38%, can be explained by the lower baseline values in the study of Verschuren et al.[39] Moreover, eight weeks of HIT prior to the school-based sports program resulted already in an increase in anaerobic performance in both sport and control group of 11%.[24] Hence, independent of baseline values, once a week sports participation improves anaerobic performance even further. Another remarkable finding is that the control group maintained its gains on anaerobic performance following the regular curriculum of six months. It is unknown which factors contribute to this sustainability, but probably youth with physical disabilities are more active in daily life compared to several years ago. A recent cross-sectional study showed that anaerobic performance increased in youth with CP between 2004 and 2014.[40] What we can conclude thought, is that children and adolescents with physical disabilities improve anaerobic performance with an extra sports session a week, even after a training period.

The sports program resulted in a positive effect on fat mass, but found no differences in BMI, whilst other studies reported positive effects on BMI after a school-based intervention program.[41,42] Both fat mass and BMI are generally known to identify adiposity, although BMI fails to distinguish between lean and fat mass and lacks in sensitivity.[43,44] This might explain why BMI remained unchanged in the current study. Possibly, a small shift from fat to lean mass has occurred in the sport group, while weight and consequently BMI remained unchanged. Besides BMI, also no changes were found in waist-circumference and waist-to-hip ratio, while other studies showed significant changes in adolescents with CP after following an exercise program.[4,6] Possibly, if participants continue with sports participation, or exercise more frequently, in the longer term these non-significant differences in health will diverge positively compared to individuals who do not exercise regularly.[45,46]

The current school-based sports program was performed once a week. Possibly, the frequency of once a week could explain why we found no significant effects on most of the outcome measures. However, once a week reflects daily life, since there is a discrepancy between the requirements from exercise physiology perspectives and the feasibility or the priority of sports participation. Although participants did not train following exercise guidelines, increasing sports participation with once a week improved anaerobic performance and fat mass. Consequently, being active is always better than being inactive[47] and is the starting point for an active and healthy adulthood.[48] The current study demonstrated that sports participation of only once a week already shows positive effects after six months and tend to induce more effect over a prolonged period. Beside sports participation, daily physical activity also consists of playing outside and active transportation. Compared to typically developing peers, the current population is also less active in these domains of physical activity.[12] Families of youth with a physical disability should therefore be encouraged to perform an additional activity in the week or weekends to optimally profit from the benefits of physical activity.

The current study examined the effects of group exercise on various outcome measures. Although we were interested in the group effects, reasons for participation may vary across individuals.[21] For example children and adolescents want to keep up with friends in playing soccer, lose weight, make friends or just have fun. In the current study, we did not measure these reasons. To establish greater and clinical relevant effects, future research should tailor outcome measures on individual needs. For example, school-based physical activity program targeted at overweight children reduce BMI and blood pressure to a greater extent compared to the general population.[41,42] Moreover, the current school-based sports program did not lead to significant changes in daily physical activity. This probably needs an intervention with a behavioural component, which we did not include. However, earlier research with a behavioural component showed also no effects on physical activity in youth with CP.[25,26] A recent review showed that both parental involvement by education or homework tasks and the inclusion of activities conducted after school time induce greater effects on physical activity and body composition.[41]

Several limitations should be taken into account. Firstly, the current study is not controlled by a randomly assigned group. This resulted in very dissimilar groups. Although we corrected for group differences at T1, it is difficult to attribute the improvement of anaerobic performance to the sports program only. A second limitation of this study is the composition of the study population comprising a large age range and a variety of diagnoses. However, this heterogeneity actually reflects real-life as they probably exercise together. Furthermore, the data showed that sex, age, level of mobility and school level were no confounders in the analyses. Thirdly, our results cover only the Dutch population of youth with physical disabilities. In the Netherlands, these children with special needs are often assigned to schools for special education, while in other countries these children follow inclusive education. Therefore, the practical implication of school-based sports programs at schools for special education may be different in other countries.

In conclusion, a school-based once a week sports program improved anaerobic performance and fat mass after six months in youth with physical disabilities. No intervention effects were found for aerobic performance, VO_2peak, strength, physical activity, blood pressure, arterial stiffness, and the metabolic profile. These effects are promising for long-term fitness and health promotion, because in the current study barriers for sports participation were eliminated by providing sports at school and only a training volume of once a week was added. Future research on school-based sports programs in this population should tailor outcome measures on individual needs and involve parents to induce greater and clinical relevant effects.

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7

Effects of a school-based sports program on psychosocial health and attention in children and adolescents with a chronic disease or physical disability: a controlled clinical trial

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ABSTRACT

Objective: To explore whether a school-based sports program affects psychosocial health and attention in children and adolescents with a chronic disease or physical disability.

Design: Controlled clinical trial.

Setting: Schools for special education in the Netherlands.

Subjects: Seventy children and adolescents (age 13.8 (2.9) years, aged 8–19 years, 54% boys) with various health conditions including cerebral palsy (37%), other neuromuscular (43%), metabolic (9%), musculoskeletal (7%) and cardiovascular (4%) disorders.

Interventions: Before recruitment and based on the presence of school-based sports, schools (n=4) were assigned as sport or control group. School-based sports were initiated and provided by motivated physical educators. The sport group (n=31) followed a school-based sports program (45 min/week) for six months, which included team sports. The control group followed the regular curriculum.

Main measures: Psychosocial health was assessed with self-perception (Self-Perception Profile for Children) and quality of life (DISABKIDS Chronic Generic Measure, DCGM-37). Attention was measured with experimental tasks on search efficiency, sustained attention and distractibility.

Results: Linear regression analyses revealed no differences between the sport and control group in any subscale of self-perception and quality of life. In addition, no effects in favor of the sport group were found for attention.

Conclusions: A school-based sports program seems to have no effect on psychosocial health and attention in children and adolescents with a chronic disease or physical disability.

INTRODUCTION

Typically developing youth who participate in sports have higher self-perception and health-related quality of life compared to their non-sporting peers.[1] This positive association between sports participation and improved psychosocial health is stronger for team sports than individual sports, probably because of the social nature and positive involvement of peers and adults.[1] Another factor positively associated with sports participation is attentional control.[2] Studies in typically developing youth have shown that enhanced physical activity has been associated with improved attention.[3,4] This relationship is supposed to be dose-responsive, meaning that greater amounts of physical activity induce larger attentional gains.[2] Moreover, the content of exercise seems to be important as well, integrating more coordinative and cognitive demands (i.e. sports).[3,5]

Children and adolescents with a chronic disease or physical disability less often participate in sports,[6] and they tend to have lower psychosocial health and attention compared to their typically developing peers.[6–9] Regarding psychosocial health, involvement in sports can provoke successful motor experiences and focus on children's abilities rather than on their inabilities.[1] For attention, exercise can affect the brains' physiology by increasing the cerebral blood flow and density of neural networks.[2] Recent research in youth with physical disabilities has shown a positive association between participation in sports and quality of life and self-perception, such as self-perceived physical appearance and global self-worth.[10–13] Studies regarding attention are still very limited in this population.

While results regarding the role of sports in psychosocial health and attention are promising, the evidence for children and adolescents with a chronic disease or physical disability is weak and limited by cross-sectional designs. Therefore, the aim of this controlled clinical trial was to explore whether a school-based sports program affects psychosocial health and attention in children and adolescents with a chronic disease or physical disability.

METHODS

This controlled clinical trial was part of the Sport-2-Stay-Fit study, which was conducted between September 2014 and July 2016 in the Netherlands. A full description of the study design has been published previously.[14] Both ethics approval and administrative site approvals were granted by the Medical Ethical Committee of the University Medical Center Utrecht in the Netherlands (#14-118). This trial was registered with the Dutch Trial Registry (NTR4698).

Participants

Children and adolescents were recruited through four schools for special education dedicated to youth with physical disabilities. These schools have similar learning objectives as regular schools, but the children receive additional attention and support. Rehabilitation medicine is provided within school hours by rehabilitation professionals. In the Netherlands 1.9% (26,500) of the children and adolescents following primary or secondary education follow these schools.[15,16] Children and adolescents were screened for eligibility by a physical therapist, physical educator or physician. Inclusion criteria were: (1) a chronic disease or physical disability (neuromuscular, musculoskeletal, metabolic or cardiovascular disorder), (2) aged between 6 and 19 years, (3) participation in sports less than twice a week during leisure time in the preceding three months or advised to participate in sports by their physical therapist or physician, and (4) ability to understand simple instructions. Exclusion criteria were: (1) a progressive disease, (2) use of a powered wheelchair, and (3) participation in other research that could possibly influence current results. Additionally, all parents, and participants from 12 years of age and older, provided informed consent prior to study initiation.

Procedure

Before recruitment, schools (n=4) were assigned as sport or control site. This was directed by current presence of school-based sports. School-based sports were initiated at some schools by motivated physical educators working at these schools before the study started. If school-based sports were already provided in addition to the regular curriculum (n=1) or if schools intended to provide school-based sports shortly (n=1), they were assigned as sport sites. If not, schools were assigned as control sites (n=2). In case school-based sports had been provided before the start of the trial, children and adolescents were only included if they had not participated during the preceding three months. In this way, all participants knew in what group they would be enrolled: interval training and school-based sports (i.e., sport group) or interval training and control (i.e., control group). Thus, regardless of the school, all participants followed a high-intensity interval training twice a week for eight weeks as an initial start-up for their fitness level and to get familiarized with exercise.[14] Outcome measures were assessed at baseline (T0), at two months after completion of the interval training (T1), and at eight months after completion of six months of school-based sports (T2). All outcome measures across all schools were assessed by the same trained researcher (MZ) together with research assistants. The assessors were not blinded for group allocation.

Intervention

The school-based sports program was provided once a week for 45 minutes by an experienced physical educator in addition to the regular schedule. Compared to physical education where body skills, teamwork, and cooperation are taught, sport is about moving more intense, sometimes at a competitive level. No instructions were given on exercise intensities, but children and adolescents were encouraged to be physically active, play the game in question and have fun. The content of the program was adapted by the physical educator, based on the children's skills- and cognitive level. The sports program could include, but was not restricted to, soccer, (wheelchair) basketball, (wheelchair) hockey, and/or easy to practice games like playing tag. The presence of the children was documented every session.

Outcome measures

Outcome measures were psychosocial functioning, operationalized by self-perception and health-related quality of life. Cognitive functioning was measured by experimental tasks of attention, because this domain of cognition was shown to be associated with exercise.[3]

Psychosocial health

The Dutch version of the self-perception profile for children (SPPC) was used to address a total of six domains: five specific domains of self-perception (i.e., scholastic competence, social acceptance, athletic competence, physical appearance, behavioral conduct), and global self-worth.[17] The questionnaire was filled out at school with one of the research assistants. All items were scored on a four item Likert-scale and sum scores were calculated for each of the six domains ranging from 6–24 points with higher scores indicating a higher self-perception.

Regarding health-related quality of life, the Dutch version of the DISABKIDS for children with chronic health conditions (DCGM-37) was filled out.[18] This was done online by either the participant alone (n=18), the participant together with one of their parents/caregivers (n=26) or the parent/caregiver alone (n=14), unknown (n=7). The questionnaire consists of 37 items which can be subdivided into six subscales: mental independence, mental emotion, social inclusion, social exclusion, physical limitation and physical treatment. All items were scored on a five item Likert-scale and per domain scores were transformed to a 0–100 scale with higher scores indicating a higher health-related quality of life.

Attention

All tests for attention were performed on a tablet PC (Asus Eee Slate Tablet, Asus Europe BV, Nieuwegein, the Netherlands) to minimize coordination problems and guaranteeing detection of subtle improvements in performance. To assess search efficiency, a digitalized object cancellation task was used and analyzed with Cancellation Tools.[19] The presented screen contained 56 targets presented as apples. The distractors consisted of 52 pears, 12 single letters, and 10 strings of letters. Children and adolescents had to tap on all apples, both red and green, until they completed all targets. No time limit was given. Search efficiency was measured using best *r*, with *r* defined as the highest absolute value of the Pearson correlation between the cancellation order and either horizontal or vertical cancellation position.[20] The best *r* increases with search efficiency. Ideally, the search is started at an extremity, for example the upper left corner, and is proceeded orthogonally, either downward and upward or rightward and leftward.

The extensive dot cancellation task consisted of 35 rows with 24 clusters of either three, four of five dots. Children were instructed to work line by line and mark all clusters of four dots (i.e., target) by tapping the tablet as fast, yet accurately as possible. Every line contained eight targets and 16 distractors. After a short practice session, the actual cancellation test was started. When a participant did not finish the test within 10 minutes, the test was stopped. Outcome variables were average speed per row (in seconds) and average accuracy of marked targets per row, calculated as a ratio score between 0 and 1. Data was excluded when test instructions were not followed properly (i.e., working line by line).

The original capture task used in Van der Stigchel & Nijboer[21] was adapted to measure distractibility. Each trial started with the presentation of a central fixation cross. When the cross disappeared, the target (i.e., an apple), appeared in one of the four quadrants. In 50% of the trials, a distractor (i.e., a pear) appeared as well. Participants were instructed to react as fast and accurate as possible to the appearance of the apple. Distractibility was represented as the difference in reaction time (ms) between both conditions.

Data analysis

Descriptive statistics were performed for group characteristics. For all outcome measures, assumptions for normal distribution were graphically checked using residual plots. All residuals were within acceptable ranges for normal distribution. To investigate if the sport group and control group changed over time, a paired sample t-test was used. To investigate the groups by time interaction following the intervention, a linear regression was performed, with outcomes measures at T2 as the dependent variable, and both T1 and group allocation as independent variables. Since the dataset was expected to contain incomplete data, we used multiple imputation to create and analyze 10 imputed datasets. Linear regression analyses were both performed on the original data and multiple imputation data. Because both models resulted in similar conclusions, only the multiple imputation model is shown here. Results are reported as means and standard deviation (SD), regression coefficients and corresponding 95% confidence intervals (CI). Statistical analyses were performed using SPSS for Windows (version 21.0, SPSS Inc, Chicago, Ill).

RESULTS

A total of 70 children and adolescents participated in the current study (Table 7.1). Sports participation at T0 did not differ between the sport (1.1 (1.1) times a week) and control group (0.8 (0.8) times a week) (p=.283), whereas at T2 participation in sports differed significantly between the sport (2.1 (1.0) times a week) and control group (0.9 (0.8) times a week) (p<.001).

Table 7.1: Participant characteristics

	Sport group (n=31)	Control group (n=39)
Age (y), mean (SD)	13.4 (3.0)	14.1 (2.8)
Sex, n male (%)	23 (74)	15 (39)
Height (cm), mean (SD)	156.4 (17.5)	157.2 (10.5)
Weight (kg), mean (SD)	54.4 (20.8)	57.0 (18.8)
Level of mobility, n (%)		
Able to run	22 (71)	14 (36)
Able to walk	6 (19)	19 (49)
Wheelchair user	3 (10)	6 (15)
Diagnoses, n (%)		
Neuromuscular	22 (71)	35 (86.5)
Cerebral palsy	10 (32)	16 (41)
Spina bifida	-	5 (13)
Psychomotor retardation	4 (13)	5 (13)
Acquired brain injury	2 (97)	1 (2.5)
Other	6 (19)	7 (18)
Cardiovascular	2 (6)	1 (2.5)
Metabolic	4 (13)	2 (5)
Musculoskeletal	3 (10)	2 (5)

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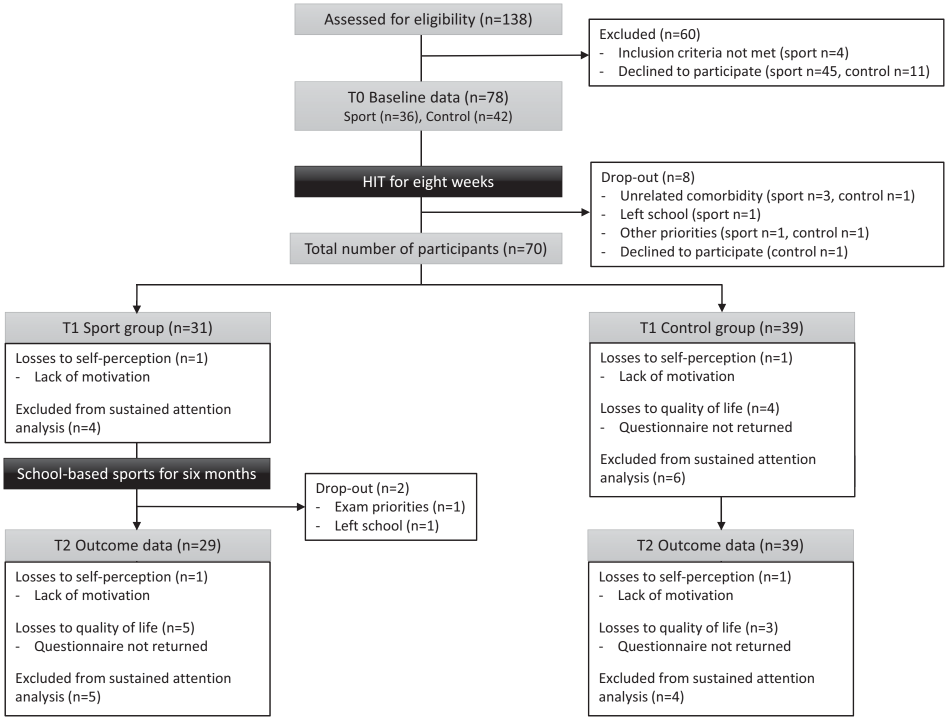

Figure 7.1: Flowchart from initial inclusion to intervention.

The number of analyzed cases are illustrated in Figure 7.1. Two participants of the sport group dropped out during the school-based sports program, because of exam priorities and leaving school. Presence at the school-based sports program was 86%, with children and adolescents attending 14.4 (4.1) sports sessions on average.

As shown in Table 7.2, no time effects were found for self-perception, quality of life and attention in either the sport nor the control group, except for physical limitation, which improved in the sport group (p=.048). Regarding the group by time interaction, no significant effects were observed in favor of the sport group for self-perception and quality of life following the school-based program of six months. In addition, no effects in favor of the sport group were found for search efficiency, sustained attention and distractibility.

Table 7.2: Mean (SD) after two months interval training (T1) and after six months school-based sports (T2), and results from linear regression analyses assessing the intervention effect on self-perception, quality of life and attention

		T1				T2				Linear regression [a]	
		n	Sport Mean (SD)	n	Control Mean (SD)	n	Sport Mean (SD)	n	Control Mean (SD)	B	95% CI
Self-perception (SPPC)	Scholastic competence (6 items, range 6–24)	30	17.4 (2.9)	38	18.1 (3.2)	28	17.5 (2.9)	38	17.9 (3.6)	-0.08	-1.66; 1.51
	Social acceptance (6 items, range 6–24)	30	18.1 (4.2)	38	17.9 (4.6)	28	18.3 (4.6)	38	17.9 (4.8)	0.15	-1.53; 1.84
	Athletic competence (6 items, range 6–24)	30	17.9 (4.3)	38	17.2 (4.0)	28	17.6 (4.2)	38	16.8 (4.2)	0.51	-1.65; 2.68
	Physical appearance (6 items, range 6–24)	30	19.4 (4.4)	38	20.0 (4.7)	28	18.7 (5.2)	38	19.4 (4.7)	-0.32	-2.19; 1.56
	Behavioral conduct (6 items, range 6–24)	30	18.2 (3.9)	38	20.3 (3.5)	28	17.4 (4.2)	38	20.0 (3.6)	-1.71	-3.61; 0.19
	Global self-worth (6 items, range 6–24)	30	20.0 (3.7)	38	20.0 (3.9)	28	19.4 (4.2)	38	20.1 (3.5)	-0.82	-2.47; 0.83
Health-related Quality of Life (DCGM-37)	Mental independence (range 0–100)	31	71.9 (14.0)	35	71.9 (12.9)	24	75.3 (14.3)	36	71.5 (15.5)	2.76	-4.93; 10.5
	Mental emotion (range 0–100)	31	65.8 (21.1)	35	78.5 (16.5)	24	74.6 (19.4)	36	77.6 (16.1)	2.87	-4.05; 9.78
	Social inclusion (range 0–100)	31	57.8 (14.6)	35	55.5 (16.0)	24	60.1 (13.1)	36	54.9 (13.4)	3.39	-1.31; 8.01
	Social exclusion (range 0–100)	31	68.0 (16.2)	35	69.5 (17.5)	24	71.7 (17.8)	36	72.5 (13.6)	-0.11	-6.34; 6.14
	Physical limitations (range 0–100)	31	62.4 (16.9)	35	62.5 (14.8)	24	70.3 (15.9)*	36	66.8 (15.0)	3.01	-2.86; 8.88
	Physical treatment (range 0–100)	12	63.2 (25.2)	14	75.0 (28.4)	8	69.3 (20.5)	10	76.3 (22.2)	NA	
	General (31 items) (range 0–100)	31	65.2 (14.0)	35	67.9 (10.9)	24	70.5 (12.3)	36	68.9 (11.8)	2.46	-2.16; 7.08
Attention	Search efficiency – Best-R (r)	31	0.66 (0.28)	39	0.58 (0.25)	29	0.67 (0.26)	39	0.61 (0.26)	0.03	-0.08; 0.14
	Sustained attention – Accuracy (range 0–1)	27	0.90 (0.14)	33	0.92 (0.10)	24	0.90 (0.12)	35	0.92 (0.09)	0.01	-0.04; 0.06
	Sustained attention – Speed (time(s)/line)	27	15.0 (5.4)	33	19.5 (7.8)	24	14.4 (5.6)	35	18.1 (7.0)	-0.15	-2.07; 1.78
	Distractibility – Time (ms)	31	64.6 (69.8)	39	86.8 (72.0)	29	65.1 (71.6)	39	74.1 (88.5)	8.71	-19.5; 36.7

B, regression coefficient; CI, confidence interval; DCGM-37, DISABKIDS chronic generic measure; SPPC, self-perception profile for children; NA, not applicable; too few participants completed this scale.
* Significant difference from T1.
[a] Multiple imputation model adjusted for baseline values.

DISCUSSION

This study explored the effects of a school-based sports program on psychosocial health and attention in children and adolescents with a chronic disease or physical disability. No differences were observed between the sport and control group after six months in any of the subscales of self-perception and health-related quality of life. In addition, attention showed no effects in favor of the sport group following the school-based once a week sports program.

Baseline scores for health-related quality of life were comparable to children and adolescents with cerebral palsy.[18] For self-perception, all scores on the subscales were comparable to those of typically developing children and adolescents.[17] A possible explanation might be peer perception; the children and adolescents included in this study attend schools for special education and are therefore surrounded by comparable peers having physical disabilities.[22]

Meaningful sports participation is a complex phenomenon, with the interaction of environmental and personal factors.[23] In the current intervention, the school-based sports program was focused on playing sports or games, experiencing success, and having fun in a familiar environment (i.e. school). Important environmental barriers, like transportation, supportive trainers, and acceptance, were deliberately eliminated by performing sports at school. Consequently, the environmental involvement from family and society is underrepresented in the current intervention. To illustrate, the most important feature for a child's perception of meaningful sports participation (i.e. social in- or exclusion) is having authentic friendships.[24] Since children and adolescents in the current study already knew each other from school, it is unknown if the intervention has contributed to strengthening their feelings of acceptance by others. Another environmental factor, family support, which can play a central role in children's motivation,[24] was not actively integrated in the current sports program. Possibly, these environmental factors are important to induce positive effects of sports participation on psychosocial health.

Furthermore, when children or adolescents are joining an adapted sports club, they consciously choose for a type of sport, and often also participate in a competition. These personal factors provide a sense of belonging to the community, expressing themselves through the specific sport, developing an identity by being part of a team, and provide the opportunity of making new friends.[24] It is unknown from the literature if these personal factors are necessary for gaining psychosocial health, since there might be a self-selection bias in youth joining sports clubs. To date, adolescents with better psychosocial functions (i.e., social skills) are more likely to join sports clubs and maintain that connection.[25] Possibly, this self-selection bias may partly account for the positive association found between sports participation and psychosocial health in cross-sectional studies.[1] In addition, a recent longitudinal study showed no additional effect of providing positive, specific and progress feedback to enhance self-perception in children with a developmental coordination disorder, following physical therapy.[26] Therefore, it is still unknown which factors are important to induce positive effects of sports participation on psychosocial health.

It is important to take into account that the sports program was designed to improve physical fitness and not attention. However, the program did include a progressive increase in task complexity, ensuring and challenging cognitive efforts, but attention did not change compared to the control group over a six-month period. This is in contrast to results presented by Hillman et al.[27] where typically developing children showed improved attention following a school-based intervention of nine months. Their program consisted of 70-minutes of physical activity, each day after school. Taking into account the dose response relation between physical activity and cognitive functioning,[2] the frequency and duration of the program could be an explanation why our intervention showed no improvements. Our dose of 45 min/week might be too low to induce effects on attention. On the other hand, by increasing physical activity levels, it is still unknown which specific factors (i.e. active ingredients) contribute to inducing attention and should be integrated in future interventions.

Besides the limitation discussed before, other limitations have to be recognized. Firstly, participants were not randomly assigned to either the sport or control group and as a consequence, assessors were not blinded for group allocation. Secondly, current sample compromised a large age range and a variety of diagnoses, which makes it implausible to discover anything other than the most major effect. However, this heterogeneity does reflect the real-life situation at schools for special education as children and adolescents participate in sports together. In addition, a recent cross-sectional study in children and adolescents with physical disabilities found that beneficial scores on psychosocial health in youth who participate in sports at least twice a week were independent of age, sex, school type (i.e., special or regular education) and diagnosis.[13]

There is a need for more research into the factors which account for the positive association between sports participation and both psychosocial health and attention in children with a chronic disease or physical disability.

Clinical messages

- A school-based sports program for children and adolescents with a chronic disease or physical disability seems not to improve psychosocial health and attention.
- Research into the important factors is needed before further resources can be given to improve sports participation for increasing psychosocial health and attention.

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Body mass index and fitness in high-functioning children and adolescents with cerebral palsy: What happened over a decade?

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ABSTRACT

Background: In recent decades, improving fitness has become an important goal in rehabilitation medicine in children and adolescents with cerebral palsy (CP).

Aims: To compare body mass index (BMI), performance-related fitness, and cardiorespiratory fitness of children with CP measured in 2014 with a comparable sample from 2004.

Methods and procedures: In total, 25 high-functioning children with CP (i.e., GMFCS I-II) measured in 2004 (13 boys; mean age 13.2 (2.6) years) were matched to 25 children measured in 2014. Outcomes included body mass and BMI, muscle power sprint test (MPST), 10x5-m sprint test, and a shuttle run test (SRT). Data of 15 participants from 2004 (10 boys; mean age 12.6 (2.5) years) were matched and analyzed for VO_2peak.

Outcomes and results: Body mass and BMI were higher (both: $p<.05$) in the 2014 cohort compared to the 2004 cohort. Further, performance-related fitness was better for the 2014 cohort on the MPST ($p=.004$), the 10x5-m sprint test ($p=.001$), and the SRT ($p<.001$). However, there were no differences for VO_2peak.

Conclusions and implications: In high-functioning children with CP, there are positive ecological time trends in performance-related fitness, but not in VO_2peak between 2004 and 2014. The substantial higher body mass and BMI is alarming and requires further investigation.

What this paper adds

In recent decades, improving fitness levels of children and adolescents with cerebral palsy (CP) has become an important goal in rehabilitation medicine. Published studies related to exercise training programs for children and adolescents with CP have shown positive effects. However, it usually takes time for scientific results to be implemented in clinical practice. This study has the unique opportunity to study, with a timeframe of 10 years, if body mass, BMI and fitness levels of children with CP has been changed. We were able to compare two datasets collected in a similar way in the same country (and regions) in children and adolescents with CP. These datasets have the same outcomes on body mass and BMI, performance-related fitness and VO_2peak. The present study shows positive ecological time trends in performance-related fitness in children and adolescents with CP between 2004 and 2014. However, the higher body mass and BMI, and the unchanged VO_2peak requires further investigation.

INTRODUCTION

Improving physical fitness has become an important aspect during rehabilitation. Over the last decades, the number of studies on physical fitness in children and adolescents with cerebral palsy (CP) has grown exponentially. It is recommended that all children with CP should engage, to the extent they are able, in aerobic, anaerobic and muscle strengthening activities.[1,2] It is generally acknowledged that children and adolescents with CP can be trained according to general exercise physiological training principles, without exacerbating spasticity.[3]

Physical fitness can be divided into subcomponents including performance-related fitness and cardiorespiratory fitness. Performance-related fitness is the combined result of coordinated exertion with a variety of physiological functions.[4] Coordination, speed, agility, and short-term muscle power are often described as outcome measures related to performance. Cardiorespiratory fitness, generally expressed as peak oxygen uptake (VO_2peak), is a strong predictor for cardiovascular disease later in life.[5]

In the typically developing population, previous ecological time trend studies showed that both performance-related fitness and cardiorespiratory fitness has declined in children and adolescents over recent decades.[6–9] Performance on both 20-meter Shuttle Run Test (SRT)[6] and 10x5-m sprint test have decreased.[7] In addition, running performance on a time trial decreased by 10% and 6% for boys and girls, respectively, in Finnish adolescents between the 1980s and 2000s.[8] Moreover, VO_2peak decreased by 8% in Norwegian adolescents from 1985 to 2002.[9]

Weight gain and the rising obesity prevalence among children and adolescents is becoming a serious health problem worldwide.[10] Physical fitness levels are negatively associated with overweight.[5] Body weight and body mass index (BMI) increased by 7% and 6%, respectively, in Norwegian adolescents from 1985 to 2002.[9] Moreover, BMI has been shown to be inversely associated with running performance, indicating higher BMIs over time may lead to poorer fitness and running capacities.[8] Since children and adolescents with CP have lower performance-related fitness and cardiorespiratory fitness compared to children who are typically developing,[11,12] they are thought to be at increased risk for overweight.

A large number of studies have shown positive effects of exercise training in children with CP on performance-related fitness[13,14] and cardiorespiratory fitness.[15] These insights in the positive effects of exercise training should ideally lead to implementation in clinical practice. The aim of the current study is to compare body mass and BMI, performance-related fitness, and cardiorespiratory fitness in a convenience sample of children with CP measured in 2014 with a comparable sample from 2004.

METHODS

For the current study, data collected in 2014[16] were compared to two datasets from two different studies performed in 2004.[14,17] Data collection was administered similarly, since two researchers (OV, TT) involved in the 2004-studies supervised data collection in 2014.

Setting and participants

All participants from both 2004 and 2014 samples were recruited from several schools for special education in the Netherlands. Children and adolescents were included if they were diagnosed with spastic CP, classified at GMFCS level I or II, and between the age of 7–18 years. Data of the 2014 study were used and matched with the data collected in 2004 according to sex, GMFCS-level and height up to three centimeter difference. These characteristics are most related to both performance-related fitness and cardiorespiratory fitness.[12] Height was used instead of age, because its more discriminative value for physical fitness in this population. Matching was done for the two different datasets from 2004 to compose separate databases containing either body mass, BMI and performance-related fitness,[14] and cardiorespiratory fitness.[17]

To assess differences in body mass and BMI, and performance-related fitness, data of 68 and 25 participants were available, collected in 2004 and 2014 respectively. We were able to match all 25 children with CP measured in 2014 to a similar sample of 25 participants from 2004. To assess differences in cardiorespiratory fitness, data of 24 and 25 children were available, collected in 2004 and 2014 respectively. Due to a sex and height differences, matching resulted in a sample of 30 participants in total; 15 measured in 2004 and 15 in 2014. Ethics approval and administrative site approvals were granted by the Medical Ethical Committee of UMC Utrecht in the Netherlands. Additionally, all parents, and participants from 12 years of age, provided informed consent.

Anthropometry

Prior to testing, each child was weighted to the nearest 100 g on electronic scales (Seca, Hamburg, Germany). Height was measured while the child was standing against a wall. The BMI was calculated as weight in kilograms divided by height in meters squared. To control for differences in height, Z-scores of BMI for height were calculated according to Dutch reference values.[18]

Performance-related fitness

Anaerobic performance

Mean and peak power were measured for anaerobic performance derived from the Muscle Power Sprint Test (MPST) as described previously.[19] Participants were instructed to complete six 15-m runs at maximum pace with a 10 second rest in between each run. Power output for each sprint was calculated using body mass and sprint time, where: power = (body mass × distance^2)/time^3. Peak power was defined as the highest calculated power, while mean power was defined as average power over the six sprints.

Agility

The 10x5-m sprint test was performed to measure agility. Participants had to sprint 10 times as fast as possible between two lines five meter apart without rest. This is a reliable field test to measure agility in children with CP.[19]

Aerobic performance

Aerobic performance was measured using the achieved level on the 10-m SRT developed for children and adolescents with CP, classified with GMCS-level I and II. This test is a reliable and valid measure in children and adolescents with CP as described previously.[17] The SRT is a field test in which a child runs or walks between two markers delineating the respective course of 10 meter at a set incremental speed determined by a signal every minute. The children have to adjust their running or walking pace to the beep-signals, until they fail to reach the line two times in a row, despite encouragements. The starting speeds for the tests are 5 and 2 km/h for participants who are classified at GMFCS levels I and II, respectively, and the speeds are increased by 0.25 km/h every minute. The last completed level, accurate to a half shuttle, was recorded and used for analysis. Heart rate was measured continuously with a heart rate monitor throughout the test. The SRT has shown to be valid, reliable and sensitive to change in children with CP.[17,19]

Cardiorespiratory fitness

Cardiorespiratory fitness was measured during the 10-m SRT by obtaining VO_2peak. Participants wore a calibrated mobile gas analysis system of Cortex Metamax (Samcon bvba, Melle, Belgium). This system is valid and reliable for measuring ventilator parameters during exercise.[20] It consists of a facemask, transmitting unit attached to a harness containing oxygen and carbon dioxide analyzers, and a receiving unit. Metabolic stress test software (Metasoft) was used to measure oxygen uptake (VO_2), carbon dioxide production, minute

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ventilation, respiratory exchange ratio (RER = VCO_2/VO_2), and heart rate (HR). Subjective criteria were used to determine if a test was maximal; signs of intense effort such as unsteady running pattern, sweating and facial flushing.

Statistical analysis

Statistical comparisons between children and adolescents with CP measured in 2004 and 2014 were tested using an independent sample T-Test. In addition, differences between GMFCS-level I and II were tested (*p*=.05). All statistical analyses were performed using SPSS for Windows (version 21.0, SPSS Inc, Chicago, IL).

RESULTS

For body mass, BMI and performance-related fitness, participant characteristics of 25 children and adolescents with CP from both the 2004 and 2014 sample are presented in Table 8.1. Sex and GMFCS-levels were equally divided; 13 boys and 15 classified at GMFCS-level I.

Anthropometry

A significantly higher body mass and BMI were found in the 2014 cohort for the total group (*p*=.010; *p*=.004) and GMFCS-level I (*p*=.026; *p*=.008). Body mass gained with 26% and 27%, and BMI was 20% and 25% higher for the total group and GMFCS-level I respectively. The same applies for the calculated BMI for height Z-scores, as shown in Table 8.1.

Performance-related fitness

Table 8.2 shows the differences for performance-related fitness. Both mean and peak power on the MPST are significantly higher, respectively 99% and 87%, over a decade (*p*=.004; *p*=.006). In addition, children with CP measured in 2014 perform significantly better on agility (23%), in the total group (*p*=.001), and for GMFCS-level I and II respectively (*p*=.007; *p*=.003). The same applies for aerobic performance as shown in Table 8.2. The 2014 cohort performed 67% better, measured with achieved level on the SRT.

Table 8.1: Participant characteristics for anthropometry and performance-related fitness

		2004 sample				2014 sample				
Participants	Variable	n	Mean	SD	Range	n	Mean	SD	Range	*p*
Total	Age (y)	25	13.2	2.6	8–17	25	14.2	2.9	8–18	.205
	Height (cm)	25	158.2	14.2	133–183	25	161.2	11.5	140–184	.406
	Weight (kg)	25	51.2	13.5	33–78	25	64.6	21.4	34–116	.010*
	BMI (kg/m^2)	25	20.2	3.8	15.4–29.7	25	24.3	5.6	17.5–39.0	.004*
	BMI for height [a]	25	0.69	1.5	-1.39–3.83	25	1.79	1.36	-.65–3.93	.009*
GMFCS I	Age (y)	15	13.9	2.5	8–17	15	14.2	3.2	8–18	.732
	Height (cm)	15	163.9	12.5	141–183	15	163.8	12.4	140–184	.970
	Weight (kg)	15	56.9	12.0	37–78	15	72.5	22.7	40–116	.026*
	BMI (kg/m^2)	15	21.2	4.4	16.5–29.7	15	26.5	5.7	18.3–39.0	.008*
	BMI for height [a]	15	0.81	1.76	-1.26–3.83	15	2.40	1.17	.46–3.93	.007*
GMFCS II	Age (y)	10	12.2	2.4	9–17	10	14.1	2.6	9–18	.100
	Height (cm)	10	149.5	12.7	133–170	10	157.4	9.3	140–169	.128
	Weight (kg)	10	42.5	11.0	33–66	10	52.7	12.3	34–74	.065
	BMI (kg/m^2)	10	18.7	2.1	15.4–22.8	10	21.0	3.3	17.5–27.9	.079
	BMI for height [a]	10	0.52	1.05	-1.39–1.91	10	0.88	1.13	-.65–2.86	.474

SD, standard deviation; GMFCS, gross motor function classification system; BMI, body mass index.
[a] Z-score, *p<.05.

Table 8.2: Performance-related fitness

		2004 sample				2014 sample				
Participants	Variable	n	Mean	SD	95% CI	n	Mean	SD	95% CI	*p*
Total	Anaerobic performance									
	Mean power (W)	25	102.4	77.7	70–134	25	203.8	148.4	142–265	.004*
	Peak power (W)	25	125.8	96.2	86–166	25	235.7	166.1	167–304	.006*
	Agility									
	10x5 (s)	25	33.7	9.6	29.7–37.6	25	25.8	5.1	23.7–27.9	≤.001*
	Aerobic performance									
	Achieved level on SRT	25	7.6	4.5	5.7–9.4	25	12.7	5.1	10.6–14.8	≤.001*
GMFCS I	Anaerobic performance									
	Mean power (W)	15	145.7	70.1	107–185	15	271.2	157.2	184–358	.009*
	Peak power (W)	15	179.9	86.5	132–227	15	311.9	174.7	215–409	.014*
	Agility									
	10x5 (s)	15	28.8	4.6	26.3–31.3	15	23.8	4.9	21.1–26.5	.007*
	Aerobic performance									
	Achieved level on SRT	15	6.3	4.0	4.1–8.5	15	11.3	5.4	8.2–14.1	.009*
GMFCS II	Anaerobic performance									
	Mean power (W)	10	37.4	24.8	20–55	10	102.6	37.3	76–129	≤.001*
	Peak power (W)	10	44.7	29.2	24–66	10	121.5	43.4	90–153	≤.001*
	Agility									
	10x5 (s)	10	41.0	10.7	33.3–48.6	10	28.9	3.8	26.2–31.7	.003*
	Aerobic performance									
	Achieved level on SRT	10	9.4	4.7	6.2–12.8	10	15.0	3.9	12.2–17.7	.011*

SD, standard deviation; CI, confidence interval; GMFCS, gross motor function classification system; SRT, shuttle run test.
*p<.05.

Cardiorespiratory fitness

Participant characteristics of 15 children and adolescents with CP from both samples are presented in Table 8.3. Sex and GMFCS-levels were equally divided; 10 boys and 8 classified at GMFCS-level I. The SRTs of both samples were quite similar regarding peak heart rate (198 (5) bpm; 183 (18) bpm) and RER (1.05 (0.11); 1.07 (0.10)) for the 2004 and 2014 sample respectively. As indicated in Table 8.4, no differences were found in VO_2peak, despite of a better performance on the SRT (*p*=.001). No differences were found for absolute (l/min) and relative (ml/kg/min) VO_2peak respectively in the total group (*p*=.408; *p*=.315), neither for GMFCS-level I (*p*=.312; *p*=.741), and GMFCS-level II (*p*=.921; *p*=.246).

DISCUSSION

This study compared body composition, performance-related fitness, and cardiorespiratory fitness in a representative cohort of high-functioning children with CP, GMFCS-level I and II, measured in 2014 with a comparable cohort from 2004. In the 2014 cohort, body mass and BMI were higher compared to the 2004 cohort. In the contrary, performance-related fitness was better for the 2014 cohort with regards to agility and both anaerobic and aerobic performance, as compared to the 2004 cohort. Cardiorespiratory fitness however, did not differ between 2004 and 2014.

Although this is the first observational study to analyze fitness trends in children and adolescents with CP, a limitation of our study is the relatively small sample size measured cross-sectionally with a considerable large age range (7–18 years). Exercise training programs for children with CP have become a contemporary focus of intervention,[15] especially at schools for special education in the Netherlands. Therefore, the results reported may not be representative for children with CP in other countries. Although several dependent outcome measures were analyzed, the two datasets differ to the extent that we assume no type I error occurred.

Body mass and BMI were 26% and 20% higher respectively in the 2014 cohort. Compared to differences of 6% and 7% in the typically developing population, this is substantial.[9] Especially when taking into account that children who develop typically and children with CP have similar body compositions.[21] Although BMI is commonly used and provides the most useful measure to estimate the prevalence of overweight, BMI does not distinguish between weight associated with muscle mass and weight associated with fat mass.[22] Since strength training may lead to hypertrophy in children and adolescents with CP,[23] they may have gained weight due to increased muscle mass in the recent decade. This could well

Table 8.3: Participant characteristics for cardiorespiratory fitness

		2004 sample				2014 sample				
Participants	Variable	n	Mean	SD	Range	n	Mean	SD	Range	*p*
Total	Age (y)	15	12.6	2.5	7–17	15	12.7	2.8	8–18	.917
	Height (cm)	15	154.5	13.9	130–175	15	157.0	10.9	140–176	.595
GMFCS I	Age (y)	8	11.9	2.8	7–16	8	12.0	2.7	8–16	.945
	Height (cm)	8	154.8	15.1	130–175	8	158.0	12.0	140–176	.643
GMFCS II	Age (y)	7	13.4	2.0	11–17	7	13.5	2.9	9–18	.936
	Height (cm)	7	154.3	13.6	135–170	7	155.9	10.3	140–166	.812

SD, standard deviation; GMFCS, gross motor function classification system.

Table 8.4: Cardiorespiratory fitness on the 10-m SRT

		2004 sample				2014 sample				
Participants	Variable	n	Mean	SD	95% CI	n	Mean	SD	95% CI	*p*
Total	Achieved level on SRT	15	9.2	2.9	7.6–10.8	15	14.2	4.8	11.6–16.9	≤.001*
	VO_2peak (l/min)	15	1.87	0.46	1.62–2.13	15	2.07	0.78	1.64–2.50	.408
	VO_2peak (ml/kg/min)	15	40.9	8.4	36.3–45.5	15	37.5	10.4	31.5–43.5	.315
GMFCS I	Achieved level on SRT	8	7.9	2.4	5.9–9.9	8	13.1	6.0	8.1–18.2	.040*
	VO_2peak (l/min)	8	1.94	0.42	1.59–2.29	8	2.33	0.96	1.52–3.13	.312
	VO_2peak (ml/kg/min)	8	41.9	6.2	36.7–47.1	8	40.2	12.6	29.7–50.8	.741
GMFCS II	Achieved level on SRT	7	10.6	2.9	7.9–13.3	7	15.5	2.6	13.1–17.9	.006*
	VO_2peak (l/min)	7	1.79	0.52	1.31–2.28	7	1.77	0.37	1.42–2.11	.921
	VO_2peak (ml/kg/min)	7	39.7	10.8	29.8–49.7	7	34.3	5.0	29.7–38.9	.246

SD, standard deviation; CI, confidence interval; GMFCS, gross motor function classification system; SRT, shuttle run test; VO_2peak, peak oxygen uptake.
*p<.05.

have caused an overestimation of the weight-gain, but nonetheless the increase in body mass and BMI is substantial. Therefore it can cause severe problems for longterm health as it increases cardiovascular risk.[24]

Compared to the 2004 cohort, VO_2peak values remain similar in children and adolescents with CP despite the higher performance on SRT. This discrepancy underlines the fact that VO_2peak and performance on SRT measure different constructs;[25] health and performance related fitness respectively. Several training studies performed between 1997 and 2007 found significant improvements in cardiorespiratory fitness ranging from 20 to 38%.[15] However, an improvement of only 11% in VO_2peak was found in a study performed in 2014.[26] In addition, a recent intervention study conducted in the Netherlands in children with CP showed no significant effect in cardiorespiratory fitness following a physical activity stimulation program consisting of fitness training, counselling and home-based therapy.[27] Possibly, standard healthcare has developed in such a way that currently baseline fitness levels have so much increased that consequently no significant training effect was induced.

Verschuren et al.[28,29] provided international reference values for performance-related fitness in children and adolescents with CP, GMFCS level I and II, in 2010. With regards to the current study, corresponding reference values of Verschuren et al. perfectly match the 2004 cohort. This implies that reference scores for mean power on the MPST, 10x5, and aerobic performance on the SRT, possibly underestimate performance outcomes of children and adolescents with CP today. Although the study population of Verschuren et al. was much larger than our study, reference values might not be up-to-date anymore.

In conclusion, the present study shows positive ecological time trends in performance-related fitness in high-functioning children and adolescents with CP over a decade. This seems promising, given the fact that clinical implementation typically takes up to 20 years. However, the higher body mass and BMI, as well as the unchanged cardiorespiratory fitness require further investigation. Future studies should consist of standardized outcome measures including waist circumference and body fat percentage measurements.

Highlights

- Performance-related fitness in children and adolescents with CP was better for the 2014 cohort as compared to the 2004 cohort
- VO_2peak values did not change over 10 years
- Currently available reference values might underestimate performance-related fitness of children and adolescents with CP today

- Body mass and BMI were substantially higher for the 2014 cohort; combination of gained muscle- and fat mass?

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9

General discussion

The main objective of this thesis was to investigate the effects of a short-term high-intensity interval training (HIT) followed by school-based sports for six months on physical fitness, cardiometabolic health, psychosocial health and attention in youth with physical disabilities. A secondary objective was to investigate ecological time trends over a decade in fitness levels of youth with cerebral palsy (CP). Furthermore, for wheelchair using youth the optimal exercise training type was examined and performance tests were evaluated.

MAIN FINDINGS

- Performing both HIT and school-based sports in a group of children and adolescents with a variety of physical disabilities and mobility levels is feasible and safe (**chapter 5** and **chapter 6**).
- Eight weeks of twice a week 30-seconds all-out HIT improved anaerobic performance, agility, aerobic performance, and both resting systolic and diastolic blood pressure, while peak oxygen uptake (VO_2peak), strength, arterial stiffness, body mass index (BMI), body composition, and the metabolic profile remained stable (**chapter 5**).
- A school-based once a week sports program improved anaerobic performance and fat mass after six months in children and adolescents with a chronic disease or physical disability in favor of those following the regular curriculum. No differences between the sport and control group were found for aerobic performance, VO_2peak, strength, physical activity, blood pressure, arterial stiffness, metabolic profile, self-perception, quality of life and attention (**chapter 6** and **chapter 7**).
- The increased agility and aerobic performance after eight weeks of HIT sustained following the school-based sports program in both the sport and control group. Anaerobic performance increased further in the sport group and remained stable in the control group (**chapter 5** and **chapter 6**). Apparently, the positive effects following an exercise training did sustain after six months in current daily life physical activity pattern.
- Between 2004 and 2014, children and adolescents with CP show positive ecological time trends in anaerobic performance, agility and aerobic performance, whereas the VO_2peak remained stable (**chapter 8**).
- Children and adolescents with CP in 2014 showed to have a higher body mass and BMI compared to a comparable sample of children with CP in 2004 (**chapter 8**).
- According to current literature, exercise training seems beneficial in improving wheelchair propulsion capacity, with interval training as the most promising training type. However, evidence is weak and limited to adults, people with spinal cord injury and able-bodied participants (**chapter 2**).

- The muscle power sprint test (MPST) and 10x5-m sprint test are both reproducible and valid field tests for measuring anaerobic performance and agility respectively, in youth with CP who self-propel a manual wheelchair (**chapter 3**).

EXERCISE TRAINING GUIDELINES

According to regular exercise recommendations for the typically developing population, children and adolescents should engage in at least 60 minutes of moderate-to-vigorous intensity activities a day, and include activities of vigorous intensity, resistance exercise, and bone loading activities on at least three days a week.[1–3] For very deconditioned children and adolescents, it is recommended to start with 1–2 sessions of vigorous activity a week.[1,4] We assumed our population to be deconditioned and provided high-intensity interval training (HIT) (i.e., vigorous activity) twice a week. Although, the HIT program has shown positive effects on exercise performance, VO_2peak and cardiovascular health did not improve. Apparently, our population was not very deconditioned at baseline and therefore the training volume was too low.

Until more is known about neuromuscular adaptations to training in children and adolescents with various physical disabilities, there is no *a priori* reason why the guidelines for children and adolescents who are developing typically cannot be used as a starting point in considering the optimal protocols for children and adolescents with a variety of chronic health conditions. This section will evaluate the extent to which the HIT program is in keeping with the recommendations for children and adolescents who are developing typically, as reflected by the American College of Sports Medicine (ACSM) guidelines. These guidelines provide more specific information, but are in accordance with the Dutch 'Beweegrichtlijnen 2017'.[1,3] The ACSM exercise guidelines for aerobic exercise follow the FITT principles (i.e., frequency, intensity, time- and type of training) and will be discussed separately (Table 9.1).[1,5] Recommendations, based on the guidelines and appropriate to youth with physical disabilities, are provided to help guide clinical practice for exercise training in this population.

Frequency

The training frequency refers to the amount of training sessions a week. The ACSM recommends children and adolescents to be at least moderately active on a daily basis including at least three days a week of vigorous intensity.[1] For very deconditioned children and adolescents, it is recommended to start with 1–2 sessions of vigorous activity a week

Table 9.1: FITT recommendations for children and adolescents who are typically developing

	Aerobic	Resistance	Bone strengthening
Frequency	Daily	≥3 days a week	≥3 days a week
Intensity	Most should be moderate-to-vigorous intensity. Include vigorous intensity ≥3 days a week.	Use of body weight as resistance or 8–15 submaximal repetitions of an exercise to the point of moderate fatigue with good mechanical form.	N/A
Time	As part of ≥60 min a day of exercise.	As part of ≥60 min a day of exercise.	As part of ≥60 min a day of exercise.
Type	Enjoyable and developmentally appropriate activities, including running, brisk walking, swimming, dancing, bicycling, and sports such as soccer, hockey, or tennis.	Muscle strengthening physical activities can be unstructured (e.g., playing on playground equipment, climbing trees or stairs) or structured (e.g., gymnastics, cycling to school, fitness).	Bone strengthening activities include running, jump rope, cycling a go-kart, hopscotch, soccer, hockey, ice skating, dancing, swimming, tennis, and fitness.

HR, heart rate. Adapted from the American College of Sports Medicine.[1]

and gradually progress to at least three days a week.[1,4] In our study, two times a week was not enough to induce effects on VO_2peak and cardiovascular health. According to the exercise guidelines and our results, at least three sessions a week of vigorous activity (i.e., HIT) are needed for children and adolescents with a chronic disease or physical disability.

Intensity

The exercise intensity should be moderate to vigorous, where for children and adolescents who are typically developing, vigorous intensities are recommended at least three days a week. Exercise intensity can be defined by the peak heart rate and the heart rate reserve (HRR); the difference between the measured or predicted maximal heart rate and resting heart rate. However, during HIT, children and adolescents were extensively encouraged to go 'all-out' within 30-seconds intervals, without need for extensive heart rate monitoring because no steady-state occurs.[6] Evaluative heart rate monitoring in several participants indicated that heart rates ≥77% of their maximal heart rates were reached easily, and following the training session near maximal (i.e., ≥96%) heart rates were achieved (Figure 9.1). These exercise intensities are in accordance with the guidelines for vigorous activity.

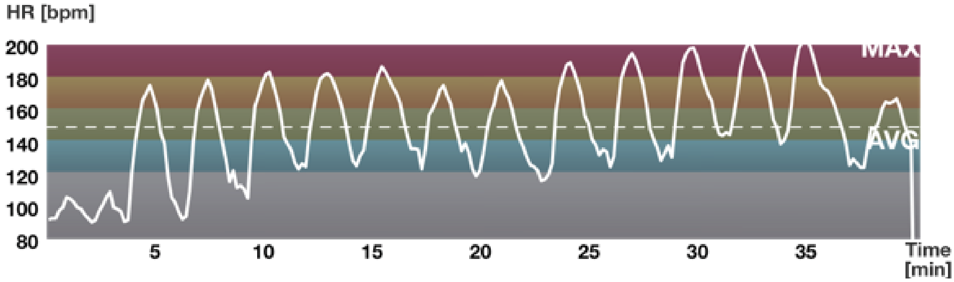

Figure 9.1: Heart rate monitoring during high-intensity interval training of a 10-year old girl.

Time

For adults, exercise should have a minimum time of 30–60 minutes for moderate exercise and 20–60 minutes for vigorous exercise. For children and adolescents, a total amount of 60 minutes a day of at least moderate exercise is recommended. It is known that only 21% of children and adolescents with a chronic disease or physical disability is physically active for at least 60 minutes a day.[2] Current HIT program was aimed to increase the amount of vigorous activity (≥20 minutes twice a week) in order to induce effects on fitness and health. However, the training volume (i.e., the product of frequency, intensity and time) of HIT was probably too low, and/or the training duration of eight weeks was too short. The duration may not have been optimal for the population included in the HIT training, assuming mechanisms of training for them are similar to those typically developing. HIT of three times a week for eight weeks or twice a week for 16 weeks should, based on a meta-analysis, elicit more effects on both fitness and health.[7]

Type

The training should consist of continuous and rhythmic exercises that involve a majority of the available muscle groups. In addition, these exercises should be enjoyable and developmentally appropriate. For children and adolescents with a chronic disease or physical disability, the type of exercise depends on their level of mobility. For HIT, especially when aiming to increase intensities in short timeframes, exercises should be easily executable and should not incorporate difficult or new skills which lower exercise intensities. In accordance with the guidelines, exercises in our study were rhythmic (i.e., running, walking or propelling a wheelchair) and involved a majority of available muscles.

Wheelchair users

Little is known about exercise training for children and adolescents who propel a manual wheelchair. The literature review (chapter 2) included only one study performed in children with disabilities (i.e., cerebral palsy and spina bifida) propelling a wheelchair. All included studies followed the ACSM guidelines with interval training as the most promising training type. Although the wheelchair users subgroup improved both agility and aerobic performance following HIT, these results should be considered exploratory because of the small sample size (n=9). More research about exercise training is needed in children and adolescents propelling a manual wheelchair.

For evaluating anaerobic performance following training, a wheelchair sprint test (e.g., muscle power sprint test (MPST)) is in accordance with a recent review on anaerobic tests for wheelchair users.[8] They suggest to use a wheelchair sprint test in people who use a wheelchair for daily locomotion instead of an arm-cranking test. This argumentation outweighs the fact that arm-cranking results in a higher mechanical efficiency compared to wheelchair propulsion, and consequently results in a higher power output.[9] The MPST intentionally incorporates both fitness variables and mechanical efficiency (i.e., energy cost and propulsion technique). Although power output is indirectly calculated using sprint time, traveled distance and weight (i.e., body mass and wheelchair mass), it is strongly correlated with the measured power output from an arm-cranking Wingate anaerobic test. When available, a more accurate measure of power output during wheelchair propulsion might be an instrumented wheel.[10] These instrumented wheels are portable and attached to a child's own wheelchair. Although more accurate, they are not used frequently in clinical practice, since they require expensive equipment and extra administration.

Reducing fat mass

As obesity is becoming a worldwide health problem,[11,12] the rising prevalence of obesity amongst children and adolescents with disabilities cannot be ignored.[13] Fat mass can be reduced following once a week school-based sports for six months, while after eight weeks of HIT fat mass remained stable. Recent studies indicate that positive effects of HIT on fat mass occur either when HIT is performed long-term (>12 weeks), and/or when participants are overweight at baseline.[14–16] Regarding our HIT program, the mean (SD) body fat percentage at baseline was 30.4 (10.4). When taking into account, independent of sex or age, participants with an extremely high body fat of ≥35% at baseline (mean 42.3% (6.2), n=20), a significant decrease of 0.8% was shown following HIT (p=.026). Although the clinical relevance of 0.8% difference is questionable, obese participants already showed improvements after eight weeks of exercise training.

In addition to exercise training, interventions targeting a reduction in energy intake as well, are more effective for reducing fat mass in children and adolescents who are typically developing.[17,18] Because of the high prevalence of malnutrition in children with CP, healthy nutrition and adequate energy intake is of paramount importance for optimum health and performance in these children.[19] Furthermore, an appropriate selection of nutrients and fluids together with the timing of intake (i.e., before and after exercise training), is also essential for exercise performance and recovery.[20] Since exercise training within rehabilitation is often compared with elite sports, rehabilitation medicine must be aware of the effects of a healthy diet. When aiming to improve physical fitness and reduce fat mass, it should be beneficial that physical therapists and sports dieticians work together as a team in clinical practice.

SUSTAINABILITY

Physical therapy

The 30-seconds all-out HIT program resulted in improvements in performance-related fitness (i.e., anaerobic performance, agility, and aerobic performance), while VO_2peak levels remained stable. With similar levels of VO_2peak, participants performed better (i.e., sprint faster, turn faster, accelerate faster). This illustrates that aerobic performance and VO_2peak measure somewhat different constructs.[21] Since peak heart rate and respiratory exchange ratio ($RER=VCO_2/VO_2$) were similar during both shuttle run/ride tests (SRTs), both exercise tests are performed at comparable maximal cardiorespiratory intensities. The increase in aerobic performance could be due to an improvement in coordination (i.e., running or walking ability, or wheelchair propulsion technique) and/or a lower energy cost for a similar activity. For rehabilitation medicine, these improvements of performance are clinically relevant. To illustrate, treatment goals within rehabilitation medicine could be: cycling for 20 minutes with family to visit grandmother (i.e., aerobic cycling performance), keeping up with peers when playing soccer, playing tag or playing wheelchair basketball (i.e., anaerobic performance and agility) or walking for 15 minutes to visit the playground (i.e., aerobic walking performance). In this way, exercise training within rehabilitation medicine should ideally be a temporary intervention or impulse aiming to achieve structural physical activity during leisure-time (i.e., participation).[22] Especially, since being physically active structurally in leisure-time at a young age is considered as a starting point for an active adulthood.[23,24] For physical therapists and other health-care professionals, it is therefore extremely important to have good connections with the child's family, to involve parents in determining treatment goals, to emphasize the focus of participation in leisure-time physical activity, and to empower parents to facilitate participation in physical activity.[25]

School-based sports

Increasing the level of sports participation once a week with 45 minutes showed positive effects after six months in anaerobic performance and fat mass. There is, and probably always will be, a discrepancy between exercise guidelines (at least three times a week) and the feasibility or priority of sports participation in families with children with physical disabilities in everyday life.[26] Parents consider, from their perspective, sports participation of once a week to be feasible and sufficient.[27] Nevertheless, still only 26% of Dutch children and adolescents with physical disabilities from schools for special education participate in sports at least once a week.[2] Although several barriers were eliminated by performing sports at school, transportation was still an issue, since parents had to arrange a one-way pick-up after school hours. Almost 90% of the parents wanted their child to participate in the school-based sports program, but arranging transportation was problematic, because of the travel distance and the business of everyday life. In the Netherlands, youth at schools for special education are often transported to and from school with taxi buses. This transportation is arranged and paid by the local municipality, and is only indicated on the basis of the school's curriculum. A postponed pick-up is therefore difficult to realize. Parents should arrange one-way transportation themselves. Although not unique for parents of youth with physical disabilities, arranging transportation might be more challenging for them. Schools can organize an opportunity to play sports in the after-school hours, however the ability of a child to participate depends on family support. In the future, involving families to prioritize sports participation for their child is one of the key factors.[25,28]

SPORTS PARTICIPATION

Next to the Sport-2-Stay-Fit study, the Health in Adapted Youth Sports (HAYS) study was performed concurrently by our research group, measuring similar outcome measures in a cross-sectional study. Results showed that sports participation at an (adapted) sports club of at least twice a week in children and adolescents with a chronic disease or physical disability is beneficial compared to peers performing no sports or only once a week. Children and adolescents who participate in sports generally have a better anaerobic performance, agility,

VO_2peak, strength, are more physically active, and have a lower fat mass.[29] In addition, youth who participate in sports have higher self-perception and quality of life.[30] These positive associations indicate that sports participation of at least twice a week is beneficial for health purposes and reveal the importance of enabling structural sports participation for this population in and by society.

In rehabilitation medicine, especially at schools for special education, improving fitness levels is an important goal. Over the past decades, youth have been encouraged to be physically active and were trained with success.[13] Schools for special education even felt responsible for these children encountering barriers within society and provided sports at school. However, for sports participation during leisure-time, society should take her responsibility. Youth should ideally have the opportunity to participate at a sports club of their choice, within the limits of their ability, in the neighborhood, because: (1) these children and adolescents can train according to current exercise guidelines,[4] (2) sports participation should not be limited to weekdays, since twice a week is more beneficial,[29,30] (3) being physically active within leisure-time will sustain towards adolescence and adulthood,[23,24] (4) they can make friendships within the neighborhood,[31] and most importantly (5) they are involved within society.[32] For effective sports participation in society, these factors have to be tackled:[33–36]

- Opportunity at sport clubs (i.e., facilities, guidance and support)
- Acceptance by society (i.e., peers, others parents and trainers)
- Family support (i.e., attitude, active engagement, and transportation)

Opportunity at sports clubs

The fact that only 26% of youth with physical disabilities from schools for special education participate in sports at an adapted or regular sports club at least once week, is partly due to the few opportunities for this population in the neighborhood.[34–36] People with physical disabilities participated at only 18% of regular sports clubs in the Netherlands in 2012.[37] According to the sports clubs without members with disabilities, several reasons are mentioned: absence of qualified trainers (31%), low number of volunteers for supervision (22%), no appropriate adapted facilities (17%), no supportive devices (15%), does not fit into our sports club (16%), no knowledge about how to implement adapted sports (15%), and never thought of it (21%).[37] Unfortunately, of all sports clubs without members with disabilities only 12% indicate no barriers for involving people with disabilities in their sports clubs in the future.[37]

Efforts have been taken by the Ministry of Health, Welfare and Sports to promote sports for youth with disabilities close to home.[38] They stated that it should also be easy for them to find sporting facilities in their neighborhood, if possible at existing sports clubs. The program stimulates inter-sectoral collaboration by setting up regional partnerships on sports and disability in which different organizations (e.g., primary healthcare providers, rehabilitation centers, schools), sport providers and local government agencies (e.g., municipalities, provinces) work together. An important role has been appointed to the neighborhood sports consultant (i.e., coaches), who have both a motivational and connecting role (i.e., match between demand and supply). Furthermore, sports clubs can provide funding to set up local projects to stimulate sports participation for youth with physical disabilities. This impulse-policy seems to work as programs like 'Special Heroes' and 'Uniek Sporten' were successfully scaled up and sustained after the funded period.[39,40] Special Heroes invites a sports club in the neighborhood to schools for special education to provide several lessons for promotion purposes. Uniek Sporten is a national online platform (www.unieksporten.nl) where the individual searching for a sports club, the neighborhood sports consultant, and all (potential) providers come together. Both the possibility to experience different sports and a central access point are important strategies to raise awareness for the options and opportunities towards promoting sports participation in society, according to a recent Delphi survey in Canada.[28]

Box 9.1: An anecdotal participant

One of the participants in the control group of the Sport-2-Stay-Fit study, a 17-year-old boy with spina bifida who propels a wheelchair, completed the HIT with success. He has been improving himself every week during the training by increasing the amount of runs within 30 seconds. He was pushing himself weekly to cross his limits and enjoyed it. It made him visibly proud to improve himself every training session. After eight weeks he improved his anaerobic performance exponentially. His physical therapist did not want him to stop exercising and improving. Since no sports program was provided at school, he encouraged him within therapy to play wheelchair basketball, learned him skills of picking up the ball from the floor, and practiced how to shoot the basket. He really loved to play wheelchair basketball and wanted to rehears every week within physical therapy. His therapist explored the opportunities of playing wheelchair basketball during leisure time at an adapted sports club in his own neighborhood. Apparently, the nearest sports club to play wheelchair basketball was a 40 km drive from his house. His parents were not able to drive him there and he was not able to go there by himself. In the end, he was not able to play his favorite sport, and did not play any sports at all.

Box 9.2: Sports club Only Friends

Only Friends is a sports club for children and adolescents with a disability. Only Friends started in Amsterdam 15 years ago and has been extended to Utrecht since September 2016.[41] Youth with physical, mental, or multiple disabilities can attend. Every Saturday morning children and adolescents can choose among different sports and games and change every 30 minutes. Their slogan is "You are good as you are", which emphasizes the focus on participation and fun, instead of competition and winning. Everyone wears the same shirt and shouts the same yell after an activity, as a team. A 10-year old boy with cerebral palsy explains the difficulty of sporting at a regular sports club: "I have played judo for a couple of months. I found it very difficult, because the other children could get me on the floor immediately. Besides, I was already tired after the warm-up".[42] Therefore he quit judo and subscribed for Only Friends Utrecht. He enjoys playing sports, and likes to play different sports: "I really like Only Friends, because I can play different sports. I have made a lot of friends and notice the physical advantage. I am getting fitter now."

Although the network for disabled sports has extensively improved, still sports are not often provided in the neighborhood as shown in the case of an anecdotal participant (Box 9.1). This is mainly caused by the absence of qualified trainers at existing sports clubs.[2] To close the knowledge gap in trainers at existing sports clubs and remove barriers to implement a program, staff and volunteers of regular sports clubs should be educated or instructed in how to train youth with physical disabilities. Additionally, sports clubs should be stimulated to recruit volunteers for supervising sports activities and clinics for this population. In the meantime, sports clubs dedicated to youth with disabilities are established with qualified trainers and appropriate facilities (Box 9.2). Despite the fact that a lot of children and adolescents can participate in sports this way and have fun, they are not involved in society where they are able to meet typically developing youth (i.e., reverse integration). In the future, more existing regular sports clubs should involve children and adolescents with physical disabilities, and integrate them into the sports club and consequently into society (Box 9.3).

Box 9.3: Hockey club Kampong

At Kampong in Utrecht, the aim is to learn the basic hockey skills within individual abilities, to have fun, and where possible to play games and tournaments.[43] The national hockey federation strives to integrate people with disabilities in regular sports clubs. They have different disciplines for individuals with mental disabilities (G-hockey), physical disabilities (LG-hockey), for those using electric wheelchairs (E-hockey), and manual wheelchairs (H-hockey).[44] For new sports clubs and trainers a LG-hockey manual is available where all relevant information about the population, the content of the training (i.e., individually based, adapted exercises, creativity, rules), and integration in the sports club is described in detail.[45] At Kampong, the training sessions of these teams take place among the regular teams of Kampong for visibility and integration. A parent of a boy with epilepsy explains her experiences at Kampong:[46] "He was very inactive, because he goes to school with a taxi bus. Therefore, at first I was afraid that he would not like it and refuse to participate. However, to our surprise he is participating, exercising and enjoying. Only the match at the end of the training remains difficult (i.e., which way to go?), but he is progressing. Every Saturday when we drive to hockey, he looks forward to it, and then so do we! I noticed that he is proud to have his own hockey club, similar to his brother. And as a parent, conversations with other parents along the hockey field are of additional value. In the end, hockey is of additional value to all of us."

Acceptance by society

Youth with a physical disability in the Netherlands are often isolated from society, because they attend schools for special education and, if at all, participate in sports at adapted (i.e., other or school-based) sports clubs. That is probably the reason why these children and adolescents most often do not have friends in their neighborhood.[47] Ideally, youth with and without a disability should participate in sports together, at least at the same sports club. A vast majority of the Dutch population agreed that this integration is beneficial for both children with disabilities (60%) and without disabilities (79%).[32] It is promising that the Dutch population is open-minded with regards to the integration of youth with physical disabilities. However, much should be changed in the organization of sports clubs in our society. The integration of youth with physical disabilities within sports clubs is often dependent on one person in the organization or even a parent. This should change, society should not exclude them and prejudice their disability, but rather include them and focus on their ability. Among

Dutch sports clubs or within society, it is often about winning and being competitive. In this way, children and adolescents with a chronic disease or physical disability will never have a fair chance. The Olympic spirit is not about winning medals, but is about the participation that counts; "the essential thing is not to have conquered but to have fought well". That spirit should be brought into society and especially into Dutch sports clubs, so that youth with a disability can participate as well, within their own possibilities.

Family support

Children and adolescents with physical disabilities are often dependent on parental or caregivers support to be able to participate in sports. Practically, there is need for assistance with medical care, transportation, financial support, and supervision or guidance during the activity itself. On the other hand, encouragements or parents' attitude towards sports participation are also important. Parents' interests and priorities about physical activity or sports very much determine a child's experiences of exercise and foster engagement and enjoyment of physical activities.[48] According to health care professionals, a families' attitude towards sports participation determines whether a child has an opportunity.[36] A family-centered approach focusing on solutions and possibilities with active engagement and involvement of parents, supports awareness of the benefits of sports participation.[25,35] This can also be partly addressed by health care professionals (e.g., physicians, physical therapists, nurses) providing attention for, and information about, an active lifestyle throughout their regular hospital visits.[35] Implementing this subject explicitly as a standard component in usual care, will highlight the importance and probably support parents.

ACTIVE LIFESTYLE

According to the exercise recommendations of typically developing youth, it is advised to engage in moderate-to-vigorous intensity physical activity every single day including at least three days a week of vigorous intensity activities (i.e., sports).[1,5] In the Netherlands, only 21% of youth with a chronic disease or physical disability meet these guidelines of daily activity.[2] In the Sport-2-Stay-Fit study, we did not monitor intensity of physical activity, but measured mode of activity objectively. Youth able to run, participate on average four minutes a day in running activities, and 19 minutes a day in cycling activities. For youth able to walk, this was 2 and 17 minutes, respectively, for running and cycling. Assuming that the other modes of activities (i.e., walking, standing, sitting and lying) are not moderate-to-vigorous activities, ambulatory youth with a physical disability are engaged in only 23 minutes (runners) and

19 minutes (walkers) of moderate-to-vigorous activities a day on average. This might be an underestimation, because walking can be of moderate intensity in some children and adolescents.[49] However, it is obvious that increasing physical activity in daily life, next to sports participation, is of importance for health purposes in this population.[50,51]

Next to physical activity levels, sedentary behavior can also have adverse health effects, even among those who exercise regularly.[52–54] Sedentary behavior is defined as any waking behavior characterized by an energy expenditure ≤1.5 metabolic equivalents (METs) while in a sitting, reclining or lying posture.[55] There is increasing evidence that limiting sedentary time impacts body composition and metabolic risk factors positively in adults, regardless of whether an individual meets the physical activity recommendations.[56–58] Ambulatory children and adolescents with CP are higher sedentary with fewer breaks compared to youth who is typically developing.[59] These insights of concurrently decreasing sedentary time, may be beneficial for children and adolescents with a chronic disease or physical disability. Although evidence is limited, it is encouraged to break-up sedentary behavior every hour by 1–5 minutes standing or walking in the general population.[60] The optimal patterns of physical activity and sedentary behavior in children and adolescents with a chronic disease or physical disability require further investigation.[61]

At school

During weekdays, most of the time is spent at school. Participants in the Sport-2-Stay-Fit study received 86 minutes of physical education on average, divided over 1.7 times a week. In general, children within cluster 3 in primary special schools receive between 63 and 78 minutes of physical education a week.[62] In our population, especially adolescents at secondary schools received physical education only once a week for 60–100 minutes, probably due to internships outside school. Because the focus of physical education is on learning skills and learning to play together, half of the time is spent on skill practice, physical education knowledge and management.[63] Within physical education, only 40% of time is dedicated to moderate-to-vigorous activities (Figure 9.2). Compared to break time (i.e., free play), children and adolescents spend 51% of time moderate-to-vigorously active. Furthermore, the highest amount of time spent moderate-to-vigorously active was during school-based sports (60%). When aiming to increase physical activity during daily life within school hours, the easiest way is to allow youth to spend more time playing outside actively. Besides, schools can provide school-based sports between lessons or during lunch breaks, focusing on playing the game or being active instead of practicing skills. Increasing physical activity does not have to be difficult or destined for a physical educator. A positive

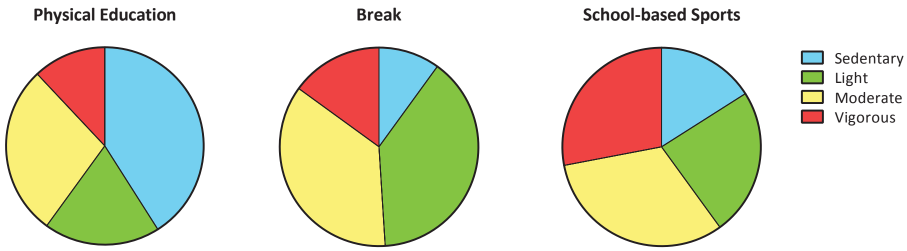

Figure 9.2: Distribution of activity levels (%) during physical education, pause, and school-based sports in children and adolescents with a chronic disease or physical disability.

and relatively easy example of increasing physical activity is 'The Daily Mile', where children are encouraged to walk or run a block around school in 10-15 minutes three times a week.[64]

In society

Children and adolescents with physical disabilities should be empowered by the community to do activities themselves when possible and to ask for help when necessary. Most of their disabilities already exist at birth or in early childhood and youth are at risk to develop learned helplessness, when parents, family, teachers, taxi drivers, students and peers help them without asking. Within the social environment of a child, concerns and fears exist associated with safety and the ability of the child to manage physical activity.[25] Therefore, parents should be involved within physical therapy to learn and recognize that they are able to facilitate participation of the same activity in their home and community. Involving parents will help them to become less protective. Another factor in stimulating physical activity is creating awareness of the value of physical activity concerning the beneficial effects on fitness and health. If families do not inherently value physical activity, they are unlikely to engage in, or sustain, physical activity behavior.[65] When parents are ready to change their physical activity behavior, they can start to focus on opportunities and abilities instead of impairments. For example, active transportation instead of using a car (i.e., walking or cycling to school, family or the supermarket) might be a good starting point in getting active structurally.

CLINICAL IMPLICATIONS

Based on the evidence of this thesis as well as clinical experience, the current section includes several tips and tricks regarding exercise training and sports for clinicians, trainers and coaches working with youth with physical disabilities.

Fun

Logically, exercise training is only beneficial if a child engages in it. To this end, individual preferences and enjoyment should be taken into account; find out what they want to do! This can be done by asking what they like and/or let them experience different sports (e.g., physical therapy, Special Heroes). Children and adolescents are more likely to enjoy short-term, high-intensity bouts of exercises, because they usually offer the necessary variety and better match their usual daily activity pattern.[66] For youth with disabilities especially, the modification of an exercise, rules and equipment are important factors for enjoyment,[67] because the experience of success will not be accomplished when exercises are too difficult. Adapting on individual abilities within a training session, allows children and adolescents to participate and be involved. In addition to the activity itself, friends and social interaction were recognized from a child's perspective as significant contributors of having fun.[68] The social character of sports (e.g., belonging to a club and team, playing a game) enhances children's feelings of inclusion and acceptance by others.

Safety

Regarding safety, in our study no exercise-related injuries occurred. In general, it is important to exercise within pain limits and to wear well-fitting orthoses. Additionally, it is advised to inform yourself about the diagnoses, especially when you are not familiar with them. Parents or physical therapists can inform you whether the child is allowed to participate in (high-intensity) physical activities without restrictions. Contraindications for exercise training, specifically for this population, are uncontrolled epilepsy, severe osteoporosis, uncontrolled congenital cardiac disease, and recent surgery.[69] In these cases, always consult their physician and/or perform a cardiopulmonary exercise test before starting exercise.

Feasibility

When possible, play regular sports with normal rules. If necessary, adapt exercises, rules and equipment to the ability of the child individually. Be creative in involving youth with physical disabilities in sports, adapt the rules and focus on participating instead of competition. In addition, to monitor children's fitness levels use available adapted physical fitness tests (e.g., shuttle run/ride test) instead of regular tests in which they always score below average.[70] In 30 seconds 'all-out' HIT, regardless of their disability, all children exercise at their own mobility level. For encouragement, children and adolescents can be instructed to count their amount of repetitions within one session and be motivated to improve themselves.

Fatigue

Compared with youth who are typically developing, those with neuromuscular disorders who are ambulatory have altered movement patterns, which results in a poor economy and high energy expenditure during walking.[49] This implies that these children achieve higher exercise intensities during walking at similar walking speed compared to typically developing peers. When dribbling a ball at 4 km/h, one child may be exercising at 50% of their maximal heart rate, when the other child is performing similar exercise at 90%. Hence, in youth with physical disabilities one size does not fit all; the same exercise can induce other training intensities or may be too difficult to coordinate. This possibility of higher fatigue values is important to keep in mind when working with a group of children and adolescents with different motor disabilities. Heart rate monitoring is therefore of paramount importance when, for example, an exercise is intended to be at low-intensity to ensure recovery time. Otherwise, the exercise should be adapted to ensure low-intensity activity and thus sufficient recovery time.

CONCLUSIONS

The main objective of the current thesis was to investigate the effects of a short-term HIT followed by a school-based sports program for six months on fitness and health in youth with physical disabilities. We found that the increased anaerobic performance, agility and aerobic performance after eight weeks of HIT sustained following school-based sports in both the sport and control group, where anaerobic performance increased further compared to the control group. Regarding cardiometabolic health, only fat mass improved in favor of the sport group. The once a week school-based sports program showed no effects on psychosocial health and attention. Although school-based sports are safe and feasible, it has been argued that youth with physically disabilities should ideally participate at sports club within society. To facilitate sports participation at sports clubs in society, trainers, staff and volunteers should be educated and instructed in how to train youth with physical disabilities (Box 9.4).

The secondary objective was to investigate ecological time trends over a decade in fitness levels of youth with CP. Between 2004 and 2014, positive time trends were shown in anaerobic performance, agility and aerobic performance, but not in VO_2peak. Furthermore, the substantial higher body mass and BMI of youth with CP in 2014 compared to a comparable sample from 2004 is alarming. Health care professionals are advised to emphasize the importance of an active and healthy lifestyle to create awareness of the benefits (Box 9.5).

Box 9.4: Recommendations for trainers

- Inform yourself (e.g., parents, child) about the diagnose and possible behavioral or medical considerations to take into account regarding exercising
- One size does not fit all: adapt exercises, rules and equipment to their ability ensuring success experiences, having fun, and intended exercise intensities
- Be aware of higher fatigue values because of poor economy of movement, use heart rate monitoring and progressively increase training volume, especially when deconditioned

In the end, they are just kids, do not unnecessarily medicalize and make it FUN!

Box 9.5: Recommendations for clinical practice

- Inquire about their daily activity pattern
- Emphasize the importance, and involve and empower parents about structural physical activity and the break-up of sedentary time
- When a child or parent is searching for a sports provider refer to http://www.unieksporten.nl
- Consult a (sports) dietician when aiming to improve body composition

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Samenvatting

Het is algemeen bekend dat regelmatig sporten belangrijk is voor jongeren. Jongeren met een fysieke beperking die naar speciaal onderwijs gaan sporten minder vergeleken met hun leeftijdsgenoten. Dit heeft negatieve gevolgen voor hun fitheid en gezondheid. Het is voor deze groep jongeren vaak lastiger om te gaan en blijven sporten bij een sportvereniging in de buurt. Steeds vaker wordt hierdoor sport op school, buiten lestijd, aangeboden. Dit proefschrift onderzoekt het effect van een intervaltraining gevolgd door (na)schoolse sport op fitheid en gezondheid bij jongeren met een fysieke beperking op speciaal onderwijs; de Sport-2-Stay-Fit studie. Allereerst zal de aanleiding en achtergrond van het onderzoek besproken worden, vervolgens zal per hoofdstuk worden toegelicht wat er onderzocht is.

Speciaal onderwijs

Leerlingen die specialistische of intensieve begeleiding nodig hebben, gaan in Nederland naar het speciaal onderwijs. Deze scholen hebben dezelfde doelen als regulier onderwijs. De leerlingen in het speciaal onderwijs krijgen wel extra begeleiding en meer tijd om deze doelen te halen. Het speciaal onderwijs is onderverdeeld in vier clusters gericht op de soort beperking of stoornis: (1) visueel, (2) auditief, (3) fysiek en (4) gedragsproblematiek. Het onderzoek in dit proefschrift richt zich op cluster 3; jongeren (8–19 jaar) met een fysieke beperking. In Nederland zijn er totaal 26.500 leerlingen die cluster 3 onderwijs volgen. Dit is 1.9% van de totale populatie leerlingen die naar basis- en middelbaar onderwijs gaan.

Doelgroep

De meest voorkomende fysieke beperking bij jongeren is cerebrale parese (CP). CP is een niet-progressieve aandoening die ontstaat door een hersenbeschadiging voor het eerste levensjaar. Jongeren met CP hebben hierdoor problemen met hun grove motoriek. Ze hebben vaak spasme in een of meerdere ledematen. Afhankelijk van de ernst van de CP kunnen zij lopen, rennen of gebruiken zij een rolstoel om zich voort te bewegen. Naast de fysieke beperking, hebben zij vaak in meer of mindere mate andere stoornissen, zoals cognitieve functiestoornissen of gedragsproblematiek. Andere aandoeningen die in de onderzoeksgroep voorkomen zijn spina bifida (open ruggetje), psychomotore ontwikkelingsachterstand en stofwisselingsziekten. De groep jongeren met een fysieke beperking op speciaal onderwijs is dus heel divers. Daarom hebben we in dit proefschrift de onderzoeksgroep ingedeeld op functieniveau aan de hand van de Functional Mobility Scale (FMS):

- Renners: jongeren die kunnen rennen met zweefmoment,
- Lopers: jongeren die kunnen lopen, maar niet kunnen rennen,
- Rolstoelrijders: jongeren die een handbewogen rolstoel gebruiken.

Fitheid

Jongeren met een fysieke beperking zijn minder actief in het dagelijks leven. Zij sporten minder vergeleken met hun leeftijdsgenoten; 26% van de kinderen met een fysieke beperking sport wekelijks tegenover 71% van hun leeftijdsgenoten. Het is voor jongeren met een fysieke beperking vaak lastiger om te sporten. Het ontbreken van bekwame trainers of begeleiders is een vaak gehoorde barrière om te gaan sporten bij een sportverenging. Ook voelen ze zich vaak niet geaccepteerd of is een geschikte sportvereniging te ver weg. Doordat jongeren met een fysieke beperking minder bewegen dan hun leeftijdsgenoten, hebben zij logischerwijs ook een lagere fitheid. Deze lage fitheid heeft negatieve gevolgen voor hun gezondheid. Ook, of mogelijk zelfs daardoor, hebben jongeren met CP op latere leeftijd een grotere kans op het krijgen van diabetes, hoge bloeddruk en een beroerte. Een goede fitheid is daarnaast sterk gerelateerd aan positieve waardes op zelfperceptie, kwaliteit van leven en een beter concentratievermogen op school.

Uit eerder onderzoek is gebleken dat jongeren met een fysieke beperking positief reageren op training; als ze gestructureerd gaan trainen (bijvoorbeeld krachttraining of conditietraining), dan gaan ze conditioneel vooruit. Helaas blijft deze vooruitgang niet behouden als het trainingsprogramma stopt. Voor deze doelgroep geldt dus ook het "use it or lose it"-principe; structureel blijven bewegen is essentieel om de fitheid te behouden. Een trainingsprogramma aangeboden tijdens de fysiotherapie is meestal tijdelijk, en de effecten hiervan zullen op lange termijn dan ook niet behouden blijven. Om te onderzoeken wat er nodig is om de trainingswinst te behouden in deze minder fitte doelgroep, hebben we in dit proefschrift onderzocht of 1x per week (na)schoolse sport voldoende is. Sporten op school, buiten school- of lestijd lijkt een goede methode om de genoemde barrières te doorbreken, waardoor deze jongeren structureel kunnen sporten.

Rolstoelrijders: trainen en testen

De beoogde doelgroep bestaat onder andere uit rolstoelrijdende jongeren. Er is nog weinig bekend over hoe deze groep het beste te trainen en te testen is. Daarom is dit eerst onderzocht in **hoofdstuk 2**. Hierin hebben we een literatuuroverzicht gemaakt met alle trainingsstudies die de fysieke conditie van het rolstoelrijden verbeteren. In dit onderzoek ging het niet om de vorm van training, maar om het effect op de conditie van het rolstoelrijden. Dat was ook het belangrijkste onderzoeksresultaat. Dit kon zowel de sprintconditie (anaerobe fitheid) als de duurconditie (aerobe fitheid) zijn. Op basis van de huidige literatuur lijkt training positieve effecten te hebben op de conditie van het rolstoelrijden, waarbij intervaltraining de meest belovende trainingsvorm is.

Om een dergelijke intervaltraining bij rolstoelrijders te kunnen evalueren, zijn er testen nodig om de fitheid vóór en na de training in kaart te brengen. Deze testen zijn ontwikkeld en onderzocht in **hoofdstuk 3**. Voor de anaerobe fitheid zijn twee sprinttesten ontwikkeld: de muscle power sprint test (MPST) en de 10x5 meter sprinttest. De MPST is een test waarbij jongeren 3x over een afstand van 15 meter moeten sprinten met tussendoor 10 seconden pauze. Bij de tweede test moeten de jongeren 10x heen en weer sprinten over een afstand van 5 meter zonder pauze. In deze test wordt dus ook de draai- en startsnelheid meegenomen. Beide testen zijn in een groep van 23 rolstoelrijdende jongeren met CP onderzocht op betrouwbaarheid en validiteit. Dit zijn eigenschappen die bepalen of een test reproduceerbaar is (herhaaldelijk hetzelfde meet) en of de test meet wat je wilt meten (validiteit). Uit het onderzoek is gebleken dat zowel de MPST als de 10x5 meter sprinttest betrouwbaar en valide zijn om te gebruiken in de praktijk.

Protocol Sport-2-Stay-Fit studie

Nu het ook voor de rolstoelgebonden groep duidelijk is welke training het meest effectief is en hoe dit is te evalueren, is de Sport-2-Stay-Fit studie opgezet. Het protocol van deze studie wordt behandeld in **hoofdstuk 4**. Het onderzoek vond plaats op vier scholen voor speciaal onderwijs in Nederland. Alle jongeren met een fysieke beperking die aan het onderzoek deelnamen volgden eerst een high-intensity interval training (HIT) om bekend te raken met inspanning en om een verhoogd fitheidsniveau te creëren. Na de HIT is de helft van de groep verdergegaan met (na)schoolse sport. De andere helft kreeg geen (na) schoolse sport aangeboden en fungeerde daardoor als controlegroep. De groepen werden ingedeeld op basis van de aanwezigheid van een (na)schools sportaanbod op school. Op twee scholen draaide al een (na)schools sportaanbod of hadden ze de intentie om dit op te starten. Het (na)schoolse sportaanbod is geïnitieerd door gemotiveerde sportdocenten die ook de sportlessen verzorgen. De lessen vinden plaats na schooltijd of in pauzes. Deelnemers wisten dus voor de start van de studie wat hen te wachten stond: HIT én (na)schoolse sport (sport groep) of HIT en controlegroep. De uitkomstmaten werden gemeten zowel voor de start van de HIT (T0), direct na acht weken HIT (T1), als na de zes maanden (na)schoolse sport of controle (T2). Naast de sprinttesten voor anaerobe fitheid, werd de aerobe fitheid (duurconditie) gemeten met de shuttle run test (piepjestest). De deelnemer loopt of rijdt tijdens deze test heen en weer tussen twee pionnen op het tempo van piepjes, waarbij de snelheid van de piepjes toeneemt. De behaalde trap zegt iets over de aerobe prestatie; hoe langer iemand de test kan volhouden, hoe hoger de aerobe prestatie. Tijdens de test werd de in- en uitgeademde lucht gemeten middels een mobiel zuurstofanalyse systeem. Hiermee is de maximale zuurstofopname (VO2piek) bepaald, een maat voor de conditie, die gerelateerd is aan veel gezondheidsparameters.

Effect van acht weken HIT

Een HIT lijkt een effectievere en efficiëntere manier van trainen vergeleken met klassieke duurtraining. Onderzoek laat zien dat bij HIT met veel minder tijd en een lager trainingsvolume dezelfde of zelfs betere resultaten behaald kunnen worden op fitheid en gezondheid. In **hoofdstuk 5** zijn de effecten van acht weken HIT onderzocht bij 70 jongeren (8–19 jaar) met een fysieke beperking. De groep bestond uit 36 renners, 25 lopers en 9 rolstoelrijders, met CP als meest voorkomende diagnose (36%). Ze trainden twee keer per week gedurende 30 minuten, waarbij ze zich tijdens 8–12 series van 30 seconden maximaal moesten inspannen. Tijdens de 30 seconden moesten ze zo vaak mogelijk heen en weer sprinten om bijvoorbeeld pittenzakjes naar de overkant te brengen. Hierna hadden ze 90 tot 120 seconden actief herstel, waarbij ze konden uitrusten, maar niet mochten gaan zitten. Na acht weken was de anaerobe fitheid (sprintconditie) verbeterd in alle groepen. Ook de behaalde trap (aerobe prestatie) op de shuttle run test verbeterde, terwijl de VO_2piek (maat voor duurconditie) gelijk bleef. Dit betekent dat jongeren met dezelfde conditie het sporten langer vol kunnen houden. De bloeddruk leek iets te dalen, maar die resultaten waren niet consistent over de verschillende groepen. Er zijn na de acht weken HIT geen effecten gevonden op kracht, aderstijfheid, vetpercentage en cholesterol.

Effect van zes maanden (na)schoolse sport

Na de HIT ging de helft van de groep door met (na)schoolse sport. De (na)schoolse sport werd 1x per week 45 minuten gegeven door een ervaren sportdocent. Voorbeelden van de (team)sporten die tijdens de (na)schoolse sport werden gedaan zijn: voetbal, (rolstoel) basketbal, (rolstoel)hockey en/of eenvoudige tikspellen. Deze sportles verschilt van een reguliere gymles, omdat de nadruk bij de gymles ligt op het 'leren bewegen', terwijl het tijdens de (na)schoolse sport om 'intensief bewegen' gaat. De resultaten van de (na)schoolse sport zijn weergegeven in **hoofdstuk 6** en **hoofdstuk 7**. Uiteindelijk zaten 31 jongeren in de sportgroep en 40 jongeren in de controlegroep. Na zes maanden 1x per week (na)schools sporten presteerde de sportgroep gemiddeld 16% beter op de MPST (sprintconditie) ten opzichte van de controlegroep. Dit positieve effect komt bovenop de toename die we zagen na de HIT. Opvallend is dat de controlegroep het positieve effect na de HIT van acht weken behoudt, terwijl we verwachtten dat deze terug zou zakken richting het beginniveau. Zes maanden (na)schoolse sport verbeterde ook het vetpercentage van de sportgroep met gemiddeld -2,8% vergeleken met de controlegroep. Er werden geen effecten gevonden op aerobe prestatie, VO2piek, kracht en fysieke activiteit. Daarnaast lieten bloeddruk, aderstijfheid en cholesterol ook geen verbeteringen op groepsniveau zien. Het lijkt erop dat vooral de jongeren die aan het begin van het sportprogramma slecht scoren op gezondheidsparameters profijt hebben van 1x per week sporten. Uitkomstmaten waar 1x per week (na)schoolse sport geen effect op lijkt te hebben zijn aandacht en psychosociaal functioneren. Doordat de sport op school wordt aangeboden zijn omgevingsfactoren weggelaten, die mogelijk een belangrijke bijdrage kunnen leveren aan het psychosociaal functioneren, zoals nieuwe vriendschappen maken, ouderlijke steun, en gevoel bij een vereniging of team te horen.

Kinderrevalidatie: vroeger en nu

Vroeger waren behandelingen bij jongeren met een fysieke beperking vooral gericht op de esthetiek van bewegen en niet op functieniveau. Pas sinds de jaren '90 weten we dat kracht- en conditietraining de aandoening of spasticiteit bij bijvoorbeeld CP niet verergert, maar juist een positieve bijdrage levert op functieniveau. De eerste grootschalige trainingsstudie bij jongeren met CP vond plaats in Nederland in de jaren '90. Sinds die tijd hebben de fysieke behandelingen binnen de kinderrevalidatie op speciaal onderwijs een vlucht genomen. De vooruitgang na acht weken HIT was minder groot dan we op basis van eerder onderzoek verwacht hadden. Zou het kunnen zijn dat jongeren met een fysieke beperking van nu fitter zijn vergeleken met vroeger? In **hoofdstuk 8** worden de baseline data van een studie uit 2004 vergeleken met de baseline data van de Sport-2-Stay-Fit studie uit 2014 op anaerobe fitheid, aerobe fitheid en body mass index (BMI). In totaal worden 25 jongeren met CP per groep meegenomen, die vergelijkbaar zijn qua geslacht, lengte en ernst van de aandoening. De resultaten laten zien dat zowel anaerobe fitheid (23–99%) als de behaalde trap op de shuttle run test (67%) enorm verbeterd zijn in 10 jaar, terwijl de VO_2piek gelijk is gebleven. De data laten ook zien dat dat het lichaamsgewicht (26%) en de BMI (20%) zijn toegenomen.

Toekomst

De bevindingen van dit proefschrift zijn in **hoofdstuk 9** in perspectief geplaatst van de huidige wetenschappelijke kennis en het sportklimaat in Nederland. Parallel aan de huidige studie werd de HAYS studie uitgevoerd, waarbij dezelfde uitkomstmaten cross-sectioneel werden gemeten. Zij vergeleken jongeren met een fysieke beperking die minstens 2x per week sporten met jongeren die maar 1x per week of helemaal niet sporten. De sportende groep heeft een betere anaerobe en aerobe fitheid, ze zijn actiever in het dagelijks leven, hebben een lager vetpercentage, en een hogere zelfperceptie en kwaliteit van leven. De (na)schoolse sport is ontstaan vanuit speciaal onderwijs doordat jongeren met een fysieke beperking niet altijd terecht kunnen bij een sportvereniging. Voor de toekomst zou de samenleving haar verantwoordelijkheid moeten nemen, zodat deze jongeren in hun vrije tijd in de buurt kunnen sporten. Op deze manier kunnen ze ook in het weekend sporten (2x per week), komen ze in contact met jongeren uit de buurt en worden ze meer betrokken in de samenleving.

Kindersamenvatting

Dit boekje gaat over kinderen met een beperking. Dat zijn kinderen die bijvoorbeeld moeite hebben met lopen, of een rolstoel gebruiken. Deze kinderen sporten veel minder dan kinderen zonder een beperking. Ze kunnen niet overal aan meedoen, omdat ze moeite hebben met de sportbewegingen. Het is ook niet altijd mogelijk om met een rolstoel bij een sportvereniging komen.

VROEGER dachten de dokters en fysiotherapeuten dat sporten voor kinderen met een beperking slecht was. De beperking zou dan erger worden. Gelukkig weten we inmiddels dat dat niet zo is. **NU** denken we dat sporten voor kinderen met een beperking heel goed is en dat ze **NU** ook meer mogen bewegen. Daarom hebben we gekeken of kinderen met een beperking van **NU** fitter zijn dan kinderen van 10 jaar geleden. En wat blijkt: kinderen met een beperking van **NU** zijn fitter dan de kinderen van **VROEGER**. Dat komt waarschijnlijk omdat ze tijdens therapie en onder schooltijd veel meer trainen en bewegen.

Om te kijken hoe goed sporten **NU** is voor kinderen met een beperking, hebben we kinderen die extra gingen sporten op school vergeleken met kinderen die niet extra gingen sporten. De extra sportles werd gegeven door de gymjuf of gymmeester in de pauze of na schooltijd. Het maakte niet uit hoeveel moeite kinderen hadden met lopen. Ook kinderen die in een rolstoel sporten, deden mee. We hebben gekeken of de kinderen sneller, sterker en gezonder werden van sporten. Na een half jaar konden de kinderen die een extra sportles volgden harder rennen vergeleken met kinderen die niet extra gingen sporten. Daarnaast hadden de kinderen van de extra sportles ook minder vet in hun lichaam. Sporten is dus ook goed voor kinderen met een beperking. Het helpt voor een gezonder en fitter lijf.

NU we weten dat sporten goed is, zouden kinderen met een beperking in de **TOEKOMST** ook gewoon moeten kunnen sporten bij een sportvereniging in de buurt. Helaas is dat **NU** vaak nog niet mogelijk en sporten ze bij een aparte sportvereniging (op school) voor kinderen met een beperking. Als ze lid zouden worden van een sportvereniging in de buurt, dan kunnen ze in het weekend ook makkelijker sporten. Dan halen ze twee keer per week sporten. Dat is waarschijnlijk nog beter.

VROEGER

NU

TOEKOMST


5

Dankwoord

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About the author

CURRICULUM VITAE

Maremka Zwinkels was born in Delft, the Netherlands, on September 4th, 1987. After completing secondary school at ISW in Naaldwijk, she studied Human Movement Sciences at the VU University in Amsterdam. During her studies, she was secretary in the board of the study association of the faculty of Human Movement Sciences (V.I.B.). In 2009 she qualified as a group fitness instructor and started working at the University Sports Centre (USC) in Amsterdam. In 2010, she completed her master thesis on the effects of electrical stimulation in spinal cord injury at Reade in Amsterdam.

After working 2.5 years as research assistant and junior researcher at the Center of Excellence for Rehabilitation Medicine (a collaboration between the University Medical Center Utrecht and De Hoogstraat Rehabilitation) she started her PhD in 2013 on the project 'From exercise training to school-based sports: the effects on fitness and health in youth with physical disabilities'. This PhD project investigated the effects of high-intensity interval training and a school-based sports program on fitness and health outcomes in youth with physical disabilities across four schools for special education. From August to December 2016, she combined her research with clinical practice at De Hoogstraat Rehabilitation, where she worked as an exercise physiologist for an innovation project. The aim of the project was to demonstrate the additional value of exercise testing in rehabilitation. During her PhD, she also followed the research education program 'Clinical and Experimental Neuroscience' at the Graduate School of Life Sciences at Utrecht University. In July 2017, she was appointed as exercise physiologist at De Hoogstraat Rehabilitation. At this center she performs exercise tests in patients with chronic health conditions like cerebral palsy, stroke and spinal cord injury, and advises rehabilitation physicians and therapists about potential exercise training.

Maremka lives together with Paul in Amsterdam. In her free time she likes to go out for a run, play indoor soccer, go out dancing, and spend quality time with friends and family.

PUBLICATIONS

International publications

Zwinkels M, Takken T, Ruyten T, Visser-Meily JMA, Verschuren O. Body mass index and fitness in high-functioning children and adolescents with cerebral palsy: what happened over a decade? Res Dev Disabil 2017; 71: 70–6.

Haapala EA, Lankhorst K, de Groot J, **Zwinkels M**, Verschuren O, Wittink H, Backx FJ, Visser-Meily A, Takken T. The associations of cardiorespiratory fitness, adiposity and sports participation with arterial stiffness in youth with chronic diseases or physical disabilities. Eur J Prev Cardiol 2017; 24: 1102–11.

Lankhorst K, Van der Ende–Kastelijn K, de Groot J, **Zwinkels M**, Verschuren O, Backx F, Visser-Meily JMA, Takken T. Health in Adapted Youth Sports study (HAYS): health effects of sports participation in children and adolescents with a chronic disease or physical disability. Springerplus 2015; 4: 796.

Zwinkels M, Verschuren O, Lankhorst K, Van der Ende-Kastelijn K, de Groot J, Backx F, Visser-Meily A, Takken T. Sport-2-Stay-Fit study: health effects of after-school sport participation in children and adolescents with a chronic disease or physical disability. BMC Sports Sci Med Rehabil 2015; 7: 22.

Zwinkels M, Verschuren O, Janssen TW, Ketelaar M, Takken T. Exercise training programs to improve hand rim wheelchair propulsion capacity: a systematic review. Clin Rehabil 2014; 28: 847–61.

Verschuren O, **Zwinkels M**, Obeid J, Kerkhof N, Ketelaar M, Takken T. Reliability and validity of short-term performance tests for wheelchair-using children and adolescents with cerebral palsy. Dev Med Child Neurol 2013; 55: 1129–35.

Smit CA, **Zwinkels M**, van Dijk T, de Groot S, Stolwijk-Swüste JM, Janssen TW. Gluteal blood flow and oxygenation during electrical stimulation-induced muscle activation versus pressure relief movements in wheelchair users with a spinal cord injury. Spinal Cord 2013; 51: 694–9.

Verschuren O, **Zwinkels M**, Ketelaar M, Reijnders-van Son F, Takken T. Reproducibility and validity of the 10-meter shuttle ride test in wheelchair-using children and adolescents with cerebral palsy. Phys Ther 2013; 93: 967–74.

Submitted publications

Zwinkels M, Verschuren O, de Groot JF, Backx FJ, Wittink H, Visser-Meily JMA, Takken T. Effects of high-intensity interval training on fitness and health in youth with physical disabilities. Submitted.

Zwinkels M, Verschuren O, Balemans A, Lankhorst K, te Velde S, van Gaalen L, de Groot J, Visser-Meily JMA, Takken T. Effects of a school-based sports program on physical fitness, physical activity and cardiometabolic health in youth with physical disabilities: data from the Sport-2-Stay-Fit study. Submitted.

Zwinkels M, Ketelaar M, Nijboer T, Verschuren O, te Velde S, de Groot J, Takken T, Visser-Meily JMA. Effects of a school-based sports program on psychosocial health and attention in children and adolescents with a chronic disease or physical disability: a controlled clinical trial. Submitted.

National publications

Tuijtelaars J, **Zwinkels M**, Lankhorst K, Duijf M, Takken T. Testen fitheid bij kinderen met een chronische aandoening of beperking. https://www.allesoversport.nl/ 3 oktober 2016.

Lankhorst K, **Zwinkels M**, De Groot J, Verschuren O, Takken T. Is sport gezond voor chronisch zieke jongeren? Sportgericht 2016 Jun 3: 26.

PRESENTATIONS

International scientific conferences and presentations

Zwinkels M. HIT Rehabilitation Medicine – High-intensity Interval Training in clinical practice. Dutch Congress of Rehabilitation Medicine (DCRM), November 9-10th 2017, Maastricht, the Netherlands.

Zwinkels M. Sport-2-Stay-Fit: school-based sports in children and adolescents with a chronic disease or physical disability. Summer School Paediatric Sport and Exercise Medicine, August 23th 2017, Utrecht, the Netherlands.

Zwinkels M, Takken T, Visser-Meily JMA, Verschuren O. High-intensity interval training (HIT) in children with a chronic disease or condition. Poster presentation. American College of Sports Medicine (ACSM) annual meeting. May 30th–June 3rd 2017, Denver, Colorado, USA.

Zwinkels M, Takken T, Ruyten T, Visser-Meily JMA, Verschuren O. Fitness trends in children and adolescents with cerebral palsy between 2004-2014: fatter & fitter? Poster presentation. European Academy for Childhood Disability. May 17–20th 2017, Amsterdam, the Netherlands.

Zwinkels M, Takken T, Visser-Meily JMA, Verschuren O. High-intensity interval training (HIT) in children with a chronic disease or condition. Poster pitch and presentation. 3rd European Workshop on Clinical Pediatric Exercise Testing. November 11–12th 2016, Zeist, the Netherlands.

Zwinkels M, Verschuren O, Ruyten T, Borcherts E, van Dinther K, Elfrink M, Steenweg G, Lankhorst K, van der Ende-Kastelijn, de Groot JF, Backx FJG, Visser-Meily JMA, Takken T. How effective is high-intensity interval training (HIT) in children with a chronic disease or condition: preliminary results. Oral presentation. Pediatric Work Physiology meeting XXIX, September 9–13th 2015, Utrecht, the Netherlands.

Zwinkels M, Verschuren O, Lankhorst K, Van der Ende-Kastelijn K, De Groot J, Backx F, Visser-Meily A, Takken T. Research design of the Sport-2-Stay-Fit study: afterschool sport participation for children with a chronic disease or condition. Poster presentation. 2nd European workshop on Clinical Pediatric Exercise Testing, November 7–8th 2014, Zeist, the Netherlands.

Zwinkels M, Verschuren O, Lankhorst K, Van der Ende-Kastelijn K, De Groot J, Backx F, Visser-Meily A, Takken T. Research design of the Sport-2-Stay-Fit study: afterschool sport program for children with a physical disability. Utrecht Summer School, April 14th 2014, Utrecht, the Netherlands.

Zwinkels M, Verschuren O, Janssen TWJ, Ketelaar M, Takken T. Exercise training programs to improve hand rim wheelchair propulsion capacity: a systematic review. Poster presentation. Rehabmove congress, April 23–25th 2014, Groningen, the Netherlands.

Zwinkels M, Verschuren O, Ketelaar M, Reijnders-Son F, Takken T. Can we measure cardio-respiratory fitness with the 10-meter shuttle ride test in wheelchair-using youth with cerebral palsy. Poster Presentation. European Academy for Childhood Disability. October 10–12th 2013, Newcastle, United Kingdom.

National scientific conferences and presentations

Balemans A, Bolster E, **Zwinkels M**. Fitheid – van klinische inspanningstest naar training. Masterclass de long centraal, Radboud UMC Health Academy en de Vereniging docenten kinderneurorevalidatie, 24 november 2017, Nijmegen.

Zwinkels M, Lankhorst K. Sporten met een beperking. Symposium Kind en Bewegen – innovaties in de zorg voor kinderen. Hogeschool Utrecht, Kinderbewegingscentrum WKZ, kenniscentrum revalidatiegeneeskunde Utrecht, 3 november 2017, Utrecht.

Zwinkels M, Takken T, Ruyten T, Visser-Meily JMA, Verschuren O. Fitness trends in kinderen met cerebrale parese. Dag van het Sportonderzoek, 10 november 2016, Groningen.

Zwinkels M. Fitheid bij kinderen met een chronische ziekte – Are we doing better over a decade? What else? Symposium Vondelgames, READE, 3 juni 2016, Amsterdam.

Zwinkels M. Verschuren O, Lankhorst K, Van der Ende-Kastelijn K, De Groot J, Backx F, Visser-Meily A, Takken T. Research design of the Sport-2-Stay-Fit study: after school sport participation for children with a chronic disease or condition. Poster presentatie. Conferentie Johanna kinderfonds (JKF), 22 mei 2015, Arnhem.

Zwinkels M, Verschuren O, Ruyten T, Borcherts E, van Dinther K, Elfrink M, Steenweg G, Lankhorst K, van der Ende-Kastelijn, de Groot JF, Backx FJG, Visser-Meily JMA, Takken T. High-intensity interval training (HIT) bij kinderen met een chronische ziekte of fysieke beperking: voorlopige resultaten. Dag van het Sportonderzoek, 29 oktober 2015, Zwolle.

Zwinkels M, Verschuren O, Lankhorst K, Van der Ende-Kastelijn K, De Groot J, Backx F, Visser-Meily A, Takken T. Sport-2-Stay-Fit (S2SF): naschools sporten voor kinderen met een chronische ziekte of aandoening. Dag van het Sportonderzoek, 30 oktober 2014, Nijmegen.

Presentations at faculties and for medical professionals

Zwinkels M. High-intensity interval training (HIT) – inzetten als therapie: hoe en wanneer? Workshop. Nederlandse Vereniging voor Kinderfysiotherapie (NVFK) congres, 23 september 2017, Zwolle.

Zwinkels M. Sport voor kinderen met een chronische ziekte of aandoening. Gastcollege en workshop. Hogeschool van Arnhem en Nijmegen (HAN), 16 mei 2017, Nijmegen en 20 mei 2015, Arnhem.

Zwinkels M. High-Intensity interval training (HIT) – resultaten Sport-2-Stay-Fit studie. Landelijke Fitkids dag, 10 mei 2017, Zeist.

Zwinkels M. Gezondheid bevorderende fysieke activiteit bij kinderen met cerebrale parese. Researchbattle. Hogeschool van Amsterdam (HvA), 19 april 2017, 9 december 2016, 18 mei 2016, Amsterdam.

Zwinkels M. High-intensity Interval Training (HIT). Workshop. Symposium Fit for the Future! Hogeschool Utrecht, 11 juni 2016, Utrecht.

Zwinkels M, van den Brink S. Naschoolse sport op het speciaal onderwijs. Workshop. Symposium MEEDOEN met sport, 24 november 2016, Berg en Dal.

Zwinkels M. Van FBW- naar PhD-student. Oriëntatie op de arbeidsmarkt, Faculteit Bewegingswetenschappen, Vrije Universiteit, 20 november 2014, Amsterdam.

Bloemen M, **Zwinkels M**, de Groot J, Verschuren O, Takken T. Fitheidstesten bij rolstoel rijdende kinderen. Workshop. Nederlandse Vereniging voor Kinderfysiotherapie (NVFK) congres, 7 juni 2013, Zwolle.

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