

The impact of (adapted) organized sports participation on health in youth with a chronic disease or physical disability

Health in adapted youth sports



UMC Utrecht Brain Center

Kristel Lankhorst

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Kristel Lankhorst
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The impact of (adapted) organized sports participation on health in youth with a chronic disease or physical disability

Health in adapted youth sports

De impact van (aangepast) georganiseerd sporten op de gezondheid van jongeren met een chronische ziekte of een fysieke beperking

Aangepast sporten en gezondheid bij jongeren

(met een samenvatting in het Nederlands)

Proefschrift

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Kristel Maria Lankhorst

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Promotor:

Prof. dr. F.J.G. Backx

Copromotoren:

Dr. T. Takken

Dr. J.F. de Groot

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Gewoon lekker sporten!

Het lijkt zo vanzelfsprekend: gewoon lekker sporten. Helaas is het in Nederland (maar ook in de rest van de wereld) nog lang niet vanzelfsprekend dat kinderen met een lichamelijke beperking wekelijks sporten en/of lid zijn van een sportvereniging. En dat terwijl sport zo veel voor je kan betekenen. Dat heb ik nadat ik op mijn 8-ste een dwarslaesie kreeg, aan den lijve ondervonden. Sport heeft mij ongelooflijk veel gebracht. Het heeft mij gemaakt tot wie ik nu ben. Ik heb geleerd wat ik wel en wat ik niet kan. Ik ben er dan ook van overtuigd dat wekelijks sporten er voor zorgt dat je ook de kracht van zelfvertrouwen leert kennen. Zelfvertrouwen dat je zo ongelooflijk hard nodig hebt om verder te komen in het leven. Ik vind het dan ook fantastisch dat Kristel haar proefschrift heeft gewijd aan de sportparticipatie onder kinderen/jongeren met een chronische ziekte of fysieke beperking.

Met mijn foundation zijn wij actief op bijna 30 sportverenigingen in Nederland. Op al die verenigingen geven wij kinderen/jongeren met een fysieke beperking de kans om 'gewoon' te sporten; net als ieder ander kind. Wij zien dat de impact die dit heeft enorm is. Regelmatig komen de kinderen met knikkende knieën bij een clinic kijken. Ze vinden het spannend en weten niet of ze het wel kunnen. Als ik dan een aantal maanden later nog een keer op een training kom kijken, en ik zie dan ondeugende, stoere kids, dan weet ik dat dat dus de kracht van sport is!

En ook ouders spelen een enorm belangrijke rol, zeker in het begin. De keuze om wel of niet te starten met een sport, wordt veelal genomen door de ouders. Vaak wordt het door de ouders in eerste instantie gezien als "weer een wekelijkse verplichting". Tot het moment dat ze zien wat sport (en samen spelen met andere kids, en onderdeel zijn van een clubje) met hun kind doet. Die glimlach op het gezicht van hun kind, het plezier wat de kinderen samen beleven, maakt dat de meeste ouders toch overstag gaan. Gelukkig maar!

Ik heb eens gelezen dat bijna 70% van de kinderen/jongeren met een lichamelijke beperking aangeeft eenzaam te zijn. Ook in het oplossen van het gevoel van eenzaamheid, wat afschuwelijk is, speelt wekelijks sporten een hele belangrijke rol. Los dus van alle fysieke voordelen die sport biedt, draagt het ook bij aan de mentale gezondheid en aan de persoonlijke ontwikkeling van de kinderen op sociaal gebied.

Ik ben ervan overtuigd dat dit proefschrift ook weer bijdraagt aan de ontwikkeling van sportparticipatie voor kinderen en jongeren met een chronische ziekte of fysieke beperking. Ik wil Kristel bedanken voor al haar inzet en haar werk. En laten we samen werken aan een toekomst vol sportende kinderen.



Esther Vergeer



1

General introduction

Kristel Lankhorst

Chronic disease and physical disability

One in four Dutch youngsters is growing up with a chronic disease or disability

In 2018, over 1.3 million children or adolescents in the Netherlands have a chronic condition. This means that 1 in 4 youngsters (26.2%) are

growing up with a chronic disease or disability¹. The group, children and adolescents with a chronic condition, represents a diverse array of medical diagnoses with varying prevalence. A chronic condition refers to diseases and disabilities that can be divided into somatic and psychological (mental) conditions. Asthma is the most common chronic disease -4.6% of children and adolescents in the Netherlands are affected, followed by anxiety and mood disorders (4.1%) or attention deficit hyperactivity disorder (ADHD) (3.6%)¹. Of all somatic disorders in youngsters, pulmonary diseases (mainly asthma) have the largest prevalence of 13.7%, followed by neurological diseases (8.9%), skeletal diseases (7.2%), heart diseases (2.1%), rheumatism (1.4%), oncological diseases (0.9%) and immunological diseases (0.5%)¹.

A chronic disorder is considerate to be a chronic condition in childhood if;

- 1) It occurs in children aged 0 up to 18 years,
- 2) The diagnosis is based on medical scientific knowledge, using reproducible and valid methods or instruments, according to professional standards,
- 3) It is not (yet) curable, and
- 4) It has been present longer than three months, or it has occurred three times or more during the past year and will probably reoccur ²⁹.

In this thesis, our research will focus on children and adolescents with chronic somatic conditions in childhood, i.e. chronic disease or physical disability (CDPD) (Table 1). For the classification of the different medical diagnoses, we used the categories of the American College of Sports Medicine (ACSM): cardiovascular disease (e.g. congenital heart disease), pulmonary disease (e.g. asthma), metabolic disease (e.g. diabetes), musculoskeletal or orthopedic disability, neuromuscular disorder (e.g. cerebral palsy, spina bifida), immunological or hematological disease, cancer and epilepsy².

Table 1: Focus on children and adolescents with chronic diseases or physical disabilities in the Health in Adapted Youth Sports study, classified according to categories of the American College of Sports Medicine (ACSM)².

Health in Adapted Youth Sports study	
Chronic disease	Physical disability
<ul style="list-style-type: none">• Pulmonary disease (asthma)• Metabolic disease (diabetes)• Immunological / hematological (rheumatism)• Epilepsy• Cancer	<ul style="list-style-type: none">• Neuromuscular disorder (cerebral palsy, spina bifida)• Musculoskeletal / orthopedic disability (amputation)• Cardiovascular (congenital heart diseases)

Model of Shephard & Bouchard

Various factors influence self-reliance in society. Shephard & Bouchard³ have described a model in which relationships between physical activity (PA), health-related fitness and health are being presumed (Figure 1) in adults, while at the same time genetics and environment play an important role in this interaction.

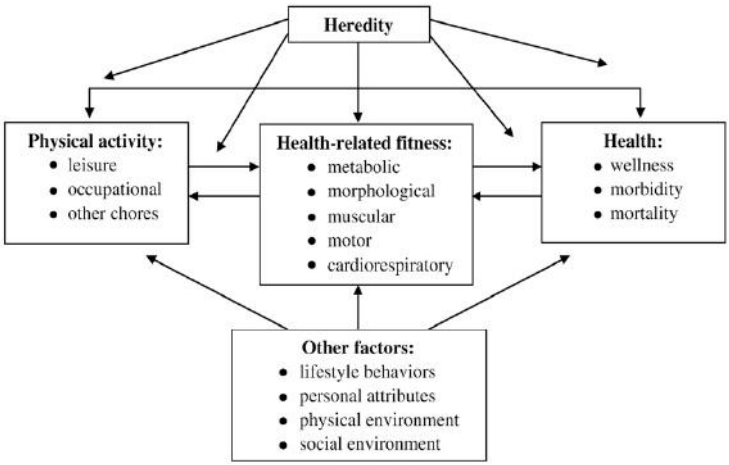


Figure 1: Principal components of fitness: relationship to physical activity and lifestyle. Model of Shephard & Bouchard, 1994.

In the model, health-related fitness is defined as a state characterized by 1) an ability to perform and sustain daily activities and 2) demonstration of traits or capacities that are associated with a low risk of premature development of diseases and conditions related to movement⁴. Health-related fitness refers to those components of fitness that are affected by PA and relate to health status. PA is any bodily movement produced by the contraction of skeletal muscles that results in energy expenditure⁵. There are several subdomains of physical activity where children and adolescents are physically active, such as: physical education lessons at school, leisure time sport and play, active transport (walking, cycling to their school), and sports participation. As a counterpart to beneficial factors like PA for health-related fitness, time spent being sedentary (inactive) contributes also to health-related fitness as a component with negative influence.

Sedentary behavior is any waking behavior characterized by an energy expenditure ≤ 1.5 metabolic equivalents (METs), while in a sitting, reclining or lying posture.

In general, this means that, when a person is sitting or lying down, they are engaging in sedentary behavior⁶.

Physical (in)activity and sports participation

For typically-developing youngsters, it is already difficult to meet recommended levels of PA; it is even more difficult for youngsters with CDPD. Such youngsters participate even less in competitive and recreational sports than their healthy peers^{7,8}. Only 26% of youngsters with CDPD participate in sports once a week, compared to 71% of their healthy peers. Several studies have reported significant barriers for youngsters with CDPD to participate in sports. These include both personal and environmental factors^{7,9}. The latter include the accessibility of the physical environment, and also attitudes from the social environment, e.g. parents, teachers, health-care professionals and policy makers. Such attitudes include thinking that sports might be too difficult, lack of knowledge of the importance of PA for youngsters with CDPD, practical considerations such as the difficulty of transportation to an adapted sports facility, or fear of sport-related injuries and/or illnesses¹⁰.

There are many opportunities for creating a healthy active lifestyle. Structured and weekly participation in (organized) sports is one of these that has the potential to maintain and/or optimize health-related fitness, physical activity and psychosocial wellbeing in youngsters with CDPD. To overcome some of these social barriers, research is needed

showing that there are positive associations between sports participation, health-related fitness, and psychosocial health, and into how far the fear of sport-related injuries and illnesses is justified for youngsters with CDPD. With this knowledge, attitudes towards the importance of sports participation for youngsters with CDPD may change. To test this hypothesis, two studies, the Health in Adapted Youth Sports (HAYS) study and Sport-2-Stay-Fit (S2SF), were initiated by the Shared Utrecht Pediatric Exercise Research Lab (SUPER-Lab) research group in Utrecht.

The S2SF study investigated whether a school-based sports program could maintain or increase the results of a High Intensity Training (HIT) program in children and adolescents with CDPD who go to schools for special education¹¹, specifically Cluster III schools, which are specifically for people with physical, mental or multiple disabilities, and people with a chronic disorder.

The aim of the HAYS study was to determine the associations between sports participation, as a specific subdomain of PA, and health outcomes in youngsters with CDPD, by comparing sports participants in organized sports to their non-sporting peers and those who participate in sports once per week. Children and adolescents of all levels of education can participate in this study, including primary and secondary education, and regular and special education.

Figure 2 is a schematic representation of the relations and risks that were investigated in the HAYS study. This is based on the model of Shephard & Bouchard for adults, as introduced and displayed in Figure 1. We adapted this model specifically for children and adolescents. We will examine the associations between participation in sports and health-related fitness, physical activity, psychosocial health and physical health (the incidence and risks of injuries and illnesses) in youngsters with CDPD. Each domain will be introduced in the following paragraphs. The impact of sports participation on 'other factors' and the personal and social environment will not be investigated in this thesis.

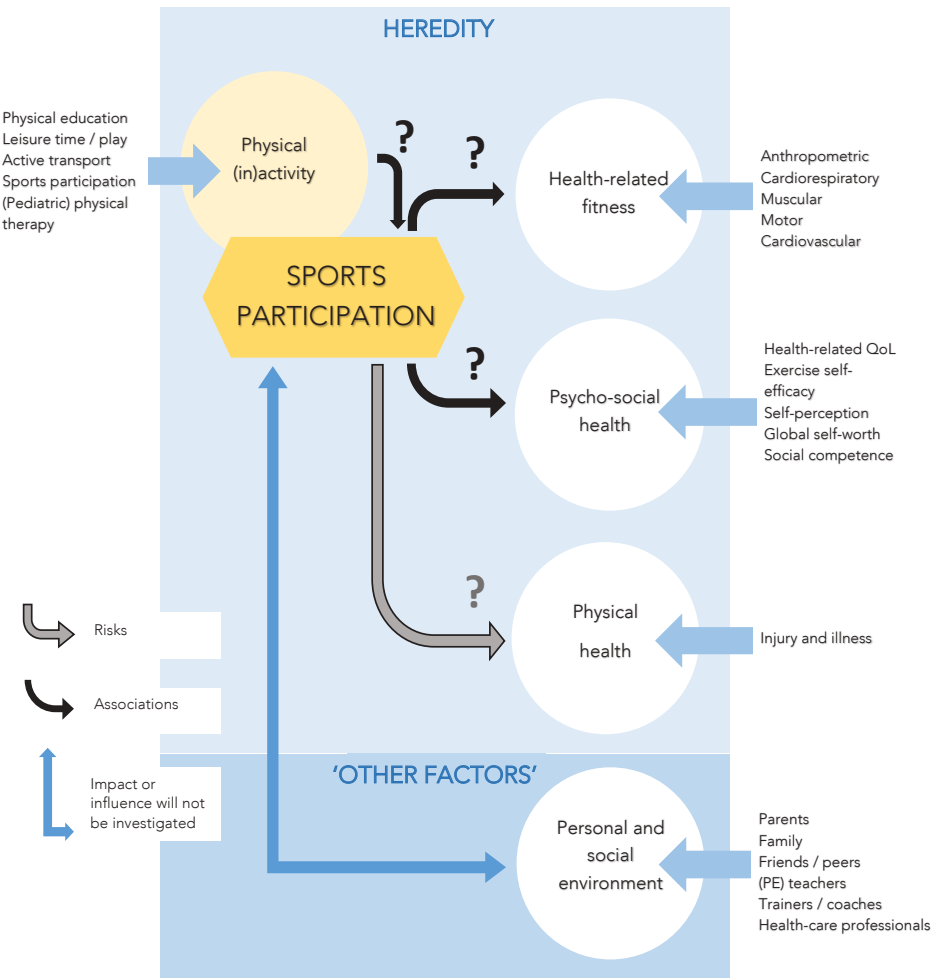


Figure 2: Schematic representation of the relationships and risks being investigated with the HAYS study. QoL: quality of life; PE: physical education.

Sports participation and health-related fitness

Youngsters with CDPD have lower fitness levels¹², lower levels of PA^{13,14} and a higher prevalence of adiposity¹⁵ than their healthy peers. The detrimental health effects of physical inactivity in healthy youngsters are well established¹⁵⁻¹⁷. In typically-developing youngsters, insufficient PA levels are highly associated with low cardiorespiratory fitness,

higher levels of obesity and increased cardiovascular risk^{18,19}. While health benefits related to sports and PA are reported in healthy youngsters, there is only limited evidence on the extent to which sports participation may affect PA and health-related fitness in youngsters with CDPD. At the same time, training studies have shown that youngsters with CDPD indeed can improve their PA level and health-related fitness through supervised intervention programs in rehabilitation settings^{12,20}. However, the positive results achieved are often not maintained in the longer term after these programs end²¹⁻²³. Regular participation in (organized) sports, as a subdomain of PA, seems therefore an important goal for youngsters with CDPD, to maintain or even improve on the results achieved with a training program or supervised intervention program, to meet PA guidelines⁷ and to develop a healthy active lifestyle.

Sports participation and psychosocial health

Supervised intervention programs in a rehabilitation environment pursue goals that are often focused on physical functioning, i.e. mobility, strength and fitness. Sports participation can contribute to one's health and wellbeing at both physical functioning and psychosocial levels. Sports participation can contribute to the development of athletic skills, and competing with others can be a positive challenge. In addition, due to its social nature, sports participation provides opportunities for social interaction and companionship and may therefore have greater benefits for psychosocial health than other domains of PA (leisure or occupational)²⁴⁻²⁶. Involvement in sports can encourage focus on children's abilities (personal attributes) rather than on their inabilities²⁴, which gives autonomy and control in their lives. Youngsters with CDPD engage less often in PA and sport compared to their healthy peers. These children and adolescents may therefore experience low levels of self-worth and quality of life due to their physical limitations and body image concerns. The current thesis will therefore focus on the potential beneficial effects of sports participation, as a specific subdomain of PA, on psychosocial health.

Sports participation and physical health

For youngsters with CDPD, it is already a challenge to reach adequate levels of PA, as mentioned before, due to existing social and environmental barriers to sports and exercise participation. It is even harder to pursue an active and healthy lifestyle through participation in sports when sports participation is associated with (fear of) sport-related

injuries or illnesses. A long-lasting injury or illness can limit participation in sports or lead to dropping out of sports or fear of return to sports^{27,28}. There is a reasonable fear among parents of children with CDPD that sports participation may lead to sport-related injuries. As a consequence of such injury, their child might experience additional limitations in their daily lives, e.g. no longer being able to perform their daily activities independently or needing more help with these from their parents or caregivers. These negative experiences and beliefs about sports participation can further limit the support given to allow their child to be active in sports. The current thesis will therefore focus on the incidence, type, severity and risks of (sports-related) injuries and illnesses among youngsters with CDPD, to see whether the fear of sport-related injuries and illnesses is justified.

Aims and outline of this thesis

The current thesis focuses on the impact of sports participation on health and fitness in youngsters with CDPD. **Chapter 2** describes the design of the Health in Adapted Youth Sports study, a cross-sectional study comparing youngsters with CDPD who participate in organized sports at least twice weekly with their non-sporting peers or those who participate in sports once a week. **Chapter 3** introduces the criterion validity of the Activ8 for quantifying and promoting physical activity in youngsters with typical development and youngsters who are ambulatory but have motor disability. **Chapter 4, 5, 6 and 7** addresses the results of the HAYS study. **Chapter 4** shows the results of sports participation on health-related fitness and physical activity in youngsters with CDPD; **Chapter 5**, the associations of sports participation with self-perception, exercise self-efficacy, and quality of life. **Chapter 6** summarizes the association of cardiorespiratory fitness, adiposity and sports participation with arterial stiffness. **Chapter 7** shows the risks of (sports-related) injuries and illnesses among youngsters with CDPD. **Chapter 8** presents a systematic review that summarizes the evidence of instruments measuring physical activity in persons who use a wheelchair. Finally, **Chapter 9** comprises the general discussion of this thesis including: the main findings, theoretical considerations, interpretation of results, methodological considerations, practical implementation, future perspectives, recommendations and conclusions.

REFERENCES

1. Hal van L, Tierolf B, Rooijen van M, Hoff van der M. Een actueel perspectief op kinderen en jongeren met een chronische aandoening in Nederland: Omvang, samenstelling en participatie. Verwey-Jonker Instituut, Utrecht April 2019; ISBN 978-90-5830-936-5.
2. Durstine LJ, Moore GE, Painter PL, Roberts SO. ACSM's exercise management for persons with chronic diseases and disabilities. 3rd ed. Champaign, IL: Human Kinetics; 2009.
3. Shephard RJ, Bouchard C. Principal components of fitness: Relationship to physical activity and lifestyle. *Can J Appl Physiol* 1994;19(2):200-214.
4. Winnick, JP & Short, FX, ed. The Brockport Physical Fitness Test Manual. Champaign, IL: Human Kinetics; 1999.
5. Caspersen CJ, Powell KE, Christenson GM. Physical activity, exercise, and physical fitness: Definitions and distinctions for health-related research. *Public Health Rep* 1985;100(2):126-131.
6. Bussmann JB, van den Berg-Emons RJ. To total amount of activity..... and beyond: Perspectives on measuring physical behavior. *Front Psychol* 2013;4:463. doi: 10.3389/fpsyg.2013.00463.
7. Burghard M, de Jong NB, Vlieger S, Takken T. 2017 Dutch Report Card+: Results from the first physical activity report card plus for Dutch youth with a chronic disease or disability. *Front Pediatr* 2018;6:122. doi: 10.3389/fped.2018.00122.
8. Burghard M, Knitel K, van Oost I, Tremblay MS, Takken T, Dutch Physical Activity Report Card Study Group. Is our youth cycling to health? Results from the Netherlands' 2016 report card on physical activity for children and youth. *J Phys Act Health* 2016;13(11 Suppl 2):S218-S224. doi: 10.1123/jpah.2016-0299.
9. Jaarsma EA, Dijkstra PU, de Blecourt AC, Geertzen JH, Dekker R. Barriers and facilitators of sports in children with physical disabilities: A mixed-method study. *Disabil Rehabil* 2015;37(18):1617-23; quiz 1624-5. doi: 10.3109/09638288.2014.972587.
10. Verschuren O, Wiart L, Hermans D, Ketelaar M. Identification of facilitators and barriers to physical activity in children and adolescents with cerebral palsy. *J Pediatr* 2012;161(3):488-494. doi: 10.1016/j.jpeds.2012.02.042.
11. Zwinkels M. From exercise training to school-based sports, the effects on fitness and health in youth with physical disabilities [Dissertation]; Utrecht University, 2018.
12. van Brussel M, van der Net J, Hulzebos E, Helders PJ, Takken T. The Utrecht approach to exercise in chronic childhood conditions: The decade in review. *Pediatr Phys Ther* 2011;23(1):2-14. doi: 10.1097/PEP.0b013e318208cb22.

13. Carlon SL, Taylor NF, Dodd KJ, Shields N. Differences in habitual physical activity levels of young people with cerebral palsy and their typically developing peers: A systematic review. *Disabil Rehabil* 2013;35(8):647-655. doi: 10.3109/09638288.2012.715721.
14. Neter JE, Schokker DF, de Jong E, Renders CM, Seidell JC, Visscher TL. The prevalence of overweight and obesity and its determinants in children with and without disabilities. *J Pediatr* 2011;158(5):735-739. doi: 10.1016/j.jpeds.2010.10.039.
15. Azevedo MR, Araujo CL, Reichert FF, Siqueira FV, da Silva MC, Hallal PC. Gender differences in leisure-time physical activity. *Int J Public Health* 2007;52(1):8-15.
16. Jimenez-Pavon D, Kelly J, Reilly JJ. Associations between objectively measured habitual physical activity and adiposity in children and adolescents: Systematic review. *Int J Pediatr Obes* 2010;5(1):3-18. Accessed 7/7/2017 9:28:46 AM. doi: 10.3109/17477160903067601.
17. LaMonte MJ, Blair SN. Physical activity, cardiorespiratory fitness, and adiposity: Contributions to disease risk. *Curr Opin Clin Nutr Metab Care* 2006;9(5):540-546. doi: 10.1097/01.mco.0000241662.92642.08.
18. Janssen I, Leblanc AG. Systematic review of the health benefits of physical activity and fitness in school-aged children and youth. *Int J Behav Nutr Phys Act* 2010;7:40-5868-7-40. doi: 10.1186/1479-5868-7-40.
19. Riddoch CJ, Mattocks C, Deere K, Saunders J, Kirkby J, Leary SD, Blair SN, Ness AR. Objective measurement of levels and patterns of physical activity. *Arch Dis Child* 2007;92(11):963-969. doi: adc.2006.112136.
20. Edouard P, Gautheron V, D'Anjou MC, Pupier L, Devillard X. Training programs for children: Literature review. *Ann Readapt Med Phys* 2007;50(6):510-9, 499-509. doi: S0168-6054(07)00129-8.
21. de Groot JF, Takken T, van Brussel M, Gooskens R, Schoenmakers M, Versteeg C, Vanhees L, Helders P. Randomized controlled study of home-based treadmill training for ambulatory children with spina bifida. *Neurorehabil Neural Repair* 2011;25(7):597-606. doi: 10.1177/1545968311400094.
22. Zwinkels M, Verschuren O, de Groot JF, Backx FJG, Wittink H, Visser-Meiley A, Takken T. Effects of high-intensity interval training on fitness and health in youth with physical disabilities. *Pediatr Phys Ther* 2019;31(1):84-93. doi: 10.1097/PEP.0000000000000560.
23. Zwinkels M, Verschuren O, Balemans A, Lankhorst K, Te Velde S, van Gaalen L, de Groot J, Visser-Meiley A, 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. *Front Pediatr* 2018;6:75. doi: 10.3389/fped.2018.00075.

24. Eime RM, Young JA, Harvey JT, Charity MJ, Payne WR. A systematic review of the psychological and social benefits of participation in sport for children and adolescents: Informing development of a conceptual model of health through sport. *Int J Behav Nutr Phys Act* 2013;10:98-5868-10-98. doi: 10.1186/1479-5868-10-98.
25. Eime RM, Harvey JT, Brown WJ, Payne WR. Does sports club participation contribute to health-related quality of life? *Med Sci Sports Exerc* 2010;42(5):1022-1028. doi: 10.1249/MSS.0b013e3181c3adaa.
26. Street G, James R, Cutt H. The relationship between organised physical recreation and mental health. *Health Promot J Austr* 2007;18(3):236-239.
27. Siesmaa EJ, Blitvich JD, Finch CF. A systematic review of the factors which are most influential in children's decisions to drop out of organised sport. In: Farelli AD, ed. *Sport participation: Health benefits, injuries and psychological effects*. Hauppauge, NY: Nova Science Publishers, Inc.; 2011:1-45.
28. Siesmaa EJ, Blitvich JD, Telford A, Finch CF. Factors that are most influential in children's continued and discontinued participation in organized sport: The role of injury and injury risk perceptions. In: Farelli AD, ed. *Sport participation: Health benefits, injuries and psychological effects*. Nova Science Publishers, Inc.; 2011:47-84.
29. Mookink L, Lee van der J, Grootenhuys M, Offringa M, Praag van B, Heymans H. Omvang en gevolgen van chronische aandoeningen bij kinderen. *Tijdschrift voor Kindergeneeskunde* Feb 2007;75:154-158.



2

Health in Adapted Youth Sports study (HAYS): Health effects of sports participation in children and adolescents with a chronic disease or physical disability

Kristel Lankhorst

Karin van der Ende-Kastelijn

Janke de Groot

Maremka Zwinkels

Olaf Verschuren

Frank Backx

Anne Visser-Meily

Tim Takken

ABSTRACT

Introduction. In typically developing children, participation in sports has been proven to be positively correlated to both physical and psychosocial health outcomes. In children and adolescents with a physical disability or chronic disease participation in both recreational and competitive sports is often reduced, while for this population an active lifestyle may be even more important in reaching optimal levels of physical and psychosocial health. Therefore, the aim of the Health in Adapted Youth Sports (HAYS) Study is to determine both negative and positive effects of sports on children and adolescents with a chronic disease or disability.

Methods. In this cross-sectional study differences will be compared in regards to physical and psychosocial health, cognitive functioning, school performance, daily physical activity and injuries between children and adolescents with a chronic disease or physical disability who participate in sports and those who do not. Children and adolescents, both ambulatory and wheelchair dependent, in the age of 10-19 years with a physical disability or chronic disease will be included. "Sports" is defined as participation in an organized sport at least two times a week for a duration of three months or more prior to the assessment. Parametric and non-parametric statistics will be used to determine the differences between the two groups.

Discussion. This study provides insight in the effects of sports participation in relation to health, psychosocial functioning, physical activity and school performance in children and adolescents (10-19 years) with a chronic disease or physical disability. Results will guide healthcare professionals working with these children to better guide this population in reaching optimal levels of health and physical activity levels.

INTRODUCTION

Children with disabilities often show reduced fitness levels and physical activity patterns and they participate less in competitive and recreational sports compared to their non-disabled peers^{1,2}. The relationship between health and physical fitness has been studied by many authors.

In the general population, low level of physical activity is highly associated with low physical fitness, an increased cardiovascular and overall mortality³⁻⁶. In typically developing children, participation in sports has been proven to be positively correlated to both physical and psychosocial health outcomes³. Also for adults with various disabilities the physical, psychological, social, and economic benefits of participation in sports and recreational activities are described previously⁸⁻¹¹. Because of the reduced fitness levels and physical activity pattern in children with a disability or chronic disease, support for an active lifestyle, including participation in sports, may be even more important in this population.

Next to the possible benefits of sports on health and fitness level, increasing evidence also shows benefits from physical activity on school performance and level of cognition could be influenced in a positive way. Children and adolescents without a medical condition are already known to perform better at school when being physically active¹²⁻¹⁵. Childhood physical fitness is also associated with higher levels of cognition and appeared to be a good predictor of school performance and level of cognition 1 year later¹⁶⁻²⁰.

Participation in sports could also have an positive influence on health related quality of life (HRQoL) and self-worth²¹. HRQoL refers to the impact of health and illness on an individual's quality of life. In relation to sports, adult athletes with cerebral palsy for example reported a positive influence of sports participation on their HRQoL^{22,23}.

When looking at competence levels, Special Olympics participants with developmental disorders showed higher levels of self-worth and perceived physical competence in comparison to nonparticipants²⁴. These findings might support the hypothesis that sports participation will enlarge the global self-worth of children and adolescents with a chronic disease and/or disability. But when motor competence is inadequate for the type of sport, a feeling of failure could predominate.

While there are many positive reasons for participating in sports or other physical activities for children with a chronic disease and/or disability, attention must be paid to the risk of acute and overuse injuries but also illness. Indeed parents and healthcare providers are wary of injuries due to participation in sports, which could further limit

their child's physical functioning. These worries are confirmed in healthy children. A recent study reported a higher absolute risk of getting injured and a high probability of sustaining the injury when adding physical education hours at school or increasing organized sports outside school²⁵⁻²⁷. At the same time though, the relative risk of injury seems higher for children with low levels of habitual physical activity²⁸. Studies also reported children with disabilities to have a higher risk of injury than children without disabilities, but these studies were limited to just one type of disability, sport, or injury²⁹. Identifying injury patterns and illness in children with disabilities is important to provide safety in sports activities and to prevent dropout in physical activities and return to an inactive lifestyle^{28,30,31}.

In summary, children with chronic disease and/or disabilities often show reduced levels of physical activity and fitness and participate less in organized sports compared to their non-disabled peers. The positive effects of sports and daily activities on the psychosocial and physical health, cognition and injury risk as depicted in Fig 1 already have been reported in a healthy population and disabled adults.

To date, however, limited evidence exists for these effects of participation in sports by children and adolescents with a physical disability or chronic disease. Therefore, the aim of the HAYS study is to determine both the positive and negative effects of sports related to health outcomes in children with disabilities and chronic childhood onset conditions. Therefore the Health in Adapted Youth Sports (HAYS) Study is designed to determine the positive and negative effects of participation in sports in this specific population.

METHODS

Design

The HAYS study will be a cross-sectional study comparing children and adolescents with a physical disability or chronic disease, age 10 to 19 years, who are actively participating in organized sports to their non-sporting peers. The subjects who participate in sports will be matched on gender, age and diagnosis to their non-sporting peers, Fig 2.

The current study is part of a larger project in which a controlled clinical trial will take place to evaluate the effectiveness of an after school sports program following a standardized interval training in children and adolescents with a chronic disease or physical disability. This study, titled The Sport-2-Stay-Fit study (S2SF; Trialregister.nl registration number: NTR4698), will use the same outcome instruments³².

Participants

Eligible for this study are all children and adolescents aged from 10 up to 19 years with a physical chronic disease or condition, including cardiovascular, pulmonary, musculoskeletal, metabolic or neuromuscular disorders. Table 1 shows the in- and exclusion criteria. Both children who are ambulatory or those propelling a wheelchair are eligible for this study. Participants have to understand the Dutch language, understand simple instructions and be able to perform a physical fitness test. Children and adolescents in electric wheelchair, having a progressive disease or participate in other research projects which might influence the current study results will be excluded. Contra-indications for performing an exercise test, based on the exercise pre-participation screening questionnaire, may lead to exclusion on the cardiopulmonary exercise test³³. Informed consent must be provided by all parents, as well as by subjects up from 12 years till 17 years. In line with Dutch law, no parental informed consent is required for subjects aged 18 years and over.

This study was approved by the Medical Ethics Committee of the University Medical Center Utrecht, the Netherlands.

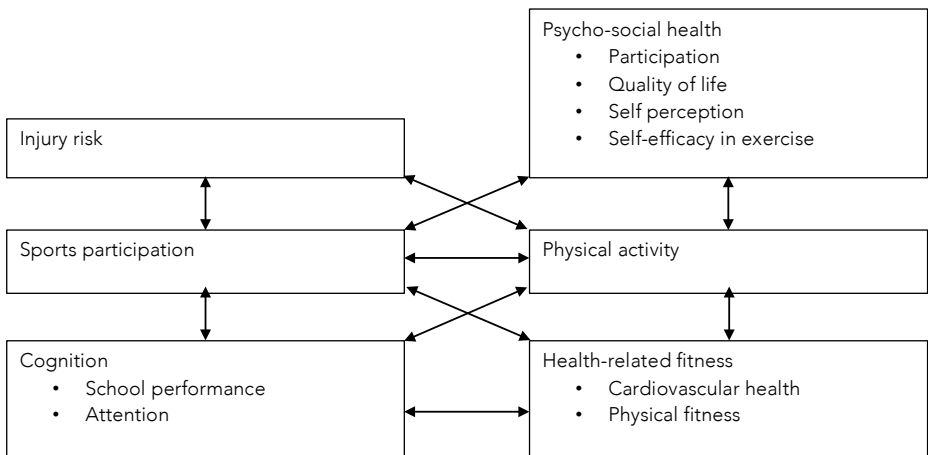


Figure 1: Overview of positive relations already established within a population of children and adolescents without a chronic disease and/or disability, and within a population of adults with and without a disability.

Table 1: Eligibility and exclusion criteria.

Eligibility	Exclusion
<ul style="list-style-type: none">• Children and adolescents with a physical disability or chronic disease: cardiovascular, pulmonary, musculoskeletal or neuromuscular disorder• Children and adolescents between the age of 10 and 19 years• Children and adolescents have to understand simple instructions• Children and adolescents are able to perform physical fitness tests	<ul style="list-style-type: none">• Children and adolescents with progressive diseases• Children and adolescents using an electric wheelchair• During the length of the study, children are not allowed to participate in other research projects which might influence the current study results• For the sporting group of the HAYS-study only: subjects who have not participated in any sports for the preceding three months.• No signed informed consent

Recruitment

The children and adolescents will be recruited in the Netherlands among different patients associations, pediatric physical therapy practices, Wilhelmina Children’s Hospital in Utrecht, De Hoogstraat Rehabilitation Center in Utrecht, Fitkids, schools for children with a disability and sports clubs. Athletes will be recruited from a broad range of participation in sports: from recreational level to high level competitive sports.

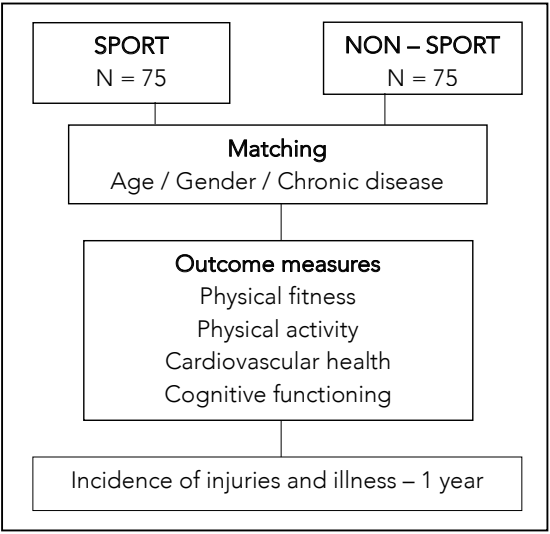


Figure 2: Overview of the design of the HAYS study.

Setting

This study is a collaboration between the exercise lab at the University of Applied Sciences Utrecht, Wilhelmina Children's Hospital and De Hoogstraat Rehabilitation Center in Utrecht, the Netherlands.

Procedures

Criteria for eligibility and exclusion of subjects are depicted in Table 1. After permission of the subject and when the subject is eligible to participate in the study, the subject is scheduled for the assessment. Thereafter a secured link to four of the questionnaires will be sent to the participants or their parents by email in order to fill out the questionnaires online 1 week before the first testing moment. The researchers will assess the subjects subsequently, once on physical fitness, cognition, psychosocial- and cardiovascular health. Physical activity will be monitored during 1 week. The incidence of injuries and illness will be monitored for the duration of 1 year, by sending a secured link to a 5-item questionnaire every 2 weeks by email. If an injury or illness is reported, a telephone conversation will follow, to get insight and to classify the sort of injury or illness. Table 2 shows the procedure for the principle researchers and the subjects in the HAYS study, from recruitment to the end of participation.

Outcome measures

Table 3 shows an overview of the outcome measures and chosen measurement instruments in this study. The participants' characteristics such as date of birth, gender, medical diagnosis, functional mobility scale (FMS) score and type and frequency of sport activity will be identified by a general questionnaire. An exercise pre-participation screening questionnaire will inquire about possible factors influencing the outcomes of the test and the participants' safety during the tests. These questionnaires will take approximately 5 minutes to be filled out by the participant and / or parent.

Table 2: Organisational and subject flow of the HAYS study.

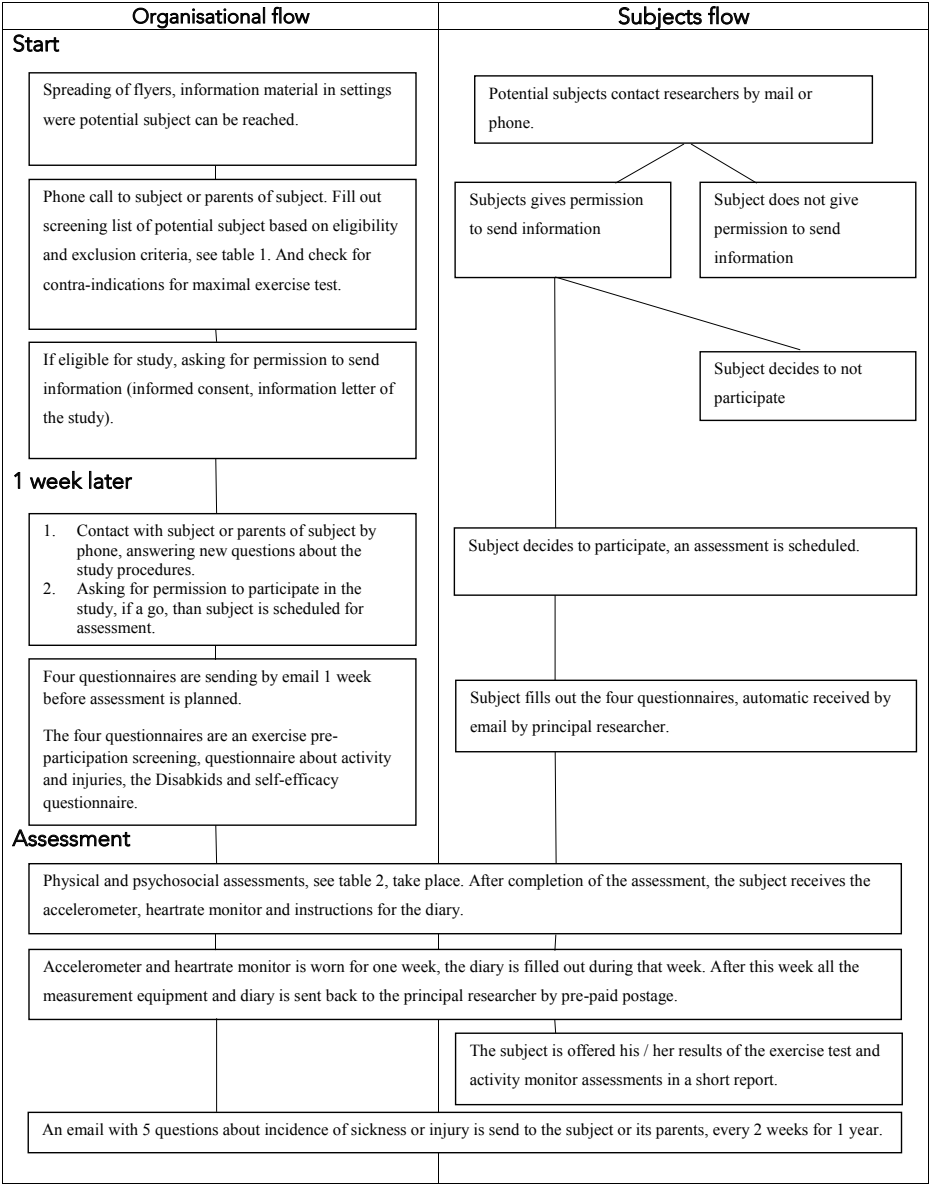


Table 3: Overview of assessments used instruments, questionnaires and time schedule.

Outcome measure	Parameter	Variable	Measurement	Time at location	Time at home
General	Demographics	DOB, Gender, Diagnosis, FMS score, activity level	General questionnaire		5 min
Cardiovascular Health	Metabolic parameters	BMI Fat Free Mass Blood pressure	Length, Weight BIA Sphygmomanometer	5 min 10 min	
Physical Fitness	Aerobic fitness	Arterial stiffness Pulse wave velocity VO _{2peak} RER	Arteriograph CPET Shuttle run/ride test	30 min	
	Anaerobic fitness	Anaerobic threshold Peak power Mean power	Bicycle test MPST	5 min	
	Muscle strength	Isometric muscle strength	Grip strength Reverse curl Seated push up	10 min	
		Explosive muscle strength	Standing broad jump or One stroke push		
	Flexibility		Modified Apley test Modified Thomas test	5 min	
	Agility		10x5 meter sprint	5 min	
Physical activity	Intensity	Heart rate	Actiheart	5 min	7 days
	Modality	Type of activity	Activ8 and Activity diary	5 min	7 days
Cognitive functioning	School performance	Educational level	Type of education	0 min	
	Attention	Focused attention Sustained attention Strategy Distractibility	Recent CITO-score Bourdon-Vos Cancellation task Capture task	15 min 5 min	
Injury and illness	Incidence injury and illness	Retrospective 3 months Longitudinal one year, every 2 weeks	Online questionnaire Online questionnaire		3 min
	Classification of injury or illness (if applicable)		Telephone conversation		5 min
Psychosocial health	Self-perception		Self-Perception Profile for Children (SPPC)	10 min	
	Quality of life		Disabkids		10 min
	Self-efficacy		Exercise Self-efficacy Scale		2 min
Total time				150 min	28 min

DOB; date of birth, FMS; functional movement score, BMI; body mass index, BIA; body impedance analysis, VO_{2peak}; peak oxygen uptake, CPET; cardio-pulmonary exercise test, RER; respiratory exchange ratio, MPST; muscle power sprint test.

General information

Cardiovascular health

Cardiovascular health is divided in several aspects in this study: fat free mass, body mass index, arterial stiffness and blood pressure.

Fat free mass will be determined with bioelectrical impedance analysis (BIA), using the Bodystat Quadscan 4000 (EuroMedix, Leuven, Belgium). BIA is a non-invasive easy test to measure lean body mass and fat by comparing conductivity and resistance in the body³⁴. Body mass index will be calculated as weight (kg)/length (m)². Weight will be measured using a wheelchair scale in case of subjects who are in the wheelchair group. In other cases a person weighing scale will be used. Height will be measured standing in case of ambulant subjects and supine in case of wheelchair bound persons. In case of spasticity, arm span width will be measured. Arm span length will be measured to the nearest centimeter from middle finger tip to finger tip. Body mass index (BMI) will then be calculated as the body mass divided by the square of arm span length that will be adjusted using arm span \times 0.95 for mid-lumbar lesions and arm span \times 0.90 for high lumbar/thoracic lesions in case of central neurological disorders³⁵.

Arterial stiffness has two independent measurement values: augmentation index: (AIX) and pulse wave velocity (PWV). Measurement will take place with the Arteriograph (Arteriograph.nl/LITRA, Amsterdam, the Netherlands). The Arteriograph measures the PWV and the AIX through the brachioradial artery using oscillometric tonometry. Each subject rests supine for at least 10 minutes before recordings are made. The measurement will take place with an inflatable cuff (similar to blood pressure measurement) at the right upper arm. Instructions to the subject are no food intake 3 hours before measurement and no talking during the measurement. Blood pressure will also be measured using the Arteriograph within the same measurement.

After the pulse wave velocity measurement, the child is allowed to eat something, before continuing with the rest of the tests.

Physical fitness

Physical fitness consists of a combination of aerobic fitness, anaerobic fitness, muscle strength, flexibility and agility. Using the FMS for 500 meters the group of subjects will be divided in two sets depending on their mode of locomotion on this distance: wheelchair users and subjects who are able to walk with or without mobility aids. For these two groups, different testing tools will be used, appropriate for the type of locomotion.

Subsequently, within the group of subjects who are ambulatory different levels of mobility can be identified in order to apply a proper testing protocol, see Table 4.

Aerobic fitness

In exercise testing peak, oxygen uptake (VO_{2peak}) is considered to be the single best indicator of the cardio-respiratory system, often referred to as aerobic fitness. A cardio-pulmonary exercise testing system, the Cortex Metamax 3X (Samcon bvba, Melle, Belgium), will be used for evaluating the respiratory gasses and VO_{2peak} .

Bhambani et al. concluded that maximal exercise testing during the main mode of ambulation elicits the highest oxygen uptake. Therefore the type of sports or daily locomotion determines whether the shuttle run, shuttle ride or a cycling test will be used³⁶.

Children who are able to walk will be tested with an (adapted) shuttle run test^{37,38}. The speed of the shuttle run test will be adjusted based on the results of the muscle power sprint test and the agility test and the level of mobility, see Table 4.

In children with a congenital cardiopulmonary disease a cycling test will be used, because of the monitoring of the heart for safety issues. The cycling test, using the Godfrey protocol³⁹, will also be applied to evaluate the aerobic fitness in children who are active on a bike in sports or daily living. Load depends on height of the child and the expected level of fitness.

A shuttle ride test will be used in children using a wheelchair. Shuttle tests are field tests in which a participant walks or runs between 2 markers. In this case they have to ride a distance of 10 meters in their own wheelchair, if applicable their sports wheelchair, between two cones. The starting speed is 2.0 km/h and the speed is increased with 0.25 km/h every minute. The children have to keep on riding, until they fail to reach the cone two times in a row, despite encouragements. This protocol has been proven valid as a maximal exercise test in youth with CP and spina bifida^{40,41}.

Regardless of the testing modality, the test will start with a resting steady state measurement for 3 minutes. To reach a total exercise time of at least 6 (children) and 8 (adolescents) minutes the protocol will be adapted based on the expected level of fitness. This might be due to the subjects high competition level for example or extreme athletic physique but also when a very low fitness is expected. In case of the cycling test, an unloaded phase of 1 minute will precede the exercise phase. Each test will be until volitional exhaustion. Usual emergency procedures are in place to ensure health and

safety in the very unlikely event of an emergency.

Anaerobic fitness

The muscle power sprint test is used to measure anaerobic fitness^{42,43}. Subjects have to complete six 15-meter runs at a maximum pace. The MPST is an intermittent sprint test, in which the child stops and starts at standardized intervals. For children who self-propel manual a wheelchair, the MPST has also been proven a reproducible test for measuring anaerobic fitness and agility in children and adolescents with CP and SB^{44,45}.

Power output will be calculated for each of the six sprints: $\text{power} = \text{weight} \times (\text{distance})^2 / (\text{time})^3$. Peak power will be defined as the highest calculated power, while mean power was defined as average power over the six sprints.

Table 4: Level of mobility for identifying testing protocol.

Category	Level of mobility
I	Subjects with a <u>very low level of mobility</u> (able to walk < 400 meter in 6 minutes)
II	Subjects with a <u>low level of mobility</u> (able to walk > 400 meter in 6 minutes)
III	Subjects with <u>average level of mobility</u> : walking indoors and outdoors and climbing stairs without limitations and able to perform gross motor skills including running and jumping.
IV	Subjects with <u>an excellent level of mobility</u> , who are used to run at a speed of 8 km/h during their practicing their sports or during competition.

Strength

To test the strength of the subjects, tests from the Brockport fitness test are chosen⁴⁶. The handgrip strength will be tested through the use of a hand held hydraulic dynamometer (HHD) as described by Beenakker et al.⁴⁷ The subjects dominant hand will be tested.

To measure the functionality of hand, wrist, and arm the reverse curl is used⁴⁸. The participant attempts to pick up a (0.5-kg) dumb-bell with the preferred arm while seated in a chair or wheelchair.

The seated push up test is designed to measure upper-body strength and endurance⁴⁸. Participants attempt to perform a seated push-up and hold it for up to 20s.

The standing Broad jump (only for the ambulatory group) will be used to evaluate the

explosive strength of the lower limbs by measuring the distance jumped with two legs together from standing position⁴⁹. In wheelchair dependent participants, the one stroke push will evaluate the explosive strength of the upper limbs by measuring the distance one can cover in a wheelchair by one push⁴⁴.

Flexibility

To measure upper-body flexibility, the modified Apley test will be applied. The participant attempts to reach back and touch with one hand the superior medial angle of the opposite scapula. The modified Thomas test is designed to assess the length of the participant's hip flexor muscles (M. Iliopsoas and M. Rectus Femoris)⁴⁸.

Agility

The 10x5 Meter Sprint Test will be used to measure agility⁴⁴. During this test, the child has to sprint (for ambulatory and wheelchair users as well) as fast as possible, 10 times, in between 2 lines that are 5 meter apart. There is no resting period, so the child/adolescent has to turn as fast as possible during this test. Time will be recorded using a stopwatch.

Physical activity

Accelerometry in combination with heart rate monitoring will be used to measure the type, duration, frequency and intensity of physical activity in daily life. The Activ8 (2M Engineering Ltd. Valkenswaard) will measure the modality of physical activity. The Activ8 system is new and has been recently be validated in cooperation with the Erasmus Medial Centre's rehabilitation department in Rotterdam (Lankhorst, K. et al. work in progress). Participants who are able to walk wear one small sensor on the dominant leg. Participants in a wheelchair have to wear 2 small sensors (chest, one wrist). Participants will wear these monitors during the tests and during 7 days in their own environment. Wearing the activity monitor will not alter the activity pattern of the participants; all activities in daily life and in sport can be performed. The Actiheart (Camntech), will be used to detect the intensity of physical activity⁵⁰. The Actiheart is a small device detecting the heart rate frequency via two electrode-stickers on the chest and has been used previously in children with Spina Bifida⁵¹. During the one week of wearing the activity monitor, the participant will also fill out an activity diary in order to interpret missing data of the activity monitor. The completion of the activity diary will only take approximately 5 minutes per day.

Injuries and illness

The incidence and type of injury and illness of the participant at T0 and 3 months in retrospective will be filled out via a digital questionnaire and takes 5 minutes to complete. The injury density (ID) will be calculated per 1000 hours of scheduled physical activity⁵². Besides the retrospective registration, the injuries and illnesses will also be monitored longitudinal for one year, every 2 weeks. The parent and / or participant receives a short digital questionnaire of 5 simple questions, send by email and takes approximately 1 minute to complete. If a question will be answered positively, there will be a telephone conversation with the child, young adult or parent, to detect the cause and severity to register the diagnosis of injury or illness. The researcher will register the injury by using a classification list. Only if the diagnosis remains unclear after the telephone conversation, a physical examination of a sports physician or a physical therapist will take place.

School performance

The type of education of the participant will be noted. If available, outcomes of a national educational achievement test (CITO) will be used for quantifying school performance, in both 'elementary school' and 'secondary school'.

Cognition

To evaluate cognitive functioning, attention will be measured. To overcome locomotive influences attention tests will be carried out on a tablet (Asus Eee Slate Tablet, with a 12.1 inch display and clock speed of 1.33 GHz).

Three types of attention will be measured: sustained attention, search strategy and distractibility. Sustained attention will be measured using an adapted digitalized version of the Bourdon-Vos task⁵³. This task is a time-limited test. Children had to continue until the whole test was finished, or stop after 10 minutes. For distractibility an object cancellation task will be used and takes 5 minutes to complete. The search strategy of the participant during this cancellation test for distractibility is computed afterwards via software of this tablet test. Another test for measuring distractibility is the capture task. This test was adapted from Van Der Stigchel & Nijboer⁵⁴. In each trial the participant is asked to focus on a central fixation cross. When the cross disappears, the target, represented as an apple, appears 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 health

To evaluate self-perception in this study, the Dutch translation of the self-perception profile scale (SPPS) for children and for adolescents will be used⁵⁵. This questionnaire has been validated to measure self-perception. The questionnaire will take approximately 10 minutes to complete.

Quality of life

To evaluate the quality of life satisfaction, the Dutch version of the Disabkids will be used. This questionnaire measures the quality of life and the independence of children with chronic health conditions. It takes approximately 10 minutes to fill out.

Self-efficacy

To assess self-efficacy specific for exercise and physical activity a Dutch questionnaire is filled out digitally at home by the child and will take approximately 2 minutes⁵⁶.

Sample size

The sample size of the HAYS-study is based on a study of Verschuren & Takken⁵⁷. In this study children and adolescents with cerebral palsy had an average peak oxygen uptake of 42 ± 8.2 ml/kg/min. To prove a difference of 10% between sporting or non-sporting subjects, with an alpha of 0.05 and beta of 0.20 (power of 0.80) a sample size of 66 subjects per group is required. When taking a failure rate of 10% into account, 146 subjects should be included in total.

Statistical analysis

Data will be checked for normal distribution before applying the right type of parametric or non-parametric tests. First, descriptive statistics will be used to describe the two samples. Independent sample T-tests or appropriate non-parametric tests will be used to determine the differences between the sport and non-sport group.

DISCUSSION

The current paper describes the rationale, design and methods of a cross-sectional study concerning participation in sports focused on children and adolescents with a physical disability or chronic disease.

To our knowledge this is the first study to evaluate this effect of sport participation on physical, social and mental health within this population. The aim is to determine both the positive and negative effects of sports related to health outcomes in children with disabilities and chronic childhood onset conditions. We hypothesize that the group participating in sports regularly will show higher levels of physical activity associated with better health outcomes, a relative lower incidence density, better cognitive functioning and school performance.

New insights gained from this study could stimulate the improvement of facilities for adapted sports by the government. Also children with a chronic disease or physical disability and their caregivers, sport coaches, physical therapists and other interested parties will be more easily convinced of the advantages of sports participation for this population.

Although we carefully chose our study settings according to its population, we are aware of some limitations of the study.

Objective measurements are chosen where possible. To achieve the best results possible, the best fitting testing modality will be chosen for each participant. The testing modality has to mimic the type of sport participated in. In children who do not participate in sport, the most common form of physical activity in daily life will be chosen as an indicator for the test modality. In general, some test modalities have been proven to elicit a higher $\text{VO}_{2\text{peak}}$ in the same subject³⁶. The test modality could therefore influence the test outcome. To minimize this bias we will pursue a comparable number of testing modality types in both groups.

In conclusion, this study will provide insight in the effects of sports participation on the health of children and adolescents with a chronic disease or physical disability.

REFERENCES

1. Murphy NA, Carbone PS, American Academy of Pediatrics Council on Children With Disabilities: Promoting the participation of children with disabilities in sports, recreation, and physical activities. *Pediatrics* 2008; 121(5):1057-1061.
2. van Brussel M, van der Net J, Hulzebos E, Helders PJ, Takken T: The Utrecht approach to exercise in chronic childhood conditions: the decade in review. *Pediatr Phys Ther* 2011; 23(1):2-14.
3. Arraiz GA, Wigle DT, Mao Y: Risk assessment of physical activity and physical fitness in the Canada Health Survey mortality follow-up study. *J Clin Epidemiol* 1992; 45(4):419-428.
4. Blair SN, Kohl HW, Paffenbarger RS, Clark DG, Cooper KH, Gibbons LW: Physical fitness and all-cause mortality: A prospective study of healthy men and women. *JAMA* 1989; 262(17):2395-2401.
5. Colditz GA, Samplins-Salgado M, Ryan CT, Dart H, Fisher L, Tokuda A, Rockhill B: Harvard Report on Cancer Prevention, Volume 5 Fulfilling the potential for cancer prevention: policy approaches. *Cancer Causes & Control* 2002; 13(3):199-212.
6. Erikssen G: Physical fitness and changes in mortality. *Sports Med*. 2001; 31(8):571-576.
7. Eime RM, Young JA, Harvey JT, Charity MJ, Payne WR: A systematic review of the psychological and social benefits of participation in sport for children and adolescents: informing development of a conceptual model of health through sport. *Int J Behav Nutr Phys Act* 2013; 10(98):16.
8. Richter KJ, Gaebler Spira D, Mushett CA: Sport and the person with spasticity of cerebral origin. *Dev Med Child Neurol* 1996; 38(9):867-870.
9. Klapwijk A: The multiple benefits of sports for the disabled. *Disabil Rehabil* 1987; 9(2):87-89.
10. Jackson RW, Davis GM: The value of sports and recreation for the physically disabled. *Orthop Clin North Am* 1983; 14(2):301-315.
11. Hutzler Y, Bar Eli M: Psychological benefits of sports for disabled people: A review. *Scand J Med Sci Sports* 1993; 3(4):217-228.
12. Basch CE: Healthier students are better learners: a missing link in school reforms to close the achievement gap. *J Sch Health* 2011; 81(10):593-598.
13. Donnelly JE, Greene JL, Gibson CA, Smith BK, Washburn RA, Sullivan DK, DuBose K, Mayo MS, Schmelzle KH, Ryan JJ: Physical Activity Across the Curriculum (PAAC): a randomized controlled trial to promote physical activity and diminish overweight and obesity in elementary school children. *Prev Med*. 2009; 49(4):336-341.

14. Sallis JF, McKenzie TL, Kolody B, Lewis M, Marshall S, Rosengard P: Effects of health-related physical education on academic achievement: Project SPARK. *Res Q Exerc Sport* 1999; 70(2):127-134.
15. Singh A, Uijtdewilligen L, Twisk JW, Van Mechelen W, Chinapaw MJ: Physical activity and performance at school: a systematic review of the literature including a methodological quality assessment. *Arch Pediatr Adolesc Med*. 2012; 166(1):49-55.
16. Chaddock L, Pontifex MB, Hillman CH, Kramer AF: A review of the relation of aerobic fitness and physical activity to brain structure and function in children. *J Int Neuropsychol Soc* 2011; 17(06):975-985.
17. Pontifex MB, Scudder MR, Drollette ES, Hillman CH: Fit and vigilant: the relationship between poorer aerobic fitness and failures in sustained attention during preadolescence. *Neuropsychology* 2012; 26(4):407.
18. Tomporowski PD, Davis CL, Miller PH, Naglieri JA: Exercise and children's intelligence, cognition, and academic achievement. *Educ Psychol Rev* 2008; 20(2):111-131.
19. Chaddock L, Hillman CH, Pontifex MB, Johnson CR, Raine LB, Kramer AF: Childhood aerobic fitness predicts cognitive performance one year later. *J Sports Sci* 2012; 30(5):421-430.
20. London RA, Castrechini S: A longitudinal examination of the link between youth physical fitness and academic achievement. *J Sch Health* 2011; 81(7):400-408.
21. Eime RM, Young JA, Harvey JT, Charity MJ, Payne WR: A systematic review of the psychological and social benefits of participation in sport for children and adolescents: informing development of a conceptual model of health through sport. *International Journal of Behavioral Nutrition & Physical Activity* 2013; 10(1).
22. Groff DG, Lundberg NR, Zabriskie RB: Influence of adapted sport on quality of life: Perceptions of athletes with cerebral palsy. *Disability & Rehabilitation* 2009; 31(4):318-326.
23. Verschuren O, Ketelaar M, Gorter JW, Helden PJ, Uiterwaal CS, Takken T: Exercise training program in children and adolescents with cerebral palsy: a randomized controlled trial. *Arch Pediatr Adolesc Med* 2007; 161(11):1075-1081.
24. Weiss J, Diamond T, Demark J, Lovald B: Involvement in Special Olympics and its relations to self-concept and actual competency in participants with developmental disabilities. *Res Dev Disabil* 2003; 24(4):281-305.
25. Adirim TA, Cheng TL: Overview of injuries in the young athlete. *Sports Med*. 2003; 33(1):75-81.
26. Trifonov Rexen C, Andersen LB, Ersboll AK, Jespersen E, Franz C, Wedderkopp N: Injuries

- in children with extra physical education in primary schools. *Med Sci Sports Exerc* 2014; 46(4):745-752.
27. World Health Organization: Excessive Physical Training in Children and Adolescents. A Position Statement from the International Federation of Sports Medicine (FIMS). *Schweiz Z Med Traumatol* 1991; 39(1):32-34.
 28. Bloemers F, Collard D, Paw MC, Van Mechelen W, Twisk J, Verhagen E: Physical inactivity is a risk factor for physical activity-related injuries in children. *Br J Sports Med* 2012; 46(9):669-674.
 29. Sinclair SA, Xiang H: Injuries among US children with different types of disabilities. *Am J Public Health* 2008; 98(8):1510-1516.
 30. Collard DC, Chinapaw MJ, Van Mechelen W: Design of the iPlay Study. *Sports Med.* 2009; 39(11):889-901.
 31. Webborn N, Willick S, Reeser JC: Injuries among disabled athletes during the 2002 Winter Paralympic Games. *Med Sci Sports Exerc* 2006; 38(5):811-815.
 32. Zwinkels M, Verschuren O, Lankhorst K, van der Ende-Kastelijn K, de Groot F, Backx F, Visser-Meily J, 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.
 33. Balady GJ, Chaitman B, Driscoll D, Foster C, Froelicher E, Gordon N, Pate R, Rippe J, Bazzarre T: Recommendations for cardiovascular screening, staffing, and emergency policies at health/fitness facilities. *Circulation* 1998; 97(22):2283-2293.
 34. Mok E, Beghin L, Gachon P, Daubrosse C, Fontan JE, Cuisset JM, Gottrand F, Hankard R: Estimating body composition in children with Duchenne muscular dystrophy: comparison of bioelectrical impedance analysis and skinfold-thickness measurement. *Am J Clin Nutr* 2006; 83(1):65-69.
 35. Dosa NP, Foley JT, Eckrich M, Woodall-Ruff D, Liptak GS: Obesity across the lifespan among persons with spina bifida. *Disability & Rehabilitation* 2009; 31(11):914-920.
 36. Bhambhani YN, Holland LJ, Steadward RD: Maximal aerobic power in cerebral palsied wheelchair athletes: validity and reliability. *Archives of Physical Medicine & Rehabilitation* 1992; 73(3):246-252.
 37. Léger LA, Lambert J: A maximal multistage 20-m shuttle run test to predict \dot{V}O₂ max. *Eur J Appl Physiol Occup Physiol* 1982; 49(1):1-12.
 38. Verschuren O, Takken T, Ketelaar M, Gorter JW, Helders PJ: Reliability and validity of data for 2 newly developed shuttle run tests in children with cerebral palsy. *Phys Ther* 2006;

86(8):1107-1117.

39. Washington RL, Bricker JT, Alpert BS, Daniels SR, Deckelbaum RJ, Fisher EA, Gidding SS, Isabel-Jones J, Kavey RE, Marx GR: Guidelines for exercise testing in the pediatric age group. From the Committee on Atherosclerosis and Hypertension in Children, Council on Cardiovascular Disease in the Young, the American Heart Association. *Circulation* 1994; 90(4):2166-2179.
40. 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(7):967-974.
41. Bloemen MA, de Groot JF, Backx FJ, Westerveld RA, Takken T: Arm cranking versus wheelchair propulsion for testing aerobic fitness in children with spina bifida who are wheelchair dependent. *J Rehabil Med* 2015; 47(5):432-437.
42. Verschuren O, Takken T, Ketelaar M, Gorter JW, Helders PJ: Reliability for running tests for measuring agility and anaerobic muscle power in children and adolescents with cerebral palsy. *Pediatr Phys Ther* 2007; 19(2):108-115.
43. 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(12):1129-1135.
44. 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(12):1129-1135.
45. Verschuren O, Bloemen M, Kruitwagen C, Takken T: Reference values for anaerobic performance and agility in ambulatory children and adolescents with cerebral palsy. *Developmental Medicine & Child Neurology* 2010; 52(10):e222-e228.
46. Vanhees L, Lefevre J, Philippaerts R, Martens M, Huygens W, Troosters T, Beunen G: How to assess physical activity? How to assess physical fitness? *Eur J Cardiovasc Prev Rehabil* 2005; 12(2):102-114.
47. Beenakker EA, van der Hoeven JH, Fock JM, Maurits NM: Reference values of maximum isometric muscle force obtained in 270 children aged 4-16 years by hand-held dynamometry. *Neuromuscul Disord* 2001; 11(5):441-446.
48. Winnick J, Short F: The Brockport physical fitness test manual: a health-related test for youths with physical and mental disabilities. Champaign, IL: Human Kinetics 1999.
49. Deitz JC, Kartin D, Kopp K: Review of the Bruininks-Oseretsky test of motor proficiency, (BOT-2). *Phys Occup Ther Pediatr* 2007; 27(4):87-102.

50. Brage S, Brage N, Franks P, Ekelund U, Wareham N: Reliability and validity of the combined heart rate and movement sensor Actiheart. *Eur J Clin Nutr* 2005; 59(4):561-570.
51. De Groot J, de Jong A, Visser T, Takken T: Validation of the Actical and Actiheart monitor in ambulatory children with Spina Bifida. *J Pediatr Rehabil Med: An Interdisciplinary Approach* 2013; 6:103-111.
52. van Mechelen W, Hlobil H, Kemper HC: Incidence, severity, aetiology and prevention of sports injuries. *Sports Med.* 1992; 14(2):82-99.
53. Vos P: De Bourdon concentratietest voor kinderen.[The Bourdon concentration test for children]. 1992 Swetz & Zeitlinger, Lisse (the Netherlands)
54. Van der Stigchel S, Nijboer TC: The imbalance of oculomotor capture in unilateral visual neglect. *Conscious Cogn* 2010;19(1):186-197.
55. Harter S: Self-perception profile for children: revision of the perceived competence scale for children. Denver, CO: University of Denver, Department of Psychology; 1985.
56. Nooijen CF, Post MW, Spijkerman D, Bergen MP, Stam HJ, van den Berg-Emons, Rita JG: Exercise self-efficacy in persons with spinal cord injury: Psychometric properties of the Dutch translation of the exercise self-efficacy scale. *J Rehabil Med* 2013; 45(4):347-350.
57. Verschuren O, Takken T: Aerobic capacity in children and adolescents with cerebral palsy. *Res Dev Disabil* 2010; 31(6):1352-1357.



3

A novel tool for quantifying and promoting physical activity in youths with typical development and youths who are ambulatory and have motor disability

Kristel Lankhorst

Rita van den Berg-Emons

Johannes Bussmann

Herwin Horemans

Janke de Groot

ABSTRACT

Background. Several device-based instruments have been validated in the pediatric population, but none of these are clinically applicable and provide real-time feedback on actual physical activity in terms of postures and movements. A new device (Activ8) is promising for that purpose.

Objective. The objective of this study was to investigate the criterion validity of the Activ8 for measuring static (sitting, standing) and dynamic (walking, bicycling, running) activities, and for separating postures and movements within basic and complex activities in youths with typical development (TD) and peers with motor disability (not typical development [NTD]).

Design. This was a criterion validation study.

Methods. Ten participants with TD (mean age = 14 years; SD = 2.5) and 10 participants with NTD (mean age = 12.9 years; SD = 2.1) performed a standardized series of basic and daily life (complex) activities. The Activ8 measured postures and movements, while camera recording served as a reference. The outcome measures were the mean time differences between the Activ8 output and video data for the merged categories "static" and "dynamic" and for the separate postures and movements.

Results. For the merged categories static and dynamic, the criterion validity was found to be excellent in both participants with TD and participants with NDT within basic activities and was found to be good to excellent in participants with TD and moderate to good in participants with NTD within complex activities. The detection of separate postures and movements was found to be poor to excellent in both groups within complex activities.

Limitations. The sample of youths with NTD was small and limited to youths who could be considered to be at least ambulatory within a household.

Conclusions. Activ8 is a valid tool when the merged categories static and dynamic are used to interpret physical activity in daily life in both youths with TD and youths with NTD and mild motor impairment. To optimize the quantification of separate postures and movements, adjustment of the existing algorithm is required.

INTRODUCTION

The level of being physically active and sedentary are 2 independent factors related to health outcomes in youths.^{1–4} Children and adolescents who are physically active have positive health outcomes. This fact is evident not only in youths with typical development (TD) but also in youths with motor disability—in whom the prevention of health-related problems is an important reason for being active.^{5–7} Therefore, a growing number of physical activity (PA) guidelines address the importance of being physically active and limiting sedentary time to reduce the risk of developing chronic diseases.^{6,8–12} In order to determine whether a child meets the PA guidelines and whether interventions targeting PA are indicated and successful, valid instruments to measure PA in daily life are needed. Self-reported instruments, like questionnaires, are frequently used to assess PA, but are not proven valid.¹³ Device-based instruments, like accelerometers, have been proven more valid to assess PA.^{14–16} Most of the currently used activity monitors¹⁷ are, however, expensive and not easy to use in clinical practice. Clinical use is limited due to complexity of the data analysis, need for additional software and related to the limited clinical applicability of the obtained information regarding PA levels. Moreover, most of the currently validated activity monitors lack a goal-setting and direct feedback function which can support physical therapists in coaching their patients toward healthy active lifestyles.

The Activ8 is a newly developed accelerometry-based activity monitor (developed by 2M Engineering). Given its low costs and easy-to-use features, it seems promising as a new outcome measure for both research and clinical purposes. Next to being a measurement instrument of PA, it also provides a feedback function that can be used as an intervention tool. The Activ8 is a 1-sensor ambulatory 3-dimensional accelerometer with dimensions of 30 × 32 × 10 mm and a weight of 20 g. The Activ8 contains a battery, a real-time clock, and a medium for data storage (up to 1 month). The Activ8 aims to detect the actual PA not in terms of movement counts, but in terms of body postures and movements, including bicycling, besides sitting, standing, walking, and running. Activ8 also has the possibility to provide real-time feedback to the user, health care professional, or both about the volume and distribution of postures and movements either through light-emitting diodes on the device itself or a smartphone. Good criterion validity of the Activ8 to quantify PA has recently been established in both healthy adults and in adults with cerebral palsy (CP)^{18,19} but not yet in youths. Because the validation of accelerometers in the adult population may not translate to children and adolescents,^{20,21}

the reestablishment of validity for this group as well is recommended. For use in clinical pediatric physical therapist practice, it is of interest whether the Activ8 is able to distinguish static (ie, sitting and standing) from dynamic activities (ie, walking, bicycling, and running), as such information can be used to monitor and encourage active behavior in youths. With children often moving in short bouts, it is also of interest whether the Activ8 is able to distinguish the separate postures and movements (ie, sitting, standing, walking, bicycling, running) during short-duration activities. Finally, it is also important to determine the validity of the Activ8 in youths with a pathological gait, such as CP and spina bifida (SB), the 2 largest patient groups within pediatric rehabilitation medicine. Because of muscle atrophy and spasticity, these children and adolescents perform activities in a different way than their peers with TD—a fact that may affect the validity of accelerometers for quantifying PA in these populations. Establishing validity in both youths without and with a pathological gait, would enable physical therapists to use this device in a wide range of patients.

The aim of this study was to investigate the criterion validity of the Activ8 using the current algorithm to quantify static and dynamic activities and to separate postures and movements within basic and complex activities in youths with TD and youths with motor disability (not typical development [NTD]) and who would be considered at least ambulatory in a household.

METHODS

Participants

Children and adolescents were recruited through pediatric physical therapists associated with the research group (Fit for the Future consortium).²² In total, 10 children and adolescents with TD and 10 children and adolescents with CP or SB (NTD) were identified as appropriate samples of the target population in which the measurement instrument will ultimately be used.²³

We used the Functional Mobility Scale (FMS)²⁴ in all participants to objectify the functional mobility at different distances. To specify the functional mobility in more detail in youths with motor disability (NTD), the Gross Motor Function Classification System was used for participants with CP^{25,26} and the adapted Hoffer classification was used for participants with SB.²⁷

The 6 inclusion criteria for both groups were as follows: age of 10 to 18 years; ability to understand the instructions necessary to perform the activity protocol; ability to perform

the activities in the activity protocol; FMS score of 6 for TD group; FMS score of ≥ 4 and Gross Motor Function Classification System 1 or 2 for youths with CP (1); and FMS score ≥ 4 and adapted Hoffer classification ≥ 4 for youths with SB.^{25–27} Person- and disease-related characteristics of the participants are described in Table 1. All participants and their parents gave written informed consent.

Table 1: Characteristics of Youths Who Had Typical Development (TD) and Youths Who Did Not Have Typical Development (NTD)^a

Participant	Age (y)	Height (cm)	Weight (kg)	Sex	Diagnosis	Classification ^b	FMS ^c
TD							
1	16	167	52	M			6
2	16	186	70	M			6
3	10	151	44	M			6
4	10	150	32	F			6
5	14	167	50	F			6
6	15	173	57	F			6
7	14	174	62	M			6
8	17	162	51	F			6
9	16	178	64	F			6
10	12	165	45	M			6
Mean (SD)	14.0 (2.5)	167.3 (11.3)	52.7 (11.1)				
NTD							
11	14	171	62	M	CP	GMFCS 1	5
12	13	167	63	M	CP	GMFCS 1	5
13	11	145	49	M	CP	GMFCS 1	5
14	12	161	48	M	CP	GMFCS 1	5
15	15	182	61	M	CP	GMFCS 1	5
16	14	172	62	M	CP	GMFCS 1	5
17	16	167	98	F	SB	Community walkers	5
18	10	142	32	M	SB	Community walkers	5
19	14	180	92	M	SB	Household walkers	4
20	10	136	34	M	SB	Household walkers	4
Mean (SD)	12.9 (2.1)	162.3 (16.1)	60.1 (21.6)				

^a Youths who did not have typical development were ambulatory and had motor disability; CP = cerebral palsy; SB = spina bifida; FMS = Functional Mobility Scale; GMFCS = Gross Motor Function Classification System. ^bGMFCS²⁵ or Hoffer classification. ^cScore on the FMS.^{24,26}

Testing Procedure

Participants performed a series of basic and complex activities according to a standardized protocol (Tab. 2) in a natural setting which was either at home or at school. We used a standardized protocol in which participants were instructed following the same way. The standardized protocol, used in previous validation studies in adults^{19,28–30} was adapted for validation in youths in close collaboration with pediatric physical therapists, and researchers working in the field of pediatric physical therapy and human movement sciences of the Activ8.²² Three activities (vacuuming, hanging laundry to dry, and doing the dishes) of the adult validation protocol were removed from the adult validation protocol since they were not considered representative for daily activities of children. In addition, we added the following new activities: setting a table, arts and crafts, walking in a slalom, dribbling a ball, and playing a ball game. According to the experts and previous studies with youths, these new activities are more in line with the movement behavior (sport and play) of children in their leisure time.

In line with the adult validation protocol, 2 important distinctions were made between basic and complex activities. Where a basic activity was defined as an activity consisting of a single body posture or movement, like sitting or walking, a complex activity was defined as a combination of postures and movements, during a short period of time. For instance, a ball game could consist of both standing (static), walking and running (dynamic).^{30,31} To ensure validity during real life activities, participants were instructed to perform the activities in their own manner. The researchers asked the participants after each performed activity whether they felt ready to continue the activity protocol; if not, further resting was allowed. The duration of rest that was needed was determined by the participants. The duration of each activity was 1 minute 30 seconds. The total measurement time of the activity protocol was approximately 45 minutes.

Activ8 Activity Monitor

Activ8 recording. The Activ8 was fixated to the front of the upper leg of the participants with Tegaderm (3M) skin tape to ensure a fixed position on the leg (Fig. 1). The postures and movements that the Activ8 can distinguish in the adult healthy population are sitting, standing, walking, bicycling and running. To determine the type of postures and movements, the Activ8 uses an algorithm that combines orientation of the sensor with respect to gravity with movement intensity. For example, during standing or walking the

orientation of the Activ8 is both mainly vertical but movement intensity for walking is higher.

An epoch window was set at 5 seconds, in which each 0.63 second a posture or a movement was detected. This approach resulted in 8 data points per 5-second epoch. Within these 5-second epochs, multiple postures and movements can be recorded. For example, within 1 epoch, 5 samples of standing still (3.1 seconds) and 3 samples of walking (1.9 seconds) can be detected separately. The Activ8 detection of non-wear is based on the absence of movement or signal variability for a time interval of >5 minutes.



Figure 1: Ventral placement of Activ8 (2M Engineering) on the dominant leg.

Video observation. During performance of the protocol activities, video recordings were made with a handheld digital video camera and used as reference method (Fig. 2). The use of video is an appropriate standard for the assessment and observation of postures and movements.^{14,28} To ensure good viewing of the activities, a standardized protocol was used in line with earlier research in adults^{19,30} and youths who (partly) used a wheelchair.²⁸ The video recordings were visually scored independently by 2 trained research assistants (1 pediatric physical therapist and 1 human movement scientist) using a standardized protocol^{19,32} of scoring the postures and movements using 1-second time intervals. The 2 research assistants were trained and supervised by an experienced researcher who is a human movement scientist and familiar with the method and analysis technique of the study.^{19,33} To assure interrater reliability of the observation and scoring of the video, 2 training sessions took place before the actual scoring of the videos. In total, 2 videotapes were used for the calculation of the interrater agreement between the 2 research assistants, in which all activities in the protocol of 1920 time points of activity scores were assessed. The intraclass correlation coefficient was 0.95. To further ensure good data collection and analysis, the 2 research assistants were monitored during the

actual data collection and data analysis was rechecked by the third trained researcher. If there was no consensus between the 2 researchers, a third person (physical therapist and human movement scientist) was asked to define the posture or movement for final allocation. The third researcher was consulted 4 times during the actual observation and scoring of the videos. In 2 participants, it was difficult to distinguish between walking at a slow pace and walking at a normal pace. In 2 other participants, the distinction between walking and standing was difficult when the participants made a turn during both the activities setting a table and the activity walking in a slalom.

Each 1-second interval was scored for the posture or movement at that moment. The video scoring categories were sitting, standing, walking, bicycling, and running. For the complex activities, multiple activities were performed during the measurement period. As with setting a table, the participant walked and stood still within 1 measurement period. During the complex activities, the posture or movement were also scored each second. Within our protocol staircase climbing was defined as walking.



Figure 2: Video observation vs Activ8 (2M Engineering) classification within static and dynamic categories and within separate postures and movements.

Data Analysis

The Activ8 epochs of 5 seconds were synchronized with the 1-second video data. To avoid the effect of protocol activities that were not directly started or that were finished before the scheduled end of the 90-second period, the first 20 seconds and the last 10 seconds from each performed protocol activity were excluded from our analysis.

This approach resulted in a 60-second period of each protocol activity, consisting of twelve 5-second epochs. Thereafter, for each participant the number of seconds of each posture or movement category was calculated for both the Activ8 recording and the video observation. This was the input for further analysis. This method is commonly used for the validation of accelerometers for the assessment of postures and movements.^{28,33}

Outcome Parameters

The relative time difference as a percentage was calculated as $[(\text{Activ8 recording} - \text{video observation}) / \text{video observation}] \times 100$ for the merged categories "static" (sitting, standing) and "dynamic" (walking, bicycling, running) within basic and complex activities. The relative time difference as a percentage was calculated as $[(\text{Activ8 recording} - \text{video observation}) / \text{video observation}] \times 100$ for the separate postures and movements (sitting, standing, walking, bicycling, running) within basic and complex activities.

Criterion validity was determined from the differences between the Activ8 recording and the video observation. We defined a priori and in line with earlier research a maximum 10% mean time difference between the Activ8 recording and the video observation as excellent criterion validity, 10% to 20% as good, 20% to 30% as moderate, and >30% as poor.^{19,28,33,34}

Role of the Funding Source

This study was funded by an unconditional grant (PRO-4-03) from the Dutch Foundation Innovation Alliance (SIA-RAAK), part of the Dutch Organization for Scientific Research (NWO). The funding source had no role in the study's design, conduct, and reporting.

RESULTS

There were no significant differences in age, weight, and height between the TD group and the NTD group. Table 2 shows the participants who were included in the analysis within basic and complex activities. The differences in numbers of participants per activity were due to the ability of a participant to perform an activity, the availability of a staircase, or correct execution of the activity according to the activity protocol.

Table 2: Basic and complex activities in the activity protocol and number of participants for whom data were available for analysis^a.

Activities	No. of Participants included in analysis ^b	
	TD	NTD
Basic ^c (n = 9)		
Sitting	10	10
Sitting slouched on a couch or chair	10	10
Standing	9 (3)	10
Walking at a slow pace ^d	10	10
Normal walking ^d	10	10
Walking at a fast pace ^d	10	10
Running over a distance of 10 m ^d	9 (5)	8 (17 and 18)
Cycling at a slow pace ^d	10	8 (18 and 20)
Cycling at a fast pace ^d	10	7 (14, 18, and 20)
Complex ^e (n = 7)		
Climbing stairs at a preferred pace (forbidden to run on the stairs)	10	8 (15 and 16)
Setting a table	9 (5)	10
Engaging in arts and crafts while sitting in a chair	10	10
Walking in a slalom ^d (6 m, at 0, 1.5, 3, 4.5, and 6 m a pawn was placed)	10	10
Dribbling a ball over a distance of 10 m	10	10
Throwing a ball to a wall (with the participant standing 3 m from the wall)	9 (3)	10
Sitting in a chair, standing up, walking 5 m, returning to the chair, and sitting down	10	10

^aNTD = not typical development (ie, ambulatory and with motor disability); TD = typical development.

^bNumbers in parentheses are the designations of participants who were excluded from the analysis.

^cActivities similar to those in the Activ8 (2M Engineering) classification. ^dParticipants performed the activities at their own preferred and constant pace, ^eActivities of daily living (including combined activities, such as walking, running, and standing during a ball game).

Merged Categories Static and Dynamic

The results concerning the merged categories static and dynamic are presented in Table 3, for both participants with TD and NTD and basic and complex activities. For the basic activities, the relative time difference between the Activ8 recordings and the video observation ranged from -1.3% to 0.7% in participants with TD and from -0.4% to 0.7% in participants with NTD. The criterion validity was therefore rated excellent in both groups for both merged categories static and dynamic. For the complex activities, the relative time difference between the Activ8 recordings and the video observation for static activities was -17%; for dynamic activities, it was 8.3%. Therefore criterion validity during complex activities was rated good to excellent in participants with TD. In participants with NTD, the relative time differences were -24.3% for static activities and 13.3% for dynamic activities; therefore, the criterion validity was rated moderate to good.

Table 3: Mean and relative time differences between video data and activ8^a output within static and dynamic activities.

Group ^b	Activity Category	Activities	Video (s)	Activ8 (s)	Mean Time Difference (s) ^c	Relative Time Difference (%) ^d	Criterion Validity Score
TD	Basic	Static ^e	1764	1741	-23	-1.3	Excellent
		Dynamic ^f	3516	3539	23	0.7	Excellent
NTD	Basic	Static ^e	1802	1815	13	0.7	Excellent
		Dynamic ^f	3178	3165	-13	-0.4	Excellent
TD	Complex	Static ^e	1515	1258	-257	-17.0	Good
		Dynamic ^f	3105	3362	257	8.3	Excellent
NTD	Complex	Static ^e	1658	1256	-402	-24.3	Moderate
		Dynamic ^f	3022	3424	402	13.3	Good

^a2M Engineering. ^bNTD = not typical development (ie, ambulatory and with motor disability); TD = typical development. ^cCalculated as (Activ8 – video). ^dCalculated as $[(\text{Activ8 time} - \text{video time}) / \text{video time}] \times 100$. ^eMerged category of sitting and standing. ^fMerged category of walking, bicycling, and running.

Separate Analysis of Postures and Movements

The results concerning the separate postures and movements are presented in Table 4. For the basic activities, the relative time difference between the Activ8 recordings and the video observation ranged from -14.7% (standing) to -0.2% (walking) in participants

with TD. This difference in standing was mainly the result of standing being sometimes classified as sitting by the Activ8.

In participants with NTD, the relative time difference between Activ8 recordings and the video observation ranged from 3.4% (running) to 39.7% (bicycling). In 1 participant (designated 20), the posture standing was detected as sitting, while for another participant (designated 16), the activity bicycling at a slow pace was detected as sitting. This resulted in an overestimation of sitting. Furthermore, in 3 participants (designated 16, 19, and 20), the activities walking at a slow pace, walking at a normal pace, and walking at a fast pace were, for several seconds, detected as bicycling. This finding resulted in an overestimation of bicycling and an underestimation of walking.

Within basic activities, the criterion validity was rated good for standing and excellent for the other activities (sitting, walking, bicycling and running) in participants with TD. In participants with NTD, the criterion validity was rated excellent for sitting (6.9%) and running (3.4%), good for standing (-11.6%), moderate for walking (-21%), and poor for the activity bicycling (39.7%).

Within complex activities, the relative time difference between Activ8 recordings and video observation varied from -42.1% (standing) to 3.4% (walking) in participants with TD. Within setting a table, ball game, and stand up from a chair, walk back and sit down, standing was underestimated and detected as walking. In 4 participants (designated 1, 3, 8, and 10), the complex activity climbing stairs was detected as bicycling instead of walking. This resulted in an overestimation of bicycling (182 seconds).

For participants with NTD, the time difference ranged from -8.1% (sitting) to 265.2% (running). Overall, in 2 participants (designated 16 and 20), the Activ8 overestimated bicycling (713 seconds) in disadvantage of walking and standing, which were underestimated (-372 seconds and -296 seconds, respectively). The underestimation of walking was seen in all complex activities that included the activity walking (setting a table, walking in a slalom, ball game, dribbling with a ball, and stand up from a chair, walk back and sit down).

Within complex activities, the criterion validity in participants with TD was rated excellent for walking, good for sitting, moderate for running and poor for standing. In participants with NTD, the criterion validity was rated excellent for sitting, good for walking and poor for standing and running.

Table 4: Mean and relative time differences between video data and activ8^a output within basic and complex activities.

Group ^b	Activity Category	Activities	Video (s)	Activ8 (s)	Mean Time Difference (s) ^c	Relative Time Difference (%) ^d	Criterion Validity Score
TD	Basic	Static ^e	1764	1741	-23	-1.3	Excellent
		Dynamic ^f	3516	3539	23	0.7	Excellent
NTD	Basic	Static ^e	1802	1815	13	0.7	Excellent
		Dynamic ^f	3178	3165	-13	-0.4	Excellent
TD	Complex	Static ^e	1515	1258	-257	-17.0	Good
		Dynamic ^f	3105	3362	257	8.3	Excellent
NTD	Complex	Static ^e	1658	1256	-402	-24.3	Moderate
		Dynamic ^f	3022	3424	402	13.3	Good

^a2M Engineering. ^bNTD = not typical development (ie, ambulatory and with motor disability); TD = typical development. ^cCalculated as (Activ8 – video). ^dCalculated as [(Activ8 time – video time)/video time] × 100. ^eMerged category of sitting and standing. ^fMerged category of walking, bicycling, and running.

DISCUSSION

The aim of the current study was to investigate the criterion validity of the Activ8, using the current algorithm to quantify (1) static and dynamic activities and (2) separate postures and movements within both basic and complex activities in youths with TD and youths with motor disability (NTD) and who would be considered at least ambulatory in a household.

The results of our study indicate that the current version of the Activ8 is a valid device for distinguishing between static and dynamic activities in daily life in both youths with TD and youths with mild motor disabilities and for quantifying separate movements and postures in youths with TD, with the exception of standing, which was often quantified as the non-dynamic posture sitting. At the same time, though, the currently used algorithm in the Activ8 is not valid for quantifying separate postures and movements during some complex activities in youths with motor disability. These data imply that the Activ8, in its current state, can be used in clinical practice and research only for the quantification of static and dynamic activities and not for the quantification of separate postures and movements in children with motor disability.

Furthermore, the Activ8 can validly distinguish separate postures and movements within basic activities in youths with TD (criterion validity: good to excellent). In youths with NTD, a more varied criterion validity was found, which ranged from poor (bicycling) to excellent (sitting and running), probably due to the presence of a so called crouched posture and gait (more degrees of hip and knee flexion in standing position or during gait) which is common in persons with CP or SB. Exclusion of the 2 participants with a pronounced crouched posture and gait from the analysis, resulted in excellent classifications of the posture standing and the activities walking and bicycling within basic activities (Supplement I). These results are in line with studies in adults with CP and patients after stroke, where Activ8 also had difficulty recognizing certain postures and movement in participants with a more pronounced crouched posture or gait, due to a different hip angle (different angular position of Activ8).^{32,35} At the same time these results indicate, that for youths with more severe motor impairments, further validation is still recommended. The existing thresholds used in the algorithm of the Activ8, developed for use in healthy adults are not yet sufficient enough to quantify separate postures and movements within complex activities or during short bouts of activities. In both youths with TD and youths with NTD, the Activ8 had difficulty to distinguish standing from other postures or movements within complex activities (Supplement II). For example, walking and standing were alternated during an activity as setting a table. If the posture standing was performed for 1 second and was immediately followed by walking, the Activ8 did not detect standing because the posture standing was too short for an adequate detection. These results are also in line with 2 earlier studies, also reporting the inaccurate detection of standing by Activ8 in free-living conditions in adults with CP and adults after stroke.^{32,35} One might question how accurately the Activ8 needs to quantify short-term activities in daily life because that affects the determined acceleration threshold used in the algorithm.^{32,35} Furthermore, climbing stairs was sometimes classified as bicycling instead of walking in both participants with TD and participants with NTD. This finding is most likely related to the height difference between children and adults resulting in a larger hip flexion during stair climbing in youths.

Comparing the criterion validity of the Activ8 with that of other accelerometers quantifying postures and movements in daily life in youths is difficult. Accelerometers that are often used to measure PA in youths focus only on estimating energy expenditure of PA rather than the type of PA.^{14,20,36–39} The activPAL (PAL Technologies) is one of the few activity monitors that measures several body postures and motions (lying, sitting,

standing, and walking) and that was evaluated for criterion validity in free-living conditions in youths.^{40,41} Studies showed an acceptable agreement of detecting sitting, standing and walking activities separately or distinguishing lying or sitting from upright activities with the activPAL compared to direct observation in youths with TD and peers with CP. In participants with a crouched posture/gait or toe walk also lower levels of agreement were found,^{40,41} similar to the validity results in our study.

The VitaMove (2M Engineering) as the manufacturer of VitaMove, which is a 3-sensor activity monitor and the precursor of the Activ8, showed good validity for detecting the types of postures and movements in youths who were ambulatory and had motor disability and in the adult clinical population.^{28,33,42,43} The use of the Activ8 is, however, preferred over the activPAL and the VitaMove because the Activ8 can measure for a longer period of time (ie, 1 month instead of 1 week), is low-priced, has easy to use data analysis and interpretation and consist of only 1 small sensor (instead of 3 for the VitaMove). In addition, a feedback tool on the device and a smartphone is available for the Activ8, while this is not available for other devices like the VitaMove or the activPAL.

Strengths and Limitations

There were some limitations in this study. The sample size was small in both groups. At the same time, this sample size is common in comparable studies evaluating criterion validity of accelerometers.^{19,32,40,43} In addition, the 1-second scoring of the video within complex activities was sometimes difficult. Postures and movements during daily life activities (complex activities) performed by the participants were difficult to allocate to 1 posture or movement category on a second-by-second basis. Finally, the sample of youths with NTD was limited to participants with mild motor impairments and those who could be considered to be at least ambulatory in a household. At the same time, including a mixed sample of both youths with and youths without motor impairments, also enlarges the generalizability of the results to a clinical population ranging from household to normal ambulatory level. Another strength was the involvement of a multidisciplinary group of experts in the adaptation of the standardized protocol used in adults to a protocol for the pediatric population. Finally, the labor intensive data recording was performed by trained researchers using robust protocols and expertise from several other validation studies in the PA field of research.

Clinical Implications and Recommendations

Our results indicate that the Activ8 can validly distinguish static from dynamic activities in both youths with TD and youths with NTD during both basic activities and complex activities. At the same time, though, the Activ8, with the current setting for adults, was not valid for distinguishing separate postures and movements during more complex activities similar to those in daily life, in both youths with TD and youths with NTD. Misdetections were related to a greater hip angle in youths, a (relative) inability to detect short-duration activities and the presence of crouched posture or gait in those with more severe motor impairments. Based on the results of this study and 2 other studies determining the validity of Activ8,^{32,35} developers of Activ8 recently adapted the algorithm for hip angle and specific thresholds to resolve the inaccurate detection of separate postures and movements when using the Activ8 in youths and in participants with a crouched posture or gait. Future validation studies should include youths with more severe motor impairment.

The advantage of the Activ8, in addition to monitoring PA in a clinician friendly way, is its possibilities to provide real-time feedback to patients using a goal-setting function. Activ8 can therefore be used in encouraging behavioral change, as part of tailored interventions targeting PA in youths with motor disability and youths without motor disability. In that perspective, using the Activ8 provides new possibilities to assess associations between PA and health, to monitor change of PA over time, and to estimate population prevalence of meeting PA guidelines in both youths with TD and youths with NTD.

In conclusion, when interested in assessment of static and dynamic activities in daily life, the Activ8 is a valid device in youths with TD and youths who are ambulatory and have mild motor disabilities for both research and clinical purposes. The existing thresholds of Activ8 to quantify separate postures and movements were not valid and the algorithm has now been adapted. Future research should study the validity of an adapted algorithm in a larger sample of participants, with special attention to those with more severe motor impairments and other pediatric diagnoses.

Supplement I: Mean and Relative Time Differences Between Video Data and Activ8^a Output Within Basic and Complex Activities in participants with NTD^b (N=8) with exclusion of 2 participants.

Activity Category	Activity	Video (s)	Activ8 (s)	Mean Time Difference (s) ^c	Relative Time Difference (%) ^d	Criterion Validity Score
Basic	Static ^e	1442	1440	-2	-0.1	Good
	Dynamic ^f	2698	2700	2	0.1	Excellent
Basic	Sitting	960	960	0	0.0	Excellent
	Standing	482	480	-2	-0.4	Excellent
	Walking	1470	1390	-80	-5.4	Excellent
	Bicycling	840	844	4	0.5	Excellent
	Running	388	466	78	20.1	Moderate
Complex	Static ^e	1368	1016	-352	-25.7	Moderate
	Dynamic ^f	2352	2704	352	15.0	Good
Complex	Sitting	1047	962	-85	-8.1	Excellent
	Standing	321	54	-267	-83.2	Poor
	Walking	2329	2604	275	11.8	Good
	Bicycling	0	76	76	NA	NA
	Running	23	24	1	4.4	Excellent

^a2M Engineering. ^bNTD = not typical development (ie, ambulatory and with motor disability); TD = typical development. ^cCalculated as (Activ8 – video). ^dCalculated as [(Activ8 time – video time)/video time] × 100. NA = not applicable.

Supplement II: Mean and relative time differences between video data and Activ8^a output within complex activities in both participants with TD and participants with NTD^b.

COMPLEX ACTIVITIES									
	Video	Activ8	Mean time difference (s) ^c	Relative time difference (%) ^d	Posture or movement	Video	Activ8	Mean time difference (s) ^c	Relative time difference (%) ^d
TD						NTD			
Climbing stairs	580	423	-157	-27.1	Walking	480	356	-124	-25.8
	0	176	176	NA	Bicycling	0	124	124	NA
	20	1	-19	-95.0	Running
Setting the table	154	82	-72	-46.8	Standing	99	26	-73	-73.7
	386	458	72	18.7	Walking	501	477	-24	-4.8
	Bicycling	0	97	97	NA
Art and craft	600	600	0	0	Sitting	600	600	0	0
work while sitting									
Walking in a slalom	600	600	0	0	Walking	600	431	-169	-28.2
	Bicycling	0	168	168	NA
	Running	0	1	1	NA
Dribbling with a ball	2	0	-2	-100	Standing				
	515	523	8	1.6	Walking	577	433	-144	-25.0
	Bicycling	0	144	144	NA
	83	77	-6	-7.2	Running	23	23	0	0
Ball game	75	50	-25	-33.3	Standing	251	28	-223	-88.8
	447	475	28	6.3	Walking	349	452	103	29.5
	0	6	6	NA	Bicycling	0	120	120	NA
	18	9	-9	-50.0	Running
Sitting in a chair, stand up, 5 meter walk and back to the chair and sit down	162	8	-154	-95.1	Sitting	108	2	-106	-98.2
	438	584	146	33.3	Walking	492	478	-14	-2.9
	Bicycling	0	60	60	NA
	0	8	8	NA	Running	0	60	60	NA

^a2M Engineering. ^bNTD = not typical development (ie, ambulatory and with motor disability); TD = typical development. ^cCalculated as (Activ8 – video). ^dCalculated as [(Activ8 time – video time)/video time] × 100. NA = not applicable.

REFERENCES

1. Jimenez-Pavon D, Kelly J, Reilly JJ. Associations between objectively measured habitual physical activity and adiposity in children and adolescents: Systematic review. *Int J Pediatr Obes* 2010;5:3-18.
2. LaMonte MJ, Blair SN. Physical activity, cardiorespiratory fitness, and adiposity: contributions to disease risk. *Curr Opin Clin Nutr Metab Care* 2006;9:540-546.
3. Riddoch CJ, Mattocks C, Deere K, Saunders J, Kirkby J, Tilling K, et al. Objective measurement of levels and patterns of physical activity. *Arch Dis Child* 2007;92:963-969.
4. Saunders TJ, Gray CE, Poitras VJ, Chaput JP, Janssen I, Katzmarzyk PT, et al. Combinations of physical activity, sedentary behaviour and sleep: relationships with health in children and youth. *Med Sci Sports Exerc* 2016;48(5 suppl 1):912.
5. Balemans ACJ, Van Wely L, Becher JG, Dallmeijer AJ. Associations between fitness and mobility capacity in school-aged children with cerebral palsy: a longitudinal analysis. *Dev Med Child Neurol* 2015;57:660-667.
6. Verschuren O, de Haan F, Mead G, Fengler B, Visser-Meily A. Characterizing energy expenditure during sedentary behavior after stroke. *Arch Phys Med Rehabil* 2016;97:232-237.
7. Schoenmakers MA, de Groot JF, Gorter JW, Hillaert JL, Helders PJ, Takken T. Muscle strength, aerobic capacity and physical activity in independent ambulating children with lumbosacral spina bifida. *Disabil Rehabil* 2009;31:259-266.
8. Biddle SJ, Gorely T, Stensel DJ. Health-enhancing physical activity and sedentary behaviour in children and adolescents. *J Sports Sci* 2004;22:679-701.
9. Penedo FJ, Dahn JR. Exercise and well-being: a review of mental and physical health benefits associated with physical activity. *Curr Opin Psychiatry* 2005;18:189-193.
10. Janssen I, Leblanc AG. Systematic review of the health benefits of physical activity and fitness in school-aged children and youth. *Int J Behav Nutr Phys Act* 2010;7:40.
11. Roman-Vinas B, Chaput JP, Katzmarzyk PT, Fogelholm M, Lambert EV, Maher C, et al. Proportion of children meeting recommendations for 24-hour movement guidelines and associations with adiposity in a 12-country study. *Int J Behav Nutr Phys Act* 2016;13:123.
12. Burghard M, Knitel K, van Oost I, Tremblay MS, Takken T; Dutch Physical Activity Report Card Study Group. Is our youth cycling to health? Results from the Netherlands' 2016 report card on physical activity for children and youth. *J Phys Act Health* 2016;13(11 suppl 2):S218-S224.

13. Hidding LM, Altenburg TM, Mokkink LB, Terwee CB, Chinapaw MJ. Systematic review of childhood sedentary behavior questionnaires: what do we know and what is next? *Sports Med* 2017;47:677–699.
14. Sirard JR, Pate RR. Physical activity assessment in children and adolescents. *Sports Med* 2001;31:439-454.
15. Aminian S, Hinckson EA. Examining the validity of the ActivPAL monitor in measuring posture and ambulatory movement in children. *Int J Behav Nutr Phys Act* 2012;9:119.
16. Basterfield L, Adamson AJ, Pearce MS, Reilly JJ. Stability of habitual physical activity and sedentary behavior monitoring by accelerometry in 6- to 8-year-olds. *J Phys Act Health* 2011;8:543-547.
17. Montoye AH, Moore RW, Bowles HR, Korycinski R, Pfeiffer KA. Reporting accelerometer methods in physical activity intervention studies: a systematic review and recommendations for authors. *Br J Sports Med* 2016. doi: 10.1136/bjsports-2015-095947.
18. Erasmus MC University Medical Center. Validation of Activ8 Activity Monitor: Detection of Body Postures and Movements. Rotterdam, the Netherlands: Erasmus MC University Medical Center; 2013.
19. Claridge EA, McPhee PG, Timmons BW, Martin Ginis KA, Macdonald MJ, Gorter JW. Quantification of physical activity and sedentary time in adults with cerebral palsy. *Med Sci Sports Exerc* 2015;47:1719-1726.
20. Strath SJ, Pfeiffer KA, Whitt-Glover M. Accelerometer use with children, older adults, and adults with functional limitations. *Med Sci Sports Exerc* 2012;44:S77-S85.
21. Hildebrand M, VAN Hees VT, Hansen BH, Ekelund U. Age group comparability of raw accelerometer output from wrist- and hip-worn monitors. *Med Sci Sports Exerc* 2014;46:1816-1824.
22. de Groot JF, Overvelde A. Healthy Active Living in Youth With Neuromotor Disability (HALYNeD) Project: a translational project with researchers, pediatric physical therapists, and patients working together toward evidence-based exercise prescription. *Pediatr Phys Ther* 2013;25:228-229.
23. de Vet HCW, Terwee CB, Mokkink LB, Knol DL. Measurement in Medicine: A Practical Guide. New York, NY: Cambridge University Press; 2011:159.
24. Graham HK, Harvey A, Rodda J, Nattrass GR, Pirpiris M. The Functional Mobility Scale (FMS). *J Pediatr Orthop* 2004;24:514-520.
25. Wood E, Rosenbaum P. The Gross Motor Function Classification System for

- cerebral palsy: a study of reliability and stability over time. *Dev Med Child Neurol* 2000;42:292-296.
26. Palisano RJ, Rosenbaum P, Bartlett D, Livingston MH. Content validity of the expanded and revised Gross Motor Function Classification System. *Dev Med Child Neurol* 2008;50:744-750.
 27. Schoenmakers MA, Gulmans VA, Gooskens RH, Pruijs JE, Helders PJ. Spinal fusion in children with spina bifida: influence on ambulation level and functional abilities. *Eur Spine J* 2005;14:415-422.
 28. Nooijen CF, de Groot JF, Stam HJ, van den Berg-Emons RJ, Bussmann HB; Fit for the Future Consortium. Validation of an activity monitor for children who are partly or completely wheelchair-dependent. *J Neuroeng Rehabil* 2015;12:11.
 29. Postma K, Bussmann J, Sluis T, Bergen M, Stam H. Validity of the detection of wheelchair propulsion as measured with an activity monitor in patients with spinal cord injury. *Spinal Cord* 2005;43:550-557.
 30. Bussmann J, Martens W, Tulen J, Schasfoort F, Van Den Berg-Emons H, Stam H. Measuring daily behavior using ambulatory accelerometry: the activity monitor. *Behav Res Methods Instrum Comput* 2001;33:349-356.
 31. Bouisset S, Do MC. Posture, dynamic stability, and voluntary movement. *Neurophysiol Clin* 2008;38:345-362.
 32. Fanchamps MHJ, Horemans HLD, Ribbers GM, Stam HJ, Bussmann JBJ. The accuracy of the detection of body postures and movements using a physical activity monitor in people after a stroke. *Sensors (Basel)* 2018;18: E2167.
 33. Bussmann JB, Tulen JH, van Herel EC, Stam HJ. Quantification of physical activities by means of ambulatory accelerometry: a validation study. *Psychophysiology* 1998;35:488-496.
 34. Altman DG, ed. *Practical Statistics for Medical Research*. Boca Raton, FL: Chapman & Hall/CRC; 1990.
 35. Claridge EA, McPhee PG, Timmons BW, Martin Ginis KA, Macdonald MJ, Gorter JW. Quantification of physical activity and sedentary time in adults with cerebral palsy. *Med Sci Sports Exerc* 2015;47:1719-1726.
 36. Corder K, Ekelund U, Steele RM, Wareham NJ, Brage S. Assessment of physical activity in youth. *J Appl Physiol* 2008;105:977-987.
 37. Oliver M, Schofield GM, Kolt GS. Physical activity in preschoolers: understanding prevalence and measurement issues. *Sports Med* 2007;37:1045-1070.

38. Reilly JJ, Penpraze V, Hislop J, Davies G, Grant S, Paton JY. Objective measurement of physical activity and sedentary behaviour: review with new data. *Arch Dis Child* 2008;93:614-619.
39. Rowlands AV. Accelerometer assessment of physical activity in children: an update. *Pediatr Exerc Sci* 2007;19:252-266.
40. Tang KT, Richardson AM, Maxwell D, Spence WD, Stansfield BW. Evaluation of an activity monitor for the objective measurement of free-living physical activity in children with cerebral palsy. *Arch Phys Med Rehabil* 2013;94:2549-2558.
41. O'Donoghue D, Kennedy N. Validity of an activity monitor in young people with cerebral palsy Gross Motor Function Classification System level I. *Physiol Meas*. 2014;35:2307-2318.
42. Bussmann JB, van de Laar YM, Neeleman MP, Stam HJ. Ambulatory accelerometry to quantify motor behaviour in patients after failed back surgery: a validation study. *Pain* 1998;74:153-161.
43. van den Berg-Emons HJ, Bussmann JB, Balk AH, Stam HJ. Validity of ambulatory accelerometry to quantify physical activity in heart failure. *Scand J Rehabil Med* 2000;32:187-192.



4

Sports participation, physical activity, and health-related fitness in youth with chronic diseases or physical disabilities: the health in adapted youth sports study

Kristel Lankhorst

Tim Takken

Maremka Zwinkels

Leendert van Gaalen

Saskia te Velde

Frank Backx

Olaf Verschuren

Harriet Wittink

Janke de Groot

ABSTRACT

Background. Youth with chronic diseases or physical disabilities (CDPD) often show reduced fitness and physical activity (PA) levels and participate less in organized sports compared to healthy peers. The purpose of this study was to examine the associations between participation in sports and health-related fitness and PA in youth with CDPD.

Methods. A total of 163 participants (mean age 14; range 8-19 years) with CDPD were included in this cross-sectional study, with 81 participating in organized sports and 82 not. Participants were recruited between October 2014 and November 2016. Aerobic and anaerobic fitness, agility and muscle strength were assessed in the lab while PA was monitored in daily life using accelerometry during one week. Linear regression analyses were used to assess the associations of sports participation (independent variable) with health-related fitness and PA (dependent variables).

Results. Results show that youth with CDPD participating in organized sports two times a week performed better on all outcome measures. They reached a higher peak oxygen uptake (difference of 4.9 ml O₂/kg/min, $P=0.001$) compared to their peers not participating in sports. Also, anaerobic fitness, agility, muscle strength and PA were all positively associated with sports participation. Moreover, the association between sports participation and aerobic fitness was mediated by PA for 31% ($P=0.045$).

Conclusion. Participation in sports is associated with both higher levels of PA and health-related fitness in youth with CDPD. Promotion and stimulation of participation in sports seems a good way to promote health-related fitness as well as a healthy active lifestyle in youth with CDPD.

INTRODUCTION

Youth with chronic diseases or physical disabilities (CDPD) have lower fitness level¹, lower levels of physical activity (PA)^{2,3} and a higher prevalence of adiposity⁴ than their healthy peers. The detrimental health effects of physical inactivity in healthy youth are well established⁵⁻⁷. In healthy youngsters, insufficient PA levels are highly associated with low cardiorespiratory fitness, higher levels of obesity and increased cardiovascular risk^{8,9}. Many studies have reported the benefits of sports in promoting PA^{10,11} and its contribution of moderate-to-vigorous physical activity (MVPA) to daily recommended PA in healthy youth¹². The current recommendations for youth (5-17 years), including those with CDPD, state that they should accumulate at least 60 min of MVPA daily for healthy development or 20 min of vigorous physical activity (VPA) for at least three times weekly to achieve the same health effect^{13,14}. Shorter exercise time with a higher intensity (VPA) support sports participation to meet PA guidelines and to develop a healthy active lifestyle.

While the health benefits related to sports and PA have been reported in healthy youth, there is only limited evidence as to what extent sports participation may affect PA and health-related fitness in youth with CDPD. A recent study did report a significant correlation between aerobic fitness and cardiovascular health and also an inverse relationship between adiposity and cardiovascular health in youth with CDPD¹⁵. Finding ways to improve health-related fitness and cardiovascular health therefore seems an important goal for youth with CDPD. At the same time, training studies have shown that youth with CDPD indeed can improve their PA level and health-related fitness through supervised intervention programs in rehabilitation settings^{1,16}. However, the positive results achieved are often not maintained in the longer term after these programs end^{17,18}. Weekly participation in sports could be, based on what we know in healthy youth, a solution to maintain and optimize PA levels and health-related fitness in youth with CDPD.

A recent study showed that youth with CDPD often do not meet the guidelines for healthy PA. While already difficult for healthy peers to meet recommended levels of PA, a Dutch publication showed that youth with CDPD participate even less in competitive and recreational sports^{13,14}. Only 26% of youth with CDPD participate in sports once a week¹⁴ compared to 71% of their healthy peers¹³.

While this low adherence to PA guidelines might be detrimental to health-related fitness and physical functioning, several studies have reported important barriers for youth with

CDPD making it difficult to participate in sports. These barriers include both personal factors, e.g. the lack of energy, fatigue and lack of 'leisure' time due to longer school hours and more time spent in activities of daily living, and environmental factors^{14,19}. The latter includes accessibility of the physical environment, but also attitudes from the social environment, e.g. parents, teachers, healthcare professionals and policy makers, including fear of injury, thinking sports might be too difficult, lack of knowledge regarding the importance of PA for youth with CDPD, or more practical considerations such as transportation to an adapted sport facility²⁰. To overcome some of these social barriers, research is needed showing that there are positive associations between sports participation and health-related fitness. With this knowledge, attitudes might change towards the importance of sports participation for youth with CDPD.

We hypothesize that youth with CDPD who participate in sports are more active and show better health-related fitness outcomes than those who are not. Therefore, the aim of the present study was to investigate the associations of sports participation with health-related fitness and PA in youth with CDPD.

METHODS

Experimental approach to the problem

This study is part of the larger Health in Adapted Youth Sports (HAYS) study. We used a cross-sectional prospective design to investigate the associations of sports participation with physical fitness, PA, cognition, cardiovascular health and quality of life in youth with CDPD. The current paper focuses on the associations of sports participation with physical activity and health-related fitness in youth with CDPD. The associations of sport participation with self-perception, exercise self-efficacy and quality of life were published previously²¹. All assessments were performed between October 2014 and November 2016. A detailed description of the research design and testing procedures has been published previously²². An overview of the outcome measures and used measurement instruments of the current study are summarized in Table 1.

Table 1: Overview of the outcome measures and chosen measurement instruments in this study.

Outcome measure	Parameter	Variable	Measurement
General			
	Demographics	DOB, gender, medical diagnosis, Activity level and participation in sports	General questionnaire
Health-related fitness			
Cardio-vascular health	Metabolic	BMI	Height, weight
		Body composition	Waist and hip circumference
		Fat Mass	BIA
		Blood pressure	Arteriograph
		Arterial stiffness	Arteriograph
		Pulse wave velocity	Arteriograph
Physical fitness	Muscle strength	Isometric muscle strength	Grip strength
		Explosive muscle strength	Standing broad jump
	Agility	Time	10x5 meter sprint
	Anaerobic fitness	Mean power	MPST
	Aerobic fitness	VO _{2peak}	CPET, Shuttle run test or Bicycle test, Godfrey protocol
		RER	
		Heartrate	
Physical activity			
	Modality	Type of activity	Activity monitor (Activ8) and Activity diary

DOB; date of birth, BMI; body mass index, BIA; body impedance analysis, VO_{2peak}; peak oxygen uptake, CPET; cardiopulmonary exercise test, RER; respiratory exchange ratio, MPST; muscle power sprint test.

Subjects

Participants were eligible for this study when they were ambulatory, aged from 8 up to 19 years with CDPD and diagnosed with either cardiovascular, pulmonary, musculoskeletal, metabolic or neuromuscular disorder. Table 2 shows the eligibility and exclusion criteria. The characteristics of the participants such as age, sex, medical diagnosis, identification of sports participation and non-sport are displayed in Table 3. The medical diagnoses

were further classified into categories according to the American College of Sports Medicine (Table 3)²³.

Written informed consent was provided by all participants and as required by Dutch law also by the parents of participants under 18 years of age. In line with Dutch law, no parental informed consent was required for participants aged 18 years and above. This study was approved by the Medical Ethics Committee of the University Medical Center Utrecht, the Netherlands (METC number: 14-332/c).

Table 2: Eligibility and exclusion criteria.

Eligibility	Exclusion
<ul style="list-style-type: none">• Children and adolescents with a physical disability or chronic disease: cardiovascular, pulmonary, musculoskeletal or neuromuscular disorder• Children and adolescents between the age of 8 and 19 years• Children and adolescents had to understand simple instructions• Children and adolescents who were able to perform physical fitness tests	<ul style="list-style-type: none">• Children and adolescents with progressive diseases• Children and adolescents using a wheelchair as main mode of mobility• During the length of the study, children were not allowed to participate in other research projects which might influenced the current study results• For the sporting group of the HAYS-study only: subjects who have not participated in any sports for the preceding three months.• No signed informed consent

Procedures

Independent variable

Sports participation Sports participation was identified using three standardized questions used by the National Institute for Public Health and the Environment (RIVM)²⁴: 1) do you participate in organized sports?, 2) what is / are the type of organized sport(s)? and, 3) what is the frequency of participation in organized sports per week?. When participants were involved in organized sports at least two times per week, they were classified as the ‘sports group’ (SG), all others were classified as the ‘non’-sporting group (NSG).

Dependent variables Physical activity and health-related fitness including cardiovascular health and physical fitness, were assessed (Table 1). Measurement instruments and tests which were valid and reliable in children and adolescents with physical disabilities were used. Specific details about the validity and reliability of the measurement instruments and tests were described in detail previously²². One week before the testing session, the general questionnaire was completed by the participant. Each assessment was

performed using a standardized test protocol. All measurements related to health-related fitness took place on one day and were carried out in the following order; height and weight, bioelectric impedance analysis, waist and hip circumference measurements, Arteriograph measurement, grip strength, standing broad jump, 10x5 meter sprint, muscle power sprint test (MPST) and cardiopulmonary exercise test. Finally, the use of the PA monitor and activity diary was explained to the participant for the PA monitoring in the home situation (Table 1). In general, instructions were given by the researcher and if needed a practice session took place before the actual test started. When this was the case, the participant received sufficient rest before the actual test took place. Between each test there was time for recovery. In addition, the researchers asked the participants after each test and recovery period if the participant felt ready to continue the test protocol; if not further resting was allowed. The need extra of recovery was determined by the participant.

Health-related fitness

Cardiovascular health Height and weight were measured, body mass index (BMI) was calculated using body weight (kg) / body height² (m). For waist and hip circumference, a measure in standing position was taken at the umbilicus and trochanter major and waist / hip ratio was defined. To control for differences in age, Z-scores were calculated of body height, weight, BMI, waist and hip circumference and waist / hip ratio according to Dutch reference values²³. Fat mass was measured in supine position with bioelectrical impedance analysis (Bodystat Quadscan 4000, EuroMedix, Leuven, Belgium).

Arterial stiffness was determined by aortic pulse wave velocity (PWVao), as a measure of arterial stiffness, and augmentation index (AIX%), as measure of peripheral arterial tone. The measurements were performed in a supine position after ten-minute rest by a non-invasive oscillometric tonometry device (Arteriograph, TensioMed Ltd, Budapest, Hungary) at the right arm. A higher PWVao indicates a higher aortic stiffness and a higher AIX% indicates a higher peripheral arterial tone. Blood pressure (systolic and diastolic) and resting heart rate were also measured using the Arteriograph within the same measurement. Instructions to the subject were no food intake three hours before measurement and no talking during the measurement. After the measurement of arterial stiffness, the child was allowed to eat something, before continuation of the other tests.

Physical fitness To test the strength of the subjects, tests from the Brockport fitness test were chosen²⁴. The isometric muscle strength was tested through the use of a hand

held hydraulic dynamometer (HHD)²⁵. The participants' dominant hand was tested and one practice session took place before the actual test started. Mean grip strength was calculated out of three attempts.

The standing broad jump was used to evaluate the explosive strength of the lower limbs by measuring the distance jumped with two legs together from standing position²⁶. Mean distance was calculated of three trials and used for analysis. One practice session took place before the actual test started.

Agility was measured using the 10x5 Meter Sprint Test²⁷ and also valid for use in children and adolescents with and without a pathological gait pattern²⁸. During this test, the child was asked to sprint as fast as possible, 10 times, in between 2 lines that were 5 meter apart. There was no resting period, so the child / adolescent had to turn as fast as possible during this test. Time was recorded using a stopwatch.

Anaerobic fitness was determined by the Muscle Power Sprint Test (MPST), which has been validated for use in children and adolescents with and without a pathological gait pattern²⁹⁻³². Subjects had to complete six 15-m runs at a maximum pace. One sprint was performed by the participant for practice, before the actual test started. The MPST is an intermittent sprint test, in which the child stops and starts at standardized intervals. Power was calculated $((\text{weight} \times \text{distance})^2 / \text{time}^3)$ for each of the six sprints and mean power was defined as the average power over the six sprints and used for further analysis.

Aerobic fitness was determined using an incremental exercise test. In exercise testing, peak oxygen uptake ($\text{VO}_{2\text{peak}}$) is considered to be the single best indicator of cardiorespiratory fitness, or aerobic fitness. A cardiopulmonary exercise testing system, the Cortex Metamax 3X (Samcon bvba, Melle, Belgium) was used for evaluation of the respiratory gasses and $\text{VO}_{2\text{peak}}$. Aerobic fitness was assessed by an adapted 10-meter incremental shuttle run test³² or by an incremental exercise test on an electronically braked cycle ergometer (Ergoline, Ergoselect 200 K, Ergoline, Bitz, Germany). In persons with disability the main mode of locomotion / mobility elicits the highest $\text{VO}_{2\text{peak}}$ ³³. Therefore the type of sports or daily locomotion determined whether the shuttle run test or a cycling ergometry test was used. In children with a congenital cardiopulmonary disease a cycling test was always used, because of the electrocardiography monitoring of the heart for safety issues.

The speed of the shuttle run test was adjusted based on the results of the muscle power

sprint test and the agility test. The cycling test, using the Godfrey protocol³⁴, was used to test the aerobic fitness in children who are active on a bike in sports or daily living. Load depended on height of the child and the expected level of fitness. Regardless of the testing modality, the test started with a resting measurement for 3 min. Participants were verbally encouraged to keep on exercising until voluntary exhaustion. Exercise tests were considered maximal and included for analysis if two out of three of the following criteria were achieved: 1) peak heart rate >180 beats/min, 2) peak Respiratory Exchange Ratio >1.0 and or 3) subjective signs of exhaustions (out of breath, sweating, or plateau of VO_2) or unable to continue the test³⁵. Cardiorespiratory fitness was defined as $\text{VO}_{2\text{peak}}$ per body weight, $\text{VO}_{2\text{peak}}$ in liter per minute and to control for differences in age, Z-scores of $\text{VO}_{2\text{peak}}$ were calculated according to Dutch reference values³⁶.

Physical activity PA was measured using an activity monitor, the Activ8 (2M Engineering BV, Valkenswaard, the Netherlands). The Activ8 is a valid one-sensor ambulatory monitoring system and has been validated for use in youth with and without motor impairments³⁷. Each subject wore the sensor on the dominant leg, fixed with Tegaderm™ (3M, Delft, the Netherlands) waterproof skin tape during seven consecutive days for 24 hours each day. In order to calculate and interpret the data of waking hours gathered with the activity monitor, sleeping time was recorded in a diary. A minimum of one weekend day and two schooldays with a minimum of 600 minutes per day was required for further analysis³⁸. The time in minutes per day were calculated for each activity (lying, sitting, standing, walking, bicycling and running) during awake hours and calculated for school and weekend days separately. The physical activity data were divided into sedentary and active categories. Sedentary activity consisted of lying and sitting, whereas active activities entailed walking, bicycling and running.

Power analysis The sample size of the HAYS-study was based on a study of Verschuren et al.³⁹. In this study children and adolescents with cerebral palsy had a $\text{VO}_{2\text{peak}}$ of 42 ml $\text{O}_2/\text{kg}/\text{min}$ ($\text{SD} \pm 8.2$). To prove a difference of 10% between the SG and NSG, with an alpha of 0.05 and beta of 0.20 (power of 0.80) a sample size of 66 subjects per group was required. When taking a failure rate of 10% into account, a minimum of 146 subjects was required in total.

Statistical analyses

All analyses were performed using the SPSS Statistics, Version 23.0 (IBM Corp., Armonk,

NY, USA). First, descriptive statistics was used to describe the outcomes in the two groups. Independent Student's t-tests, the Mann-Whitney U-test and chi-square test were used to determine the differences between the SG and NSG. Linear regression analyses were used to assess the relationship between sports participation (independent variable) with health-related fitness and PA (dependent variables). Assumptions regarding normality of residuals were checked and met the assumptions. The regression models were adjusted for potential confounders such as age, sex and diagnosis.

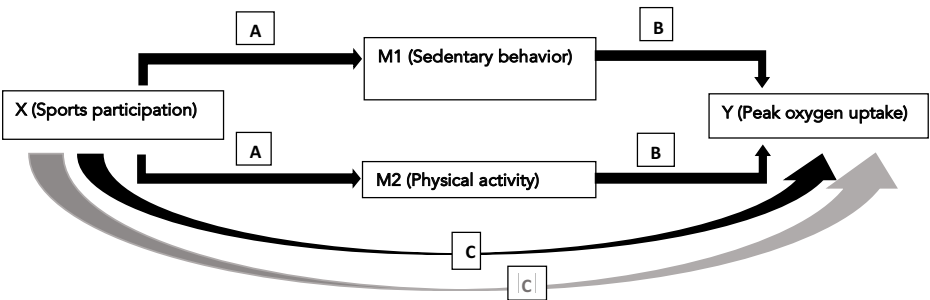


Figure 1: Mediation analysis.

To determine whether the association between sports participation and the primary outcome variable VO_{2peak} was mediated by PA, mediation regression analyses were conducted using the product-of-coefficient method, see figure 1 and Table 8⁴⁰. Firstly, the overall relationship between participation in sports and the outcome variable VO_{2peak} was estimated (the C coefficient). Secondly, the relationship between participation in sports and the potential mediator variables, the two categories; M1) sedentary and M2) active was estimated (the A coefficient). Thirdly, the relationship between the mediator variables and the outcome VO_{2peak} was estimated (the B coefficient). Finally, the effect of participation in sports on VO_{2peak} adjusted for the mediator was estimated (the C' coefficient or mediator adjusted effect). The proportion of the total effect of participation in sports on VO_{2peak} measure that was mediated by any of the potential mediators tested was also calculated (dividing the mediated effect by the total effect (A coefficient*B coefficient / C coefficient). Mediation was considered partial when the mediator adjusted association between participation in sports and the outcome (the C' coefficient) remained significant. The PROCESS module in SPSS was used to conduct the mediation analyses⁴¹. The mean time in minutes of the total week (school and weekend days) for the clustered categories

sedentary and active were calculated and used for this mediation regression analyses. For each coefficient estimated, 95% bootstrapped confidence intervals (CIs), P values and adjusted R square were calculated. A P value < 0.05 was considered statistically significant.

RESULTS

Participants' characteristics

A total of 163 ambulatory participants with CDPD were included for this study between September 2014 and October 2016 (96 boys (mean age 14.3 years, SD±2.8) and 67 girls (mean age 14.2 years, SD±3.0). Descriptive statistics of the sample are presented in Table 3.

Table 3: Characteristics of the non-sport (NSG) and sport group (SG).

	Total N = 163	Non-sport group (SD) N = 82	Sport group (SD) N = 81	P-value
Boys / Girls ^a	96 / 67	38 / 44	58 / 23	0.001*
Age in years ^b		14.31 (3.1)	14.15 (2.7)	0.725
Diagnose group in % ^a				0.532
Cardiovascular disease	12.9	17.1	8.6	
Pulmonary disease	4.9	3.7	6.2	
Metabolic disease	9.8	8.5	11.1	
Musculoskeletal / orthopedic disability	6.1	4.9	7.4	
Neuromuscular disorder	49.1	51.2	46.9	
Immunological / hematological disease	10.4	11.0	9.9	
Cancer	1.8	1.2	2.5	
Epilepsy	4.9	2.4	7.4	

Age is presented as mean and standard deviations (SD), diagnose group is presented as percentages and the P-values from the t-test for independent samples for continuous variables with normal distribution. ^a = Chi-square test for sex and prevalence of diseases or disabilities, ^b = Mann Whitney-U test for continuous variables with skewed distribution. *significant difference. Diagnose group classifications according to the guidelines of the American College of Sports Medicine (24).

Health-related fitness

Youth who participated in sports had a significant higher VO_{2peak} , a higher power on the MPST, were faster on the 10x5 meter sprint test, had a higher mean grip strength and were able to jump horizontally further compared with the NSG (Table 2). In addition, the participants in the SG had a significant lower waist / hip circumference ratio SDS and lower percentage of fat mass compared with the NSG (Table 4). There were no significant differences between participants in the SG and NSG for blood pressure (systolic and diastolic), resting heart rate and the arterial stiffness (PWV and AIX) (Table 4). The number of participants who performed an incremental shuttle run test or an incremental exercise test on a cycle ergometer were equally distributed between the NSG and SG.

Physical Activity

Youth participating in sports were more active than those who did not during both school and weekend days. The mean total minutes of bicycling and running were significantly higher for the SG compared with the NSG (Table 5). In addition, the total active time (walking, bicycling and running) was significantly higher in the SG compared with the NSG. Sedentary time (lying and sitting) did not significantly differ between the groups during both the weekend and school days (Table 5).

Table 4: Health-related fitness in relation to outcome variables of the non-sport (NSG) and sport group (SG).

Metabolic variables N = 162	NSG (SD) N = 81	SG (SD) N = 81	P-value
Body mass index (kg/m^2)	21.28 (4.82)	20.33 (3.72)	0.159
Body mass index (kg/m^2) SDS	0.75 (1.55)	0.61 (1.23)	0.537
Waist circumference (cm)	77.16 (14.4)	74.4 (12.2)	0.195
Waist circumference SDS	1.05 (1.4)	0.72 (1.26)	0.123
Hip circumference (cm)	87.6 (12.5)	86.9 (11.2)	0.696
Hip circumference SDS	0.40 (1.34)	0.41 (1.11)	0.937
Waist / hip ratio	0.88 (0.10)	0.86 (0.08)	0.087
Waist / hip ratio SDS	1.17 (1.38)	0.63 (1.40)	0.013 *
Fat mass (%)	25.4 (9.2)	20.8 (8.1)	0.001 *

Aerobic fitness variables N = 151[^]	NSG (SD) N = 74	SG (SD) N = 77	P-value
Peak oxygen uptake (L/min)	2.16 (0.7)	2.48 (0.9)	0.019 *
Peak oxygen uptake (L/min) SDS	-0.91 (1.3)	-0.29 (1.3)	0.005 ^a *
Peak oxygen uptake (ml/kg/min)	39.2 (7.7)	45.6 (9.6)	0.000 ^a *
Peak oxygen uptake (ml/kg/min) SDS	-0.97 (1.3)	-0.21 (1.5)	0.001 ^a *
Maximal heart rate	191 (10.6) n=72	192 (12.5)	0.723
RER	1.10 (0.07)	1.10 (0.10)	0.943
Cardio-vascular variables N = 160	NSG (SD) N = 79	SG (SD) N = 79	P-value
Systolic blood pressure (mmHg)	124.5 (14.0)	126.8 (16.6)	0.348
Diastolic blood pressure (mmHg)	69.1 (10.1)	67.7 (10.3)	0.392
AIX (%)	9.5 (8.3) n = 76	8.3 (6.8) n = 76	0.331
PWV (m/s)	6.1 (0.9) n = 76	5.8 (0.9) n = 76	0.119
Resting heart frequency	71 (11.0) n = 75	69 (12.3) n = 81	0.334
Motor variables N = 160	NSG (SD) N = 82	SG (SD) N = 78	P-value
MPST – mean power (Watt)	230.9 (145.6)	301.7 (180.7)	0.007 ^b *
10x5 (sec)	24.2 (4.7)	22.0 (4.6) n = 77	0.003 *
Mean grip strength (Newton)	212.5 (114.4)	267.9 (127.1) n = 77	0.004 *
Mean standing broad jump (cm)	103.5 (39.8) n = 81	124.9 (41.5) n = 78	0.001 *

Outcome variables are presented as mean or standard deviation scores (SDS; Z-score) and standard deviations (SD) with the P-values from the students t-test for independent samples for continuous variables with normal distribution or a Mann-Whitney U test for non-normal distributed data ^a. ^b equal variances not assumed, *significant difference. NSG; non-sport, SG; sport group, RER; respiratory exchange ratio, AIX; augmentation index, PWV; pulse wave velocity, MPST; muscle power sprint test. [^] Drop-out of seven participants during cardio-pulmonary exercise test, reasons; four participants stopped earlier due to musculoskeletal pain, and three did not wanted to do the test on beforehand. Of five participants data was inaccurate due to device problems.

Table 5: Objective measured physical activity during school days and weekend-days in the non-sport group (NSG) and sport group (SG).

Physical activity during school days in minutes per day; N = 126	NSG (SD) N = 65	SG (SD) N = 61	P-value
Sedentary time (lying – sitting)	588.0 (89.2)	581.0 (71.1)	0.627
- Lying	4.3 (6.8)	6.1 (11.7)	0.301
- Sitting	583.7 (88.7)	574.9 (72.8)	0.546
Standing	115.7 (43.8)	119.2 (42.0)	0.651
Active time (walking – cycling - running)	155.9 (50.0)	186.8 (44.0)	0.000 *
- Walking	128.0 (42.3)	141.7 (43.4)	0.074
- Cycling	24.8 (24.5)	37.5 (25.0)	0.005 *
- Running	3.1 (3.2)	7.5 (5.3)	0.000 ^a *
Physical activity during weekend days in minutes per day; N = 116	NSG (SD) N = 59	SG (SD) N = 57	P-value
Sedentary time (lying – sitting)	543.2 (100.8)	561.8 (102.7)	0.327
- Lying	6.0 (9.4)	11.6 (20.9)	0.060 ^a
- Sitting	537.2 (100.3)	550.1 (101.2)	0.490
Standing	119.2 (57.5)	123.2 (47.3)	0.681
Active time (walking – cycling - running)	134.3 (62.9)	159.0 (62.0)	0.035*
- Walking	115.4 (57.1)	123.6 (53.3)	0.424
- Cycling	17.3 (14.6)	28.4 (24.1)	0.004 ^a *
- Running	1.6 (2.0)	7.0 (8.2)	0.000 ^a *

*Outcome variables are presented as mean and standard deviations (SD) with the P-values from the students t-test for independent samples for continuous variables with normal distribution. Criteria for analysis; 1) > 600 minutes awake time per day 2) at least one measured weekend-day and at least two school days. ^a equal variances not assumed, *statistical difference, NSG; non-sport group, SG; sport group.*

Sports participation associated with health-related fitness

Sports participants showed a higher $\text{VO}_{2\text{peak}}$. We found significant positive associations for both boys and girls between participation in sports and $\text{VO}_{2\text{peak}}$. Boys had a higher $\text{VO}_{2\text{peak}}$ compared with girls. The effect of sports participation was independent of the medical diagnosis (Table 6).

The mean power on the MPST, the time on the 10x5 meter sprint test, the mean grip strength and the mean standing broad jump were all positively associated with sports participation (Table 6). Overall, participation in sports, sex and age were associated with a better performance on all these outcome measurements. Participants with a pathological gait pattern scored lower on all outcomes (see motoric gait function, Table 6).

Youth participating in sports had a lower percentage of fat mass compared to peers not participating in sports. (Table 6). Youth with a pathological gait pattern had a significantly higher percentage of fat and higher waist/hip circumference SDS. Sports participation, sex and age was positively associated with the waist/hip circumference SDS.

Sports participation associated with physical activity

Sports participation was positively associated with the amount of active time during both school and weekend days (Table 7). Age was negatively associated with the amount of active time during weekend days. Participants with a pathological gait pattern spent a lower number of minutes in active time during school days compared with peers without a pathological gait pattern.

Sports participation was positively associated with both bicycling and running during school days. The motoric gait function was negatively associated with bicycling, while sex and age was negatively associated with running during school days. In addition, sports participation was positively associated with bicycling during weekend days, while age was negatively associated with bicycling. Youth in the SG ran more in the weekend compared to peers from the NSG.

Mediation regression analysis

PA mediated the association between sports participation and $\text{VO}_{2\text{peak}}$ (Table 8). The mediating effect of PA on $\text{VO}_{2\text{peak}}$ was 0.033 ml O_2 /kg/min. For every additional minute of PA, $\text{VO}_{2\text{peak}}$ increased with 0.033 ml/kg/min. The proportion mediated was 31%. Sedentary time did not mediate the association between sports participation and $\text{VO}_{2\text{peak}}$.

Table 6: Associations of sports participation with health-related fitness in youth with chronic diseases or physical disabilities.

Health-related fitness	B (SD)	95% CI	P-value	Adjusted R Square
Peak oxygen uptake (ml/kg/min); N = 151				
Constant	42.438 (1.215)	40.036 to 44.840	0.000*	
Sports participation	4.891 (1.389)	2.145 to 7.636	0.001*	
Gender	-6.242 (1.419)	-9.046 to -3.437	0.000*	0.213
Peak oxygen uptake (L/min); N = 151				
Constant	-0.125 (0.252)	-6.23 to 3.74	0.622	
Sports participation	0.248 (0.094)	0.062 to 0.433	0.009*	
Gender	-0.667 (0.095)	-0.856 to -0.478	0.000*	
Age in years	0.180 (0.016)	-0.148 to 0.212	0.000*	0.559
MPST – mean power (W); N = 160				
Constant	-175.516 (46.409)	-267.191 to -83.841	0.000*	
Sports participation	41.203 (17.872)	5.899 to 76.508	0.022*	
Gender	-124.794 (18.104)	-160.556 to -89.032	0.000*	
Age in years	35.245 (2.983)	29.355 to 41.135	0.000*	
Motoric gait function	-60.422 (17.263)	-94.523 to -26.322	0.001*	0.575
10x5 meter sprint test (sec); N = 159				
Constant	31.924 (1.602)	28.759 to 35.089	0.000*	
Sports participation	-1.718 (0.617)	28.759 to 35.099	0.006*	
Gender	1.719 (0.626)	0.483 to 2.956	0.007*	
Age in years	-0.724 (0.103)	-0.927 to -0.521	0.000*	
Motoric gait function	3.424 (0.594)	2.250 to 4.597	0.000*	0.389

Mean grip strength (Newton); N = 159

Constant	-90.542 (35.263)	-160.203 to -20.880	0.011*	
Sports participation	40.384 (13.584)	13.549 to 67.219	0.003*	
Gender	-59.404 (13.751)	-86.569 to -32.240	0.000*	
Age in years	25.965 (2.263)	21.493 to 30.436	0.000*	
Motoric gait function	-71.497 (13.131)	-97.436 to -45.557	0.000*	0.553

Mean standing broad jump (cm); N = 157

Constant	44.308 (12.477)	19.657 to 68.960	0.001*	
Sports participation	17.447 (4.801)	7.962 to 26.933	0.000*	
Gender	-23.056 (4.857)	-32.652 to -13.459	0.000*	
Age in years	6.224 (0.805)	4.634 to 7.814	0.000*	
Motoric gait function	-35.073 (4.652)	-44.264 to -25.882	0.000*	0.504

Fat mass (%)

Constant	25.740 (3.369)	19.086 to 32.394	0.000*	
Sports participation	-3.303 (1.285)	-5.840 to -0.765	0.011*	
Gender	4.583 (1.303)	2.009 to 7.157	0.001*	
Age in years	-0.395 (0.216)	-0.821 to 0.031	0.069*	
Motoric gait function	5.777 (1.239)	3.131 to 8.024	0.000*	0.223

Waist / hip ratio SDS

Constant	0.968 (0.035)	0.900 to 1.036	0.000*	
Sports participation	-0.033 (0.013)	-0.059 to -0.007	0.014*	
Gender	-0.037 (0.013)	-0.064 to -0.011	0.006*	
Age in years	-0.007 (0.002)	-0.011 to -0.002	0.003*	
Motoric gait function	0.056 (0.013)	0.031 to 0.081	0.000*	0.178

The data are standardized regression coefficients (B) with standard deviation (SD) and their 95% confidence intervals (CI), P-values and explained variance (adjusted R-square). Outcome variables are adjusted for sports participation, sex, age and motoric gait function. MPST; muscle power sprint test, SDS; standard deviation scores (Z-score).

Table 7: Associations of sports participation with physical activity in youth with chronic diseases or physical disabilities.

Mean PA during school days in minutes per day; N = 126	B (SD)	95% CI	P-value	Adjusted R Square
Cycling				
Constant	30.491 (3.907)	22.758 to 38.225	0.000*	
Sports participation	11.956 (4.351)	3.343 to 20.569	0.007*	
Motoric gait function	-9.965 (4.358)	-18.590 to -1.339	0.024*	0.087
Running				
Constant	11.210 (1.871)	7.506 to 14.914	0.000*	
Sports participation	4.187 (0.744)	2.715 to 5.659	0.000*	
Gender	-1.724 (0.752)	-3.212 to -0.245	0.024*	
Age in years	-0.511 (0.125)	-0.759 to -0.264	0.000*	0.315
Active time (walking – cycling - running)				
Constant	167.933 (7.418)	153.250 to 182.616	0.000*	
Sports participation	29.321 (8.261)	12.968 to 45.674	0.001*	
Motoric gait function	-21.228 (8.274)	-37.605 to -4.850	0.012*	0.130
Mean PA during weekend in minutes per day N = 116	B (SD)	95% CI	P-value	Adjusted R Square
Cycling				
Constant	37.658 (9.628)	18.583 to 56.733	0.000*	
Sports participation	11.368 (3.627)	4.182 to 18.555	0.002*	
Age years	-1.429 (0.653)	-2.722 to -0.137	0.031*	0.095
Running				
Constant	1.563 (0.770)	0.038 to 3.089	0.045*	
Sports participation	5.452 (1.099)	3.275 to 7.628	0.000*	0.170
Active time (walking – cycling - running)				
Constant	246.000 (28.966)	168.613 to 303.386	0.000*	
Sports participation	26.488 (10.913)	4.868 to 48.108	0.017*	
Age in years	-7.851 (1.963)	-11.731 to -3.961	0.000*	

The data are standardized regression coefficients (B) with standard deviation (SD) and their 95% confidence intervals (CI), P-values and explained variance (adjusted R-square). Outcome variables are adjusted for sports participation, sex, age and motoric gait function.

Table 8: Mediation analysis; association between sports participation (yes/no) and $VO_{2\text{ peak}}$ via the mediator variables of sedentary behavior and physical activity (N = 106). *†‡

Mediator	Measure	A coefficient (SE)	P-value	B coefficient (SE)	P-value	C coefficient (SE)	P-value	C' coefficient (SE)	P-value	Mediated effect (a*b) (95% CI)	Proportion mediated
M1 = Sedentary behavior min/week	$VO_{2\text{ peak}}$	10.3 (30.0)	0.732	0.008 (0.01)	0.203					0.09 (-0.34; 1.13)	0.02
M2 = Physical activity min/week	$VO_{2\text{ peak}}$	46.8 (17.68)	0.009	0.033 (0.01)	0.003	5.044 (1.65)	0.003	3.406 (1.69)	0.045	1.55 (0.33; 3.65)	0.31

* $VO_{2\text{ peak}}$; peak oxygen uptake, CI; bootstrapped confidence interval.

† A coefficient refers to the effect of sports participation on the mediator variable (total minutes of sedentary behavior or physical activity during one week).

B coefficient; effect of the mediator variable on the outcome variable ($VO_{2\text{ peak}}$), adjusted for sports participation (X).

C coefficient; effect of sports participation on the outcome variable ($VO_{2\text{ peak}}$).

C' coefficient; effect of sports participation on the outcome ($VO_{2\text{ peak}}$), adjusted for the mediator variables.

Mediated effect refers to indirect effect of X on Y through M, calculated as the product-of-coefficients (a x b)

‡ All models were adjusted for sex, age and motoric gait function.

DISCUSSION

The results from this study demonstrate that sports participation was strongly associated with better health-related fitness and increased PA level in children and adolescents with CDPD. Youth participating at least twice weekly in organized sports performed better on all outcome measures. Our results also suggest that the association between sports participation and aerobic fitness was mediated for 31% by an increased PA level. We did not observe any significant association between sports participation and sedentary time. We found significant positive associations for both boys and girls between participation in sports and VO_{2peak} . These findings are in line with current literature. Earlier studies have shown an association between fitness and sports and exercise in both girls and boys^{42,43}. We found levels of aerobic fitness comparable to values observed in healthy youth (mean values 41.5 ml/kg/min and 46-49 ml/kg/min for females and males respectively)³⁶. Previous studies have reported lower aerobic fitness in youth with a neuromuscular disorder^{44,45}. A large proportion (49.1%) in our study population consisted of participants with neuromuscular disorders. Despite this condition, sports participants with neuromuscular disorders in our study show that they can achieve VO_{2peak} values within the normal range for healthy Dutch peers. From a health perspective is this a remarkable and promising finding, because it shows that sports seems effective in optimizing aerobic fitness and this is currently not achieved in the longer term with rehabilitation programs in youth with CDPD^{46,47}.

Youth participating in sports also performed significantly better on outcomes of anaerobic fitness, agility and strength measurements. Other cross-sectional studies have reported similar results with in healthy peers, where those participation in recreational sports scored higher in anaerobic fitness and muscular strength than youth those who did not⁴⁸. Short bursts of high intensity sprints are required in youth sports, which may have resulted in a higher anaerobic fitness than their non-sporting peers. In contrast to aerobic fitness, the medical diagnosis did influence the outcomes of anaerobic fitness, agility and muscle strength in our study. Participants with a neuromuscular disorder like Cerebral Palsy or Spina Bifida scored lower on all anaerobic fitness and performance measures compared with those without a neuromuscular disorder. These results are in line with existing evidence which shows that a motor impairment affect anaerobic fitness negatively and the motor impairment influenced anaerobic fitness more strongly compared to aerobic fitness^{45,49}. A muscle impairment results in a reduced muscle strength and coordination in muscles that have been affected due to the neuromuscular

disorder. Due to lower level of coordination, youth with motor impairment often need more time to complete motor tasks. Tasks including stops, turns and accelerations, as part of the agility measures will therefore take more time in youth with motor impairment compared to those without. While youth with a neuromuscular disorder perform less well because of their medical condition, our study showed that participation in sports can significantly contribute to better muscle function and better performance on the anaerobic fitness and performance tests compared to their non-sporting peers with a neuromuscular disorder.

In our study, youth with CDPD who participated in sports were more active during both school and weekend days. At the same time, this higher physical activity level explained 31% of the association between sports participation and VO_{2peak} . A total of 47 minutes more active time per week resulted in a higher VO_{2peak} of 1.55 ml/kg/min. In contrast with the current finding, studies in healthy youth have shown only small correlations between VO_{2peak} and MVPA^{50,51}. Youth with CDPD are less active compared with their healthy peers, but do achieve positive effects on health-related fitness when they become active.

This study was the first large study to evaluate the associations between sports and health-related fitness and PA. Strong points are the objective direct measurement of both VO_{2peak} and accelerometry-based PA. Obviously, this study also has some limitations. Firstly, the cross-sectional design limits the ability to establish causality and direction. At the same time establishing associations is an important first step for further research. Secondly, we only included ambulatory youth with CDPD and therefore our results are limited to ambulatory youth with CDPD. Moreover, there were more boys in the SG compared to the NSG, which we did correct for in the statistical modelling. Sex stratification is recommended for future studies as outcomes do differ among sex. Finally, using the standardized questionnaire on sports, we did not have information regarding the training history of the sports group (e.g. how many years they have been doing sports). We only recorded whether they have participated in organized sports at least twice weekly during the past 3 months. We would recommend future research to include information of the number of years of sports participation as well as the duration, type and characteristics of the sports activities, as it give more insight into the long-term effects of sports. Despite these limitations, this study still demonstrates positive associations of sports participation with PA and health-related fitness independent of age, sex and motoric gait function in ambulatory youth with CDPD.

Practical applications

While in healthy youth only a weak relationship has been demonstrated between PA and aerobic fitness⁵², physical activity was a strong predictor of aerobic fitness in youth with CDPD, as shown with the mediation regression analysis in our study. Our data shows that youth with CDPD who participate in organized sports at least twice weekly benefit from the positive effects of sports on health-related fitness and physical activity. Especially in children and adolescents with CP or SB with muscle weakness, which is a major problem, organized sports contributes to a better muscular function.

As described earlier, youth with CDPD are able to improve their PA level and health-related fitness through supervised intervention programs in rehabilitation settings^{1,16}, however achieved results are often not maintained in the longer term^{17,18}. Data of the current study suggest that participation in sports is an excellent way to improve or maintain health-related physical fitness in youth with CDPD.

Therefore, this study also shows it is important to focus on reducing barriers to sports participation for youth with CDPD, with special attention for girls⁵³ and adolescents. In this group the drop-out from sports is high¹¹, physical activity decreases and sedentary time increases⁵⁴. This will most likely involve a multidisciplinary approach, with professionals from the medical, social and educational domain working together with the youth with CDPD to overcome barriers and find the possibilities to participate in sports.

For professionals, the training guidelines developed for healthy youth⁵⁵ are mostly applicable for youth with CDPD, but some characteristics and physical possibilities for certain types of sports should be taken into account. Adapted guidelines for both exercise testing and training have been published to guide coaches and trainers of the young athlete with CDPD⁵⁶. In addition, it is advisable to use relevant registration and assessment tools to monitor training loads. All measurement instruments and performance tests used in this study are feasible, easy to use and proven valid and reliable in youth with CDPD. The measurement outcomes of all tests of the current study could be used as a reference, which makes it possible to put the test outcome of your young patient or sports participant with CDPD in perspective with their sporting and non-sporting peers.

In conclusion, participation in sports is associated with both higher levels of PA and health-related fitness in youth with CDPD. Promotion and stimulation of healthy active lifestyles including sports participation is therefore highly recommended in this special population.

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REFERENCES

1. van Brussel M, van der Net J, Hulzebos E, Helders PJ, Takken T. The Utrecht approach to exercise in chronic childhood conditions: the decade in review. *Pediatr Phys Ther* 2011 Spring;23(1):2-14.
2. Carlon SL, Taylor NF, Dodd KJ, Shields N. Differences in habitual physical activity levels of young people with cerebral palsy and their typically developing peers: a systematic review. *Disabil Rehabil* 2013 Apr;35(8):647-655.
3. Neter JE, Schokker DF, de Jong E, Renders CM, Seidell JC, Visscher TL. The prevalence of overweight and obesity and its determinants in children with and without disabilities. *J Pediatr* 2011 May;158(5):735-739.
4. Kotte EM, Winkler AM, Takken T. Fitkids exercise therapy program in the Netherlands. *Pediatr Phys Ther* 2013 Spring;25(1):7-13.
5. Dencker M, Andersen LB. Health-related aspects of objectively measured daily physical activity in children. *Clin Physiol Funct Imaging* 2008 May;28(3):133-144.
6. Jimenez-Pavon D, Kelly J, Reilly JJ. Associations between objectively measured habitual physical activity and adiposity in children and adolescents: Systematic review. *Int J Pediatr Obes* 2010;5(1):3-18.
7. LaMonte MJ, Blair SN. Physical activity, cardiorespiratory fitness, and adiposity: contributions to disease risk. *Curr Opin Clin Nutr Metab Care* 2006 Sep;9(5):540-546.
8. Janssen I, Leblanc AG. Systematic review of the health benefits of physical activity and fitness in school-aged children and youth. *Int J Behav Nutr Phys Act* 2010 May 11;7:40-5868-7-40.
9. Riddoch CJ, Mattocks C, Deere K, Saunders J, Kirkby J, Tilling K, et al. Objective measurement of levels and patterns of physical activity. *Arch Dis Child* 2007 Nov;92(11):963-969.
10. Kjonniksen L, Anderssen N, Wold B. Organized youth sport as a predictor of physical activity in adulthood. *Scand J Med Sci Sports* 2009 Oct;19(5):646-654.
11. Vella SA, Schranz NK, Davern M, Hardy LL, Hills AP, Morgan PJ, et al. The contribution

- of organised sports to physical activity in Australia: Results and directions from the Active Healthy Kids Australia 2014 Report Card on physical activity for children and young people. *J Sci Med Sport* 2016 May;19(5):407-412.
12. Telford RM, Telford RD, Cochrane T, Cunningham RB, Olive LS, Davey R. The influence of sport club participation on physical activity, fitness and body fat during childhood and adolescence: The LOOK Longitudinal Study. *J Sci Med Sport* 2016 May;19(5):400-406.
 13. Burghard M, Knitel K, van Oost I, Tremblay MS, Takken T. Dutch physical activity report card study group. Is our youth cycling to health? Results from The Netherlands'2016 report card on physical activity for children and youth. *J Phys Act Health* 13:S218-S224, 2016.
 14. Burghard M, de Jong NB, Vlieger S, Takken T. 2017 Dutch Report Card(+): Results From the First Physical Activity Report Card Plus for Dutch Youth With a Chronic Disease or Disability. *Front Pediatr* 2018 Apr 30;6:122.
 15. Haapala EA, Lankhorst K, de Groot J, Zwinkels M, Verschuren O, Wittink H, et al. 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 Jul;24(10):1102-1111.
 16. Edouard P, Gautheron V, D'Anjou MC, Pupier L, Devillard X. Training programs for children: literature review. *Ann Readapt Med Phys* 2007 Jul;50(6):510-9, 499-509.
 17. de Groot JF, Takken T, van Brussel M, Gooskens R, Schoenmakers M, Versteeg C, et al. Randomized controlled study of home-based treadmill training for ambulatory children with spina bifida. *Neurorehabil Neural Repair* 2011 Sep;25(7):597-606.
 18. Verschuren O, Ketelaar M, Gorter JW, Helders PJ, Uiterwaal CS, Takken T. Exercise training program in children and adolescents with cerebral palsy: a randomized controlled trial. *Arch Pediatr Adolesc Med* 2007 Nov;161(11):1075-1081.
 19. Jaarsma EA, Dijkstra PU, de Blecourt AC, Geertzen JH, Dekker R. Barriers and facilitators of sports in children with physical disabilities: a mixed-method study. *Disabil Rehabil* 2015;37(18):1617-23; quiz 1624-5.
 20. Verschuren O, Wiaart L, Hermans D, Ketelaar M. Identification of facilitators and barriers to physical activity in children and adolescents with cerebral palsy. *J Pediatr* 2012 Sep;161(3):488-494.
 21. Te Velde SJ, Lankhorst K, Zwinkels M, Verschuren O, Takken T, de Groot J, et al. Associations of sport participation with self-perception, exercise self-efficacy and quality of life among children and adolescents with a physical disability or chronic disease-a cross-sectional study. *Sports Med Open* 2018 Aug 15;4(1):38-018-0152-1.

22. Lankhorst K, van der Ende-Kastelijin K, de Groot J, Zwinkels M, Verschuren O, Backx F, et al. 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 Dec 22;4:796-015-1589-z. eCollection 2015.
23. Talma H. Growth charts 2010: A manual to measure and weigh children and completing growth charts, Delft: TNO. 2010.
24. Winnick, J.P. & Short, F.X. editor. The Brockport Physical Fitness Test Manual. : Campaign, IL: Human Kinetics; 1999.
25. Beenakker KG, Ling CH, Meskers CG, de Craen AJ, Stijnen T, Westendorp RG, et al. Patterns of muscle strength loss with age in the general population and patients with a chronic inflammatory state. Ageing Res Rev 2010 Oct;9(4):431-436.
26. Deitz JC, Kartin D, Kopp K. Review of the Bruininks-Oseretsky Test of Motor Proficiency, Second Edition (BOT-2). Phys Occup Ther Pediatr 2007;27(4):87-102.
27. Verschuren O, Takken T, Ketelaar M, Gorter JW, Helders PJ. Reliability for running tests for measuring agility and anaerobic muscle power in children and adolescents with cerebral palsy. Pediatr Phys Ther 2007 Summer;19(2):108-115.
28. Vrijkotte S, de Vries S, Jongert T. Fitheidstesten voor de jeugd. Leiden;TNO Kwaliteit van Leven. 2007;031.10038.
29. Douma-van Riet D, Verschuren O, Jelsma D, Kruitwagen C, Smits-Engelsman B, Takken T. Reference values for the muscle power sprint test in 6- to 12-year-old children. Pediatr Phys Ther 2012 Winter;24(4):327-332.
30. Steenman K, Verschuren O, Rameckers E, Douma-van Riet D, Takken T. Extended Reference Values for the Muscle Power Sprint Test in 6- to 18-Year-Old Children. Pediatr Phys Ther 2016 Spring;28(1):78-84.
31. Verschuren O, Bongers BC, Obeid J, Ruyten T, Takken T. Validity of the muscle power sprint test in ambulatory youth with cerebral palsy. Pediatr Phys Ther 2013 Spring;25(1):25-28.
32. Verschuren O, Takken T, Ketelaar M, Gorter JW, Helders PJ. Reliability and validity of data for 2 newly developed shuttle run tests in children with cerebral palsy. Phys Ther 2006 Aug;86(8):1107-1117.
33. Bhambhani YN, Holland LJ, Steadward RD. Maximal aerobic power in cerebral palsied wheelchair athletes: validity and reliability. Arch Phys Med Rehabil 1992 Mar;73(3):246-252.
34. Godfrey S, Davies CT, Wozniak E, Barnes CA. Cardio-respiratory response to exercise in normal children. Clin Sci 1971 May;40(5):419-431.
35. Verschuren O, Maltais DB, Takken T. The 220-age equation does not predict maximum

- heart rate in children and adolescents. *Dev Med Child Neurol* 2011 Sep;53(9):861-864.
36. Bongers B, Hulzebos E, van Brussel M, Takken T. *Pediatric Norms for Cardiopulmonary Exercise Testing*. 2nd ed. Den Bosch: Boxpress; 2015.
37. Lankhorst K, Berg Emons van den R, Bussmann J, Horemans H, de Groot JF. A novel tool for quantifying and promoting physical activity in youths with typical development and youths who are ambulatory and have motor disability. *Phys Ther* 99:1-10, 2019.
38. Montoye AH, Moore RW, Bowles HR, Korycinski R, Pfeiffer KA. Reporting accelerometer methods in physical activity intervention studies: a systematic review and recommendations for authors. *Br J Sports Med* 2016.
39. Verschuren O, Takken T. Aerobic capacity in children and adolescents with cerebral palsy. *Res Dev Disabil* 2010 Nov-Dec;31(6):1352-1357.
40. Hayes AF, Preacher KJ. Statistical mediation analysis with a multicategorical independent variable. *Br J Math Stat Psychol* 2014 Nov;67(3):451-470.
41. Hayes AF. The Process macro for SPSS and SAS. 2016; Available at: <http://afhayes.com/index.html>.
42. Armstrong N, Tomkinson G, Ekelund U. Aerobic fitness and its relationship to sport, exercise training and habitual physical activity during youth. *Br J Sports Med* 2011 Sep;45(11):849-858.
43. Armstong N, McNarry M. Aerobic Fitness and Trainability in Healthy Youth: Gaps in Our Knowledge. *Pediatr Exerc Sci* 2016 May;28(2):171-177.
44. Verschuren O, Ketelaar M, Gorter JW, Helders PJ, Takken T. Relation between physical fitness and gross motor capacity in children and adolescents with cerebral palsy. *Dev Med Child Neurol* 2009 Nov;51(11):866-871.
45. Schoenmakers MA, de Groot JF, Gorter JW, Hillaert JL, Helders PJ, Takken T. Muscle strength, aerobic capacity and physical activity in independent ambulating children with lumbosacral spina bifida. *Disabil Rehabil* 2009;31(4):259-266.
46. Zwinkels M, Takken T, Visser-Meiley JMA, Verschuren O. Effects of high-intensity interval training on fitness and health in youth with physical disabilities. *Pediatr Phys Ther* 31:84-93, 2019.
47. Bloemen M, Van Wely L, Mollema J, Dallmeijer A, de Groot J. Evidence for increasing physical activity in children with physical disabilities: a systematic review. *Dev Med Child Neurol* 2017 Oct;59(10):1004-1010.
48. Hoffman JR, Kang J, Faigenbaum AD, Ratamess NA. Recreational sports participation is associated with enhanced physical fitness in children. *Res Sports Med* 2005 Apr-Jun;13(2):149-161.

49. Verschuren O, Maltais DB, Douma-van Riet D, Kruitwagen C, Ketelaar M. Anaerobic performance in children with cerebral palsy compared to children with typical development. *Pediatr Phys Ther* 2013 Winter;25(4):409-413.
50. Armstrong N. Aerobic fitness and physical activity in children. *Pediatr Exerc Sci* 2013 Nov;25(4):548-560.
51. Poitras VJ, Gray CE, Borghese MM, Carson V, Chaput JP, Janssen I, et al. Systematic review of the relationships between objectively measured physical activity and health indicators in school-aged children and youth. *Appl Physiol Nutr Metab* 2016 Jun;41(6 Suppl 3):S197-239.
52. McManus AM, Armstrong N. Maximal oxygen uptake. . In: Rowland TW, editor. *Cardiopulmonary Exercise Testing in Children and Adolescents*: Champaign IL, Human Kinetics.; 2018.
53. Azevedo MR, Araujo CL, Reichert FF, Siqueira FV, da Silva MC, Hallal PC. Gender differences in leisure-time physical activity. *Int J Public Health* 2007;52(1):8-15.
54. Ortega FB, Konstabel K, Pasquali E, Ruiz JR, Hurtig-Wennlof A, Maestu J, et al. Objectively measured physical activity and sedentary time during childhood, adolescence and young adulthood: a cohort study. *PLoS One* 2013 Apr 23;8(4):e60871.
55. Lloyd RS, Cronin JB, Faigenbaum AD, Haff GG, Howard R, Kraemer WJ, et al. National Strength and Conditioning Association Position Statement on Long-Term Athletic Development. *J Strength Cond Res* 2016 Jun;30(6):1491-1509.
56. Durstine LJ, Moore GE, Painter P, Roberts S,O. *ACSM's Exercise Management for Persons with Chronic Diseases and Disabilities*. 3rd ed.: Human Kinetics; Champaign, IL, 2009.



5

Associations of sport participation with self-perception, exercise self-efficacy and quality of life among children and adolescents with a physical disability or chronic disease: a cross-sectional study

Saskia te Velde

Kristel Lankhorst

Maremka Zwinkels

Olaf Verschuren

Tim Takken

Janke de Groot

ABSTRACT

Background. Little evidence is available about how sports participation influences psychosocial health and quality of life in children and adolescents with a disability or chronic disease. Therefore, the aim of the current study is to assess the association of sports participation with psychosocial health and with quality of life, among children and adolescents with a disability.

Methods. In a cross-sectional study 195 children and adolescents with physical disabilities or chronic diseases (11% cardiovascular, 5% pulmonary, 8% metabolic, 8% musculoskeletal/-orthopaedic, 52% neuromuscular and 9% immunological diseases and 1% with cancer), aged 10-19 years, completed questionnaires to assess sports participation, health related quality of life (DCGM-37), self-perceptions and global self-worth (SPPC or SPPA) and exercise self-efficacy.

Results. Regression analyses showed that those who reported to participate in sports at least twice a week, had more beneficial scores on the various indicators compared to their peers who did not participate in sport or less than twice a week. Those participating in sports scored better on all scales of the DCGM-37 scale, on the scales for feelings of athletic competence and children but not adolescents participating in sports reported greater social acceptance. Finally, we found a strong association between sport participation and exercise self-efficacy.

Conclusion. This study provides the first indications that participating in sports is beneficial for psychosocial health among children and adolescents with a disability. However, more insight is needed in the direction of the relationships.

Key points

- Children and adolescents with a physical disability or chronic disease who are participating in sports scored better on all scales of the DCGM-37 scale reflecting a better health related quality of life
- Participating in sports was associated with feelings of athletic competence in children and adolescents with a disability or chronic disease. In addition, children, but not adolescents, participating in sports reported higher feelings of social acceptance.
- Sport participation among children and adolescents with a physical disability or chronic disease was strongly positively associated with exercise self-efficacy.

INTRODUCTION

Physical activity and sports participation have several health benefits for people of all ages¹. Engaging in sufficient levels of physical activity improves cardiopulmonary health, strength, flexibility, and endurance and has been related to reduced risks for cardiovascular diseases and specific cancers^{2,3}. Moreover, physical activity, especially sports, exercise and leisure time activities, has been related to reductions in mortality⁴. In addition, due to its social nature, sports participation provides opportunities for social interaction, companionship and may therefore have greater benefits for social and mental well-being than other domains of physical activity⁵⁻⁸. Furthermore, sports participation may enhance health-related quality of life in adults as well as in children and adolescents^{9,10}. Health-related quality of life is a broadly defined construct evaluating the health status from the person's perspective covering physical, emotional, mental, social and functional domains¹¹ and has been used in evaluations of sport and exercise interventions^{12,13}.

The benefits of sport and physical activity for psychosocial health, i.e. one's mental processes, self-reflections and interactions with others, are universal and not restricted to healthy adults and typically developing children and adolescents. In the contrary, children and adolescents with a chronic disease or physical disability (CDPD) may benefit even more¹⁴. Both children with a physical disability as well as children with a chronic disease may experience similar barriers for sport participation. They both have lower fitness levels, lower levels of physical activity and a higher prevalence of overweight and obesity¹⁵⁻¹⁷. Sport and physical activity does not only improve the physical functioning and physical independence in both groups, it may enhance inclusion and well-being. For example, a review about leisure time activities and quality of life among children and adolescents with neurological disabilities reported that participation in active leisure time activities was associated with better physical well-being, improved sense of self, emotional well-being and promote social well-being¹⁸. Unfortunately, children and adolescents with CDPD engage less often in physical activities and sports^{17,19,20}. Furthermore, these children and adolescents may experience low levels of self-worth and quality of life due to their physical limitations and body image concerns²¹⁻²⁴. Therefore, insight in the potential beneficial effects of physical activity and in particular sports participation, as a specific sub-domain of physical activity, are important for subsequent interventions promoting sports participation among children and adolescents with disabilities.

So far, only a few studies addressed the association of sports participation with psychosocial health and quality of life in children and adolescents in general or in children and adolescents with CDPD. Dinomais and colleagues²⁵ showed that young

people with disabilities who participated in competitive sports scored high on social functioning, which is in line with the review by Sahlin and Lexel⁹ who concluded that children and adolescents with a disability who engaged in sports reported similar scores on self-concept than non-disabled athletes. However, it remains unclear whether positive associations were the result of the physical activity itself, or of the social interaction and learning environment of the sports club. A study among 15-69 year-old French men and women showed that across all levels of physical activity, sports participation was positively associated with quality of life²⁶. In addition, the review by Eime and colleagues⁵ reported that club-based and team-based sports participation resulted in better psychosocial health outcomes than individual forms of sports.

The aforementioned studies used different indicators, each measuring different but related aspects of psychosocial health. Positive self-worth, self-perceptions, self-esteem, social support and self-efficacy are acknowledged indicators of psychosocial health and quality of life^{23,27}. Involvement in sports, and the interaction with others in this context, allows children and adolescents to develop their self-concept, especially related to the physical and social domains²⁸. A systematic review found that physical activity was strongly associated with perceived (athletic) competence²⁹. A meta-analysis supported the proposition that physical activity positively influences body image, and this effect was larger in adolescents compared to students, adults and older adults³⁰. In summary, participation in physical activity may have an impact on many aspects of psychosocial health, however the evidence available about how sports participation influences psychosocial health and quality of life in children and adolescents with a disability is still scarce. Therefore, the aim of the current study is to assess the association of sports participation with psychosocial health, specifically, self-perceptions, exercise self-efficacy and quality of life, among children and adolescents with a disability. Based on previous studies, we hypothesize that children and adolescents who participate in sport have more favourable scores on these indicators of psychosocial health and quality of life.

METHODS

Design and sample

Cross-sectional data from two related studies using identical outcomes were combined. Firstly, the cross sectional Health in Adapted Youth Sports (HAYS) study that includes children and adolescents aged 10-19 years old with a chronic disease or physical disability (CDPD)³¹. Secondly, the baseline data from the Sports-2-Stay Fit (S2SF) study which is a clinically controlled trial among children and adolescents aged 6-19 years with a CDPD³². For both studies, children and adolescents were recruited through patient organisations,

pediatric physical therapy practices, Wilhelmina Children's Hospital in Utrecht, De Hoogstraat Rehabilitation Center, Fitkids and schools (for special education) in the Netherlands.

Inclusion criteria were: having a physical disability or a chronic disease (cardiovascular, pulmonary, musculoskeletal or neuromuscular disorder), aged between 10 and 19 years (HAYS) or between 6 and 19 years (S2SF), ability to understand simple instructions, able to perform physical fitness tests. Children and adolescents were not eligible for participation in these studies if they had a progressive disease, used an electric wheelchair, participated in other (research) projects that may influence the results of the current studies, or did not sign the informed consent form.

For the current study, children and adolescents were included in the analyses if they had valid data on sports participation and valid data on at least one of the outcome variables (quality of life, self-efficacy, self-perceptions, general self-worth). Eight children and adolescents from the S2SF-study did not complete the online questionnaires during their first assessment, but did during the second assessment (8 weeks later). For that reason, we included data from these eight children or adolescents from their second assessment. We assumed that even though their scores may have been improved in that 8 week period, this will not affect the association between sports participation and these variables.

In total, 197 participants had complete data on sports participation, of whom two had no valid data on the outcome variables. Of the remaining 195 participants, 177 participants had complete data on all outcome variables, another 13 participants had valid data on two of the three outcome variables and 5 participants had valid data on only one outcome variable. The distribution of valid data on the outcome variables did not differ by sport participation status ($p=0.428$) or by diagnosis (ACSM category)($p=0.346$) or by gender ($p=0.393$). We included 195 children and adolescents who had complete data on sports participation and at least one outcome variable. Of those included children and adolescents, 145 participated in the HAYS study, while 50 participated in the S2SF study.

Procedures

The procedures and protocols for the HAYS and the S2SF study have been described elsewhere in more detail^{31,32}. Briefly, participants who agreed to participate and met the inclusion criteria were scheduled for an assessment at the lab (University of Applied Science, Utrecht). One week before this visit to the laboratory, the participants or their parents received a secured link to an online questionnaires assessing exercise self-efficacy and quality of life. These questionnaires took about 10 -15 minutes to complete. When the children visited the laboratory, they were first asked to complete the questionnaires

on self-perceptions and global self-worth in the presence of one of the researchers or research assistants, which took about 10 minutes. When they finished the questionnaires their physical fitness, cognition, and cardiovascular health was assessed, which took on average about two hours^{31,32}.

The studies were approved by the Medical Ethics Committee of the University Medical Center Utrecht, the Netherlands. (METC number: 14-332/c and 14-118/m). All participants and the parents of participants under 18 years of age provided their informed written consent. Studies were conducted in accordance with the Helsinki Declaration.

Measurements

Independent variable

Sports participation

Sports participation was assessed by means of a questionnaire that was completed before the start of the physical fitness tests. Sports participation was identified using three standardized questions used by the National Institute for Public Health and the Environment (RIVM)³³ 1) "do you participate in organized sports?" 2) "what is / are the type of organized sport(s)?" and, 3) "what is the frequency of participation in organized sports per week?". When participants indicated that they participate in organized sports at least 2 times per week, they were classified as 'participating in sports', all others were classified as 'non-sporting'. This cut-off was based on the Consensus statement for the Dutch Guidelines for Physical Activity for youth (< 18 years old)³⁴. This guideline was developed for typically developing children and was in place at the time of the start of the current research project. It advises that children and adolescents should be physically active for at least one hour per day at least a moderate intensity. In addition, children and adolescents should engage in activities that specifically address physical fitness at least two times a week. Sport activities are typically activities in which physical fitness, including strength, flexibility and coordination, are addressed. However, there are no universally accepted guidelines for children and adolescents with a chronic disease of physical disability and Van Brussel and colleagues³⁵ advise to use a training frequency of minimally 2 times per week. This is in line with recommendations for people with Cerebral Palsy that were based on a comprehensive literature review, expert opinions and extensive clinical experience³⁶.

Dependent variables

Health-related Quality of Life

To evaluate the quality of life, the Dutch version of the Disabkids (DCGM-37) was used, which was completed online by either the participant alone (n=45), together with one of the parents/caregivers (n=54) or the parent alone (n=9) (unknown for n=89). This questionnaire measures the quality of life and the independence of children and adolescents with chronic health conditions. The questionnaire has been used in other paediatric populations and has been tested for internal consistency and validity³⁷⁻³⁹. The questionnaire includes 37 items that cover six subscales, i.e. mental independence, mental emotion (inner strength), social inclusion, social exclusion (social equality), physical limitations (physical ability) and physical treatment (i.e. the impact of taking medication, receiving injections, etc.)³⁸. All items were scored on a 5-point Likert scale ranging from 1 = never to 5 = always. For each scale a sum score was calculated by following the instructions of the developers of the DCGM-37 and the provided syntax file³⁸. If one item was missing, this missing value was substituted by the mean of the non-missing values. If more than 1 item of a domain was missing, no sum score was calculated. These scores were transformed so that they had a range from 0 – 100 with higher scores indicating a higher perceived quality of life.

Self-perception and global self-worth

To evaluate self-perception, the Dutch translations of the self-perception profile for children (SPPC) and for adolescents (SPPA) were used⁴⁰. We used the children's version for children of 12 years old and younger, or with cognitive capabilities of this age group, and the adolescent version for those older than 12 years. Based on the researcher's expertise, an older child was provided the children's version or when the child clearly was not able to complete the adolescent version or did not understand the questions. The children's version uses 36 items to measure five domains, i.e. 'scholastic competence', 'social acceptance', 'athletic competence', 'physical appearance', 'Behavioural conduct', and the general concept 'global self-worth'^{41,42}. All six scales consist of 6 items that include bipolar statements (e.g. 'some kids feel they are very good at school, but other kids worry about whether they do the schoolwork assigned to them'). The children have to choose which of the two statements resembles them, and how much, i.e. 'sort of true for me' or 'really true for me'. All items were scored on a 4-point scale and sum scores were calculated. The questionnaire has been tested in a Dutch norm group and showed reasonable to good internal consistency and test-retest reliability⁴².

The adolescent version includes 35 items which make up 7 scales; i.e. 'scholastic competence', 'social acceptance', 'athletic competence', 'physical appearance',

'behavioural conduct', 'close friendship' and the general concept 'global self-worth'. Similar to the child version of the questionnaire, all items included bipolar statements that were scored on a 4-point scale by which the adolescents indicated which statement resembled them 'sort of true' or 'really true'⁴³.

If one item of the scale was missing, the scale score was not calculated ($n=3$). All scores on each scale were compared to Dutch norm scores to indicate whether the participants scored 'below average', i.e. below the 15th percentile, or above average, i.e. at or higher than the 85th percentile. For the child version, different norms have been formulated for boys and girls⁴¹. For the adolescent version different norms were formulated by school level for the scales 'scholastic competence' and 'behavioural conduct' and a distinction by sex was made for all scales except 'social acceptance'⁴³. In the current study, results regarding these norm scores were only used for descriptive purposes.

Exercise Self-Efficacy

To assess whether sport participation was associated with exercise self-efficacy, the Dutch version of the Exercise Self-Efficacy Scale^{44,45} was filled out digitally at home by the child. Self-efficacy is a well-known behavioural determinant and described in several behavioural theories such as the Social Cognitive Theory by Bandura⁴⁶ in which self-efficacy is seen as important influencer of behaviour. In the current study we hypothesise that behaviour, in this study engaging in sport, can have a positive impact on self-efficacy in relation to physical activity and exercise. This hypothesis is based on the fact that mastery experiences and vicarious experiences are important sources of self-efficacy⁴⁷ and when participating in sport, children may learn new skills and may see others like themselves performing specific tasks or behaviours. The questionnaire takes approximately 2 min to complete. The questionnaire consists of 10 items about the level of self-confidence with regard to performing regular physical activities and exercise that could be rated on a 4-point Likert scale ranging from 'not true at all' to 'always'. A sample item is: "I am confident that I can overcome barriers and challenges with regard to physical activity and exercise if I try hard enough". A sum score was calculated when all 10 items were answered. Internal consistency of the scale was high, Cronbach's alpha = 0.99. A higher score indicates a higher exercise self-efficacy. Validity and reliability of the scale have been tested in a sample with spinal cord injury and showed good validity and adequate reliability⁴⁴.

Potential confounders and co-variables

Demographic variables

The demographic questionnaire that included questions about date of birth, sex, and school level.

Medical Diagnosis

The general questionnaire assessed the medical diagnosis. The medical diagnosis were further classified in categories according to The American College of Sports Medicine (ACSM)⁴⁸: 1. Cardiovascular (e.g. ventricular septal defect, tetralogy of Fallot, cardiomyopathy), 2. Pulmonary (Asthma), 3. Metabolic (Diabetes), 4. Musculoskeletal or orthopaedic (Amputation, clubfoot, Hereditary Multiple Exostoses - Multiple Osteochondromas, congenital anomalies), 5. Neuromuscular (Cerebral palsy, spina bifida, neurofibromatosis, Kabuki syndrome, centronuclear myopathy, psychomotor retardation, Martsolf syndrome, acquired brain injury), 6. Immunological or haematological (Rheumatism, Fanconi anaemia), 7. Cancer (Tumor in hypophysis), 8. Epilepsy. In the regression analyses categories were merged distinguishing disabilities or disease related to the metabolic system or oxygen transport (1. Cardiovascular, 2 Pulmonary, and 3. Metabolic) from the other disabilities or diseases, mostly related to the musculoskeletal or neurological system.

School type

Children participating in the current study attended either regular education or special education for children with physical disabilities (so called mytyl schools). These schools, dedicated to children with CDPD, have similar learning objectives as regular schools, but the children receive additional attention and support. Children attending special education may have different references for social comparison which may influence how they perceive themselves. Therefore, school type was added as a potential confounder in the analyses.

Statistical analyses

Continuous data was described by means or medians and standard deviations or interquartile ranges and categorical data by frequencies and proportions. Crude comparisons on the outcome variables between the sporting and non-sporting participants were made using ANOVA. Adjusted associations between sports participation and the outcome variables were estimated by linear (or logistic) regression analyses. Assumptions were checked and residuals showed an acceptable normal distribution, except for the Exercise Self-Efficacy scale. Therefore the scale was dichotomized at the third tertile (score ≥ 37) and associations with sports participation were assessed by means of logistic regression analyses. Sensitivity analyses were run with and without outliers, i.e. those with standardized residual scores below -3 or higher than 3.

Different models will be presented, unadjusted as well as adjusted for potential confounders sex, age, school type, diagnosis or ACSM category. All analyses were

checked for potential interaction by age. In case of significant interactions, results will be presented for two age groups. For the analyses, the ACSM categories were merged into two categories, i.e. one including those with cardiovascular, pulmonary and metabolic conditions, and another including those with musculoskeletal, neuromuscular, immunological, cancer or epileptic conditions. Results are reported as regression coefficients or odds ratios and their 95% confidence intervals.

RESULTS

Of the 195 children included in the current study, 96 were classified as participating in sports (see Table 1). Based on the ACSM classification, most children (n=104 (52.8%)) had a neuromuscular condition (Table 1), followed by cardiovascular disease (n=22 (11.3%)). One third of the children attended special education.

Table 1: Characteristics of the study sample (N=195).

	mean	Standard deviation
Age (years)	14.3	2.8
	n	%
Study ¹ (HAYS)	145	74.4
Sex (boys)	116	59.5
Sports participation ² (yes)	96	49.2
ACSM		
Cardiovascular	22	11.3
Pulmonary	9	4.6
Metabolic	16	8.2
Musculoskeletal or orthopedic	16	8.2
Neuromuscular	103	52.8
Immunological or heamatological	18	9.2
Cancer	3	1.5
Epileptic	8	4.1
Education type ³		
Special-education	65	33.3

¹ participating in either the HAYS or the S2SF study; ² sport participation is defined here as participating in sport at least 2 times a week; ³ Education type could be regular education or special education.

Table 2 shows the descriptive statistics regarding the outcome variables. The mean scores on the different subscales for the DCGM-37 varied, it was lowest for the social exclusion scale (62.9 (17.7)) and highest for the mental emotion (77.9 (17.8)) and physical treatment (77.1 (22.1)). When looking at the norm scores for the SPPC and SPPA, a relative highly proportion of children scores below the norm for social acceptance (SPPA: 21.3%) and athletic competence (SPPC: 25%, SPPA: 23.45). On the other hand, relatively many children score above the 85th percentile for scholastic competence (SPPA: 29.2%), social acceptance (SPPC: 25.7%), athletic competence (SPPC, 23.6%) and behavioural conduct (SPPC: 35.0%, SPPA: 33.3%).

Table 3 displays the unadjusted comparisons on the outcome variables between sporting and non-sporting participants. Those participating in sports scored higher on all subscales of the DCGM-37, except on the physical treatment scale. The significant differences in the unadjusted analyses remained statistically significant after adjustment for potential confounders, indicating that school type and medical diagnosis did not substantially confound the association. (Table 4).

Children participating in sports reported higher scores on social acceptance and athletic competence than their non-sporting peers, while no differences were observed for the other scales of the SPPC (Table 3). These differences became slightly stronger after adjustment for potential confounders (Table 4).

The adolescents who participated in sports, scored higher on athletic competence than the non-sporting adolescents (SPPA, Table 3). Moreover, this difference became slightly larger after adjustment of age, gender, school type and ACSM classification (Table 4). In addition, after adjustment for potential confounders, the scores on the scale for close friendships was significantly higher for adolescents participating in sports compared to their non-sporting peers.

Those participating in sports scored more often in the upper tertile on the exercise self-efficacy scale than their non-sporting peers (Table 3 and Table 4). After adjustment for school type and medical diagnosis the odds ratio decreased slightly, but remained significant. Those who participate in sports were 2.55 more likely to score in the upper tertile for exercise self-efficacy, independent of sex, school type or medical diagnosis. (Table 4).

Table 2: Descriptive statistics for quality of life, self-perceptions, global self-worth and self-efficacy.

DISABKIDS (DCGM-37) ¹	N	Mean	SD	Median	p ₂₅	p ₇₅	min	max
Mental independence (range 0 - 100)	187	75.8	14.6	79.2	66.7	87.5	33.3	100
Mental emotion (range 0 - 100)	187	77.9	17.8	78.6	67.9	92.9	28.6	100
Social inclusion (range 0 - 100)	187	62.9	17.7	66.7	50.0	75.0	16.7	100
Social exclusion (range 0 - 100)	187	75.8	17.9	79.2	66.7	91.7	12.5	100
Physical limitations (range 0 - 100)	187	68.0	18.5	70.8	54.2	83.3	12.5	100.0
Physical treatment (range 0 - 100)	87	77.1	22.1	83.3	66.7	91.7	20.8	100
General (31 items) (range 0 - 100)	187	72.3	1.5	72.6	61.3	83.9	29.0	100
Self-perception profile for children (SPPC)								
	N	Mean	SD	Median	p ₂₅	p ₇₅	% < p ₁₅ ²	% > p ₈₅ ³
Scholastic competence (6 items, range 6-24)	140	17.1	4.0	17	14	20	16.4	18.6
Social acceptance (6 items, range 6-24)	140	18.2	4.2	18	16	22	15.0	25.7
Athletic competence (6 items, range 6-24)	140	17.6	4.4	18	15	21	25.0	23.6
Physical appearance (6 items, range 6-24)	140	19.3	4.4	20	17	23	15.7	19.3
Behavioural conduct (6 items, range 6-24)	140	18.8	3.8	19	17	22	11.4	35.0
Global self-worth (6 items, range 6-24)	139	20.2	3.3	21	18	23	13.7	22.3
Self-perception profile for adolescents (SPPA)								
	N	Mean	SD	Median	p ₂₅	p ₇₅	% < p ₁₅	% > p ₈₅
Scholastic competence (5 items, range 5-20)	48	14.8	2.9	15	13	17	14.6	29.2
Social acceptance (5 items, range 5-20)	47	14.8	2.9	15	13	17	21.3	6.4
Athletic competence (5 items, range 5-20)	47	13.1	4.0	14	10	16	23.4	14.9
Physical appearance (5 items, range 5-20)	47	14.0	3.2	14	12	16	12.8	10.6
Behavioural conduct (5 items, range 5-20)	48	15.9	2.9	16	15	18	4.2	33.3
Close friendship (5 items, range 5-20)	47	17.6	2.2	18	17	19	4.2	10.4
Global self-worth (5 items, range 5-20)	46	15.7	2.7	16	14	18	10.9	10.9
Exercise self-efficacy (sumscore 10 items)								
	N	Mean	SD	Median	p ₂₅	p ₇₅	min	Max
Exercise self-efficacy (sumscore 10 items)	187	33.8	5.3	35	31	38	14	40
Exercise self-efficacy (dichotomized, > 3 rd tertile)								
	n		%					
Exercise self-efficacy (dichotomized, > 3 rd tertile)	187	71.0	38.0					

¹ All subscales are scored from 0 to 100 with higher scores indicating higher self-perceived health-related quality of life; p₂₅ – 25th percentile; p₇₅ – 75th percentile; ² < p₁₅ based on a reference sample; ³ > p₈₅ based on a reference sample

Table 3 : Unadjusted comparisons on quality of life, self-perceptions, global self-worth and self-efficacy between sporting and non-sporting participants.

DISABKIDS (DCGM-37) ¹	Sporting				Non-sporting				P-value
	N	Mean	SD	Median	N	Mean	SD	Median	
Mental independence (range 0 - 100)	93	79.2	13.8	79.2	94	72.4	14.6	75.0	0.001
Mental emotion (range 0 - 100)	93	81.9	16.7	85.7	94	73.9	18.1	75.0	0.002
Social inclusion (range 0 - 100)	93	68.6	17.5	70.8	94	57.2	16.0	58.3	<0.001
Social exclusion (range 0 - 100)	93	79.9	15.9	83.3	94	71.8	18.8	75.0	0.002
Physical limitations (range 0 - 100)	93	72.0	18.0	70.8	94	64.1	18.3	62.5	0.003
Physical treatment (range 0 - 100)	48	78.6	20.0	83.3	39	75.2	24.6	83.3	0.474
General (31 items) (range 0 - 100)	93	76.5	13.9	79.8	94	68.1	14.0	69.4	<0.001
Self-perception profile for children (SPPC)									
Scholastic competence (6 items, range 6-24)	66	17.0	4.0	17.0	74	17.3	3.9	18.0	0.639
Social acceptance (6 items, range 6-24)	66	19.3	3.3	19.0	74	17.2	4.7	18.0	0.003
Athletic competence (6 items, range 6-24)	66	19.1	3.9	20.0	74	16.2	4.5	17.0	<0.001
Physical appearance (6 items, range 6-24)	66	19.5	4.0	20.0	74	19.1	4.7	20.0	0.601
Behavioural conduct (6 items, range 6-24)	66	18.8	3.6	19.0	74	18.8	3.9	19.0	0.989
Global self-worth (6 items, range 6-24)	66	20.3	3.3	21.0	73	20.0	3.2	21.0	0.582
Self-perception profile for adolescents (SPPA)									
Scholastic competence (5 items, range 5-20)	27	15.1	2.6	16	21	14.4	3.3	15	0.453
Social acceptance (5 items, range 5-20)	26	14.7	3.5	16	21	14.8	2.0	15	0.928
Athletic competence (5 items, range 5-20)	27	14.3	3.2	14	20	11.6	4.5	11.5	0.022
Physical appearance (5 items, range 5-20)	27	14.3	2.8	15	20	13.5	3.8	13.5	0.409
Behavioural conduct (5 items, range 5-20)	27	15.8	3.0	16	21	16	2.8	16	0.749
Close friendship (5 items, range 5-20)	28	17.8	2.1	18	20	17.4	2.5	17.5	0.560
Global self-worth (5 items, range 5-20)	25	16.0	2.4	17	21	15.4	3.1	15	0.485
Exercise self-efficacy (sumscore 10 items)	93	35.2	4.3	36	94	32.4	5.8	33.5	<0.001
	n	%			n	%			
Exercise self-efficacy (dichotomized, 3 rd tertile)	93	46	49.5		94	25	26.6		0.002 ²

¹ All subscales are scored from 0 to 100 with higher scores indicating higher self-perceived health-related quality of life; ² tested with a Chi-square test

Table 4: Results from regression analyses comparing sporting (1) and non-sporting participants (0) on health-related quality of life, self-perceptions, global self-worth and exercise self-efficacy.

	model 1 ^a		model 2 ^b		model 3 ^c	
	b	95%CI	b	95%CI	b	95%CI
DISABKIDS (DCGM-37)						
Mental independence (range 0 - 100)	6.06	(1.83; 10.3)	4.68	(0.18; 9.18)	5.75	(1.41; 10.1)
Mental emotion (range 0 - 100)	6.32	(1.19; 11.5)	6.24	(0.74; 11.7)	7.06	(1.60; 12.5)
Social inclusion (range 0 - 100)	11.0	(5.99; 16.0)	7.76	(2.56; 13.0)	8.86	(3.80; 13.9)
Social exclusion (range 0 - 100)	7.56	(2.35; 12.8)	5.63	(0.11; 11.1)	6.79	(1.42; 12.2)
Physical limitations (range 0 - 100)	6.75	(1.35; 12.1)	5.82	(0.05; 11.6)	7.02	(1.39; 12.6)
Physical treatment (range 0 - 100)	n.a.		n.a.		n.a.	
General (31 items) (range 0 - 100)	7.50	(3.35; 11.7)	6.03	(1.63; 10.4)	7.09	(2.86; 11.3)
Self-perception profile for children (SPPC)						
Scholastic competence (6 items, range 6-24)	-0.86	(-2.17; 0.45)	-0.36	(-1.76; 1.03)	-0.42	(-1.81; 0.96)
Social acceptance (6 items, range 6-24)	1.63	(0.26; 3.01)	2.00	(0.52; 3.48)	2.01	(0.52; 3.50)
Athletic competence (6 items, range 6-24)	1.90	(0.49; 3.31)	2.69	(1.21; 4.17)	2.65	(1.17; 4.12)
Physical appearance (6 items, range 6-24)	-0.50	(-1.95; 0.94)	-0.23	(-1.79; 1.33)	-0.26	(-1.82; 1.31)
Behavioural conduct (6 items, range 6-24)	0.25	(-0.87; 1.38)	0.48	(-0.74; 1.69)	0.48	(-0.74; 1.70)
Global self-worth (6 items, range 6-24)	0.15	(-0.93; 1.23)	0.50	(-0.65; 1.66)	0.47	(-0.68; 1.63)
Self-perception profile for adolescents (SPPA)						
Scholastic competence (5 items, range 5-20)	0.623	(-1.249; 2.496)	0.504	(-1.393; 2.401)	0.849	(-1.242; 2.940)
Social acceptance (5 items, range 5-20)	0.058	(-1.856; 1.973)	0.112	(-1.844; 2.067)	1.173	(-0.925; 3.270)
Athletic competence (5 items, range 5-20)	2.385	(-0.075; 4.845)	2.542	(0.043; 5.041)	3.319	(0.647; 5.992)
Physical appearance (5 items, range 5-20)	-0.013	(-2.029; 2.004)	-0.132	(-2.183; 1.919)	-0.030	(-2.283; 2.222)
Behavioural conduct (5 items, range 5-20)	-0.241	(-1.791; 1.309)	-0.401	(-1.946; 1.144)	-0.181	(-1.881; 1.520)
Close friendship (5 items, range 5-20)	0.849	(-0.592; 2.289)	0.676	(-0.755; 2.108)	1.636	(0.269; 3.002)
Global self-worth (5 items, range 5-20)	-0.666	(-2.545; 1.213)	-0.832	(-2.723; 1.059)	-0.360	(-2.473; 1.753)
Exercise self-efficacy (dichotomized, 3rd tertile)						
	OR	95%CI	OR	95%CI	OR	95%CI
	2.72	(1.47; 5.02)	2.24	(1.17; 4.27)	2.55	(1.30; 4.99)

^a Only adjusted for sex and age; ^b further adjusted for school type; ^c further adjusted for ACSM classification (recoded into two categories (cardiovascular, pulmonary, metabolic impairments vs musculoskeletal, neuromuscular, immunological, cancer, epileptic impairments/diseases)); b – regression coefficient, reflects the difference in score between non-sporting (0) and sporting (1) children; 95%CI – 95% confidence interval; n.a. – not applicable, too few children who completed this scale

DISCUSSION

The current study aimed to assess the associations between sports participation and psychosocial health among children and adolescents with a disability. In general, those who reported to participate in sports at least twice a week, had more beneficial scores on the various indicators compared to their peers who did not participate in sports or less than twice a week. This was independent of age, sex, school type or medical diagnosis and largely in line with our hypotheses.

Those participating in sports scored better on all scales of the DCGM-37 scale. The effect sizes were quite substantial, for the total score the difference between the groups was about 7 points. Although the fact that the DCGM-37 distinguishes six sub-scales reflecting different concepts, these sub-scales were strongly correlated in the current sample. Therefore, it is not surprising that the groups differed on all sub-scales. Unfortunately, it is difficult to compare these findings with other studies, due to the fact that other instruments to assess quality of life were used, or that other studies did not made comparisons between sporting and non-sporting participants. However, our findings support the hypothesis that participating in organized sports by children and adolescents with a CDPD can contribute to all domains of quality of life and are in line with two studies among adults^{49,50}.

Furthermore, our findings regarding the self-perceptions are in line with the existing reviews among adults, children and adolescents^{29,30}, and show that participating in organized sports contributes to feelings of athletic competence in children and adolescents with a CDPD. In addition, children, but not adolescents, participating in sports reported higher feelings of social acceptance. It may be that adolescents have more networks besides a sports club that influence their feelings of social acceptance, while children have a more controlled and limited network and that participating in sports with other children therefore has a greater influence on their feelings of social acceptance. On the other hand, the effect estimate was in favour of the sporting adolescents and it may be that for these analyses not enough participants were included to show a significant difference. In addition, a study among high school students found a positive association between sports participation and self-belief, that included feelings of social acceptance⁵¹. Lastly, it may be that children who do not feel accepted withdraw from sport participation. That we did not find significant differences between the groups on most of the other self-perception scales, may be due to the fact that our study population scored relatively high on most scales, at least when compared with the 15th and 85th percentile norm values. However, compared to a study sample described by Shapiro and colleagues, the current sample scored lower on the social acceptance, athletic competence and physical appearance scales⁵². That in the current

study no associations between sport participation and self-perception concepts were found may be because of other factors than sports participation play a more crucial role in these scales or that different types of sports may have different effects that could not be detected in the current study design.

Finally, despite the fact that the self-efficacy scale was dichotomized which may have led to reduced power, we found a strong association between sports participation and exercise self-efficacy. Self-efficacy is a well known behavioural determinant of physical activity⁵³ assuming that exercise self-efficacy influences physical activity. In the current study, we hypothesized another causal pathway, i.e. that sports participation would result in beneficial scores on a range of psychosocial health indicators, including exercise self-efficacy. Sources of influence of self-efficacy are vicarious experiences or modelling and past experiences⁴⁶, therefore it can be expected that those who participate in sports see others performing sports, and also experience themselves that they are able to perform sports, which both positively impact on their exercise self-efficacy.

An important limitation of the current study is its cross sectional design limiting the conclusions regarding causality. It may as well be that those who experience a better quality of life, or who feel socially accepted and athletically competent, are more likely to participate in organized sports. Therefore, experimental and longitudinal studies are required to study causal or reciprocal relationships. These longitudinal designs would also allow for mediation analyses to study underlying pathways. For instance, in a longitudinal design we could analyse whether changes in sport participation result in changes in intermediate variables (e.g. self-perception) that in turn result in changes in health-related quality of life. Another limitation is the crude measure of sports participation, which did not include information on duration, quality and type of sports. It may be that team sports have different impact on psychosocial health indicators than individual sports. Furthermore, we did not report whether they participated in regular sports or in adapted forms of sports. The advantage of participation in adapted forms of sports is that children can better focus on their abilities rather than their inabilities⁴⁹. Additionally, the DCGM-37 was in a few cases ($n=9$) completed by the parent, which may have biased the findings as parents tend to report lower scores than children^{37,54}. However, if this is a systematic bias, it will not affect the association between sport participation and health related quality of life. Finally, those defined as participating in sports may perform sports at different intensities and frequencies. In conclusion, the group defined as participating in sport is very heterogeneous and this may have biased the results such that potential associations between sport participation and the outcome may not have been detected. Therefore, future studies should take these aspects into account and may also investigate how the level of sport participation is related to self-

perceptions. However, even with the use of this crude measure for sports participation, significant associations with psychosocial health and quality of life were detected.

CONCLUSIONS

Despite some limitations and considering the fact that this is (one of) the first studies addressing this topic in children and adolescents with a CDPD, the current study provides the first indications that participating in sports is beneficial in this population for their psychosocial health. A next step would be to perform experimental and longitudinal studies to see what type of sports, duration and intensities are most promising for improving psychosocial health.

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REFERENCES

1. Warburton DER, Nicol CW, Bredin SSD. Health benefits of physical activity: the evidence. *CMAJ* 2006;174:801–9.
2. Kubota Y, Evenson KR, MacLehose RF, Roetker NS, Joshu CE, Folsom AR. Physical Activity and Lifetime Risk of Cardiovascular Disease and Cancer. *Med Sci Sport Exerc* 2017;1.
3. Alves AJ, Viana JL, Cavalcante SL, Oliveira NL, Duarte JA, Mota J, et al. Physical activity in primary and secondary prevention of cardiovascular disease: Overview updated. *World J Cardiol* 2016;8:575–83.
4. Samitz G, Egger M, Zwahlen M. Domains of physical activity and all-cause mortality: Systematic review and dose-response meta-analysis of cohort studies. *Int J Epidemiol* 2011;40:1382–400.
5. Eime RM, Harvey JT, Brown WJ, Payne WR. Does Sports Club Participation Contribute to Health-related Quality of Life? *Med Sci Sport Exerc* 2010;42:1022–8.
6. Street G, James R, Cutt H. The relationship between organised physical recreation and mental health. *Heal Promot J Aust* 2007;18:236–9.
7. Seippel Ø. The meanings of sport: fun, health, beauty or community? *Sport Soc* 2017;9:51–70.
8. Eime RM, Young JA, Harvey JT, Charity MJ, Payne WR. A systematic review of the psychological and social benefits of participation in sport for adults: informing development of a conceptual model of health through sport. *Int J Behav Nutr Phys Act* 2013;10:135.
9. Sahlin KB, Lexell J. Impact of Organized Sports on Activity, Participation, and Quality of Life in People With Neurologic Disabilities. *PM R. Am J Phys Med Rehabil*; 2015;7:1081–8.
10. Mitchell TMDF, Barlow CEMS. Review of the Role of Exercise in Improving Quality of Life in Healthy Individuals and in Those With Chronic Diseases. *Curr Sports Med Rep* 2011;10:211–6.
11. Bullinger M, Schmidt S, Petersen C. Assessing quality of life of children with chronic health conditions and disabilities: a European approach. *Int J Rehabil Res* 2002;25:197–206.
12. Crosbie A. The Effect of Physical Training in Children With Asthma on Pulmonary Function, Aerobic Capacity and Health-Related Quality of Life: A Systematic Review of Randomized Control Trials. *Pediatr Exerc Sci* 2012;24:472–89.
13. Kotte EMW, De Groot JF, Winkler AMF, Huijgen BCH, Takken T. Effects of the Fitkids Exercise Therapy Program on Health-Related Fitness, Walking Capacity, and Health-related Quality of Life. *Phys Ther* 2014;94:1306–18.
14. Koldoff EA, Holtzclaw BJ. Physical Activity Among Adolescents with Cerebral Palsy: An Integrative Review. *J Pediatr. Nurs Elsevier Inc.*; 2015;30:e105–17.

15. Kotte EMW, Winkler AMF, Takken T. Fitkids exercise therapy program in the Netherlands. *Pediatr Phys Ther* 2013;25:7–13.
16. Neter JE, Schokker DF, de Jong E, Renders CM, Seidell JC, Visscher TLS. The Prevalence of Overweight and Obesity and Its Determinants in Children with and without Disabilities. *J Pediatr* 2011;158:735–9.
17. Carlon SL, Taylor NF, Dodd KJ, Shields N. Differences in habitual physical activity levels of young people with cerebral palsy and their typically developing peers: a systematic review. *Disabil Rehabil* 2013;35:647–55.
18. Dahan-Oliel N, Shikako-Thomas K, Majnemer A. Quality of life and leisure participation in children with neurodevelopmental disabilities: A thematic analysis of the literature. *Qual Life Res* 2012;21:427–39.
19. Rimmer JA, Rowland JL, Rimmer JA, Rowland JL. Physical activity for youth with disabilities : A critical need in an underserved population. *Dev Neurorehabil*. 2008;11:141–8.
20. Buffart LM, Roebroek ME, Rol M, Stam HJ, Van Den Berg-Emons RJG, Netherlands Transition Research Group South-West. Triad of Physical Activity, Aerobic Fitness and Obesity in Adolescents and Young Adults with Myelomeningocele *J Rehabil Med* 2008;40:70–5.
21. Rimmer JH, Riley B, Wang E, Rauworth A, Jurkowski J. Physical Activity Participation Among Persons with Disabilities. *Am J Prev Med* 2004;26:419–425.
22. Sawin KJ, Bellin MH. Quality of life in individuals with spina bifida: A research update. *Dev Disabil Res Rev* 2010;16:47–59.
23. Ferguson MK, Kovacs AH. Quality of life in children and young adults with cardiac conditions. *Curr Opin Cardiol* 2013;28:115–21.
24. Houben-van Herten M, Bai G, Hafkamp E, Landgraf JM, Raat H. Determinants of health-related quality of life in school-aged children: A general population study in the Netherlands. *PLoS One* 2015;10:1–13.
25. Dinomais M, Gambart G, Bruneau A, Bontoux L, Deries X, Tessiot C, Richard I. Social functioning and self-esteem in young people with disabilities participating in adapted competitive sport. *Neuropediatrics* 2010;41:49–54.
26. Abdou Y, Erpelding OM. Contribution of taking part in sport to the association between physical activity and quality of life. *Qual Life Res* 2013;22:2021–9.
27. Drakouli M, Petsios K, Giannakopoulou M, Patiraki E, Matziou V, M. D, et al. Determinants of quality of life in children and adolescents with CHD: a systematic review. *Cardiol Young* 2015;1027–36.
28. Balaguer I, Atienza-Gonzalez FL, Duda JL. Self-Perceptions, Self-Worth and Sport Participation in Adolescents. *Span J Psychol* 2012;15:624–30.

29. Babic MJ, Morgan PJ, Plotnikoff RC, Lonsdale C, White RL, Lubans DR. Physical Activity and Physical Self-Concept in Youth: Systematic Review and Meta-Analysis. *Sport Med* 2014;44:1589–601.
30. Hausenblas HA, Fallon EA. Exercise and body image: a meta-analysis. *Psychol Health* 2006;21:33–47.
31. Lankhorst K, van der Ende-Kastelijin K, de Groot J, Zwinkels M, Verschuren O, Backx F, et al. Health in Adapted Youth Sports Study (HAYS): health effects of sports participation in children and adolescents with a chronic disease or physical disability. Springerplus Springer International Publishing; 2015;4:796.
32. Zwinkels M, Verschuren O, Lankhorst K, van der Ende-Kastelijin K, de Groot J, Backx F, et al. 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.
33. de Hollander EL, Milder I, Proper K. Beweeg- en sportgedrag van mensen met een chronische aandoening of lichamelijke beperking. Bilthoven; RIVM 2015.
34. Kemper HCG, Ooijendijk WT., Stiggelbout M. Consensus over de Nederlandse norm voor gezond bewegen (consensus about the Dutch norm for physical activity). *Sport en Geneeskunde*. 2000;78:180–3.
35. 3Van Brussel M, Van Der Net J, Hulzebos E, Helders PJM, Takken T. The Utrecht approach to exercise in chronic childhood conditions: The decade in review. *Pediatr Phys Ther* 2011;23(1):2-14.
36. Verschuren O, Peterson MD, Balemans ACJ, Hurvitz EA. Exercise and physical activity recommendations for people with cerebral palsy. *Dev Med Child Neurol* 2016;58(8):798-808.
37. Mueller-Godeffroy E, Thyen U, Bullinger M. Health-Related Quality of Life in Children and Adolescents with Cerebral Palsy: A Secondary Analysis of the DISABKIDS Questionnaire in the Field-Study Cerebral Palsy Subgroup. *Neuropediatrics* 2016;47:97–106.
38. Froisland DH, Markestad T, Wentzel-Larsen T, Skrivarhaug T, Dahl-Jorgensen K, Graue M. Reliability and validity of the Norwegian child and parent versions of the DISABKIDS Chronic Generic Module (DCGM-37) and Diabetes-Specific Module (DSM-10). *Health Qual. Life Outcomes*. BioMed Central Ltd; 2012;10:19.
39. Simeoni MC, Schmidt S, Muehlan H, Debensason D, Bullinger M, Petersen C, et al. Field testing of a European quality of life instrument for children and adolescents with chronic conditions: The 37-item DISABKIDS Chronic Generic Module. *Qual Life Res* 2007;16:881–93.

40. Harter S. The Self-Perception Profile for Children: Revision of the Perceived Competence Scale for Children. Denver, CO; 1985.
41. Veerman JWE, Treffers DA, Van der Bergh BRH, Ten Brink LT. Competentiebelevingsschaal voor kinderen. Amsterdam: Harcourt Test Publishers; 2004.
42. eerman JW, Ten Brink LT. Measuring childrens self-concept Factorial validity. *J Per. Assess* 1996;67:142–54.
43. Treffers, Ph. D, Goedhardt AW, Veerman JW, Van den Bergh BRH, Ackaert L, de Rycke L. Handleiding Competentie Belevingsschaal voor Adolescenten. Lisse: Swets Test Publishers; 2002.
44. Nooijen CFJ, Post MWM, Spijkerman DCM, Bergen MP, Stam HJ, Van Den Berg-Emons RJG. Exercise self-efficacy in persons with spinal cord injury: Psychometric properties of the Dutch translation of the exercise self-efficacy scale. *J Rehabil Med* 2013;45:347–50.
45. Kroll T, Kehn M, Ho P-S, Groah S. The SCI Exercise Self-Efficacy Scale (ESES): development and psychometric properties. *Int J Behav Nutr Phys Act* 2007;4:34.
46. Bandura A. Health promotion from the perspective of social cognitive theory. 2012;37–41.
47. Bandura A. Self-efficacy. *The Exercise of control*. New York: Freeman; 1997.
48. Durstine LJ, Moore GE, Painter P, Roberts SO. ACSM's exercise management for persons with chronic diseases and disabilities. 3rd ed. Humen Kinetics; Champaign, IL, 2009.
49. Yazicioglu K, Yavuz F, Goktepe AS, Tan AK. Influence of adapted sports on quality of life and life satisfaction in sport participants and non-sport participants with physical disabilities. *Disabil Health J* 2012: 249–53.
50. Groff DG, Lundberg NR, Zabriskie RB. Influence of adapted sport on quality of life: perceptions of athletes with cerebral palsy. *Disabil Rehabil* 2009;31:318–26.
51. Dodge T, Lambert ÆSF. Positive Self-Beliefs as a Mediator of the Relationship Between Adolescents' Sports Participation and Health in Young Adulthood. *J Youth Adolesc* 2009;38:813–25.
52. Shapiro DR, Martin JJ. The relationships among sport self-perceptions and social well-being in athletes with physical disabilities. *Disabil Health J Elsevier Inc*; 2014;7:42–8.
53. Plotnikoff RC, Costigan SA, Karunamuni N, Lubans DR. Social cognitive theories used to explain physical activity behavior in adolescents: a systematic review and meta-analysis. *Prev Med*. 2013;56:245–53.
54. Baca CB, Vickrey BG, Hays RD, Vassar SD, Berg AT. Differences in child versus parent reports of the child's health-related quality of life in children with epilepsy and healthy siblings. *Value Heal. International Society for Pharmacoeconomics and Outcomes Research (ISPOR)*; 2010;13:778–86.



The associations of cardiorespiratory fitness, adiposity and sports participation with arterial stiffness in youth with chronic diseases or physical disabilities

Kristel Lankhorst

Eero Haapala

Janke de Groot

Maremka Zwinkels

Olaf Verschuren

Harriet Wittink

Frank Backx

Anne Visser-Meily

Tim Takken

ABSTRACT

Background. The evidence on the associations of cardiorespiratory fitness, body adiposity, and sports participation with arterial stiffness in children and adolescents with chronic diseases or physical disabilities is limited.

Design. Cross-sectional.

Methods. Altogether 140 children and adolescents with chronic diseases or physical disabilities participated in the study. Cardiorespiratory fitness was assessed using maximal exercise test with respiratory gas analyses either using shuttle run, shuttle ride, or cycle ergometer test. Cardiorespiratory fitness was defined as peak oxygen uptake (VO_{2peak}) by body weight or fat free mass (FFM). Body adiposity was assessed using waist circumference, body mass index standard-deviation score (BMI-SDS), and body fat percentage. Sports participation was assessed by a questionnaire. Aortic pulse wave velocity PWV (PWVao) and augmentation index (AIX%) were assessed by a non-invasive oscillometric tonometry device.

Results. VO_{2peak} /body weight (standardized regression coefficient $\beta=-0.222$, 95% CI=-0.386 to -0.059, $P=0.002$) and VO_{2peak} /FFM ($\beta=-0.173$, 95% CI=-0.329 to -0.017, $P=0.030$) were inversely and waist circumference directly ($\beta=0.245$, 95% confidence interval (CI)=0.093 to 0.414, $P=0.002$) associated with PWVao. However, the associations of the measures of cardiorespiratory fitness with PWVao were attenuated after further adjustment for waist circumference. A higher waist circumference ($\beta=-0.215$, 95% CI=-0.381 to -0.049, $P=0.012$) and a higher BMI-SDS ($\beta=0.218$, 95% CI=-0.382 to -0.054, $P=0.010$) were related to lower AIX%.

Conclusions. Poor cardiorespiratory fitness and higher waist circumference were associated with increased arterial stiffness in children and adolescents with chronic diseases and physical disabilities. The association between cardiorespiratory fitness and arterial stiffness was partly explained by waist circumference.

INTRODUCTION

Arteriosclerotic cardiovascular diseases are one of the leading causes of morbidity and mortality and the costs related to arteriosclerosis demonstrate a considerable economic burden¹. Arterial stiffness and endothelial dysfunction are the first signs of arteriosclerosis and they have been established already in children and adolescents^{2,3}. In adults, increased arterial stiffness has been found to predict subsequent clinical cardiovascular events⁴. Therefore, early identification of children and adolescents with increased arterial stiffness is of importance in order to prevent arteriosclerotic cardiovascular diseases in later years¹.

Poor cardiorespiratory fitness has been associated with higher carotid and femoral artery stiffness and aortic intima media thickness (IMT) and elasticity in adolescents^{5,6}. Better cardiorespiratory fitness in adolescence also has been linked to lower femoral artery stiffness and carotid IMT at the age of 36⁷. Furthermore, obesity has been consistently related to stiffer carotid and aortic arteries among youth⁸. Higher levels of habitual physical activity have been associated with lower arterial stiffness, lower aortic IMT, and improved endothelial function in children and adolescents in some⁹⁻¹¹, but not all studies¹²⁻¹⁵. Furthermore, constantly high levels of vigorous physical activity between ages 13 and 36 have been linked to lower arterial stiffness at the age of 36 year¹⁶. Finally, the results of some previous studies suggest that exercise training has favorable effects on flow-mediated dilation as a marker of endothelial function in overweight and obese children and adolescents¹⁷.

Children and adolescents with chronic diseases or physical disabilities may have an increased risk of arteriosclerosis¹⁸. The evidence suggest that children and adolescents with chronic diseases or physical disabilities have lower cardiorespiratory fitness¹⁹, higher prevalence of overweight and obesity^{19,20}, lower levels of physical activity^{20,21}, and they participate less often in organized sports²⁰, than their apparently healthy or normally developing peers. Children and adolescents with chronic disease or disabilities may also have increased arterial stiffness²². Studies among adults also suggest that arterial stiffness is particularly important marker of subsequent cardiovascular morbidity and mortality among those with chronic cardiovascular or metabolic diseases⁴. However, one small study found no differences in arterial structure and function between children with cerebral palsy (CP) and normally developing children with similar levels of physical activity and body adiposity²³. Nevertheless, there are no studies on the associations of cardiorespiratory fitness, body adiposity, and sports participation with arterial stiffness in

a large sample of children and adolescents with chronic diseases or physical disabilities. The aim of the present study was to investigate the associations of cardiorespiratory fitness, body adiposity, and sports participation with arterial stiffness in children and adolescents with chronic diseases or physical disabilities. We also studied if there are thresholds for cardiorespiratory fitness and the measures of body adiposity that are associated with increased arterial stiffness.

METHODS

Participants

The present analyses are based on the data from the Health in Adapted Youth Sports (HAYS) Study and the Sport-2-Stay-Fit (S2SF) Study. These studies are designed to investigate the associations of sports participation and exercise training with physical fitness, physical activity, body adiposity, cognition, cardiovascular health, and quality of life in children and adolescents with chronic diseases or physical disabilities. The study designs are published in detail elsewhere^{24,25}.

The children and adolescents for the HAYS Study and S2SF Study were recruited in the Netherlands among different patients' associations, pediatric physical therapy practices, Wilhelmina Children's Hospital in Utrecht, De Hoogstraat Rehabilitation Center in Utrecht, Fitkids practices, Schools for special education, and adapted sports organizations. The inclusion criteria for the HAYS Study and the S2SF Study was that participants had to understand the Dutch language, understand simple instructions, and be able to perform physical fitness tests. All ambulatory children and adolescents and wheelchair users were eligible to participate. Children and adolescents using an electric wheelchair, having a progressive disease, who participated in other research projects, or who had contra-indications for performing a maximal exercise test were excluded.

Ethical approval

The study protocols were approved by the Medical Ethics Committee of the University Medical Center Utrecht, the Netherlands (METC number: 14-332/c and 14-118/m). All participants and the parents of participants under 18-years-of-age provided their informed written consent. Studies were conducted in accordance to the Helsinki declaration.

Assessment of cardiorespiratory fitness

Peak oxygen uptake ($\text{VO}_{2\text{peak}}$) as a measure of cardiorespiratory fitness was assessed either by an adapted 10-meter incremental shuttle run test, by a 10-meter incremental shuttle ride test, or by an incremental cardiopulmonary exercise test on an electronically braked Ergoline cycle ergometer (Ergoselect 200 K, Ergoline, Bitz, Germany)²⁴. The test modality was based on the evaluation of children's ambulatory ability, mode of daily locomotion, type of sports, and safety issues. All children with a congenital cardiopulmonary disease underwent an incremental cardiopulmonary exercise test with continuous electrocardiography (ECG) monitoring on a cycle ergometer. Participants who were able to walk or run performed an incremental shuttle run test and wheelchair users performed a shuttle ride test. Altogether 64% of the participants performed a shuttle run test, 11% a shuttle ride test, and 24% a cycle ergometer test. All the test procedures have been described in detail previously²⁴.

Respiratory gases were collected using pediatric masks (Hans-Rudolph, Shawnee, Kansas, USA) during the test by a calibrated metabolic cart (Cortex Metamax, Samcon bvba, Melle, Belgium). Regardless of testing modality, respiratory gases were measured directly by the breath-by-breath method from the 3-min resting steady-state period to the post-exercise rest and were averaged over consecutive 10-s periods.

Children and adolescents were verbally encouraged to exercise until voluntary exhaustion. The exercise test was considered maximal, if the subjective and objective criteria indicated maximal effort and maximal cardiorespiratory capacity (i.e. flushing, sweating, heart rate >180 , RER >1.0 , or plateau of VO_2). Cardiorespiratory fitness was defined as $\text{VO}_{2\text{peak}}$ per body weight and $\text{VO}_{2\text{peak}}$ per kg fat free mass (FFM).

Assessment of body size and body adiposity

Body size and composition were measured after emptying the bladder. Body height was measured by stadiometer (Seca, Hamburg, Germany) in ambulant participants. Among participants using wheelchair the body height was measured in a supine position using a measuring tape. In case of spasticity of the lower limbs, arm span width was measured to the nearest centimeter from middle fingertip to fingertip. Body weight was measured by standard scale (Seca, Hamburg, Germany). Participants who used a wheelchair and who were not able to stand on a scale were measured using a wheelchair scale (Stimag B.V., Lisse, the Netherlands). Body mass index (BMI) was calculated as body weight (kg) / body height (m)² for ambulant participants and as body weight (kg) / the arm

span length. We used arm span adjustment of $\times 0.95$ and $\times 0.90$ for participants with central neurological disorders in mid lumbar lesions and high lumbar/thoracic lesions, respectively²⁶. Waist circumference was measured by an unstretchable measuring tape at the level of the navel. BMI-standard deviation score (SDS) and waist circumference-SDS were computed using national reference values²⁷. Body fat percentage and FFM were measured by bioelectrical impedance analyses using Bodystat Quadscan 4000 device (EuroMedix, Leuven, Belgium).

Assessment of sports participation

To allow a comparison between children and adolescents with chronic diseases or physical disabilities who participated in organized sports and those without regular sports participation we recruited sports participants from a broad range of participation in sports including recreational level as well as high level competitive sports²⁴. The study participants reported how often they participated in organized sports on a questionnaire. In the present study, we defined regular sports participation as any involvement in organized sports at least 2 times per week.

Assessment of arterial stiffness

Aortic PWV (PWVao), as a measure of arterial stiffness, and augmentation index (AIX%), as measure of peripheral arterial tone, were assessed by non-invasive oscillometric tonometry device (Arteriograph, TensioMed Ltd, Budapest, Hungary) from the right arm. PWVao and AIX% derived from the Arteriograph analyses have been validated against an invasive method in adults. The correlation of invasively assessed PWVao and AIX% to PWVao and AIX% measured by the Arteriograph has been found to be strong ($r > 0.90$) with a reasonable agreement between oscillometric tonometry and invasive methods²⁸. Before the measurement, participants were asked to rest in a supine position for 10 minutes. Age and sex-specific SDS-norm values for PWVao and AIX% were calculated based on the data of over a 4500 Caucasian children and adolescents^{29,30}. A higher PWVao indicates a higher aortic stiffness and a higher AIX% indicates a higher peripheral arterial tone.

Systolic blood pressure was also assessed by the Arteriograph device (TensioMed Ltd, Budapest, Hungary) in a supine position after ten-minute rest.

Statistical methods

Basic characteristics between boys and girls were compared using the Student's t-test, the Mann-Whitney U-test, or the Chi Square-test. The associations of cardiorespiratory fitness and body adiposity as independent variables with PWVao and AIX% as dependent variables were studied using linear regression analyses adjusted for age and sex. The differences in PWVao and AIX% between sport participants and non-sport participants were investigated using general linear models (GLM) adjusted for age and sex. The data on the associations of cardiorespiratory fitness, body adiposity, and sports participation with PWVao and AIX% were also mutually adjusted.

All data were additionally controlled for systolic blood pressure, the mode of exercise testing, and diagnose (cardiovascular disease vs. other).

Receiver operating characteristics (ROC) curves were used to investigate the optimal cutoff for $\text{VO}_{2\text{peak}}$, waist circumference, BMI-SDS, and body fat percentage to identify children and adolescents at or over +1SD of PWVao and AIX% reference values^{29,30}. The area under the curve (AUC) is considered a measure of the effectiveness of the predictor variable to correctly identify children and adolescents at or above +1SD of PWVao and AIX% (sensitivity) and to correctly identify participants (specificity) below +1SD of PWVao and AIX%. An AUC of 1 represents the ability to perfectly identify children and adolescents at or above +1SD of PWVao and AIX% from other participants, whereas an AUC of 0.5 indicates no greater predictive ability than chance alone.

The optimal cutoff was determined by the Youden index³¹, which is the maximum value of J that is computed as: sensitivity + specificity - 1.

The effect modification of sex was investigated by general linear models (GLM). Because we found no statistically significant sex-interactions between the measures of cardiorespiratory fitness and body adiposity and the outcome variables, we performed all analyses with data of boys and girls combined.

Student's t-test, the Mann-Whitney U-test, the Chi Square-test, GLM analyses, and the linear regression analyses were performed using the SPSS Statistics, Version 23.0 (IBM Corp., Armonk, NY, USA). The ROC curve analyses were performed using MedCalc Statistical Software, Version 16.1 (MedCalc Software bvba, Ostend, Belgium). A P-value of < 0.05 was considered as statistically significant.

RESULTS

Descriptive characteristics

Complete data on variables used in the present analyses were available for 140 children and adolescents with chronic diseases or physical disabilities (86 boys, 54 girls). Children and adolescents who were excluded (N=37, 20 boys, 17 girls) from the present analyses because of the missing data did not differ from those who were included in waist circumference or BMI-SDS. Children and adolescents who were included in the present analyses were slightly older than those who were excluded from the present analyses ($P=0.029$).

Among the included children and adolescents, boys were taller, had a lower body fat percentage, higher VO_{2peak} , and a lower AIX% than girls (Table 1). Seventy-six (56%) of the included children and adolescents participated in sports. Altogether 17 children and adolescents had a cardiovascular disease, four had a pulmonary disease, 10 had a metabolic disease, 11 had a musculoskeletal/orthopedic disability, 82 had a neuromuscular disease/disability, six had an immunological/hematological disease, three had cancer, and seven had epilepsy (Table 1). There were no age-differences among the children and adolescents in different diagnosis groups ($P=0.682$).

Table 1: Descriptive characteristics of the study population.

	All	Boys (N=84)	Girls (N=56)	P-value
Age (years)	14.3 (2.7)	14.1 (2.7)	14.7 (2.9)	0.078
Body height (cm)	160 (14.3)	162.7 (15.9)	157.9 (10.9)	0.033
Body weight (kg)	54.8 (16.6)	56.7 (18.4)	51.7 (12.7)	0.059
Body mass index* (kg/m ²)	20.0 (5.2)	20.2 (5.6)	19.7 (5.1)	0.681
Body mass index-standard deviation score	0.68 (1.3)	0.83 (1.3)	0.43 (1.3)	0.078
Prevalence of overweight (%)	39.3	43	33.3	0.253
Waist circumference (cm)	75.7 (13.3)	75.6 (14.2)	75.9 (12.1)	0.888
Waist circumference- standard deviation score	0.8 (1.3)	0.7 (1.5)	1.1 (1.1)	0.062
Body fat percentage (%)	23.9 (9.7)	21.2 (9.4)	28.1 (8.7)	<0.001
Peak oxygen uptake (L/min)	2.1 (1.0)	2.5 (0.9)	1.8 (0.4)	<0.001
Peak oxygen uptake (ml/kg/min)*	40.0 (15.0)	44.0 (15.0)	36.0 (11.0)	<0.001
Peak oxygen uptake (ml/FFM/min)*	52.9 (13.7)	56.5 (14.8)	51.1 (8.1)	<0.001
Aortic pulse wave velocity (m/s)*	5.8 (1.3)	5.8 (1.2)	5.8 (1.1)	0.098
Aortic pulse wave velocity-standard deviation score*	-0.17 (1.71)	-0.21 (1.7)	-0.04 (1.7)	0.189
Aortic augmentation index (%)*	9.0 (10.4)	7.7 (11.4)	10.1 (10.3)	0.005
Aortic augmentation index (%) -standard deviation score *	0.19 (1.23)	0.17 (1.2)	0.25 (1.2)	0.245

FFM, fat free mass. The data are mean (standard deviations), median (interquartile range*), or percentages and the P-values from the t-test for independent samples for continuous variables with normal distribution and Mann-Whitney U-test for continuous variables with skewed distribution, or Chi-square for prevalence of overweight.

Associations of cardiorespiratory fitness body adiposity, and sports participation with arterial stiffness

VO_{2peak} per body weight and VO_{2peak} per FFM were inversely and waist circumference directly associated with PWVao after adjustment for age and sex (Table 2). However, the relationship of VO_{2peak} per body weight (β =-0.133, 95% CI = -0.315 to 0.050, P=0.152) and VO_{2peak} per FFM (β =-0.137, 95% CI = -0.291 to 0.017, P=0.082) to PWVao were no longer statistically significant after further adjustment for waist circumference. Additional adjustment for systolic blood pressure, the mode of exercise testing, or diagnose had no effect on these associations (data not shown).

Table 2: Associations of cardiorespiratory fitness and body adiposity with arterial stiffness in 140 children and adolescents with chronic diseases or physical disabilities.

	Aortic pulse wave velocity (m/s)			Aortic augmentation index (%)		
	B	95% CI	P	B	95% CI	P
Peak oxygen uptake (mL/kg/min)	-0.222	-0.386 to -0.059	0.008	-0.100	-0.271 to 0.070	0.247
Peak oxygen uptake (mL/fat free mass/min)	-0.173	-0.329 to -0.017	0.030	-0.120	-0.281 to 0.041	0.142
Waist circumference (cm)	0.254	0.093 to 0.414	0.002	-0.215	-0.381 to -0.049	0.012
Body mass index-standard deviation score	0.141	-0.026 to 0.307	0.097	-0.218	-0.382 to -0.054	0.010
Body fat percentage (%)	0.036	-0.127 to 0.198	0.664	0.016	-0.150 to 0.183	0.847

The data are standardized regression coefficients and their 95% confidence intervals adjusted for age and sex.

Waist circumference and BMI-SDS were inversely associated with AIX% after adjustment for age and sex. Further adjustment for the measures of cardiorespiratory fitness or sports participation had no effect on these associations (data not shown). However, further adjustment for systolic blood pressure slightly attenuated the association between waist circumference and AIX% ($\beta=-0.166$, 95% CI = -0.343 to 0.010, $P=0.064$). Additional adjustment for the mode of exercise testing, or diagnose had no effect on these associations (data not shown).

Sports participation was not associated with PWVao or AIX% after adjustment for age and sex ($P>0.40$). The associations of cardiorespiratory fitness, body adiposity, sports participation with PWVao and AIX% remained similar when PWVao-SDS and AIX%-SDS were used as outcome measures or waist circumference-SDS was used as an independent variable (data not shown).

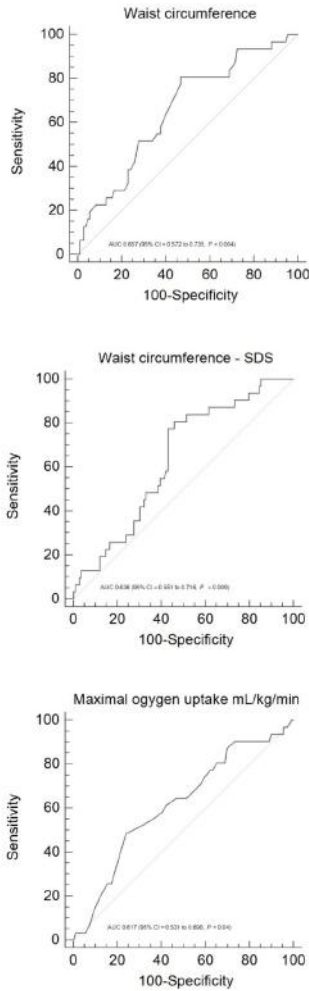


Figure 1: ROC curves of the efficacy of the measures of cardiorespiratory fitness and body adiposity to identify children and adolescents with chronic diseases or physical disabilities with increased aortic pulse wave velocity (≥ 1 standard deviation from reference values²⁸. AUC indicates the area under the curve (95% confidence interval, CI).

Ability of cardiorespiratory fitness and body adiposity to identify children and adolescents with increased arterial stiffness

The ROC curve analyses revealed that the optimal cutoff for waist circumference to identify children and adolescents ≥ 1 SD of PWV-SDS was >73 cm (95% CI = 66 to 78) with a sensitivity of 81%, a specificity of 53%, and the Youden index of 0.3386 (Figure 1.). The corresponding cutoff for waist circumference-SDS was >-0.05 units (95% CI = -0.09 to -0.02) with a sensitivity of 81%, a specificity 54%, and the Youden index of 0.3477. The optimal cutoff for VO_{2peak} per body weight was <35 ml/kg/min (95% CI 27 to 40) with a sensitivity of 48%, a specificity of 76%, and the Youden index of 0.2453. These analyses indicated that children and adolescents with a higher waist circumference and a lower VO_{2peak} per body weight were more likely to have increased arterial stiffness.

The optimal cutoff for VO_{2peak} per FFM to identify children and adolescents ≥ 1 SD of AIX%-SDS was <51 ml/kg FFM/min (95% CI 46 to 66) with a sensitivity of 64%, a specificity of 66%, and the Youden index of 0.3036 suggesting that children and adolescents with poorer cardiorespiratory fitness had increased AIX% compared to those with better cardiorespiratory fitness (Figure 2.).

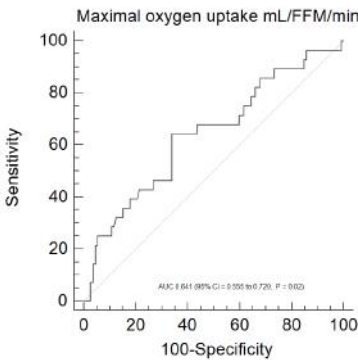


Figure 2: ROC curve of the efficacy of cardiorespiratory fitness to identify children and adolescents with chronic diseases or physical disabilities with increased peripheral arterial tone (augmentation index (AIX%) ≥ 1 standard deviation from reference values²⁹. AUC indicates the area under the curve (95% confidence interval, CI). FFM = fat free mass.

DISCUSSION

In the present study, we found inverse associations of the measures of cardiorespiratory fitness with aortic stiffness among children and adolescents with chronic disease or physical disabilities. However, the relationships between cardiorespiratory fitness and aortic stiffness were partly explained by waist circumference. We also found that a higher waist circumference had strong and consistent relationship to a higher aortic stiffness. Furthermore, higher waist circumference and BMI-SDS were associated with a lower peripheral arterial tone as indicated by a lower AIX%. Finally, we observed no relationship between sports participation and arterial stiffness.

To the best of our knowledge, this is the first study on the associations of directly assessed $\text{VO}_{2\text{peak}}$ with arterial stiffness among children and adolescents with chronic diseases or physical disabilities. In conjunction with some other studies, we found that better cardiorespiratory fitness was linked to lower aortic stiffness^{5,6}. However, the associations of $\text{VO}_{2\text{peak}}$ per body weight and $\text{VO}_{2\text{peak}}$ per FFM with arterial stiffness were partly explained by waist circumference. Similarly, one study found that an inverse association between cardiorespiratory fitness and arterial stiffness was explained by body adiposity among children aged 10 years³². These results are in line with the observations that the associations between $\text{VO}_{2\text{peak}}$ relative to body weight and cardiometabolic risk factors are confounded by body adiposity^{33,34}.

In our previous study, maximal work load by FFM as a measure of cardiorespiratory fitness, was inversely associated with arterial stiffness in children aged 6–8-years independent of body fat percentage^{34,35}. We found that $\text{VO}_{2\text{peak}}$ relative to FFM was attenuated after further adjustment for waist circumference, but remained at borderline significance. Furthermore, controlling for BMI-SDS or body fat percentage had no effect on the relationship between $\text{VO}_{2\text{peak}}$ relative to FFM and arterial stiffness. These results together indicate that better cardiorespiratory fitness is related to more compliant arteries and that this relationship is not completely due to lower levels of body adiposity in subjects with higher cardiorespiratory fitness. However, more research on the associations of cardiorespiratory fitness with arterial fitness with an appropriate scaling of $\text{VO}_{2\text{peak}}$ are warranted to further clarify the independent role of higher cardiorespiratory fitness of reduced arterial stiffness in children and adolescents.

Nevertheless, our results suggest in addition to weight management, improvements in cardiorespiratory fitness may improve arterial stiffness in children and adolescents with chronic diseases or physical disabilities.

Previous studies have suggested a threshold of 35–46 mL/kg/min for $\text{VO}_{2\text{peak}}$ to identify apparently healthy children with increased cardiometabolic risk³⁶. We found that a $\text{VO}_{2\text{peak}}$ per body weight lower than 35 mL/kg/min was the optimal threshold for identifying those with increased PWVao and the threshold for identifying children and adolescents with increased AIX% was 51 mL/FFM/min. These slight differences in the cutoffs are probably due to the differences in the outcome measures, methods used to assess cardiorespiratory fitness and to measure or estimate $\text{VO}_{2\text{peak}}$ as well as the populations studied. We also analyzed boys and girls together to achieve a better statistical power whereas other studies have provided separate cutoffs for boys and girls. However, our results along with the results from previous studies suggest that $\text{VO}_{2\text{peak}}$ below 35–40 mL/kg/min is related to increased cardiometabolic risk in children and adolescents.

Obesity has been consistently linked to increased arterial stiffness in children and adolescents⁸. Similarly, we found that particularly waist circumference was directly associated with arterial stiffness in children and adolescents with chronic diseases and physical disabilities independent of cardiorespiratory fitness. However, we found relatively weak associations of BMI-SDS and body fat percentage with arterial stiffness. An explanation for these inconsistent findings may be that abdominal adiposity has a pronounced negative impact on arterial health in children and adolescents³⁷. Furthermore, we found that higher levels of body adiposity were related to lower arterial tone at rest. We³⁵ and others³⁸ have previously observed inverse associations between body adiposity and peripheral arterial tone in children and adolescents. One reason for these observations may be that overweight may increase arterial diameter and thereby compensate the negative effects of body adiposity on arterial stiffness³⁸.

We observed that waist circumference exceeding 73 cm and waist circumference-SDS above -0.05 units were the optimal cut-offs for identifying children and adolescents with increased arterial stiffness. These cut-offs corresponding to an average waist circumference in the Dutch population, suggest that waist circumference already at normal range at the population level is associated with increased arterial stiffness in children and adolescents with chronic diseases or physical disabilities. These findings are supported by the study showing an increased cardiovascular mortality over a 40

year follow-up already among those who had a BMI at the mid-normal range during adolescence³⁹.

Previous studies have found either inverse or no association between habitual physical activity and the measures of arterial stiffness in children and adolescents^{13,40,41}. We observed no differences in arterial stiffness between sports participants and non-sports participants. One reason for our observations may be that sports participation among children and adolescents with chronic diseases or physical disabilities is insufficient in frequency, duration, or intensity to elicit favorable effects on arterial stiffness. Accordingly, there is some evidence that despite the high prevalence of sports participation, the majority of children and adolescents with chronic diseases and physical disabilities fail to meet the physical activity recommendations⁴². Another explanation may be that there may not be a large difference in total habitual physical activity among sports participants and non-participants²³.

The strengths of the present study include valid and reproducible methods used to assess cardiorespiratory fitness, body composition, and arterial stiffness in a relatively large sample of children and adolescents with chronic diseases or physical disabilities. This study had few limitations. We were not able to assess pubertal status of the participants, nor were dietary factors recorded. We also used sports participation as a proxy for physical activity instead of objectively measured physical activity. We defined chronic disease using a non-categorical approach that considers chronically-ill young people as one population rather than specific disease classes⁴³. The current sample size prohibited a sub-group analysis in specifying the disease classes. Furthermore, we analyzed boys and girls combined because of the limited statistical power to analyze boys and girls separately. We found no evidence on the effect modification of sex, but the results of some previous studies suggest that the association between cardiorespiratory fitness and arterial health is stronger in boys than in girls⁷. Furthermore, longitudinal studies are needed to study whether the effects of arterial stiffness during childhood and adolescence on cardiovascular diseases in adulthood are different in boys and girls. In conclusion, lower cardiorespiratory fitness and a higher waist circumference were associated with a higher arterial stiffness in children and adolescents with chronic diseases or physical disabilities in the present study. However, the associations of cardiorespiratory fitness were partly explained by waist circumference. Furthermore, we found no association between sports participation and arterial stiffness. We also found that thresholds of $<35 \text{ mL/kg/min}$ for $\text{VO}_{2\text{peak}}$, $>73\text{cm}$ for waist circumference, and >-0.05

units for waist circumference-SDS were associated with increased arterial stiffness. Taken together, these results suggest that interventions aiming to decrease body adiposity and to improve cardiorespiratory fitness may improve arterial stiffness children and adolescents with chronic diseases or physical disabilities. However, intervention studies are warranted to confirm the findings of the present study.

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REFERENCES

1. Piepoli MF, Hoes AW, Agewall S, Albus C, Brotons C, Catapano AL, Cooney MT, Corra U, Cosyns B, Deaton C, Graham I, Hall MS, Hobbs FDR, Lochen ML, Lollgen H, Marques-Vidal P, Perk J, Prescott E, Redon J, Richter DJ, Sattar N, Smulders Y, Tiberi M, van der Worp HB, van Dis I, Verschuren WMM, Binno S. 2016 european guidelines on cardiovascular disease prevention in clinical practice: The sixth joint task force of the european society of cardiology and other societies on cardiovascular disease prevention in clinical practice (constituted by representatives of 10 societies and by invited experts)developed with the special contribution of the european association for cardiovascular prevention & rehabilitation (EACPR). *Eur Heart J* 2016;37(29):2315-2381. doi: 10.1093/eurheartj/ehw106.
2. McGill HC,Jr, McMahan CA, Herderick EE, Malcom GT, Tracy RE, Strong JP. Origin of atherosclerosis in childhood and adolescence. *Am J Clin Nutr* 2000;72(5 Suppl):1307S-1315S. doi: 10.1093/ajcn/72.5.1307s.
3. Fernhall B, Agiovlasitis S. Arterial function in youth: Window into cardiovascular risk. *J Appl Physiol* (1985). 2008;105(1):325-333. doi: 10.1152/japplphysiol.00001.2008.
4. Vlachopoulos C, Aznaouridis K, Stefanadis C. Prediction of cardiovascular events and all-cause mortality with arterial stiffness: A systematic review and meta-analysis. *J Am Coll Cardiol* 2010;55(13):1318-1327. doi: 10.1016/j.jacc.2009.10.061.
5. Pahkala K, Laitinen TT, Heinonen OJ, et al. Association of fitness with vascular intima-media thickness and elasticity in adolescence. *Pediatrics* 2013;132(1):e77-84. doi: 10.1542/peds.2013-0041.
6. Ferreira I, Twisk JW, Stehouwer CD, van Mechelen W, Kemper HC. Longitudinal changes

- in .VO2max: Associations with carotid IMT and arterial stiffness. *Med Sci Sports Exerc* 2003;35(10):1670-1678. doi: 10.1249/01.MSS.0000089247.37563.4B.
7. Ferreira I, Twisk JW, Van Mechelen W, Kemper HC, Stehouwer CD, Amsterdam Growth and Health Longitudinal Study. Current and adolescent levels of cardiopulmonary fitness are related to large artery properties at age 36: The amsterdam growth and health longitudinal study. *Eur J Clin Invest* 2002;32(10):723-731. doi: 1066.
 8. Cote AT, Phillips AA, Harris KC, Sandor GG, Panagiotopoulos C, Devlin AM. Obesity and arterial stiffness in children: Systematic review and meta-analysis. *Arterioscler Thromb Vasc Biol* 2015;35(4):1038-1044. doi: 10.1161/atvhaba.114.305062.
 9. Pahkala K, Heinonen OJ, Lagstrom H, et al. Vascular endothelial function and leisure-time physical activity in adolescents. *Circulation* 2008;118(23):2353-2359. doi: 10.1161/circulationaha.108.791988.
 10. Pahkala K, Heinonen OJ, Simell O, et al. Association of physical activity with vascular endothelial function and intima-media thickness. *Circulation* 2011;124(18):1956-1963. doi: 10.1161/circulationaha.111.043851.
 11. Ried-Larsen M, Grontved A, Kristensen PL, Froberg K, Andersen LB. Moderate-and-vigorous physical activity from adolescence to adulthood and subclinical atherosclerosis in adulthood: Prospective observations from the european youth heart study. *Br J Sports Med* 2015;49(2):107-112. doi: 10.1136/bjsports-2013-092409.
 12. Walker DJ, MacIntosh A, Kozyrskyj A, Becker A, McGavock J. The associations between cardiovascular risk factors, physical activity, and arterial stiffness in youth. *J Phys Act Health* 2013;10(2):198-204. doi: 2011-0121.
 13. Melo X, Santa-Clara H, Pimenta NM, et al. Intima-media thickness in 11- to 13-year-old children: Variation attributed to sedentary behavior, physical activity, cardiorespiratory fitness, and waist circumference. *J Phys Act Health*. 2015;12(5):610-617. doi: 10.1123/jpah.2013-0501.
 14. Reed KE, Warburton DE, Lewanczuk RZ, et al. Arterial compliance in young children: The role of aerobic fitness. *Eur J Cardiovasc Prev Rehabil* 2005;12(5):492-497. doi: 00149831-200510000-00012.
 15. Idris NS, Evelein AM, Geerts CC, Sastroasmoro S, Grobbee DE, Uiterwaal CS. Effect of physical activity on vascular characteristics in young children. *Eur J Prev Cardiol* 2015;22(5):656-664. doi: 10.1177/2047487314524869.
 16. van de Laar RJ, Ferreira I, van Mechelen W, Prins MH, Twisk JW, Stehouwer CD. Lifetime vigorous but not light-to-moderate habitual physical activity impacts favorably on carotid

- stiffness in young adults: The amsterdam growth and health longitudinal study. *Hypertension* 2010;55(1):33-39. doi: 10.1161/HYPERTENSIONAHA.109.138289.
17. Dias KA, Green DJ, Ingul CB, Pavey TG, Coombes JS. Exercise and vascular function in child obesity: A meta-analysis. *Pediatrics* 2015;136(3):e648-59. doi: 10.1542/peds.2015-0616.
18. Rimmer JA, Rowland JL. Physical activity for youth with disabilities: A critical need in an underserved population. *Dev Neurorehabil* 2008;11(2):141-148. doi: 10.1080/17518420701688649.
19. Kotte EM, Winkler AM, Takken T. Fitkids exercise therapy program in the Netherlands. *Pediatr Phys Ther* 2013;25(1):7-13. Accessed 7/7/2017 9:21:13 AM. doi: 10.1097/PEP.0b013e318276c9bf.
20. Neter JE, Schokker DF, de Jong E, Renders CM, Seidell JC, Visscher TL. The prevalence of overweight and obesity and its determinants in children with and without disabilities. *J Pediatr* 2011;158(5):735-739. doi: 10.1016/j.jpeds.2010.10.039.
21. Carlon SL, Taylor NF, Dodd KJ, Shields N. Differences in habitual physical activity levels of young people with cerebral palsy and their typically developing peers: A systematic review. *Disabil Rehabil*. 2013;35(8):647-655. doi: 10.3109/09638288.2012.715721.
22. Cheung YF. Arterial stiffness in the young: Assessment, determinants, and implications. *Korean Circ J* 2010;40(4):153-162. doi: 10.4070/kcj.2010.40.4.153.
23. Martin AA, Cotie LM, Timmons BW, Gorter JW, Macdonald MJ. Arterial structure and function in ambulatory adolescents with cerebral palsy are not different from healthy controls. *Int J Pediatr* 2012;2012:168209. doi: 10.1155/2012/168209.
24. Lankhorst K, van der Ende-Kastelijin K, de Groot J, et al. 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;4:796-015-1589-z. eCollection 2015. doi: 10.1186/s40064-015-1589-z.
25. Zwinkels M, Verschuren O, Lankhorst K, et al. 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;7:22-015-0016-7. eCollection 2015. doi: 10.1186/s13102-015-0016-7.
26. Dosa NP, Foley JT, Eckrich M, Woodall-Ruff D, Liptak GS. Obesity across the lifespan among persons with spina bifida. *Disabil Rehabil* 2009;31(11):914-920. doi: 10.1080/09638280802356476.
27. Schonbeck Y, Talma H, van Dommelen P, et al. Increase in prevalence of overweight in

- dutch children and adolescents: A comparison of nationwide growth studies in 1980, 1997 and 2009. *PLoS One* 2011;6(11):e27608. doi: 10.1371/journal.pone.0027608.
28. Horvath IG, Nemeth A, Lenkey Z, et al. Invasive validation of a new oscillometric device (arteriograph) for measuring augmentation index, central blood pressure and aortic pulse wave velocity. *J Hypertens* 2010;28(10):2068-2075. doi: 10.1097/HJH.0b013e32833c8a1a.
 29. Hidvegi EV, Illyes M, Benczur B, et al. Reference values of aortic pulse wave velocity in a large healthy population aged between 3 and 18 years. *J Hypertens* 2012;30(12):2314-2321. doi: 10.1097/HJH.0b013e328359562c.
 30. Hidvegi EV, Illyes M, Molnar FT, Cziraki A. Influence of body height on aortic systolic pressure augmentation and wave reflection in childhood. *J Hum Hypertens* 2015;29(8):495-501. doi: 10.1038/jhh.2014.118.
 31. Perkins NJ, Schisterman EF. The inconsistency of “optimal” cutpoints obtained using two criteria based on the receiver operating characteristic curve. *Am J Epidemiol* 2006;163(7):670-675. doi: kwj063.
 32. Sakuragi S, Abhayaratna K, Gravenmaker KJ, et al. Influence of adiposity and physical activity on arterial stiffness in healthy children: The lifestyle of our kids study. *Hypertension* 2009;53(4):611-616. doi: 10.1161/HYPERTENSIONAHA.108.123364.
 33. Shaibi GQ, Cruz ML, Ball GD, et al. Cardiovascular fitness and the metabolic syndrome in overweight latino youths. *Med Sci Sports Exerc* 2005;37(6):922-928. doi: 00005768-200506000-00004.
 34. Tompuri T, Lintu N, Savonen K, et al. Measures of cardiorespiratory fitness in relation to measures of body size and composition among children. *Clin Physiol Funct Imaging* 2015;35(6):469-477. doi: 10.1111/cpf.12185.
 35. Veijalainen A, Tompuri T, Haapala EA, et al. Associations of cardiorespiratory fitness, physical activity, and adiposity with arterial stiffness in children. *Scand J Med Sci Sports* 2016;26(8):943-950. doi: 10.1111/sms.12523.
 36. Adegboye AR, Anderssen SA, Froberg K, et al. Recommended aerobic fitness level for metabolic health in children and adolescents: A study of diagnostic accuracy. *Br J Sports Med* 2011;45(9):722-728. doi: 10.1136/bjism.2009.068346.
 37. Arnberg K, Larnkjaer A, Michaelsen KF, Molgaard C. Central adiposity and protein intake are associated with arterial stiffness in overweight children. *J Nutr* 2012;142(5):878-885. doi: 10.3945/jn.111.150672.
 38. Cote AT, Harris KC, Panagiotopoulos C, Sandor GG, Devlin AM. Childhood obesity and cardiovascular dysfunction. *J Am Coll Cardiol* 2013;62(15):1309-1319. doi: 10.1016/j.

- jacc.2013.07.042.
39. Twigg G, Yaniv G, Levine H, et al. Body-mass index in 2.3 million adolescents and cardiovascular death in adulthood. *N Engl J Med* 2016;374(25):2430-2440. doi: 10.1056/NEJMoa1503840.
 40. Palve KS, Pahkala K, Magnussen CG, et al. Association of physical activity in childhood and early adulthood with carotid artery elasticity 21 years later: The cardiovascular risk in young finns study. *J Am Heart Assoc* 2014;3(2):e000594. doi: 10.1161/JAHA.113.000594.
 41. Haapala EA, Vaisto J, Veijalainen A, et al. Associations of objectively measured physical activity and sedentary time with arterial stiffness in pre-pubertal children. *Pediatr Exerc Sci* 2017;29(3):326-335. doi: 10.1123/pes.2016-0168.
 42. Ng K, Rintala P, Tynjala J, et al. Physical activity trends of finnish adolescents with long-term illnesses or disabilities from 2002-2014. *J Phys Act Health* 2016;13(8):816-821. doi: 10.1123/jpah.2015-0539.
 43. Stein RE, Jessop DJ. A noncategorical approach to chronic childhood illness. *Public Health Rep* 1982;97(4):354-362.



7

Sports participation related to injuries and illnesses among youth with chronic diseases: results of the health in adapted youth sports study

Kristel Lankhorst

Janke de Groot

Tim Takken

Frank Backx

ABSTRACT

Background. Although sports participation leads to important health enhancement for youth with chronic diseases or physical disabilities (CDPD), it may pose an increased risk for injury or illness. This study investigated the incidence, type, severity and risks to (sports-related) injuries and illnesses among youth with CDPD.

Methods. For one calendar year, every two weeks, the characteristics of injuries and illnesses were registered by an online questionnaire and phone-based interview. Physical activity level was measured with the Activ8 during one week. Complete data was available of 103 youngsters with CDPD (61 boys, 42 girls), with a mean age of 14.4 (SD=2.7) years. The personal characteristics, the injury and illness rates per 1000 hours of PA were investigated per group of organized sports participation per week (0, 1 or ≥ 2 times p/wk).

Results. Almost half of the youngsters sustains one or more injuries or illnesses during one year, 46% and 42% resp. The injury rate per 1000 hours of PA between 0, 1 and ≥ 2 times per week of sports participation was 0.84, 1.88, 133 resp. and the illness rate was 1.87, 1.88 and 1.18 resp. The rates were not statistically different. Most reported health problems had no subsequent restriction (49%) or other minor consequences (21%) in school, physical education or sports participation. Most reported health problems were contusions (41%) at the lower extremity (74%) and flue plus fever (58%).

Conclusions. Participation in sports ≥ 2 times per week does not pose a significant increased risk in the incidence of injury or illness per 1000 hours of PA in youth with CDPD compared to once weekly or no sports participation. Athletes who participate in sports at least twice weekly get injured mostly during their sporting activities, while peers who do participate in sports once a week or not at all, get injured during less intense physical activities during physical education lessons, ADL or non-organized sports and play in leisure time. The social impact of injuries or illnesses was limited.

Single statement summarizing the clinical relevance:

Participation in sports at least twice weekly gives no higher risk in the incidence of injury per 1000 hours of physical activity in youth with CDPD compared with once or no participation in sports per week. Parents should encourage and support their children with CDPD to participate in sports, especially from a physical and psychosocial health perspective. Personalized strategies are needed and should be implemented into pediatric health care, PE and into the sports context to minimize the risk of injury during leisure time physical activities including organized sports participation in youth with CDPD.

INTRODUCTION

Sports participation has important health benefits in both healthy youth and peers with chronic diseases or physical disabilities (CDPD)¹⁻⁵. Studies has shown strong associations between being member of a sports club and the amount of moderate-vigorous physical activity (MVPA) and vigorous activity levels (VPA) in youth with CDPD; those youngsters with CDPD are twice likely to meet international physical activity (PA) recommendations⁵⁻⁷. Meeting adequate PA levels supports a healthier lifestyle. In addition, sports participation also has major positive health effects. It contributes to a better fitness, a higher degree of PA, a higher quality of life, a better self-image and children find themselves more athletic skilled^{4,5}. At the same time, sports participation is also known to lead to sports-related injuries and illnesses, as shown in adults, healthy youth and among youth with CDPD⁷⁻⁹.

Sports injuries among healthy youth have a considerable impact on their participation and performance in subsequent activities^{8,10}. A longer existing injury or illness can limit participation in sports or lead to dropping out of sports or fear to return to sports among healthy youth¹¹. Moreover, research among healthy youth shows that a low level of PA entails a higher risk of being injured when they become more active, while there is only limited evidence regarding sports participation and injuries in youth with CDPD^{12,13}. For youngsters with CDPD it is already a challenge to reach adequate levels of PA due to the existing social and environmental barriers for sports and exercise participation^{14,15}. It is even harder to pursue an active and healthy lifestyle through participation in sports, when sports participation is associated with (fear of) injuries and / or illnesses. In addition, there is a reasonable fear among parents of children with CDPD that sports participation undoubtedly leads to sport-related injuries. As a consequence of that injury, their child could experience additional limitations in their daily lives, i.e. being unable to perform their daily activities independently anymore or need more help with it from their parents or caregivers¹⁶. These negative experiences and believes with regard to sports participation can further limit the support to allow their child to be active in sports. In addition, a previous systematic review among adult athletes with disabilities recommends longitudinal studies to gain more understanding and insight in factors influencing injuries and illnesses¹⁷⁻²⁰.

Therefore the aim of this study is to investigate the incidence, type, severity and risks of (sports-related) injuries and illnesses among youth with CDPD using a longitudinal approach.

METHODS

Study design

The present study is part of the Health in Adapted Youth Sports (HAYS) Study, which involved an analysis of data about health-related fitness, PA and psychosocial health in youth with CDPD^{3-5,21}. This study was approved by the Medical Ethics Committee of the University Medical Center Utrecht, Utrecht, the Netherlands (METC number: 14-332/c). The present sub study is a longitudinal cohort study specifically evaluating injuries and illnesses related to sports participation. The procedures and protocols of the HAYS Study have been published previously in more detail²¹.

This study has two objectives; 1) to determine the incidence, type and severity of injuries or illnesses, 2) to calculate the injury and illness rates per 1000 hours of PA per group among youth with CDPD participating in organized sports once or twice a week and peers who do not participate in sports at all. These objectives are researched by means of an injury and illness registration system for one calendar year among youth with CDPD. This longitudinal cohort study followed Strengthening the Reporting of Observational studies in Epidemiology (STROBE) guidelines²².

Participants

Youngsters with CDPD were recruited for participation in the HAYS study between October 2014 and October 2016. The follow-up period and data collection of injuries and illnesses lasted till October 2017. The children and adolescents were recruited in the Netherlands among different patients associations, pediatric therapy practices, Wilhelmina Children's Hospital in Utrecht, De Hoogstraat Rehabilitation Center in Utrecht, Fitkids network, schools for special education for children with a disability, and sports clubs. Young athletes were recruited from a broad range of participation in sports: from recreational level to high level competitive sports.

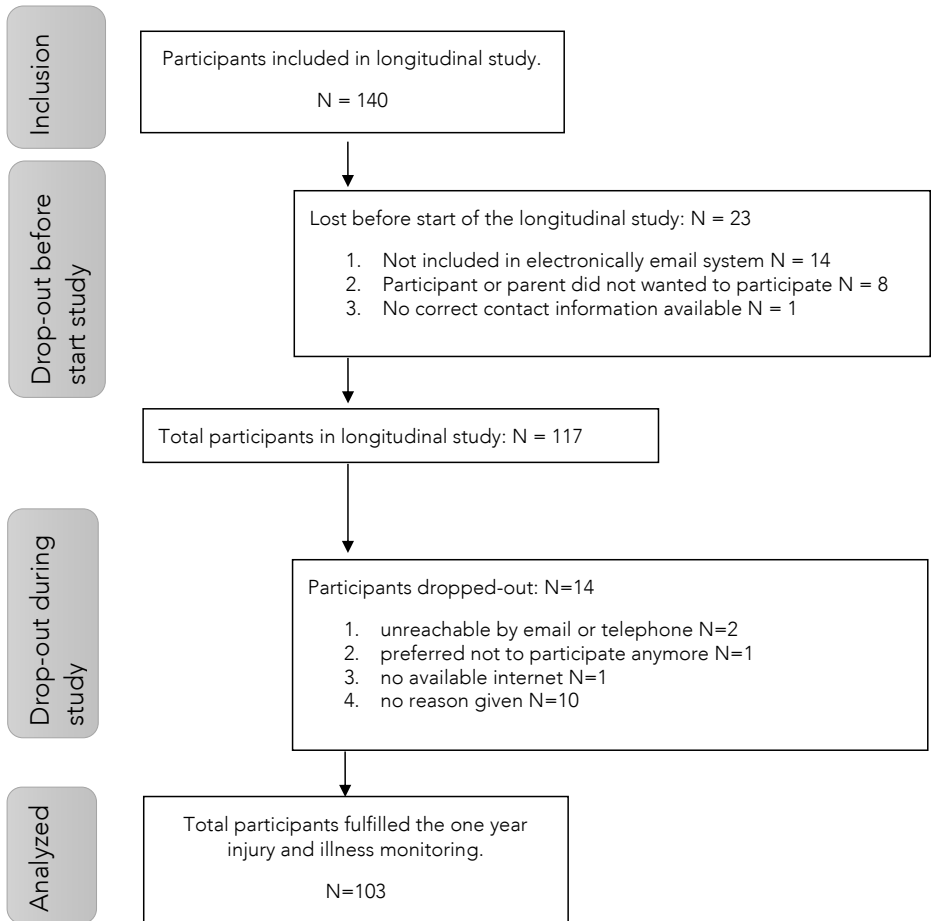
Participants were eligible for this study when they were ambulatory, aged from 8 to 19 years with CDPD, and diagnosed with one or more cardiovascular, pulmonary, musculoskeletal, metabolic or neuromuscular disorders according to the classification of the American College of Sports Medicine²³.

Informed consent was provided by all participants and by the parents of participants under 18 years of age. In line with Dutch law, no parental informed consent was required for participants aged 18 years and above.

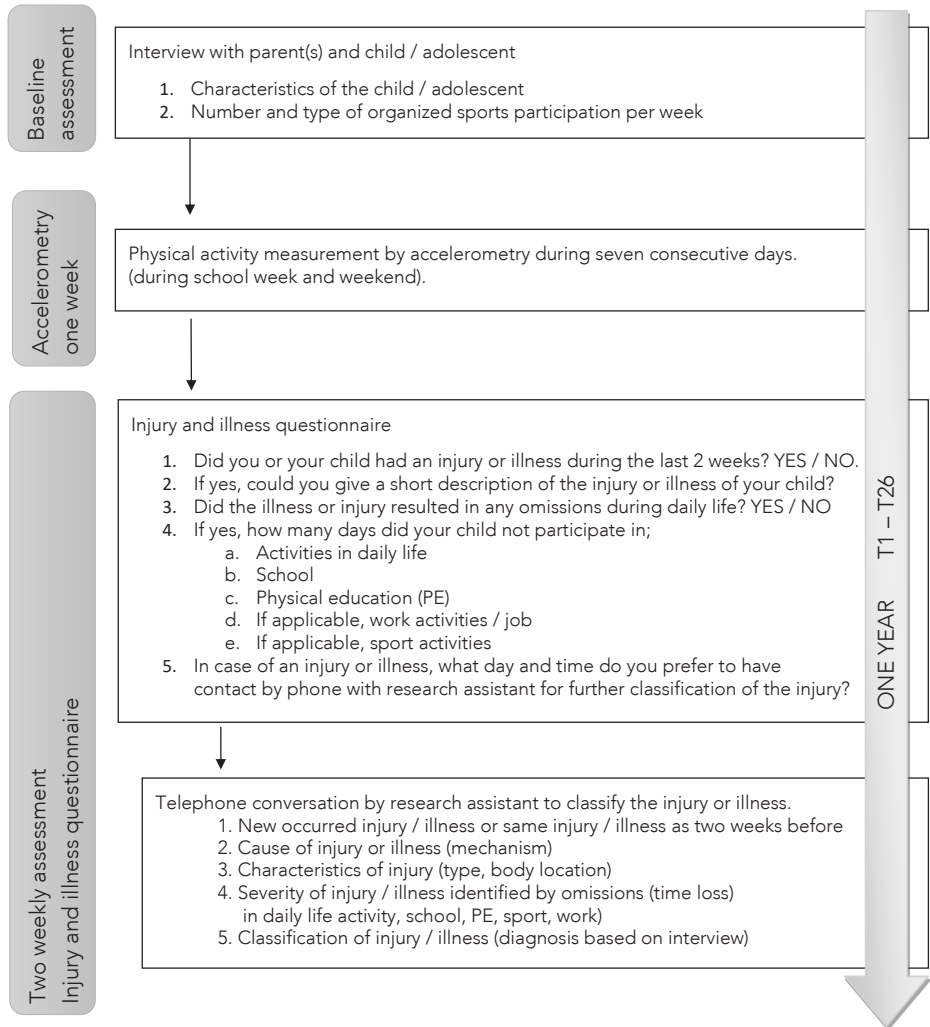
The HAYS cross-sectional sample involved 140 participants with CDPD, with a mean age (SD) of 14.4 (2.7) years (Table 1 and flowchart 1).

Procedure and data collection

An overview of the followed procedures and data collection of the study are summarized in flowchart 2. Data collection consisted of; 1) a baseline assessment with the child or young adult and parent(s), 2) objectively measured PA level of the participants during one school week and, 3) a two weekly registration of each injury or illness for one calendar year that followed after the baseline assessment.

Flowchart 1: Number of participants included and dropped-out in the longitudinal study.

Flowchart 2: Online questionnaire and phone based assessment of injury or illness.



The characteristics of the child or young adult were collected by the baseline assessment, including age, sex, medical diagnosis, participation in physical education (PE) and the number and type of sports participation in organized sports per week. The medical diagnoses were further classified into categories according to the American College of Sports Medicine (23). Sports participation was identified using the following three standardized questions used by the Dutch Institute of Health and Environment²⁴:

- 1) do you participate in sports?
- 2) what is / are the type of organized sport(s)?
- 3) what is the frequency of participation in organized sports per week?

In this study, participants were classified by the frequency of participation in organized sports per week to gain insight into sports-related injuries, non-sports-related injuries i.e. injuries related to PA during leisure time. This resulted in the following three groups: group 0 = no participation in organized sport at all; group 1 = sports participation at sport club one time per week, group 2 = sports participation at sport club two or more times per week.

We also investigated whether the more serious sports participants (two or more times of sports participation p/wk.) run a higher risk of being injured or ill compared to the less serious peers who participate in sports once per week (group 1) and not at all (group 0). Group 1 and 0 were taken together as one group for this additional analysis. PA was measured using an activity monitor, the Activ8 (2M Engineering Ltd. Valkenswaard, The Netherlands). The Activ8 is a valid one-sensor ambulatory monitoring system and has been validated for use in youth with and without motor impairments²⁵. Each subject wore the sensor on the dominant leg, fixed with Tegaderm™ (3M, Delft, the Netherlands) waterproof skin tape during seven consecutive days for 24 hours each day. The type, duration and frequency of PA in daily life (active vs inactive) were measured during a school week and one weekend. In order to calculate and interpret the data of waking hours gathered with the Activ8, also sleeping time was recorded in a diary. The total active time in minutes per day during leisure time, PE and organized sports per week was used for calculations. The analysis and results of the PA data of the studied children and adolescents of the HAYS study are described elsewhere in more detail¹⁵.

The monitoring of injuries and / or illnesses during the one calendar year follow-up period was measured by use of an online questionnaire. This questionnaire was developed

based on recommendations of the Dutch Ministry of Health, Welfare and Sport (VWS) (26) and designed in Formdesk, an online web-based tool (Innovero Software Solutions B.V., Wassenaar, The Netherlands). Following the baseline assessment, the participants and / or their parents received an email with a hyperlink to the online questionnaire every two weeks during one calendar year (flowchart 2). The questionnaire included five simple questions concerning suffered injuries and / or illnesses during the past two weeks, resulting in 26 measurements per participant. The participants received a reminder by e-mail when the questionnaire was not completed within three days. After three more days, the research assistant contacted the participant by telephone when the questionnaire was not completed, preventing an incomplete dataset. We performed a structured interview by phone in case of an injury or illness reported in the online questionnaire. In a personal interview we registered the type and body location of the injury or type of illness and the severity of the injury or illness.

Definitions of injury and illness

The injury definition according to the National Athletic Injury Registration System (NAIRS)²⁰ was adapted for use in this study and was defined as follows:

'Any new musculoskeletal pain, feeling or injury resulting from participation in recreational PA or sports and cause changes in physical activities including sports activities, regardless of whether or not time is lost from PA, sports training or competition.'

An illness was defined and adapted for use in this study as:

*'Any new illness that cause changes in physical activities including sports activities, regardless of whether or not time is lost from PA or sports training or competition.'*²⁷

Injuries and illnesses were further classified by the number of days of participation restriction in school and PE, in agreement with previous injury surveillance study²⁸ and according to the NAIRS²⁰. Reported injuries or illnesses were classified as non-time loss (NTL) injuries or illnesses when the injury or illness complaint only had a participation limitation on the same day of the injury or illness complaint and not the day after²⁸. Reported injuries or illnesses were classified as time-loss (TL) injuries or illnesses when there was a participation restriction of at least one day after the day of onset of the injury or illness complaint. To indicate the severity of the TL injuries and illnesses, the TL injury or illness were further subcategorized as follows: minor (1 -7 days lost), moderate (8 – 21 days lost) or severe (> 21 days)^{20,28}.

Primary outcome – absolute incidence, characteristics and severity of injuries and illnesses

The data regarding the occurrence of injuries (body location and type) and illnesses (type) and consequences (time-loss in days of return to school, PE and participation in sports) were collected for all three groups.

Secondary outcome - injury and illness rates per 1000 hours PA

The injury and illness rates per 1000 hours of PA was calculated for each group (group 0, 1 and 2). An injury or illness rate indicates how many injuries or illnesses occur per episodes of exposure to sport and / or PA.

The injury and illness rate is calculated by the following formula:

$$\text{Injury or illness rate} = \frac{(\text{the number of injuries or illnesses during one calendar year})}{(\text{the total exposure to PA during one calendar year})} \times 1000 \text{ hours.}$$

Statistical analysis

Descriptive analyses were used to profile characteristics of the three groups. ANOVA and post-hoc tests were used determining the differences between the groups for age, length, weight, BMI and BMI-age standard deviation score (SDS). The Kruskal-Wallis test was used determining the differences between the three groups for sex and prevalence of diseases or disabilities. Determining the differences between the three groups for the incidence of injuries and illnesses (injury and illness rates) during one calendar year follow-up the chi-square test was used. The dependent variable is the occurrence of injuries and illnesses within one calendar year and the amount of organized sports participation per week as independent variables. Analyses were performed using the SPSS Statistics, Version 23.0 (IBM Corp., Armonk, NY, USA), calculation of the 95% confidence intervals for injury and illness rates were performed using MedCalc for Windows, version 17.9 (MedCalc Software, Ostend, Belgium). A p value < 0.05 was considered statistically significant.

RESULTS

Participants' characteristics

Table 1: Characteristics of the participants per (sports)-group.

Sports participation per week	Group 0 (non-sports)	Group 1 (sports 1x/wk)	Group 2 (sports ≥2x/wk)	Total	<i>p</i> - value
Number of participants	18	21	64	103	
Number of boys (%)†	9 (50)	8 (38)	44 (69)	61 (59)	0.032§
Age in years (SD) ‡	15.4 (2.7)	14.2 (2.8)	14.1 (2.7)	14.4 (2.7)	0.219
Height in cm (SD) ‡	165.3 (9.5)	159.2 (13.2)	161.2 (14.7)	161.9 (12.5)	0.361
Weight in kg (SD) ‡	58.2 (13.5)	50.7 (16.4)	53.7 (17.6)	54.2 (15.8)	0.385
BMI (SD) ‡	21.1 (3.7)	19.7 (4.6)	20.2 (3.9)	20.2 (4.1)	0.546
BMI – age SDS (SD) ‡	0.59 (1.2)	0.11 (1.9)	0.51 (1.2)	0.44 (1.4)	0.449
Medical diagnosis (%)†					0.411
-Cardiovascular disease	3 (17)	4 (19)	4 (6)	11 (11)	
-Pulmonary disease	0	2 (9)	5 (8)	7 (7)	
-Metabolic disease	1 (6)	1 (5)	6 (9)	8 (8)	
-Musculoskeletal / orthopedic disability	1 (6)	1 (5)	6 (9)	8 (8)	
-Neuromuscular disease / disability	6 (33)	9 (43)	31 (49)	46 (46)	
-Immunological / hematological disease	5 (28)	4 (19)	6 (9)	15 (15)	
-Cancer	0	0	1 (2)	1 (1)	
-Epilepsy	2 (10)	0	5 (8)	7 (7)	

BMI; body mass index, SD; standard deviation, SDS; standard deviation score, † Kruskal-Wallis test for sex and prevalence of diseases or disabilities per group. ‡ ANOVA for age, length, weight, BMI and BMI-age SDS. § Significant difference.

Of 140 children and adolescents with CDPD who were invited to participate, 103 completed the prospective one calendar year injuries and illnesses monitoring and only their data were analyzed for the current study. Reasons for dropout during the one calendar year prospective monitoring are presented in flowchart 1. We lost 23 (16%) participants before the start of the longitudinal study mainly caused by faults in our electronically email system or participants / parents did not wanted to participate. Further, fourteen participants (10%) dropped-out during the follow-up period of the study. The drop-out of participants was not related to personal characteristics (i.e. diagnosis, age, sex) and was random divided between the three sub-groups.

The participants' characteristics (sex, age, medical diagnosis, height, weight, BMI and BMI SDS) and P-value per group are displayed in Table 1.

The vast majority of the group had a neuromuscular disease (46 out of 103), with Gross Motor Function Classification System (GMFCS) classification of 1 or 2. There was a significant difference between the number of participants per group and for sex in group 2, 44 boys versus 20 girls (P-value = 0.031).

Injury incidence, characteristics and severity

In total, 46% (N=47) of the participants with CDPD reported 86 injuries during one calendar year follow-up. Most of the registered injuries (70%) occurred in group 2 (exercising at least twice a week in sports). Almost half of the registered injuries in group 2 resulted in minor (1-7 days) and moderate (8-21 days) time loss in sports participation. While in group 0 and group 1 resp. 89% and 59% of the reported injuries could be classified as NTL, this was different in group 2 with only 40% being classified as NTL. Most of the injuries were articular contusions or distortions (41%), followed by muscles strains (24%), no severe injuries like concussions were reported (Table 3). The majority of the reported injuries were located at the lower extremity (74%). A third of the injuries occurred during organized sports (31%) and only in group 2.

Table 2: Incidence and severity of injuries and illnesses categorized according return to play (PE or sports) per group over one calendar year.

Sports participation per week	Group 0 (Non-sports)	Group 1 (Sports 1x/wk)	Group 2 (Sports ≥2x/wk)	Total (%)
Number of participants	18	21	64	103
INJURIES				
Number of participants with an injury (%)	5 (28)	7 (33)	35 (55)	47 (46)
Total number of injuries registered (%)	9 (10)	17 (20)	60 (70)	86
<i>Severity of injuries</i>				
No time loss (NTL) (%)	8 (89)	10 (59)	24 (40)	42 (49)
Minor (1-7 days) (%)	0	3 (18)	15 (25)	18 (21)
Moderate (8-21 days) (%)	0	1 (6)	13 (22)	14 (16)
Severe (>21 days) (%)	1 (11) *	3 (17)	8 (13)	12 (14)
ILLNESSES				
Number of participants with illnesses (%)	9 (50)	9 (43)	25 (39)	43 (42)
Total number of illnesses registered (%)	20; range 1-7 (22)	17; range 1-3 (19)	53 range; 1-6 (59)	90
<i>Severity of illnesses with restriction for return to school (N=90)</i>				
No time loss (NTL) (%)	5 (25)	2 (12)	10 (19)	17 (20)
Minor (1-7 days) (%)	12 (60)	12 (71)	31 (59)	55 (64)
Moderate (8-21 days) (%)	1 (5)	3 (18)	6 (11)	10 (12)
Severe (>21 days) (%)	2 (10)	0	6 (11)	8 (9)
<i>Severity of illnesses for return to PE / Sports* (N=90)</i>				
No time loss (NTL) (%)	15 (75)	10 (59)	9 (17)	34 (40)
Minor (1-7 days) (%)	2 (10)	4 (23)	30 (57)	36 (42)
Moderate (8-21 days) (%)	2 (10)	3 (18)	7 (13)	12 (14)
Severe (>21 days) (%)	1 (5)	0	7 (13)	8 (8)

Group 0 = non-sports group, group 1 = once a week of sports participation, group 2 = at least two times per week of sports participation. NTL; no time loss, i.e. injuries resulted in a participation restriction on the same day of injury complaint but no subsequent restriction, TL (minor, moderate and severe); injuries or illness resulted in restriction of participation of at least one day subsequent of the date of injury or illness complaint. *Severity of injury or illness; for group 0 it is operationalized as return to physical education, for group 1 and group 2 return to sports.

Table 3: Type, body location and context in which the injury occurred, categorized per group.

Type of injury	Group 0 (Non-sports)	Group 1 (Sports 1x/wk)	Group 2 (Sports ≥2x/wk)	Total (%)
Total number of injuries registered	9	17	60	86
Articular contusion / distortion (%)	3 (33)	4 (24)	28 (47)	35 (41)
Muscles strains (%)	2 (22)	7 (41)	12 (20)	21 (24)
Muscle, tendon (partial) rupture, hematoma / edema (%)	-	1 (6)	5 (8)	6 (7)
Epicondylitis / tendinitis, inflammation (%)	-	3 (17)	4 (7)	7 (9)
Open wound / blister (%)	2 (22)	1 (6)	4 (7)	7 (9)
(sub)luxation (%)	-	-	3 (5)	3 (3)
Fracture (%)	1 (11)	-	2 (3)	3 (3)
Other (chondropathy, ossification) (%)	1 (11)	1 (6)	2 (3)	4 (4)
Body location	Group 0 (Non-sports)	Group 1 (sports 1x/wk)	Group 2 (sports ≥2x/wk)	Total (%)
Total number of injuries registered	9	17	60	86
Lower extremity (n=64)				
Hip – upper leg (%)	-	-	11 (18)	11 (13)
Knee – lower leg (%)	4 (44)	7 (41)	15 (25)	26 (30)
Ankle (%)	-	2 (12)	12 (20)	14 (16)
Foot (%)	4 (44)	2 (12)	7 (12)	13 (15)
Upper extremity (N=11)				
Shoulder (%)	-	-	1 (2)	1 (1)
Fingers (%)	-	4 (23)	6 (10)	10 (12)
Spine (N=10)				
Upper (%)	1 (12)	-	5 (8)	6 (7)
Lower (%)	-	2 (12)	2 (3)	4 (5)
Other (face) (N=1)	-	-	1 (2)	1 (1)
Context in which injury occurred	Group 0 (Non-sports)	Group 1 (sports 1x/wk)	Group 2 (sports ≥2x/wk)	Total (%)
Total number of injuries registered	9	17	60	86
Organized sports (%)				27 (31)
Team (contact) sports ^a (%)	-	-	19 (32)	
Individual sports ^b (%)	-	-	8 (13)	
Non-organized sports and play in leisure time ^c (%)	6 (67)	3 (18)	14 (23)	23 (27)
Physical education (%)	1 (11)	6 (35)	5 (9)	12 (14)
Activities in daily living (walking, bicycling) (%)	2 (22)	8 (47)	14 (23)	24 (28)

^a Soccer, ice hockey, adapted volleyball, basketball, water polo, ^b road cycling, swimming, athletics, fitness, ^c jumping the trampoline, rowing, sailing, skiing.

Illness incidence, characteristics and severity

A total of 90 illnesses during one calendar year were reported by 43 participants (42%) (Table 2). Most of the illnesses were reported in group 2 (59%). Overall, the majority of the illnesses (64%) resulted in a minor time loss (1-7 days) for participation in school, no differences were seen between the three groups. Only 9% of the illnesses were severe (>21 days). The severity of the illness for return to sports was the highest for group 2 (57%; 1-7 days), compared to group 1 and group 0 for which most of the illnesses resulted in no time loss for return to PE or sports (75% and 59% resp). Flu and fever were the most commonly reported illnesses (58%) followed by symptoms of fatigue (18%). In addition, there were single cases of inflammation, asthma, migraine, epileptic attack, sleeping apnea, shingles and pertussis, which together accounted for 24% of the total reported illnesses.

Injury and illness rate per 1000 hours of PA

The calculated injury rate per 1000 hours of PA was for group 0, 1 and 2; 0.84, 1.88 and 1.33 resp. The illness rates per 1000 hours of PA was 1.87, 1.88 and 1.18 resp. for participation restriction on the same day of injury or illness complaint but no subsequent restriction. The differences between the three groups were not statistical different (Table 4 and 5). The illness rate per 1000 hours of PA differed significant between group 2 (N=64) compared to group 0 + group 1 taken together (N=39), $P = 0.028$.

Table 4: Injury rate per 1000 hours of physical activity and comparison of injury rates per group.

Group comparisons	Mean minutes PA per day (range min-max)	Cumulative hours of PA during one year per group	Number of injuries (95 % CI)	Injury rate (95 % CI)	IRD (95% CI) IRR (95% CI)	p-value†
Group 0 (non-sports) + group 1 (sports 1x/wk) Vs.	134.6 (59-244)	19693	26	1.32 (0.9 to 1.9)	IRD -0.015 (-0.6 to 0.6) IRR 0.99 (0.6 to 1.6)	0.962
Group 2 (sports ≥2x/wk)	171.8 (73-292)	44937	60 (45.8 to 77.2)	1.33 (1.0 to 1.7)		
Group 0 (non-sports) Vs.	146.2 (80-244)	10674	9 (4.1 to 17.1)	0.84 (0.38 to 1.6)	IRD 1.04 (-17.1 to 17.7)	0.275
Group 1 (sports 1x/wk)	123.6 (59-182)	9019	17 (9.9 to 27.2)	1.88 (1.1 to 3.1)		
Group 1 (sports 1x/wk) Vs.	123.6 (59-182)	9019	17 (9.9 to 27.2)	1.88 (1.1 to 3.1)	IRD 0.55 (-6.3 to 17.2)	0.387
Group 2 (sports ≥2x/wk)	171.8 (73-292)	44937	60 (45.8 to 77.2)	1.33 (1.0 to 1.7)		
Group 2 (sports ≥2x/wk) Vs.	171.8 (73-292)	44937	60 (45.8 to 77.2)	1.33 (1.0 to 1.7)	IRD 0.49 (-17.6 to 7.2)	0.295
Group 0 (non-sports)	146.2 (80-244)	10674	9 (4.1 to 17.1)	0.84 (0.38 to 1.6)		

PA; physical activity, CI; confidence interval, vs; versus, IRD; incidence rate difference, IRR; incidence rate ratio, † Determined with Chi-square test, * significant difference P-value ≤ 0.05.

Table 5: Illness rate per 1000 hours of physical activity and comparison of illness rates per group.

Group comparisons	Mean minutes PA per day (range min-max)	Cumulative hours of PA during one year per group	Number of illnesses (95 % CI)	Illness rate (95 % CI)	IRD (95% CI) IRR (95% CI)	p-value†
Group 0 (non-sports) + group 1 (sports 1x/wk) Vs.	134.6 (59-244)	19693	37 (26.1 to 51)	1.88 (1.4 to 1.2)	IRD = 0.7 (0.07 to 1.31) IRR = 1.6 (1.0 to 2.5)	0.028*
Group 2 (sports ≥2x/wk)	171.8 (73-292)	44937	53 (39.7 to 69.3)	1.18 (1.1 to 1.6)		
Group 0 (non-sports) Vs.	146.2 (80-244)	10674	20 (12.2 to 30.9)	1.87 (1.9 to 1.3)	IRD = -1.12 (-1.23 to 1.20) IRR = 0.99 (0.5 to 2.0)	0.986
Group 1 (sports 1x/wk)	123.6 (59-182)	9019	17 (9.9 to 27.2)	1.88 (1.9 to 1.3)		
Group 1 (sports 1x/wk) Vs.	123.6 (59-182)	9019	17 (9.9 to 27.2)	1.88 (1.9 to 1.3)	IRD = 0.7 (-0.1 to 1.5) IRR = 1.59 (0.9 to 2.8)	0.090
Group 2 (sports ≥2x/wk)	171.8 (73-292)	44937	53 (39.7 to 69.3)	1.18 (1.1 to 1.6)		
Group 2 (sports ≥2x/wk) Vs.	171.8 (73-292)	44937	53 (39.7 to 69.3)	1.18 (1.1 to 1.6)	IRD = 0.7 (1.5 to 0.1) IRR = 0.63 (0.4 to 1.1)	0.075
Group 0 (non-sports)	146.2 (80-244)	10674	20 (12.2 to 30.9)	1.87 (1.9 to 1.3)		

PA; physical activity, CI; confidence interval, vs; versus, IRD; incidence rate difference, IRR; incidence rate ratio, † Determined with Chi-square test, * significant difference P-value ≤ 0.05.

DISCUSSION

The aim of this study was to investigate the incidence, type, severity and risks of (sports-related) injuries and illnesses among youth with CDPD using a longitudinal approach. We found that participation in sports ≥ 2 times per week does not pose an increased risk in the incidence of injury or illness per 1000 hours of PA in youth with CDPD compared to once weekly or no sports participation.

Children and adolescents with CDPD are increasingly stimulated to participate in sports and exercise²⁹. Actual participation minimizes physical inactivity, optimizes physical functioning, promotes inclusion in society and enhances overall well-being in youth with CDPD^{4,5}. Given all these benefits, fear of injury frequently remains a barrier to participate in sports¹¹. The question rises whether all the major physical and psychosocial health benefits outweigh the risk of getting injured or ill through sports participation.

To the best of our knowledge this is the first longitudinal study on identifying and comparing injuries and illnesses among sporting and non-sporting youth with CDPD. We found no presumptive evidence that sports participation results in significant higher injury and illness rates among youth with CDPD. A current systematic review reported inconsistent research outcomes about the impact of sports participation on injury rates among healthy youth (age 6 to 15 years)³⁰. Some studies concluded that sports participation is the most risky to get or sustain an injury, while current literature in healthy youth without CDPD show that the absolute number of injuries occurring during leisure time and PE are consequently as high as during sports³⁰.

Our study showed comparable results according to absolute numbers of injuries and context where injuries occurred. Logically, when you do not participate in sports, you are not exposed to a higher risk that sports entails, only to risks related to daily life situations which is in line with our study. Non-sporting participants also get injured, not from sports participation but they get injured by less intense physical activities; PE lessons, ADL or non-organized sports and play in leisure time. Interestingly, youth participating in sport ≥ 2 times per week were less likely to get injured in daily life situations. We can suggest, they have better motor skills or participate in low-risk sports, which makes them less vulnerable to an injury.

Injury and illness type and severity

Although the absolute number of injuries and illnesses are high in sports participants, the types of injuries and illnesses reported in the current study have no or minimal

impact on being able to participate in school, PE or sports. The most reported injuries in our study were contusions / distortions or muscles strains, and located at the lower extremities. Our findings are in line with results of previous studies in both healthy youth and high school athletes with disabilities^{17,31}, while in Paralympic athletes most of the injuries were related to overuse (tissue inflammation and pain)³². According to time loss by sustaining an injury, our findings are similar with several studies, in which sports injuries suffered by high school athletes with disabilities and Paralympic adult athletes resulted in no or minor loss of training time^{18,31-33}. According to the severity of illnesses, preliminary results in Paralympic adult athletes shows that a higher training load results in a higher number of illnesses (infections) and the type of locomotion seems related to the incidence of illness, i.e. wheelchair athletes reported a high number of upper respiratory tract infections compared to able-bodied athletes³². The illness severity in the study of Fahger et al. (2017) was minor, 1-3 days of time loss of training, which is comparable with our study findings, although these studies are not comparative on study design, participants' age, medical diagnose and sports level with our research. For instance, the majority of the study population of Ramirez et al. (2008) had a mental disorder like autism and were on average older (18 years), whereas the study population of Fagher et al. (2017) were adult athletes with visual impairments, or were wheelchair dependent. The studies of Blauwet et al. (2016) and Derman et al. (2013) followed the athletes of the London 2012 Paralympic games during 14 days. All these studies, however, show similar results, that the severity of injuries and illnesses are low for peoples' ADL and / or sports participation. Remarkably, in our study, the incidence illness rate is the lowest in youngsters who participates in sports at least two times per week compared to their peers without any sports participation or only once. This result might suggests that regular participation in sports of at least two times a week could have a protective effect evolving illnesses.

Strengths and limitations

This study was the first large study to evaluate injuries and illnesses among sporting and non-sporting youth with CDPD during one calendar year. Strong points are the prospective longitudinal data monitoring to obtain all necessary data and the objective direct measurement of PA by using accelerometry. In addition, the incidence of injuries and illnesses are based on exposure of PA, which is a key factor^{34,35}.

Obviously, this study also has some limitations. We measured PA during one school

week and weekend, assuming that this measurement is a representative week. The injury and illness rate formula assumes that the random sample of seven days (school week) objectively measured PA distributed over the full study period represents the PA experience of the whole sample and can be used to derive estimates of the population experience over that period. Use of the PA measurements during one school week and extrapolated for one calendar year is a limitation of the study. Future research should measure PA during a longer period of time and during holidays, were PA levels are probably different from PA during school weeks. Further, the injuries and illnesses were subjective reported by the parent and / or child; not objectified by a physician or physical therapist as ideally recommended (34). Participants from all over the Netherlands were enrolled in our study, therefore a physical consultation of injury or illness was not feasible. As a solution, we choose a structured interview by phone to further analyze the injury or illness reported in the online questionnaire. Even so, feasibility of this methodology was time-consuming and required a high degree of precise working, i.e. checking the incoming questionnaires and the presence of an injury or illness. In a pilot study of Fagher et al, the use of a novel eHealth-based application for self-report in Paralympic athletes was generally feasible and usable³², and seemed less time-consuming compared to our technique. The use of new methods developed for Paralympic athletes may be recommended for use in future recreational-level research to collect this type of data. Moreover there were more boys compared to girls in our sport-2 group. This may have influenced our results as evidence in healthy youth shows that girls are at increased risk of injuries while participating in PA compared to boys. Low levels of PA and/or physical fitness seemed to increase injury incidence levels, but the exact mechanisms remains unclear³⁰. Future studies should take aspects like training history, training status and physical fitness level into account and may also investigate how the level of sport participation is related to injury and illness risks in order to develop risk profiles and injury prevention programs in more detail for youth with CDPD^{32,34}.

CONCLUSIONS

Participation in sports twice a week does not pose an increased risk in the incidence of injury or illness per 1000 hours of PA in youth with CDPD compared to once or no participation in sports per week. The impact of injuries or illnesses was only minor. Given the evident health benefits for youth with CDPD without obvious risks, sports participation at least twice weekly in youth with CDPD is highly recommended. A next

step would be to conduct studies to identify specific variables (i.e. physical fitness, type and training history of sports) that could prevent injuries and illnesses.

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REFERENCES

1. Janssen I, Leblanc AG. Systematic review of the health benefits of physical activity and fitness in school-aged children and youth. *Int J Behav Nutr Phys Act* 2010 May 11;7:40-5868-7-40.
2. Khan KM, Thompson AM, Blair SN, Sallis JF, Powell KE, Bull FC, et al. Sport and exercise as contributors to the health of nations. *Lancet* 2012 Jul 7;380(9836):59-64.
3. Haapala EA, Lankhorst K, de Groot J, Zwinkels M, Verschuren O, Wittink H, et al. 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 Jul;24(10):1102-1111.
4. Te Velde SJ, Lankhorst K, Zwinkels M, Verschuren O, Takken T, de Groot J, et al. Associations of sport participation with self-perception, exercise self-efficacy and quality of life among children and adolescents with a physical disability or chronic disease-a cross-sectional study. *Sports Med Open* 2018 Aug 15;4(1):38-018-0152-1.
5. Lankhorst K, Takken T, Zwinkels M, van Gaalen L, te Velde S, Backx F, et al. Sports participation, physical activity, and health-related fitness in youth with chronic diseases or physical disabilities: The Health in Adapted Youth Sports study. *J Strength Cond Res* 2019;Published Ahead-of-Print - Last Updated: May 02, 2019.
6. Ng K, Tynjala J, Sigmundova D, Augustine L, Sentenac M, Rintala P, et al. Physical activity among adolescents with Long-term illnesses or disabilities in 15 European Countries. *Adapt Phys Activ Q* 2017 Oct 06:1-10.
7. Ng KW, Tynjala J, Rintala P, Kokko S, Kannas L. Do adolescents with long-term

- illnesses and disabilities have increased risks of sports related injuries? *Inj Epidemiol* 2017 Dec;4(1):13-017-0112-0. Epub 2017 May 1.
8. Finch C, Cassell E. The public health impact of injury during sport and active recreation. *J Sci Med Sport* 2006 Dec;9(6):490-497.
 9. Emery CA. Risk factors for injury in child and adolescent sport: a systematic review of the literature. *Clin J Sport Med* 2003 Jul;13(4):256-268.
 10. Siesmaa EJ, Blitvich JD, Finch CF. A systematic review of the factors which are most influential in children's decisions to drop out of organised sport. In: Farelli AD (ed). *Sport Participation: Health Benefits, Injuries and Psychological Effects*. NY: Nova Science Publishers, Inc.; 2011.
 11. Siesmaa EJ, Blitvich JD, Telford A, Finch CF. Factors that are most influential in children's continued and discontinued participation in organized sport: The role of injury and injury risk perceptions. In: Farelli AD, editor. *Sport Participation: Health Benefits, Injuries and Psychological Effects*.. Nova Science Publishers, Inc.; 2011. p. 47-84.
 12. Bloemers F, Collard D, Paw MC, Van Mechelen W, Twisk J, Verhagen E. Physical inactivity is a risk factor for physical activity-related injuries in children. *Br J Sports Med* 2012 Jul;46(9):669-674.
 13. Verhagen E, Collard D, Paw MC, van Mechelen W. A prospective cohort study on physical activity and sports-related injuries in 10-12-year-old children. *Br J Sports Med* 2009 Dec;43(13):1031-1035.
 14. Bloemen MA, Backx FJ, Takken T, Wittink H, Benner J, Mollema J, et al. Factors associated with physical activity in children and adolescents with a physical disability: a systematic review. *Dev Med Child Neurol* 2015 Feb;57(2):137-148.
 15. Burghard M, de Jong NB, Vlieger S, Takken T. 2017 Dutch Report Card(+): Results From the first physical activity report card plus for Dutch youth with a chronic disease or disability. *Front Pediatr* 2018 Apr 30;6:122.
 16. Bloemen MA, Verschuren O, van Mechelen C, Borst HE, de Leeuw AJ, van der Hoef M, et al. Personal and environmental factors to consider when aiming to improve participation in physical activity in children with Spina Bifida: a qualitative study. *BMC Neurol* 2015 Feb 10;15:11-015-0265-9.
 17. Fagher K, Lexell J. Sports-related injuries in athletes with disabilities. *Scand J Med Sci Sports* 2014 Oct;24(5):e320-31.
 18. Blauwet CA, Cushman D, Emery C, Willick SE, Webborn N, Derman W, et al. Risk of injuries in Paralympic track and field differs by impairment and event discipline: A pro-

- spective cohort study at the London 2012 Paralympic Games. *Am J Sports Med* 2016 Jun;44(6):1455-1462.
19. Shi X, Shi J, Wheeler KK, Stallones L, Ameratunga S, Shakespeare T, et al. Unintentional injuries in children with disabilities: a systematic review and meta-analysis. *Inj Epidemiol* 2015 Dec;2(1):21-015-0053-4. Epub 2015 Sep 15.
20. van Mechelen W, Hlobil H, Kemper HC. Incidence, severity, aetiology and prevention of sports injuries. A review of concepts. *Sports Med* 1992 Aug;14(2):82-99.
21. Lankhorst K, van der Ende-Kastelijin K, de Groot J, Zwinkels M, Verschuren O, Backx F, et al. 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 Dec 22;4:796-015-1589-z. eCollection 2015.
22. von Elm E, Altman DG, Egger M, Pocock SJ, Gotsche PC, Vandenbroucke JP, et al. The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) Statement: guidelines for reporting observational studies. *Int J Surg* 2014 Dec;12(12):1495-1499.
23. Durstine LJ, Moore GE, Painter P, Roberts S,O. ACSM's Exercise Management for Persons with Chronic Diseases and Disabilities. 3rd ed.: Human Kinetics, Champaign, IL; 2009.
24. Hollander deL, Milder IE, Proper KI. Beweeg- en sportgedrag van mensen met een chronische aandoening of lichamelijke beperking, RIVM rapport, Bilthoven; 2015;2015-0064.
25. Lankhorst K, van den Berg-Emons RJ, Bussmann JBJ, Horemans HLD, de Groot JF. A novel tool for quantifying and promoting physical activity in youths with typical development and youths who are ambulatory and have motor disability. *Phys Ther* 2019 Mar 1;99(3):354-363.
26. Valkenburg H, van Nunen M, Ormel W, Vriend I. Handboek Epidemiologie Sportblessures, Stichting Consument en Veiligheid, Amsterdam, 2010;4.1.
27. Timpka T, Alonso JM, Jacobsson J, Junge A, Branco P, Clarsen B, et al. Injury and illness definitions and data collection procedures for use in epidemiological studies in Athletics (track and field): consensus statement. *Br J Sports Med* 2014 Apr;48(7):483-490.
28. Dompier TP, Powell JW, Barron MJ, Moore MT. Time-loss and non-time-loss injuries in youth football players. *J Athl Train* 2007 Jul-Sep;42(3):395-402.
29. Murphy NA, Carbone PS, American Academy of Pediatrics Council on Children With Disabilities. Promoting the participation of children with disabilities in sports, recreation, and physical activities. *Pediatrics* 2008 May;121(5):1057-1061.
30. Nauta J, Martin-Diener E, Martin BW, van Mechelen W, Verhagen E. Injury risk during

- different physical activity behaviours in children: a systematic review with bias assessment. *Sports Med* 2015 Mar;45(3):327-336.
31. Ramirez Marizen M. Sports injuries to high school athletes with disabilities. *Pediatrics* 2009-2;123(2):690-6.
 32. Fagher K, Jacobsson J, Dahlstrom O, Timpka T, Lexell J. An eHealth application of self-reported sports-related injuries and illnesses in Paralympic Sport: Pilot feasibility and usability study. *JMIR Hum Factors* 2017 Nov 29;4(4):e30.
 33. Derman W, Schwellnus M, Jordaan E, Blauwet CA, Emery C, Pit-Grosheide P, et al. Illness and injury in athletes during the competition period at the London 2012 Paralympic Games: development and implementation of a web-based surveillance system (WEB-IISS) for team medical staff. *Br J Sports Med* 2013 May;47(7):420-425.
 34. Weiler R, Van Mechelen W, Fuller C, Verhagen E. Sport Injuries Sustained by Athletes with Disability: A Systematic Review. *Sports Med* 2016 Aug;46(8):1141-1153.
 35. inch C. A new framework for research leading to sports injury prevention. *J Sci Med Sport* 2006 May;9(1-2):3-9; discussion 10.



Instruments measuring physical activity in persons who use a wheelchair: a systematic review of measurement properties

Kristel Lankhorst

Michiel Oerbekke

Rita van den Berg-Emons

Tim Takken

Janke de Groot

ABSTRACT

Objective. To systematically review the evidence evaluating validity or reliability of self-reported and device-based instruments to measure PA in individuals who use a wheelchair and making recommendations for the selection of PA outcomes tools .

Data Sources. Pubmed, Embase, and CINAHL were systematically searched.

Study Selection. Studies reporting measurement properties of instruments to assess PA in individuals who use a wheelchair.

Data Extraction. The COSMIN checklist was used to assess the methodological quality of the included studies and the measurement properties of instruments assessing PA were examined.

Data Synthesis. The search yielded 5341 records, 61 were considered relevant, 21 articles were included. A best evidence synthesis was performed on nine studies including four self-reported instruments and thirteen studies including eight device-based instruments. One study evaluated both self-reported and device-based instruments. The overall methodological quality of all studies ranged from poor to excellent. Variable levels of evidence were found for both the validity and reliability for self-reported instruments and for criterion validity for device-based instruments.

Conclusion. The PASIPD and PARA-SCI seems the most promising self-reported instruments for measuring the intensity of PA. Device-based instruments that can be used for measuring both the intensity and type of PA are the GENEactive, Actigraph GT3X+, Actiheart or the PAMS, showing moderate evidence for a positive rating of criterion validity. For measuring the type of PA the PAMS and VitaMove are suitable, showing both good evidence for a positive rating of criterion validity.

INTRODUCTION

A low level of physical activity (PA) is a well-known risk factor for many diseases, for example for developing cardiovascular diseases, osteoporosis and type 2 diabetes¹. Studies have shown that in comparison with the able-bodied population, manual wheelchair users (MWU) might even be more at risk for developing disease related to unfavourable PA, due to the long periods of sitting time in the wheelchair²⁻⁴. At the same time, it is often hypothesized that low levels of PA lead to a vicious cycle of deconditioning, even more decreased mobility and further decline of PA in daily life⁵.

Current standards promote a healthy lifestyle that consists optimal levels of PA⁶⁻⁸. Therefore optimizing levels of PA in MWU is an important treatment goal for health care professionals working with this population⁵. To further evaluate the effectiveness of interventions targeting PA, relationship between PA and health outcomes in MWU, valid instruments are needed to assess PA.

While both self-reported (subjective measurement) and device-based (objective measurement) instruments measuring PA have been developed and validated for clinical populations who are ambulatory, the use and the psychometric properties of these instruments are not that easily transferable to those who use a manual wheelchair for daily mobility.

There are two important factors of influence on the usage and the validity of instruments; 1) the sensor location and 2) applicability of current predictive models for manual wheelchair application. The sensor wearing placement of the device and sensor configuration in the ambulatory population, i.e. leg or waist may not be suitable for assessment of movement in manual wheelchair activities like walking versus manual wheelchair propulsion, or cycling versus hand biking. For recognizing active propulsion, at least two sensors are necessary to record movement of hand and wheel independently⁹. In addition, the use of energy expenditure (EE) prediction models derived from raw accelerometer data or estimation of the metabolic estimation of task (MET) from questionnaires to identify intensity (light, moderate, vigorous, high) are developed for ambulatory persons.

While several instruments are commonly used to measure PA in research, measurement properties (e.g. validity or reliability) are not always known for wheelchair activities¹⁰⁻¹². In addition, clear guidelines or recommendations are lacking about how to assess PA, including the intensity and type of activity in a valid and reliable manner.

Therefore the aim of this systematic review was to summarize the evidence on measurement properties (content, construct and criterion validity or reliability)

of instruments measuring the intensity and / or type of PA in MWU and making recommendations for the selection of PA outcomes tools.

METHODS

Data Sources and Searches

A literature search was conducted up to July 2019 in the following electronic databases: MEDLINE/Pubmed, CINAHL and Embase. The complete search strategy of MEDLINE/Pubmed, CINAHL and Embase can be found in supplemental appendix 1, where the complete search string is displayed. The general topics areas searched were; physical activity (sedentary behaviour, inactive lifestyle, activity), energy expenditure, instrument (accelerometry, questionnaire, sensor), mode of mobility (wheelchair, non-ambulant). The search string was based on a validated search filter for finding studies on measurement properties as published by the COSMIN group¹³. No date restrictions were applied to the search. The protocol for the search was not published a priori. Overall methodology of this systematic review is based on the guideline for systematic reviews of outcome measurement instruments^{14,15}.

Study Selection

Initial screening of titles was performed by one of the reviewers (MO) to exclude obvious non fitting titles and duplicate references by use of RefWorks © 2019 ProQuest LLC. A pragmatic strategy of combining auto- and hand-searching duplicates by use of RefWorks © 2019 ProQuest LLC was applied¹⁶. Titles, abstracts and full-text were then independently reviewed for eligibility by two reviewers (MO, KL). Any discrepancies in the agreement were discussed with a third reviewer (JdG) until consensus was reached. Finally, the reference lists of included studies were manually searched for additional studies.

Articles were eligible if; 1) the study sample contained individuals who use manual wheelchairs, 2) two or more activities of daily living were evaluated, 3) at least one measurement property of an instrument measuring PA was assessed, 4) self-reported (subjective measures) or device-based instruments (objective measures) were evaluated, 5) the instruments are for sale and currently supported by the manufacturer for use. The choice for inclusion of two or more types of activities (i.e. lying, sitting, hand biking, wheelchair propulsion) or activities at different intensities (light, moderate, vigorous) was done in order to evaluate PA in a broader spectrum and to give optimal recommendations

to clinicians about valid and reliable instruments to monitor PA in daily live. The type of PA is defined as the identification of body postures (e.g., sitting, standing) and body movements (e.g., walking, cycling). The intensity of PA is defined as the estimation of energy expenditure / MET's. Both perspectives of PA are based on movement counts¹⁷. Articles were excluded when written in another language than English or Dutch, were (systematic) reviews, study proposals or protocols, case studies or series, books or book chapters, poster abstracts, conference abstracts, correspondence or commentaries.

Data Extraction and Quality Assessment

One reviewer (MO) extracted data using a standard extraction form, this was checked by the second reviewer (KL). Table 1a and 1b shows the data extracted from the included articles, the extracted data were: 1) type and administration of instruments; 2) author and year; 3) characteristics of study sample, i.e. number of wheelchair users in study population, diagnosis and mean age or range; 4) studied measurement property, i.e. validity or reliability in table 1a, and used instrument for criterion validity in table 1b; 5) study outcomes. The studied measurement property for self-reported instruments were content validity, structural validity, criterion validity, hypothesis testing, the internal consistency and the test – retest reliability. The studied measurement property for device-based instruments was overall criterion validity; one study evaluated hypothesis testing and internal consistency for two device-based instruments.

Table 1a: Characteristics and study outcomes of studies evaluating self-reported and two device-based instruments on measurement properties.

Self-reported instruments							
Instru- ment	Author (Year)	n = total sample (n= WCD)	Diagnosis	Mean age (SD) or Range in years	Validity (comparative instrument)	Reliability	Study outcomes
13-item PASIPD	Washburn (2002)	N = 372 (NST)	Postpolio, paraplegia, quadriplegia, CP, SCI, amputation, muscular dystrophy, SB, visual impairment, auditory impairment, diabetes, epilepsy, hemophilia, Crohn's disease.	Male: 50.7 (12.9) Female: 48.4 (12.6)	Content & Construct (structural)	Internal Consistency	Cumulative explained variance = 62.9% Factor's Cronbach's alpha = 0.37-0.65
13-item PASIPD	Tanhouffer (2012)	N = 14	SCI	40 (13)	Criterion (DLW)	NA	TDEE r = 0.73, PA index in MET-h/day and PAEE in kJ r = 0.13 Predictive methods, Bland Altman plots; TDEE -51 ± 1748 kJ/day for DLW and PASIPD PAEE -46 ±1574 kJ/day for DLW and PASIPD (93% of the data point fell within the ±1.96 SD boundaries).
11-item PASIPD	Van den Berg- Emons (2011)	N =124 N = 51(†)	Bilateral CP, MMC, SCI.	31.2 (11.2)	Criterion (Activity Monitor)	NA	Spearman's r = 0.20 with Activity Monitor's duration of activities r = 0.23 with Activity Monitor's intensity

11-item PASIPD	De Groot (2010)	N = 139(± (158)	SCI	41.6 (14.1)	Construct (structural) & Hypothesis testing	Internal consistency	Physical capacity outcomes (peak oxygen uptake, peak power output and muscular strength): Cumulative variance = 61.77% Spearman's $r = 0.15$ -0.35 Activity outcomes (wheelchair skills, Utrecht Activity List, mobility range and social behavior subscales of the SIP68): $r = -0.41$ -0.51; Cronbach's $\alpha = 0.63$
PARA- SCI	Latimer (2006)	N = 158 (158)	SCI	38.5 (11.1)	Hypothesis testing	NA	Muscle strength $r = -0.07$ -0.36 Aerobic fitness $r = -0.09$ -0.35 LTPA score: men > women LTPA score: young person > old person LPTA score: sport > no sport LTPA score: high participation frequency > low participation frequency; Lifestyle activity heavy intensity: less disabled < more disabled Lifestyle activity minutes in mild intensity: work or school > no work or school MVPA minutes per hour of wake time $r = 0.37$ MVPA minutes per day $r = 0.40$ LPA minutes per hour of wake time $r = 0.03$ LPA minutes per day $r = 0.00$ Pearson's $r = 0.27$ -0.88 for intensity scales with; $r = 0.79$ for total score with indirect calorimetry; Reliability total activities: ICC = 0.79 (95% CI: 0.70- 0.85), LTPA ICC = 0.72 (95% CI: 0.60-0.80), LA ICC = 0.78 (95% CI: 0.68-0.84)
PARA- SCI	Claridge (2014)	N = 42 N = 18(±)	CP GMFCS level 1 t/m 5	33.5 (12) Range: 18-75	Hypothesis testing (acceleromet er)	NA	
PARA- SCI	Martin Ginis (2005)	N = 137(±)	SCI	33.9(7.3) – 49.0(17.1)	Content (N=8) & Criterion (N=9) (indirect calorimetry)	Test – Retest (N=102)	
PARA- SCI	Tanhoffer (2012)	N = 14	SCI	40 (13)	Criterion (DLW)	NA	TDEE $r = 0.74$, PAEE in kJ; $r = 0.50$, PAEE in min/day; $r = 0.62$ Predictive methods, Bland Altman plots;

TDEE: -557 ±1275 kJ/day for DLW and PARA-SCI (all data point fell within the ±1.96 SD boundaries.)
 PAEE: -502 ±1147 kJ/day for DLW and PARA-SCI (all data point fell within the ±1.96 SD boundaries.)

LTPAQ-SCI	Martin Ginis (2012)	N = 103(†)	SCI	48.1(12.7)	Hypothesis testing (PARA-SCI)	Test - Retest	<p>r = 0.27-0.54 for intensity scales with intensity within LTPA PARA-SCI scale; Correlation between total score and total score of PARA-SCI LPTA scale r = 0.46 Mild intensity: ICC = 0.74 (95% CI: 0.51-0.86); Moderate intensity: ICC = 0.62 (95% CI: 0.36-0.79); Heavy intensity: ICC = 0.93 (95% CI: 0.87-0.97); Total: ICC = 0.83 (95% CI: 0.69-0.91)</p>
PAI-SCI	Butler (2008)	N = 43(†)	SCI	54.2(16.6)	Hypothesis testing (Activity monitor Actual)	Test – Retest & Internal consistency	<p>r = 0.42; Significant difference between PAI-SCI scores from lower and upper level injuries; Correlation between level of injury and sedentary time r = -0.44, Light activity r = 0.34, Moderate activity r = 0.36; Subscales Cronbach's α = 0.35-0.90; Subscales ICC = 0.28-0.59</p>

†NS: Not Stated, however it can be deduced the subjects were wheelchair users in the study population; ‡: Exact number wheelchair users not stated, though it can be deduced

Table 1b: Characteristics and study outcomes of studies evaluating device-based instruments on measurement properties.

Device-based instruments						
Instrument	Instrument placement + activity protocol	Author (Year)	N = (using WC)	Diagnosis	Mean age (SD) or Range	Used instrument for criterion validity
Intensity of physical activity						
ActiGraph GT3X	Waist, chest, dominant or non-dominant wrist	García-Massó (2013)	N = 20 (†)	SCI	40(10.6)	Indirect calorimetry
	Activity protocol: lying down, body transfers, moving items, mopping, working on a computer, watching television, arm-ergometer exercise, passive propulsion, slow and fast propulsion.					Waist-worn $r = 0.67$; Chest-worn $r = 0.66$; Dominant wrist $r = 0.86$; Non-dominant wrist $r = 0.86$.
	Waist, right upper arm, wrist	Nightingale (2014)	N = 15 (15)	SCI, fibromyalgia, CRPS, SB	36(11)	Indirect calorimetry
ActiGraph GT3X+	Activity protocol: desk work, wheelchair propulsion at 2, 4, 6 and 8 km/h on a motorised wheelchair treadmill.					Waist worn sensor $r = 0.73$; Upper arm $r = 0.87$; Wrist worn sensor $r = 0.93$;
ActiGraph GT3X+	Right upper arm, wrist	Nightingale (2015)	N = 17 (17)	SCI, CP, SB, Scoliosis.	36(10) Range: 19-50	Indirect calorimetry
	Activity protocol: resting, folding clothes, propulsion on a motorised wheelchair treadmill on a 1% gradient (3.4,5.6 and 7 km/h) and propulsion at 4 km/h on 2% and 3% gradient.					Upper arm $r = 0.68$; Wrist $r = 0.82$;
RT3	waist-worn (RT3) Activity protocol: resting, wheelchair propulsion, arm-ergometer exercise and deskwork.	Hiremath & Ding (2011)	N = 24(†)	SCI	41.4(11.4)	Indirect calorimetry
						RT3 Waist worn ICC = 0.64 [95% CI: 0.51-0.73]

PAMS	<p>A two-axis gyroscope between the spokes of the wheelchair and one accelerometer on the right upper arm and wrist.</p> <p>Activity protocol; wheelchair propelling on a tile, surface and pile carpet at self-selected medium and fast pace, propelling up and down a ramp, being pushed in a wheelchair, playing basketball, folding laundry, deskwork, playing darts, using a resistance band, exercising on an arm-ergometer.</p> <p>In a lab/semi-structured/home environment.</p>	Hirmath (2016)	N = 45 (45)	SCI	41(12.6)	Indirect calorimetry	<p>PAMS-arm ICC=0.82, 95% CI 0.79-0.85, P<0.05</p> <p>PAMS-wrist accuracy ICC=0.89, 95% CI 0.89-0.91, P<0.05</p>
GENEActiv	<p>Right upper arm, wrist</p> <p>Activity protocol; resting, folding clothes, propulsion on a motorised wheelchair treadmill on a 1% gradient (3.4, 5.6 and 7 km/h) and propulsion at 4 km/h on 2% and 3% gradient.</p>	Nightingale (2015)	N = 17 (17)	SCI, CP, SB, Scoliosis	36(10) Range: 19-50	Indirect calorimetry	<p>Upper arm $r = 0.76$</p> <p>Wrist correlation $r = 0.88$</p>
Actiheart	<p>Chest</p> <p>10 activities: resting, folding clothes, wheelchair propulsion on a 1% gradient (3.4, 5.6 and 7 km/h) and propulsion at 4 km/h (with an additional 8% of body mass, 2% and 3% gradient) on a motorised wheelchair treadmill.</p> <p>Resting, incremental arm crank ergometry, 4-hour record / normal daily activities. (in 8 participants, diary versus PAEE estimation by Actiheart).</p>	Nightingale (2015)	N = 15(15)	SCI, n=8, SB; n=3, scoliosis; n=1, CP; n=1, amputation =1, wheelchair basketball player; n=1	36(11) Range: 19-50	Indirect calorimetry	<p>Predicted with criterion PAEE generic group, $r=0.76$ and individual heart rate calibration, $r=0.95$.</p> <p>The absolute bias $\pm 95\%$ limits of agreement were 0.51 ± 1.90 for generic group and -0.22 ± 0.49 kcal/min for individual heart rate calibration</p> <p>Mean absolute errors across the activity protocol were for generic group $51.4 \pm 38.9\%$ and $16.8 \pm 15.8\%$ for using the individual heart rate calibration.</p>
Polar RS800RX	<p>Wearing the heart rate monitor over 2 days, including a 'more physically active day' and a 'less physical active day'. During waking hours of at least 12-hour duration.</p> <p>To estimate EE from HR, each individual's HR / VO_2 slope and intercept regression equation was determined.</p>	Tanhofer (2012)	N = 12(12)	SCI	40 (± 13)	Doubly labelled water technique	<p>TDEE: $r = 0.68$; PAEE: $r = 0.30$</p> <p>Predictive methods by use of Bland Altman plots:</p> <p>TDEE: mean bias was -860 ± 1397 kJ/day (all data point fell within 1.96 SD boundaries.)</p> <p>PAEE: mean bias was 461 ± 1527 kJ/day (all data point fell within 1.96 SD boundaries.)</p>

Instrument	Instrument placement + activity protocol	Author (Year)	N = (using WC)	Diagnosis	Mean age (SD) or Range	Used instrument	Study outcomes
Type of physical activity							
VitaPort	Sternum, each thigh, wrist together	Postma (2005)	N = 10(4)	SCI	Range: 19-63	Video recordings	Overall detection of hand biking and propulsion: Sensitivity = 87%, Specificity = 92% Agreement = 92%
	Activity protocol; a series of representative daily life activities (wheelchair propulsion and non-wheelchair propulsion activities), hand biking.						Detection when poor triceps strength: Sensitivity = 81% Specificity = 94% Agreement = 92% Detection when good triceps strength: Sensitivity = 95%, Specificity = 90% Agreement = 91% Mean accuracy = 84% Mean sensitivity = 80% Mean specificity = 85%
VitaMove	Sternum, each wrist and one on the lateral side of the mid-thigh (for partially ambulatory children)	Nooljen (2015)	N = 12 (12)	SB, CP	14(4) Adolescents	Video recordings	
PAMS	Activity protocol; a series of representative daily life activities (wheelchair propulsion and non-wheelchair propulsion activities), hand biking.						
	A two-axis gyroscope between the spokes of the wheelchair and one accelerometer on the right upper arm or wrist.	Hiremath (2015)	N = 45 (45)	SCI	41(12.6)	Video recordings	PAMS-arm accuracy = 89.26% PAMS-wrist accuracy = 88.47% Accuracy arm wocket = 70.38% Accuracy wrist wocket = 74.55%
	Activity protocol; wheelchair propelling on a tile, surface and pile carpet at self-selected medium and fast pace, propelling up and down a ramp, being pushed in a wheelchair, playing basketball, folding laundry, deskwork, playing darts, using a resistance band, exercising on an arm-ergometer. In a lab/semi-structured/home environment.						

ActiWatch	Wrist-worn Activity protocol for three 7-minute periods in wheelchair pushing indoor, upper extremity range-of-motion exercise, sitting quietly, sitting at desk doing keyboarding activities.	Wams & Belza (2004)	N = 22(†)	SCI	Phase 1: 37.3(8.4) Phase 2: Phase 3: 43.2(11.3)	Physical Activity Report	Wrist-worn r = 0.60 (sleep included) Wrist-worn r = 0.40 (sleep excluded)
ActiGraph GT3X	Dominant, non-dominant wrist, chest, waist Individual activities; lying down, body transfers, moving items, mopping, working on a computer, watching television, arm-ergometer exercise, passive propulsion, slow and fast propulsion. Grouped activities; sedentary, transfers, housework, moderate PA and locomotion.	García-Massó (2015)	N = 20 (20)	SCI	40.03 (10.6)	Machine learning algorithms	Individual activities with machine learning Algorithms 55.0-72.5%; Grouped activities with machine learning Algorithms 83.2-93.6%
ActiWatch Score vs PRO-diary	Wrist-worn Hypothesis testing (PRO-diary) Internal consistency	Murphy (2019)	N = 38 (19)	SCI and ambulatory participants	49 (10.6)	PRO-diary for hypothesis testing	Correlations between activities with Actiwatch Score and PRO-diary ranged from 0.78 to 0.97. For walking—wheeling normally, mean activity counts per minute were 3.3 times higher for Actiwatch Score, t(37) = -8.36, p = .0001, and 4 times higher for PRO-Diary, t(37) = -8.16, p = .0001, for those with SCI compared with those without SCI. For walking—wheeling briskly, mean activity counts per minute were 3.5 times higher for Actiwatch Score, t(37) = -8.31, p = .0001, and 3.4 times higher for PRO-Diary, t(37) = -7.38, p = .0001, for those with (compared with without) SCI. Group comparisons of physical activity levels for all other lab tasks were non-significant (all < 0.11).

†NS: Not Stated, however it can be deduced there were wheelchair users in the study population; ‡: Exact number not stated, though it can be deduced

Assessment of the methodological quality of the included studies

Two reviewers (KL and MO) independently assessed the methodological quality of the included studies using the Consensus-based Standards for the selection of health Measurement Instruments (COSMIN) checklist¹⁸. The COSMIN checklist is a modular tool and contains 12 boxes: ten boxes can be used to assess the methodological quality of studies on measurement properties, and 2 boxes contain general requirements. The four-point Consensus-based standard for the selection of COSMIN checklist was chosen to assess the methodological quality of studies on measurement properties¹⁹. The items of each box were rated with the four-point scoring system: 'excellent', 'good', 'fair' and 'poor'. To rate the methodological quality, the lowest rating of any item in a box was taken ('worse score counts' method) according to the COSMIN guidelines. Disagreement between the authors regarding the 'worse score count' of an article was resolved with a third reviewer (JdG) until consensus was reached. There is one item about sample size in every box, with minimal requirement of N=30 for an adequate sample size. Since the COSMIN guidelines were originally developed for self-reported outcomes, these sample size requirements do not necessarily apply for device-based instruments where a smaller sample size for evaluation of criterion validity is often appropriate and common^{20,21}. A lower sample size was only approved if an adequate power analysis or post hoc analysis was described. The scoring procedure for the item about sample size was only modified for evaluation of criterion validity in device-based measurement instruments and not for the self-reported outcomes. This scoring modification is in line with an article by Mitchell et al.²²

Assessment of the outcome of measurement properties

The outcome measurement properties could be qualified as 'positive' (+), 'indeterminate' (?), and 'negative' (-) as shown in table 2a¹⁹. As some studies in the review assessed the sensitivity and specificity of a device-based instrument, an additional rating was added for criterion validity (table 2b). For example, when there are convincing arguments that gold standard is 'gold' AND (correlation OR intraclass correlation (ICC) with gold standard is ≥ 0.70) OR sensitivity / specificity or accuracy is 80-100%, the outcome measurement properties were qualified as 'positive' (+).

Table 2a: Quality criteria for study outcomes - patient related outcomes
(Based on Terwee et al. 2007)⁹.

Property	Rating	Outcome quality criteria (OQ)
Validity		
Content validity	+	The target population considers all items in the questionnaire to be relevant AND considers the questionnaire to be complete
	?	No target population involvement
	-	The target population considers items in the questionnaire to be irrelevant OR considers the questionnaire to be incomplete
<i>Construct validity</i>		
• Structural validity	+	Factors should at least explain 50% of the variance
	?	Explained variance not mentioned
	-	Factors explain <50% of the variance
• Hypothesis testing	+	(Correlation with an instrument measuring the same construct must be ≥ 0.50 OR at least 75% of the results are in accordance with the hypotheses) AND correlation with related constructs is higher than with unrelated constructs
	?	Solely correlations determined with unrelated constructs OR no correlations reported
	-	(Correlation with an instrument measuring the same construct is < 0.50 OR $< 75\%$ of the results are in accordance with the hypotheses) AND correlation with related constructs is higher than with unrelated constructs
Criterion validity	+	Convincing arguments that gold standard is "gold" AND (correlation OR ICC with gold standard is ≥ 0.70). OR sensitivity / specificity or accuracy is 80-100%*.
	?	No convincing arguments that gold standard is "gold" OR no correlations have been calculated. Sensitivity and specificity are not displayed OR only the accuracy is displayed*.
	-	Correlation with gold standard is < 0.70 , despite adequate design and methods OR sensitivity / specificity or accuracy is $< 70\%*$.
Reliability		
Internal consistency	+	(Sub)scale unidimensionality AND Cronbach's alpha(s) ≥ 0.70 AND < 0.95
	?	Dimensionality not known OR Cronbach's alpha not determined
	-	(Sub)scale not unidimensional OR Cronbach's alpha(s) < 0.70
Reliability	+	ICC/weighted Kappa is ≥ 0.70 OR (Pearson's or Spearman's r is ≥ 0.80 AND evidence provided that no systematic change has occurred)
	?	Neither ICC/weighted Kappa, Pearson's or Spearman's r is determined
	-	ICC/weighted Kappa is < 0.70 OR (Pearson's or Spearman's r is < 0.80 OR no evidence provided that no systematic change has occurred)

ICC = intraclass correlation coefficient

+: positive rating

?: indeterminate rating

-: negative rating

***Note**

For device-based instrument measurements, assessing the sensitivity and specificity or accuracy of a device are normally rated as; excellent (++), good (+), moderate (~), weak (-) or not clear (?). In order to be clear and consistent with the description of the ratings for self-reported and device-based instruments together displayed in the tables and text, we use the same ratings for the sensitivity and specificity outcome measure according to Terwee et al. 2007. This makes that the outcome quality of sensitivity and specificity rated as excellent (++) and good (+) are merged as positive (+); moderate (~) and weak (-) as negative (-) and not clear (?) as indeterminate (?).

Table 2b: Levels of evidence for the quality of the measurement property.

Level	Rating	Criteria
strong	+++ or ---	Consistent findings in multiple studies of good methodological quality OR in one study of excellent methodological quality
moderate	++ or --	Consistent findings in multiple studies of fair methodological quality OR in one study of good methodological quality
limited	+ or -	One study of fair methodological quality
conflicting	+/-	Conflicting findings
unknown	?	Only studies of poor methodological quality

Rating of the outcome quality of device-based measurement, assessing the sensitivity, specificity or accuracy.

Rating outcome quality	Category	Explanation of rating
++	>90%	Excellent
+	80-90%	Good
~	70-80%	Moderate
-	<70%	Weak
?	Not clear	No information about sensitivity and specificity or accuracy of activities during wheelchair protocol.
<p>Note* For device-based measurement, assessing the sensitivity and specificity OR the accuracy of an instrument are normally rated as; excellent (++), good (+), moderate (~), weak (-) or not clear (?). In order to be clear and consistent with the description of the ratings for self-reported and device-based instruments together displayed in the tables and text, we use the same ratings for the sensitivity and specificity or accuracy outcome measure according to Tenwee et al. 2007. This makes that the outcome quality of sensitivity and specificity or accuracy rated as excellent (++) and good (+) are merged as positive (+); moderate (~) and weak (-) as negative (-) and not clear (?) as indeterminate (?).</p>		

Best evidence synthesis

A best evidence synthesis was performed because methodology of pooling of measurement properties was not possible with many different outcome measures.^{13,19}

Data for the best evidence synthesis were interpreted by three authors, KL (PhD student and physical therapist), MO (physical therapist and health scientist) and JdG (PhD, physical therapist and medical physiologist). As proposed by the Cochrane Back Review Group, the best evidence synthesis is generated by combining the level of evidence (strong, moderate, limited, conflicting, unknown, see table 2b) and the study outcomes (positive (+), indeterminate (?) and negative (-), see table 2a).

For example, the estimation of energy expenditure (EE) of the GeneActiv with wearing position at the wrist shows a correlation of 0.88 with indirect calorimetry which results in a 'positive' (+) study outcome rating (table 2a) and the methodological quality of the study was rated 'good' (table 2b). There is one study with good methodological quality of the GeneActiv for wearing position at the wrist, which results in a moderate level of evidence (table 2b). In conclusion, there is a moderate level of positive evidence for criterion validity of the GeneActiv for wearing position at the wrist.

RESULTS

Included studies

The search strategy identified 5341 potentially relevant articles from three databases after removing duplicates, 4024 records were screened (figure 1). In total, 56 full-text articles were deemed relevant based on title and abstract. The reference tracking of the included studies did generate five additional relevant studies and 41 articles were excluded based on full text. One article was added by an expert, this article was not found by use of our search strategy because the used words in the title, abstract and key words did not match the search strategy. In total, 61 full-text articles were assessed for eligibility. Finally, 21 studies met the inclusion criteria for this systematic review.

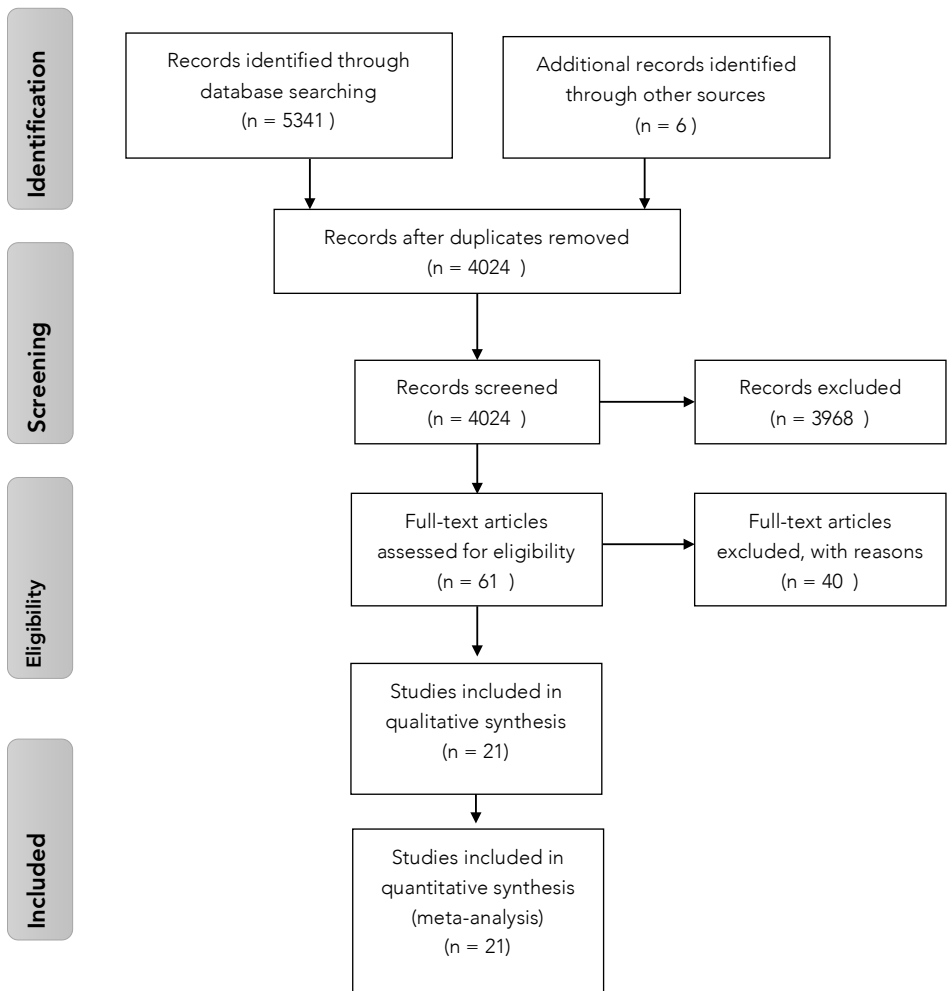


Figure 1: Flowchart identification, screening, eligibility and included studies.

The characteristics of self-reported and device-based instruments in studies reporting measurement properties are displayed in table 1a and table 1b resp. The methodological quality of the studies, the outcome quality of the measurement property and the level of evidence using the COSMIN checklist and criteria are reported in table 3 and table 4. A best evidence synthesis was performed on nine studies including four self-reported instruments (table 3) and on thirteen studies including eight device-based instruments (table 4). One study evaluated both self-reported and device-based instruments ²³. The overall methodological quality of all studies ranged from poor to excellent. First, the self-reported outcome measures will be discussed, followed by device-based outcome measures for assessment organized by either the intensity or type of PA in MWU. Supplemental appendix 2 gives a description of each of the included instruments in this systematic review.

Table 3: The methodological quality of the studies and outcome quality of the measurement property of self-reported instruments and two objective instruments using the COSMIN checklist.

Instrument	Study	Validity				Reliability	
		Content	Construct (structural)	Construct (hypothesis testing)	Criterion	Internal consistency	Test-retest
Self-reported instruments							
PASIPD	Wasburn (2002) Van den Berg-Emons (2011) De Groot (2010) Tanhoffer (2012)	Excellent (+) . . Strong	Excellent (+) . Fair (+) Strong	. . Fair (-) Limited	. Fair (-) . Good** (-) Moderate	Excellent (-) . Fair (-) . Strong
Level of Evidence							
PARA-SCI	Claridge (2014) Latimer (2006) Martin Ginis (2005) Tanhoffer (2012)	. . Good (+) . Moderate	Fair (-) Poor (-) . . Limited	. . Good* (+) Good** (+) Strong Fair (+) . Limited
Level of Evidence							
LTPAQ-SCI	Martin Ginis (2012)	. Moderate	. Good (+) Moderate	Fair (+) Limited
Level of Evidence							
PAI-SCI	Butler (2008) Unknown	Poor (-) Unknown	. .	Poor (-) Unknown	Fair (-) Limited
*:Adequate description of power analysis, but drop-out of 5 participants, **: Doubly labelled water technique as gold standard, Methodological Quality: Excellent, Good, Fair, Poor; Outcome Quality: + = positive, ? = indeterminate, - = negative Rating level of evidence (combination of methodological and outcome quality): strong, moderate, conflicting, limited, unknown.							

*, Adequate description of power analysis, but drop-out of 5 participants, **, Doubly labelled water technique as gold standard, Methodological Quality:

Excellent, Good, Fair, Poor; Outcome Quality: + = positive, ? = indeterminate, - = negative

Rating level of evidence (combination of methodological and outcome quality): strong, moderate, conflicting, limited, unknown.

Table 4: The methodological quality of the studies and outcome quality of the measurement property of device-based instruments on criterion validity using the COSMIN checklist.

Instrument	Study	Outcome	Quality	Level of evidence
ActiGraph GT3X+				
Chest	García-Massó (2013)	r = 0.68	Fair (-)	Limited
Waist	García-Massó (2013)	r = 0.67	Fair (-)	Conflicting
	Nightingale (2014)	r = 0.73	Fair (+)	
Right upper arm	Nightingale (2015)	r = 0.68	Good (-)	Conflicting
	Nightingale (2014)	r = 0.87	Fair (+)	
Right wrist	Nightingale (2015)	r = 0.82	Good (+)	Moderate
	Nightingale (2014)	r = 0.93	Fair (+)	
Non-dominant wrist	García-Massó (2013)	r = 0.86	Fair (+)	Limited
Actiheart				
Generic group	Nightingale (2015)	r=0.76	Good (+)	Moderate
Individual heart rate calibration group		r=0.95	Good (+)	
Polar RS800RX				
TDEE**	Tanhoffer (2012)	r = 0.68	Good (-)	Moderate
PAEE**		r = 0.30	Good (-)	
GeneActiv				
Right upper arm	Nightingale (2015)	r = 0.76	Good (+)	Moderate
Right wrist		r = 0.88	Good (+)	
RT3				
Waist	Hiremath & Ding (2011)	ICC = 0.64 (95% CI: 0.51-0.73)	Good (-)	Moderate
PAMS				
Right upper arm	Hiremath (2016)	ICC=0.82, 95% CI 0.79-0.85, P<0.05	Good (+)	Moderate
Right wrist		Hiremath (2016)	ICC=0.89, 95% CI 0.89-0.91, P<0.05	

Instrument	Study	Outcome	Quality	Level of evidence
Vitaport / Vitamove				
Sternum/thigh/wrist	Postma (2005)	Sensitivity = 87% Specificity = 92% Agreement = 92%	Good (+)	Strong
	Nooijen (2015)	Mean accuracy = 84% Mean sensitivity = 80% Mean specificity = 85%	Good (+)	
PAMS				
Spokes/upper arm	Hiremath (2015)	PAMS-arm accuracy = 89.26%	Excellent (+)	Strong
Spokes/wrist		PAMS-wrist accuracy = 88.47%	Excellent (+)	Strong
Right upper arm	Hiremath (2015)	Accuracy arm wocket = 70.38%	Excellent (+)	
Right wrist		Hiremath (2015)	Accuracy wrist wocket = 74.55%	
Actigraph GT3X				
Individual activities	García-Massó (2015)			Limited
Dominant wrist		55.0-61.4%	Fair (-)	
Non-dominant wrist		61.5-63.3%	Fair (-)	
All (wrists, chest, waist)		65.9-72.5%	Fair (-)	
Grouped activities	García-Massó (2015)			Limited
Dominant wrist		83.2-85.9%	Fair (+)	
Non-dominant wrist		83.9-87.0%	Fair (+)	
All (wrists, chest, waist)		89.5-93.6%	Fair (+)	
Actiwatch				
Wrist	Warms & Belza (2004)	r = 0.60 (sleep included) r = 0.40 (sleep excluded)	Poor (-)	Unknown
Actiwatch vs PRO-diary ^a	Murphy (2019)	r = 0.78 - 0.97	Fair (+)	Limited

^a:Adequate description of power calculation, **Criterion: Doubly labelled water technique. *:hypothesis testing and internal consistency, comparison activity counts wheelchair use and ambulatory activities. Methodological Quality: Excellent, Good, Fair, Poor; Outcome Quality: + = positive, ? = indeterminate, - = negative

Self-reported instruments assessing the intensity of PA

PASIPD

Four studies investigated the content validity, construct validity (structural validity and hypothesis testing), criterion validity and internal consistency²³⁻²⁶ of the 11- or 13-item Physical Activity Scale for individuals with Physical Disabilities (PASIPD). Double labeled water was used as criterion in one study²³, whereas an activity monitor was used as criterion in another study²⁵. Two studies investigated properties of the 11 item PASIPD^{24,25}. Because it was not reported if there were missing items and if there were missing items, how they were handled, the methodological quality was rated as fair for these two studies. Combining methodology and outcomes, best evidence synthesis for the

PASIPD shows 1) strong levels of positive evidence for content and structural validity, 2) moderate negative evidence on criterion validity, 3) limited negative evidence on hypothesis testing and 4) strong negative evidence on internal consistency.

PARA-SCI

The criterion validity, content validity, construct validity (hypothesis testing) and reliability (test-retest) were examined for the Physical Activity Recall Assessment for People with Spinal Cord Injury (PARA-SCI) in four studies ^{23,27-29}. The methodological quality of one reliability study ²⁸ was rated fair resulting in a limited level of positive evidence for reliability (ICC total activities = 0.79). There is limited negative evidence on hypothesis testing ^{27,29}, a strong level of positive evidence for criterion validity ^{23,28} and a moderate level of positive evidence for content validity ²⁸. The PARA-SCI has not been evaluated for structural validity (construct) and internal consistency (reliability).

LTPAQ-SCI

Only one study evaluated the Leisure Time Physical Activity Questionnaire for people with Spinal Cord Injury (LTPAQ-SCI) on construct validity (hypothesis testing) and reliability (test-retest) ³⁰. The correlation between the total score of the PARA-SCI and the PARA-SCI LPTA scale was 0.46. Therefore there is moderate positive evidence on hypothesis testing. The ICC's of the different intensity scales of the LPTA varied between 0.62 (moderate intensity) and 0.93 (heavy intensity). There is a limited level of positive evidence for test-retest reliability.

PAI-SCI

One study evaluated construct validity (hypothesis testing), internal consistency and reliability (test-retest) for the Physical Activity Inventory for patients with Spinal Cord Injury (PAI-SCI) ³¹. The evidence for hypothesis testing and internal consistency is unknown because of a poor methodology of the study and there is a limited level of negative evidence for test-retest reliability.

Measurement properties of device-based instruments

For device-based instruments only criterion validity was evaluated (table 4). For intensity assessment doubly labelled water technique was used in one study as criterion ²³, while the other seven studies used indirect calorimetry as a criterion. For assessment of type

of PA, video recordings, physical activity report or machine learning algorithms were used as criteria. Double labeled water, indirect calorimetry, video recording and machine learning algorithms were considered appropriate criteria for validation of the device³².

Device-based instruments measuring the intensity of activity

Actigraph GT3X+

The validity of the assessment of the intensity of activities measured by the Actigraph GT3X+ was evaluated in three studies³³⁻³⁵. There were five different placements of the Actigraph at the body which were evaluated (non-dominant wrist, waist, chest, right upper arm, right wrist). For measuring the intensity of activities with the Actigraph GT3X+, there is moderate levels of positive evidence (two studies, one with fair and the other with good methodology) for validity ($r = 0.82$ and 0.93) for placement at the right wrist^{33,34} and limited positive evidence on validity ($r = 0.82$) for placement at the non-dominant wrist³⁵. There is a limited level of negative evidence for validity ($r = 0.68$) using placement at the chest³⁵. There are conflicting findings for the placement at the right upper arm and the waist. The quality of the methodology of the studies were rated fair to good and the correlations varied from 0.67 to 0.87 ^{34,35}.

GENEActive

In one study, the criterion validity of the GENEActive was determined for resting, folding clothes, propulsion on a motorized wheelchair treadmill at five different speeds and three different gradients³³. Placement of the device at both the right upper arm and wrist was examined. The methodological quality of this study was good and outcomes positive ($r = 0.76$ and 0.88). Therefore there is moderate positive evidence on validity of the GENEActive for both locations.

RT3

The validity of the waist worn RT3 accelerometer was determined for resting, wheelchair propulsion (at 3 different speeds), exercise on an arm ergometer (at 3 different speeds and workloads), and deskwork³⁶. The methodological quality was rated as good and outcome

quality as negative ($ICC = 0.64$), therefore there is a moderate level of negative evidence for criterion validity for the RT3 at the waist.

PAMS

Hiremath et al. 2016 developed a Physical Activity Monitor System (PAMS), a two-axis gyroscope in combination with an accelerometer ³⁷. In the study, the criterion validity of two placements (right wrist and upper arm) was evaluated by use of indirect calorimetry as criterion during various physical activities in people with SCI using a wheelchair. There is moderate positive evidence on criterion validity for placements both on the right upper arm (ICC = 0.82) as right wrist (ICC = 0.89).

Heartrate derived measures

Two studies looked at HR derived measures instead of accelerometry to predict EE during PA. The Actiheart (a combined accelerometer and heartrate monitor) was evaluated in one study ³⁸. There is a moderate level of positive evidence for validity of the Actiheart when using the individual heartrate calibration option of the device in combination with the acceleration signals of the device. The found correlations with indirect calorimetry were 0.76 and 0.95.

At the same time, a study regarding a heartrate monitor (Polar RS800RX) ²³, using the DLW technique as criterion, showed moderate negative evidence on validity ($r = 0.68$ and 0.30) for the Polar RS800RX to predict EE during PA. Polar measures the heartbeat only, not in combination with acceleration signals like the Actiheart.

Device-based instruments measuring the type of activity

Two studies evaluated the criterion validity of the Vitaport / VitaMove. The VitaMove is the successor of the Vitaport, using the same algorithms, therefore the quality of the measurement property were taken together. With a standard protocol consisting of a series of representative daily life activities (wheelchair propulsion, non-wheelchair propulsion activities and hand biking), in a semi-natural setting, the output of the Vitaport was compared to video recordings in both adults ³⁹ and youth ⁴⁰. The overall agreement was 92% and the mean accuracy was 84%. The methodology quality was rated good. Therefore there is a strong level of positive evidence for validity detecting the type of activities with the Vitamove.

PAMS

Hiremath et al. 2015 evaluated four test conditions for the validation of the PAMS, a combination of a two-axis gyroscope, attached to the spokes of the wheelchair and

one accelerometer placed on the right upper arm or at the wrist of the individual who use a wheelchair ⁴¹. In addition, the accelerometer placement at the wrist or the upper arm were evaluated separately. In this study, the subjects performed ten activities in a lab setting (table 2b) and executed the same ten activities in a semi structured home environment. With use of the video recordings, the overall accuracy of the placement of the PAMS at the spokes at the wheelchair in combination with the arm and wrist placements were respectively 89.3% and 88.5%. Overall, there is strong levels of positive evidence for criterion validity for detecting the type of PA with the PAMS.

Actigraph GT3X

One study assessed the criterion validity of the Actigraph GT3X with use of a machine learning algorithm ⁴². Ten different activities (table 2b) were evaluated, with a variable accuracy of individual activities between 55.0-69.3% and an accuracy of grouped activities between 83.2-93.6%. The methodological quality was rated fair. Therefore, there is limited negative evidence on criterion validity regarding measuring individual activities and limited positive evidence on criterion validity for measuring grouped activities with the Actigraph GT3X.

Actiwatch

The criterion validity of the wrist-worn Actiwatch with a four day physical activity record was examined in one study ⁴³. The methodological quality was rated poor because the criterion used was a physical activity report. Therefore the level of evidence is unknown for the rating of criterion validity for this device. Furthermore, the construct validity (hypothesis testing) and reliability (internal consistency) was evaluated in one other study⁴⁴. There is limited positive evidence on internal consistency and construct validity for both the Actiwatch and the comparative instrument the PRO-diary.

DISCUSSION

The purpose of this review was to summarize the level of evidence for different measurement properties of instruments measuring PA in MWU and making recommendations for the selection of PA outcomes tools. A systematic literature search yielded twenty-one studies that included four self-reported instruments (questionnaires) and eight device-based instruments (accelerometers and gyroscopes). The self-reported

instruments identified measured the intensity and duration of PA.

Self-reported instruments

We found four questionnaires, two with reasonable level of evidence, the PASIPD and the PARA-SCI, and two not, the LTPAQ-SCI and the PAI-SCI. Three questionnaires (PARA-SCI, LTPAQ-SCI, PAI-SCI) were specifically developed for persons with SCI and not tested in other populations. None of the found questionnaires were tested in youth, only in adults who use a wheelchair. Two questionnaires (LTPAQ-SCI, PAI-SCI) focused solely on leisure time physical activity.

The PAI-SCI was assessed in one study³¹. Because of a poor methodology of the study we cannot recommend the use of this questionnaire at this time.

Although the PASIPD showed strong levels of positive evidence for content and construct validity (structural), the estimation of physical activity energy expenditure (PAEE) by use of the PASIPD is not valid because in one study, the correlation was 0.13 between the estimated PAEE with the criterion DLW technique²³. We found based on two studies^{23,25}, moderate negative evidence on criterion validity to measure PA in daily life. Construct validation is often considered to be less powerful than criterion validation⁴⁵. Therefore, it is questionable to use the PASIPD to measure PA. More research is needed to give a stronger level of evidence based recommendation for usage of this instrument.

The PARA-SCI seems valid to measure PA^{23,28}. This is based on the number and the quality of studies evaluating measurement properties of this instrument. However, the usage of this questionnaire could only be recommended in persons with SCI who use a wheelchair because this questionnaire is specifically developed and evaluated in and for this population. Future research is necessary to give a more specific and evidence based recommendation on whether or not to use this questionnaire in a wider population MWU (i.e. children, other diagnosis groups rather than SCI only) and should focus on both the reliability (test-retest)²⁸ and the generalizability of the PARA-SCI.

The LTPAQ-SCI is developed by use of the subscale 'leisure time physical activity' of the PARA-SCI. The structural validity (construct validity) and internal consistency (reliability) of the PARA-SCI were not evaluated, therefore we do not know the uni-dimensionality of the subscales (the LTPAQ-SCI) or the correlations between the subscales of the PARA-SCI. It is therefore questionable to use just one subscale of the PARA-SCI as a self-contained questionnaire (LTPAQ-SCI). In addition, the LTPAQ-SCI showed overall a low test-retest reliability correlation with activities with a low intensity while on the subscales

of mild and heavy intensity a high correlation was detected ³⁰. This suggest that the LTPAQ-SCI is less able to measure and monitor low intensity of PA during leisure time. In addition, not all studies looking at validity of questionnaires used appropriate criteria measures, e.g. the gold standard double labelled water, direct or indirect calorimetry^{18,35}. Based on above mentioned results, future research could compare all four questionnaires in the same group and relate them to device-based instruments measuring PA in MWU in order to know which self-reported instrument could be used the best to measure PA. According to the results of this systematic review, the PASIPD seems to be the most researched and the most promising self-reported instruments to measure PA in MWU (the PASIPD) and the PARA-SCI to assess PA in persons with SCI who use a wheelchair ²⁴⁻²⁹.

Device-based instruments

For the device-based instruments the criterion validity was mainly evaluated. In one study, the construct validity of the Actiwatch and reliability between the Actiwatch and PRO-diary was evaluated. Because the criterion validity is unknown; these devices will not be further discussed. The PAMS and the Actigraph GT3X are the only devices that have been evaluated on criterion validity for both the intensity level of activities and classifying the type of activities. Of the device-based instruments included in this systematic review, only the PAMS seems valid to assess both intensity and the type of PA ^{37,41,46}.

Measuring the intensity of physical activity

Six devices were identified for measuring the intensity of PA. The Actigraph GT3X+, GENEactiv, PAMS and Actiheart are recommended for assessment of the intensity of PA in adults who use a wheelchair ^{33,34,37,38}. The GENEactiv was validated in adults with cerebral palsy (CP), spina bifida (SB), spinal cord injury (SCI) and persons with scoliosis who use a wheelchair. The best wearing position seems to be on the wrist or the upper arm ³³. The Actigraph GT3X+ is a similar device, the same best-evidence was found, and best wearing position for this device is also at the wrist ^{33,34}. To measure the intensity of PA with the PAMS, two sensors are needed with one sensor placed at the wrist or upper arm and one at the spokes of the wheelchair. This can be complicated when the person switches wheelchairs during sports activities. A different approach is the use of HR derived outcomes. Simply using raw HR data measured with a HR monitor is not valid to predict PAEE in people who use a wheelchair. There is a large degree of inter

individual variance in cardiovascular function and response to exercise (the HR-PAEE relationship)²³. Based on HR only, the PAEE cannot be estimated properly in people who use a wheelchair³⁸. The Actiheart, a combined accelerometer and HR monitor is proven valid to estimate the PAEE, when the individual heart rate calibration algorithm of the device is used instead of the general algorithm³⁸. The combination of a HR monitor and an accelerometer to estimate EE is also supported by a recently published article, which evaluated the ReSense, a newly developed accelerometer with integrated heartrate monitor, in MWU with SCI⁴⁷.

No studies were identified in this review evaluating devices for measuring EE in children or young adults who use a wheelchair for daily mobility. Therefore, there is a need for studies which specifically focus on the criterion validation of devices in this population.

Measuring the type of physical activity

Five devices were identified for measuring the type of PA. Both the PAMS and Vitamove are recommended for use, detecting the type of PA in the population who use a wheelchair. The PAMS was validated in adults with SCI⁴¹. A recommendation for future research is to validate the PAMS for a broader population MWU, with all kind of disabilities and especially for younger age groups. The Vitamove device showed its validity in both adults with SCI and youth with CP and SB^{39,40}.

While many studies use different instruments to report on PA in clinical populations, the vast majority of studies were conducted with individuals with SCI, which is a highly select client group. This is a considerable bias. The developed and validated algorithms for the SCI population is not necessarily also recommended and generalizable for use in other MWU / diagnosis groups. There are differences in participants' metabolic response to exercise, which may be attributable to differences in body composition, spinal cord lesion level and amount of body muscle mass. These factors influences the estimation of physical activity EE. In addition, there might be variation between participants in biomechanical efficiency due to differences in wheeling experience, skill level, and strength of the muscles involved in wheelchair propulsion. The latter has a particular influence on the detection of wheelchair activities, the type of PA. Future research should therefore be focused at validating instruments for objective measurements in other diagnosis groups than just those with a SCI.

Most of the studies in this systematic review evaluated the validity of accelerometers measuring the intensity or type of PA in a lab environment, instead of a real-life or semi-

natural environment like the two studies of the VitaMove^{39,40}, which makes the external validity unknown. In addition, measuring both the intensity and the type of activity in MWU has the advantage that advice regarding a healthy lifestyle can be made. It is currently unclear how much a person must be physically active for a healthy lifestyle. Wheeling 3 km per day for a healthy lifestyle in MWU with an SCI is recommended⁴⁷, which is based on a validated algorithm identifying the type and estimation of the intensity of different activities in a lab setting, but is not yet validated for a real-world setting. Future research should focus on developing algorithms measuring both the type and intensity of physical activity of daily living outside the lab as well for development of physical activity guidelines in wheelchair users.

Strengths and limitations

A strength of this systematic review is the use of a highly sensitive and validated search filter (97,4%) for finding studies on measurement properties, developed by Terwee et al. (2009)¹³. The use of a validated search filter also ensures the reproducibility of the search methods.

While the COSMIN checklist was originally developed for self-reported instruments¹⁹ and not for performance outcomes, it is commonly used for reviews looking at measurement properties of the latter^{48,49}. For future research, there is a need for a valid tool to critically appraise methodological quality of measurement outcomes. In addition, many studies in this systematic review have not provided a clear description of how missing items were handled in the analysis. This means that the study received a lower score for the methodological quality of the study which ultimately influenced the level of evidence negatively.

The established inclusion criteria have proved to be a limitation of the study. We were unable to include a number of studies with good methodological and outcome quality. The Sensewear⁵⁰⁻⁵² and ReSense⁴⁷ are objective measurement instruments that have been evaluated in studies and are recommended for use in daily practice and for research purposes. Unfortunately, the Sensewear is no longer for sale and is not supported by the manufacturer anymore. While the ReSense has been developed and validated by a research group and is not yet available widely to consumers. Therefore we had to exclude these studies from this systematic review. In addition, we were also unable to include an article from Leving at al.⁵³ this research group has developed and

validated an accelerometer (Activ8), which seems a promising tool, that can detect the types of wheelchair activity and the instrument contains a feedback tool for the user of daily PA. However, they performed the validation study with ambulatory adults (who are not familiar with wheelchair use) instead of people who depend on a wheelchair in daily life and are used to and experienced in wheelchair propulsion. For that reason we had to exclude this article. Overall, the use of accelerometers to measure PA for research goals or practical implications is complicated because in a short time many different accelerometers have become available and are still being developed whose validity is often unknown.

CONCLUSION

The PASIPD seems to be the most researched and the most promising self-reported instruments to measure PA in MWU and the PARA-SCI to assess PA in MWU with SCI. Device-based instruments that can be used for determining intensity of PA in adults who use a wheelchair are the GENEactive, Actigraph GT3X+, Actiheart and the PAMS. For measuring the type of PA, the PAMS is a valid device in adult who use a wheelchair and the VitaMove is proven valid in both adults with SCI as in the pediatric population MWU with SB or CP. It is important to realize that most of the instruments are restricted to measuring either intensity or the type of PA; this is important when choosing an instrument. Only the PAMS has been shown valid for assessment of both constructs.

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Abbreviations

CINAHL; Cumulative Index to Nursing and Allied Health Literature
 COSMIN; COnsensus-based Standards for the selection of health Measurement Instruments
 CP; Cerebral Palsy
 CRPS; Chronic Regional Pain Syndrome
 DLW; doubly labelled water technique
 EE; Energy Expenditure
 HR; Heartrate
 HR-PAEE; Heart Rate – Physical Activity Energy Expenditure
 ICC; Intraclass Correlation Coefficient;
 JdG; Janke de Groot
 KL; Kristel Lankhorst
 LA; Lifestyle Activity
 LPA; Light Physical Activity
 LTPA; Leisure Time Physical Activity
 LTPAQ-SCI; Leisure Time Physical Activity Questionnaire for people with Spinal Cord Injury
 MO; Michiel Oerbekke
 MMC; meningomyelocele
 MSE; mean signed percent error
 MVPA; Moderate to vigorous physical activity
 MWU; manual wheelchair users
 N; Number
 NA; Not applicable
 PA; Physical Activity
 PAI-SCI; Physical Activity Inventory for patients with Spinal Cord Injury
 PAEE; physical activity-associated energy expenditure
 PAMS; Physical Activity Monitor System
 PARA-SCI; Physical Activity Recall Assessment for People with Spinal Cord Injury
 PASIPD; Physical Activity Scale for individuals with Physical Disabilities
 r; Correlation
 SB: Spina Bifida
 SCI; Spinal Cord Injury
 TDEE; total daily energy expenditure

Supplement 1: Description of included instruments and website links to download or purchase the instruments.

Measurement instruments (developer or manufacturer)	Description
Included self-reported instruments	
Physical Activity Scale for Individuals with Physical Disabilities	<p>Captures information about leisure, household, and work related physical activity. Respondents have to recall the number of days and how many hours per day they participated in PA on average over the past seven days. It is developed for individuals with visual / auditory and locomotor or SCI disabilities. There is a 13-item and 11-item PASIPD. In the 13-item PASIPD, a score is calculated over 12 items (1 item is used for training) by multiplying the average hours per day for each activity by a Metabolic Equivalent of Task (MET) value associated with that activity, with a maximum of 199.6 MET hour per day. The 11-item PASIPD is the Dutch adapted translation of the 13-item PASIPD. Consequently, the maximum score is 182.3 MET hour per day. No online availability of this questionnaire. This questionnaire is available for purchase at: https://scireproject.com/outcome-measures/outcome-measure-tool/physical-activity-scale-for-individuals-with-physical-disabilities-pasipd/#1467983894177-6b9fb7a3-f550</p> <p>The PARA-SCI is an interviewer-administered three-day recall questionnaire for people with SCI using a wheelchair as their primary mode of mobility. Days are split in eight periods (e.g. morning, lunch, evening) on which the respondent is asked by a flowchart-assisted interviewer to recall activities. Time spent on an activity in daily life is registered as lifestyle activity, while activities done in leisure time are labeled as leisure time physical activity. Respondents rate the intensity of every activity as 'nothing at all', 'mild', 'moderate', or 'heavy'. The PARA-SCI is scored by averaging the sum of minutes per subscale or by minutes per intensity level within subscales. This questionnaire is available for purchase at: http://www.rehabmeasures.org/Lists/RehabMeasures/PrintView.aspx?ID=1051</p>
The Leisure Time Physical Activity Questionnaire for people with Spinal Cord Injury (LTPAQ-SCI)	<p>The LTPAQ-SCI is an interviewer-administered seven-day recall questionnaire regarding activities done during leisure time and uses the same intensity classifications as the PARA-SCI. The questionnaire can be downloaded via: http://sclactioncanada.ca/docs/research-publications/leisure-time-physical-activity-questionnaire%20.pdf</p>
The Physical Activity Inventory for patients with Spinal Cord Injury (PAI-SCI)	<p>The PAI-SCI is a revision of the PASIPD, and is a 7-day recall, 14-item questionnaire, which focuses on activities done by persons with SCI. Respondents have to recall the activities per day in which they participated in the last week. Also they have to indicate the number of minutes or hours spent in an activity to calculate the sum of average daily hours at a particular (MET). In the article of Butler et al. (3) a brief explanation in the method section is stated about the content of the PAI-SCI. https://www.ncbi.nlm.nih.gov/pmc/articles/PMC4151181/</p>

Included device-based instruments	
Actigraph GT3X (Actigraph)	<p>The wGT3X-BT is an activity monitor, to capture and record continuous, high resolution physical activity and sleep/wake information. The Bluetooth® Smart wGT3X-BT features ActiGraph's validated 3-axis accelerometer and digital filtering technology and includes integrated wear time and ambient light sensors. Specifications can be found at: http://actigraphcorp.com/products-showcase/activity-monitors/actigraph-wgt3x-bt/</p>
Actiheart	<p>The Actiheart is typically worn on two standard ECG pads on the chest. The small size of the recorder and the use of standard biocompatible ECG electrodes ensure that the Actiheart monitor is securely but comfortably attached. The Actiheart is designed for multiple day use. It is water resistant and is compatible with daily activities. https://www.camntech.com/products/actiheart/wearing-the-actiheart</p>
Polar	<p>Polar is a company that develops sensors to monitor heart rate. Polar heart rate sensors are compatible with top fitness apps, gym equipment and many other Bluetooth devices. You can even monitor your heart rate in water. Polar equipment has built-in memory for heart rate data. The data can be transferred to Polar's free fitness and training app. https://www.polar.com/en</p>
GeneActiv (Activinsights)	<p>GENEActiv is a wrist-worn, raw data accelerometer for behavioral measurement. Designed for 24-hour wear and both free-living and clinical studies measuring activities and movement frequencies. Specifications can be found at: https://www.geneactiv.org/products/</p>
RT3 Accelerometer (Stayhealthy, Inc.)	<p>The RT3 accelerometer used as an experimental tool for measuring physical activity. It is worn and clipped to the waistband as an 'accessory' during waking hours. The sensor in the RT3 is an accelerometer sensitive along three orthogonal axes (X, Y and Z). the acceleration is measured periodically, converted to a digital representation, and processed to obtain an 'activity count', which is stored in memory. Specifications of this device can be found at: http://www.stayhealthy.com/en_us/main/index.html</p>
Physical Activity Monitor System (PAMS) (no manufacturer)	<p>The PAMS is a two-axis gyroscope in combination with an accelerometer. A two-axis gyroscope is attached to the spokes of the wheelchair with an accelerometer placed on the right upper arm or at the wrist of the wheelchair user. No specification can be found online. More information is described in the article of Hiremath et al.</p>

Vitaport / Vitamove (ZIM Engineering)	<p>The Vitaport and VitaMove are multisensory systems. Several accelerometers are connected to a monitor. In WCD persons one sensor is attached to the sternum and one on each wrist. Recorded data of the Vitaport and VitaMove provides information about body postures and movements (i.e. in persons who are WCD; lying, sitting, wheelchair propulsion and hand cycling) transitions between postures, motions, frequency, duration, and intensity in daily life.</p> <p>Specification of this device can be found at: http://www.vitamove.nl/</p> <p>Specifications and further details of this device can be found at: http://www.usa.philips.com/healthcare/product/HC1046964/actiwatch-spectrum-activity-monitor</p> <p>Specifications and further details of this device can be found at: https://www.camntech.com/products/pro-diary/pro-diary-specifications</p>
Actiwatch (Philips)	
PRO-Diary (CamNtech)	
SCI: Spinal Cord Injury, MET: Metabolic Estimation of Task, WCD: Wheelchair Dependent	

REFERENCES

1. Despres JP. Physical activity, sedentary behaviours, and cardiovascular health: When will cardiorespiratory fitness become a vital sign? *Can J Cardiol* 2016;32(4):505-513. doi: 10.1016/j.cjca.2015.12.006.
2. Flank P, Wahman K, Levi R, Fahlstrom M. Prevalence of risk factors for cardiovascular disease stratified by body mass index categories in patients with wheelchair-dependent paraplegia after spinal cord injury. *J Rehabil Med* 2012;44(5):440-443. doi: 10.2340/16501977-0964.
3. Flank P, Fahlstrom M, Bostrom C, Lewis JE, Levi R, Wahman K. Self-reported physical activity and risk markers for cardiovascular disease after spinal cord injury. *J Rehabil Med* 2014;46:886-890. doi: 10.2340/16501977-1857.
4. Liang H, Chen D, Wang Y, Rimmer JH, Braunschweig CL. Different risk factor patterns for metabolic syndrome in men with spinal cord injury compared with able-bodied men despite similar prevalence rates. *Arch Phys Med Rehabil* 2007;88(9):1198-1204. doi: S0003-9993(07)00384-X.
5. Durstine JL, Painter P, Franklin BA, Morgan D, Pitetti KH, Roberts SO. Physical activity for the chronically ill and disabled. *Sports Med* 2000;30(3):207-219.
6. Proper KI, Singh AS, van Mechelen W, Chinapaw MJ. Sedentary behaviors and health outcomes among adults: A systematic review of prospective studies. *Am J Prev Med* 2011;40(2):174-182. doi: 10.1016/j.amepre.2010.10.015.
7. Thorp AA, Owen N, Neuhaus M, Dunstan DW. Sedentary behaviors and subsequent health outcomes in adults a systematic review of longitudinal studies, 1996-2011. *Am J Prev Med* 2011;41(2):207-215. doi: 10.1016/j.amepre.2011.05.004.
8. Wilmot EG, Edwardson CL, Achana FA, Davies MJ, Gray LJ, Khunti K, Yates T, Biddle SJ. Sedentary time in adults and the association with diabetes, cardiovascular disease and death: Systematic review and meta-analysis. *Diabetologia* 2012;55(11):2895-2905. doi: 10.1007/s00125-012-2677-z.
9. Kooijmans H, Horemans HL, Stam HJ, Bussmann JB. Valid detection of self-propelled wheelchair driving with two accelerometers. *Physiol Mea*. 2014;35(11):2297-2306. doi: 10.1088/0967-3334/35/11/2297.
10. Maher CA, Williams MT, Olds T, Lane AE. Physical and sedentary activity in adolescents with cerebral palsy. *Dev Med Child Neurol* 2007;49(6):450-457. doi: DMCN450.
11. Shkedy Rabani A, Harries N, Namoor A, Al-Jarrah MD, Karniel A, Bar-Haim S. Duration and patterns of habitual physical activity in adolescents and young adults with cerebral palsy. *Dev Med Child Neurol* 2014;56(7):673-680. doi: 10.1111/dmcn.12394.

12. Warms CA, Whitney JD, Belza B. Measurement and description of physical activity in adult manual wheelchair users(). *Disabil health journal* 2008;1(4):236-244. doi: 10.1016/j.dhjo.2008.07.002.
13. Terwee CB, Jansma EP, Riphagen II, de Vet HC. Development of a methodological PubMed search filter for finding studies on measurement properties of measurement instruments. *Qual Life Res* 2009;18(8):1115-1123. doi: 10.1007/s11136-009-9528-5.
14. Vet de, H, Terwee C, Mokkink, LB, Knol, DL. *Measurement in medicine* (3th ed.) New York: Cambridge University Press; 2014.
15. The COSMIN group. Guideline for systematic reviews of outcome measurement instruments. <https://www.cosmin.nl/tools/guideline-conducting-systematic-review-outcome-measures/>.
16. Qi X, Yang M, Ren W, Jia J, Wang J, Han G, Fan D. Find duplicates among the PubMed, EMBASE, and cochrane library databases in systematic review. *PLoS One* 2013;8(8):e71838. doi: 10.1371/journal.pone.0071838.
17. Bussmann JB, van den Berg-Emons RJ. To total amount of activity..... and beyond: Perspectives on measuring physical behavior. *Front Psychol* 2013;4:463. doi: 10.3389/fpsyg.2013.00463.
18. Cooper RA, Koontz AM, Ding D, Kelleher A, Rice I, Cooper R. Manual wheeled mobility - current and future developments from the human engineering research laboratories. *Disabil Rehabil* 2010;32:2210-2221. doi: 10.3109/09638288.2010.517599.
19. Terwee CB, Bot SD, de Boer MR, et al. Quality criteria were proposed for measurement properties of health status questionnaires. *J Clin Epidemiol* 2007;60(1):34-42. doi: S0895-4356(06)00174-0.
20. Leving M.T., Horemans H.L.D., Vegter R.J.K., De GS, Bussmann J.B.J., van der WL. Validity of consumer-grade activity monitor to identify manual wheelchair propulsion in standardized activities of daily living. *PLoS ONE* 2018;13(4). doi: 10.1371/journal.pone.0194864.
21. Fanchamps MHJ, Horemans HLD, Ribbers GM, Stam HJ, Bussmann JBJ. The accuracy of the detection of body postures and movements using a physical activity monitor in people after a stroke. *Sensors (Basel)* 2018;18(7):10.3390/s18072167. doi: E2167.
22. Mitchell LE, Ziviani J, Oftedal S, Boyd RN. A systematic review of the clinimetric properties of measures of habitual physical activity in primary school aged children with cerebral palsy. *Res Dev Disabil* 2013;34(8):2419-2432. doi: 10.1016/j.ridd.2013.04.013.
23. Tanhoffer RA, Tanhoffer AI, Raymond J, Hills AP, Davis GM. Comparison of methods to

- assess energy expenditure and physical activity in people with spinal cord injury. *J Spinal Cord Med* 2012;35(1):35-45. doi: 10.1179/2045772311Y.00000000046.
24. de Groot S, van der Woude LH, Niezen A, Smit CA, Post MW. Evaluation of the physical activity scale for individuals with physical disabilities in people with spinal cord injury. *Spinal Cord* 2010;48(7):542-547. doi: 10.1038/sc.2009.178.
 25. van den Berg-Emons RJ, L'Ortye AA, Buffart LM, et al. Validation of the physical activity scale for individuals with physical disabilities. *Arch Phys Med Rehabil* 2011;92:923-928. doi: 10.1016/j.apmr.2010.12.006.
 26. Washburn RA, Zhu W, McAuley E, Frogley M, Figoni SF. The physical activity scale for individuals with physical disabilities: Development and evaluation. *Arch Phys Med Rehabil* 2002;83(2):193-200. doi: <http://dx.doi.org/10.1053/apmr.2002.27467>.
 27. Claridge EA, McPhee PG, Timmons BW, Ginis KA, MacDonald MJ, Gorter JW. Quantification of physical activity and sedentary time in adults with cerebral palsy. *Med Sci Sports Exerc* 2014. doi: 10.1249/mss.0000000000000589.
 28. Ginis KA, Latimer AE, Hicks AL, Craven BC. Development and evaluation of an activity measure for people with spinal cord injury. *Med Sci Sports Exerc* 2005;37(7):1099-1111. doi: 00005768-200507000-00004.
 29. Latimer AE, Ginis KA, Craven BC, Hicks AL. The physical activity recall assessment for people with spinal cord injury: Validity. *Med Sci Sports Exerc* 2006;38(2):208-216. doi: 10.1249/01.mss.0000183851.94261.d2.
 30. Martin Ginis KA, Phang SH, Latimer AE, Arbour-Nicitopoulos K. Reliability and validity tests of the leisure time physical activity questionnaire for people with spinal cord injury. *Arch Phys Med Rehabil* 2012;93:677-682. doi: 10.1016/j.apmr.2011.11.005.
 31. Butler JA, Miller T, O'Connell S, Jelinek C, Collins EG. Physical activity inventory for patients with spinal cord injury. *SCI Nursing* 2008;25:20-29.
 32. Kelly P, Fitzsimons C, Baker G. Should we reframe how we think about physical activity and sedentary behaviour measurement? validity and reliability reconsidered. *Int J Behav Nutr Phys Act* 2016;13:32-016-0351-4. doi: 10.1186/s12966-016-0351-4.
 33. Nightingale TE, Walhin JP, Thompson D, Bilzon JL. Influence of accelerometer type and placement on physical activity energy expenditure prediction in manual wheelchair users. *PLoS One* 2015;10(5):e0126086. doi: 10.1371/journal.pone.0126086.
 34. Nightingale TE, Walhim JP, Thompson D, Bilzon JL. Predicting physical activity energy expenditure in manual wheelchair users. *Med Sci Sports Exerc* 2014;46:1849-1858. doi: 10.1249/mss.0000000000000291.

35. Garcia-Masso X, Serra-Ano P, Garcia-Raffi L, Sanchez-Perez E, Lopez-Pascual J, Gonzalez LM. Validation of the use of actigraph GT3X accelerometers to estimate energy expenditure in full time manual wheelchair users with spinal cord injury. *Spinal Cord* 2013;51:898-903.
36. Hiremath SV, Ding D. 3066485; evaluation of activity monitors in manual wheelchair users with paraplegia. *J Spinal Cord Med* 2011;34:110-117. doi: 10.1179/107902610x12911165975142.
37. Hiremath SV, Intille SS, Kelleher A, Cooper RA, Ding D. Estimation of energy expenditure for wheelchair users using a physical activity monitoring system. *Arch Phys Med Rehabil* 2016;97(7):1146-1153.e1. doi: 10.1016/j.apmr.2016.02.016.
38. Nightingale TE, Walhin JP, Thompson D, Bilzon JL. Predicting physical activity energy expenditure in wheelchair users with a multisensor device. *BMJ Open Sport Exerc Med* 2015;1(1):10.1136/bmjsem-2015-000008. eCollection 2015. doi: bmjsem-2015-000008.
39. Postma K, Bussmann J, Sluis T, Bergen M, Stam H. Validity of the detection of wheelchair propulsion as measured with an activity monitor in patients with spinal cord injury. *Spinal Cord* 2005;43(9):550-557.
40. Nooijen CFJ, De Groot JF, Stam HJ, Van DB, Bussmann HBJ. Validation of an activity monitor for children who are partly or completely wheelchair-dependent. *J Neuroeng Rehabil* 2015;12.
41. Hiremath SV, Intille SS, Kelleher A, Cooper RA, Ding D. Detection of physical activities using a physical activity monitor system for wheelchair users. *Med Eng Phys* 2015;37:68-76. doi: 10.1016/j.medengphy.2014.10.009.
42. Garcia-Masso X, Serra-Ano P, Gonzalez LM, Ye-Lin Y, Prats-Boluda G, Garcia-Casado J. Identifying physical activity type in manual wheelchair users with spinal cord injury by means of accelerometers. *Spinal Cord* 2015; 53, 772-777.
43. Warms CA, Belza BL. Actigraphy as a measure of physical activity for wheelchair users with spinal cord injury. *Nurs Res* 2004;53:136-143.
44. Murphy SL, Kratz AL, Zynda AJ. Measuring physical activity in spinal cord injury using wrist-worn accelerometers. *Am J Occup Ther* 2019;73(1):7301205090p1-7301205090p10. doi: 10.5014/ajot.2019.027748.
45. Vet dH, Terwee C, Mookink L, Knol D. 6.4 criterion validity. in: measurement in medicine. practical guide to biostatistics and epidemiology. In: 3th ed. New York, United States of America: Cambridge University Press; 2013:159.
46. Hiremath SV, Ding D, Cooper RA. Development and evaluation of a gyroscope-based

- wheel rotation monitor for manual wheelchair users. *J Spinal Cord Me.* 2013;36:347-356. doi: 10.1179/2045772313y.0000000113.
47. Popp WL, Richner L, Brogioli M, et al. Estimation of energy expenditure in wheelchair-bound spinal cord injured individuals using inertial measurement units. *Front Neurol* 2018;9:478. doi: 10.3389/fneur.2018.00478.
48. Bartels B, de Groot JF, Terwee CB. The six-minute walk test in chronic pediatric conditions: A systematic review of measurement properties. *Phys Ther* 2013;93(4):529-541. doi: 10.2522/ptj.20120210.
49. Dobson F, Hinman RS, Hall M, Terwee CB, Roos EM, Bennell KL. Measurement properties of performance-based measures to assess physical function in hip and knee osteoarthritis: A systematic review. *Osteoarthritis Cartilage* 2012;20(12):1548-1562. doi: 10.1016/j.joca.2012.08.015.
50. Tsang K, Hiremath SV, Cooper RA, Ding D. Evaluation of custom energy expenditure models for SenseWear armband in manual wheelchair users. *J Rehabil Res Dev* 2015;52(7):793-803. doi: 10.1682/JRRD.2014.08.0188.
51. Hiremath SV, Ding D, Farringdon J, Vyas N, Cooper RA. Physical activity classification utilizing SenseWear activity monitor in manual wheelchair users with spinal cord injury. *Spinal Cord* 2013;51:705-709. doi: 10.1038/sc.2013.39.
52. Hiremath SV, Ding D, Farringdon J, Cooper RA. Predicting energy expenditure of manual wheelchair users with spinal cord injury using a multisensor-based activity monitor. *Arch Phys Med Rehabil* 2012;93:1937-1943. doi: 10.1016/j.apmr.2012.05.004.
53. Leving MT, Horemans HLD, Vegter RJK, de Groot S, Bussmann JBJ, van der Woude LHV. Validity of consumer-grade activity monitor to identify manual wheelchair propulsion in standardized activities of daily living. *PLoS One* 2018;13(4):e0194864. doi: 10.1371/journal.pone.0194864.



9

General discussion

Kristel Lankhorst

The main objective of this study was to investigate the association of (adapted) sports participation with health-related fitness, physical activity, psychosocial health, and injuries and illness among youngsters with CDPD. A secondary objective was to develop and validate an instrument to measure physical activity in ambulatory youngsters with and without motor disability. Furthermore, based on scientific evidence, recommendations were made for instruments measuring physical activities in daily life in youngsters who use a wheelchair.

In the following paragraphs, the main findings, theoretical considerations, interpretation of results, methodological considerations, practical implementation, future perspectives, recommendations and conclusions are summarized and discussed.

MAIN FINDINGS

In summary:

- Youngsters with CDPD participating at least twice weekly in (adapted) organized sports performed better on all health-related outcome parameters, compared with their non-sporting peers and those who participated in sports once a week. They had significant higher $\text{VO}_{2\text{peak}}$, achieved higher power on the muscle power sprint test (MPST), were faster on the 10 x 5 m sprint test, had higher mean grip strength, and were able to jump further horizontally (**Chapter 4**).
- Youngsters with CDPD participating at least twice weekly had significant lower waist: hip circumference ratio standard deviation scores (SDS) and lower percentages of fat mass, compared with their non-sporting peers and those who participated in sports once a week (**Chapter 4**).
- Youngsters with CDPD participating at least twice weekly in sports were more physically active during school days (> 30 minutes per day) and during the weekend (25 minutes per day), compared to their non-sporting peers and those who participated in sports once per week (**Chapter 4**).
- The positive association between sports participation and peak oxygen uptake was influenced by an increased physical activity (PA) level (31%). We did not observe any significant association between sports participation and sedentary time (**Chapter 4**).
- Participation in (adapted) organized sports at least twice weekly in youngsters with CDPD contributes to all domains of health-related quality of life and

feelings of athletic competence (**Chapter 5**).

- Children (10 – 12 years of age), but not adolescents (13 to 18 years of age), participating in sports reported higher feelings of social acceptance. Sports participation was strongly associated with exercise self-efficacy in both children and adolescents (**Chapter 5**).
- Lower cardiorespiratory fitness and a higher waist circumference were associated with a higher arterial stiffness in youngsters with CDPD. The association between cardiorespiratory fitness and arterial stiffness was no longer statistically significant after adjustment of waist circumference. No association between sports participation and arterial stiffness was found (**Chapter 6**).
- Thresholds of less than 35 ml/kg/min for VO_{2peak} , more than 73 cm for waist circumference and over -0.05 units for waist circumference SDS were associated with increased arterial stiffness (**Chapter 6**). These results suggest that waist circumference already at normal range at the population level is associated with increased arterial stiffness. Interventions aiming to decrease body adiposity and to improve cardiorespiratory fitness may improve arterial stiffness among youngsters with CDPD.
- Participation in sports at least two times a week does not pose a significant increased risk in the incidence of injury or illness per 1000 hours of PA in youngsters with CDPD compared to once weekly or no sports participation (**Chapter 7**).
- Sports participants who participate in sports at least two times a week get injured mostly during their sporting activities, while peers who participate in sports once a week or not at all, get injured during less intense physical activities like physical education (PE) lessons, activities of daily living (ADL) or non-organized sports and play in leisure time (**Chapter 7**).
- The Activ8 is a valid tool measuring the type of physical (in)activity in daily life in youngsters with typical development and peers with motor disability, and can be used to monitor and evaluate interventions targeting PA (**Chapter 3**).
- According to current literature, the PASIPD and PARA-SCI seems the most promising self-reported instruments for measuring the intensity of PA in people who use a wheelchair (**Chapter 8**).
- The GENEactive, Actigraph GT3X+, Actiheart and the PAMS, showed positive criterion validity to measure the intensity of PA in people who use a wheelchair.

For measuring the type of PA the PAMS and VitaMove are suitable, showing both good evidence for criterion validity. The practical usage of each of these instruments in daily life varies (**Chapter 8**). Use of a heart rate monitor with individual calibration function, is an useful, easy and inexpensive option.

Theoretical considerations

The associations studied in this thesis are based on the model of Shephard & Bouchard¹, as described in the general introduction in which associations between health-related fitness and morbidity and mortality are assumed. In children and adolescents, however, these life-threatening diseases or behaviors are not directly addressed by the model. Our underlying motivation therefore is the prevention of life-threatening diseases at a later age by intervention at an earlier age. At the start of this study, our knowledge of preventive strategies such as increasing physical activity by regular sports participation among youngsters with physical disability or chronic disease (CDPD) was limited. Figure 1 is a schematic representation of the relations studied in this thesis, based on the model of Shephard & Bouchard for adults, but adapted for children and adolescents.

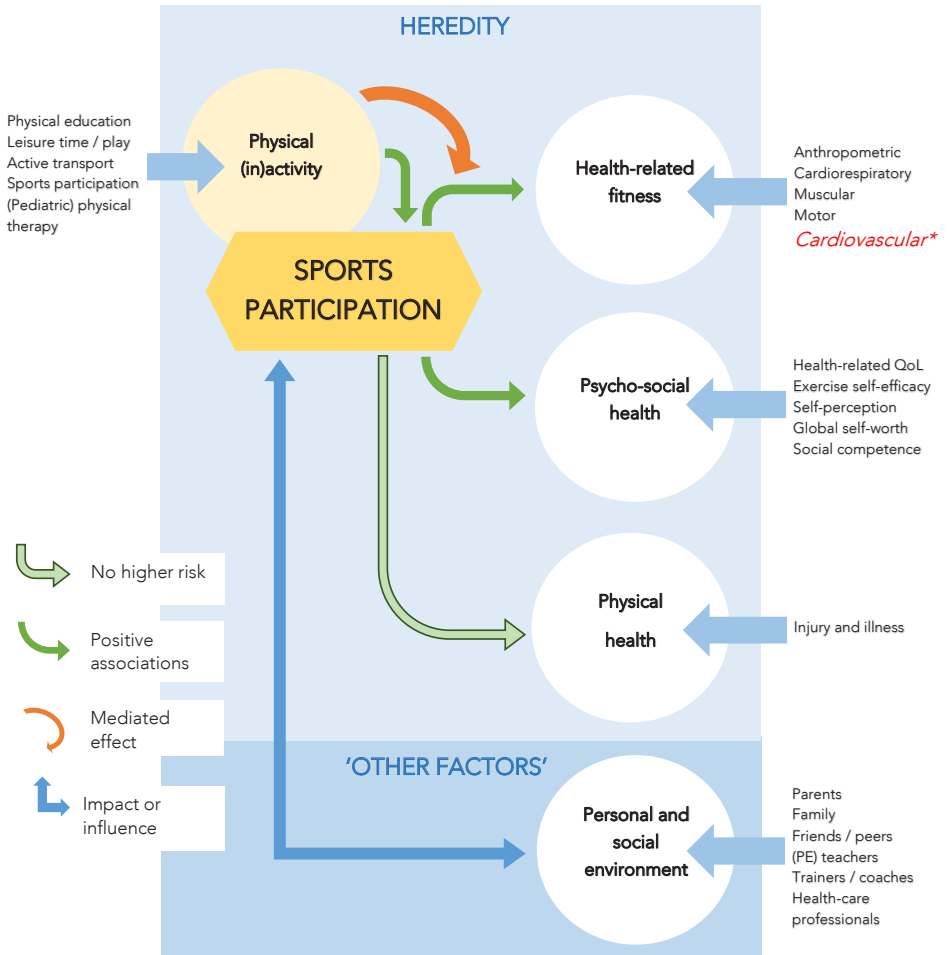


Figure 1: Schematic representation of the relations studied in this thesis.

*Sports participation was not associated with cardiovascular outcome parameters (arterial stiffness).

QoL: quality of life; PE: physical education.

INTERPRETATIONS OF RESULTS

Study population

The population studied in the current thesis is heterogeneous, based on medical diagnoses. Therefore, the population enrolled in this study cannot be representative of the total population of children and adolescents with chronic diseases or physical disability (CDPD) in the Netherlands. National data show pulmonary diseases (mainly asthma) to have the greatest prevalence (13.7%), followed by neurological diseases (8.9%)². Almost half of the participants studied in this thesis have a neuromuscular disorder, i.e. cerebral palsy (CP) or spina bifida (SB), the largest patient group seen within pediatric rehabilitation medicine.

CP and SB are permanent disorders in terms of the development of movement and posture. Signs and symptoms of these disorders vary across individuals and for each diagnosis. Individuals with neuromuscular disorders often experience problems with daily functioning, having poor fundamental movement skills due to muscle impairments. These muscle impairments result in reduced strength and coordination in affected muscles, so more time is needed to complete motor tasks^{3,4}. In addition, problems with cognitive functioning and mental health are common in individuals with CP or SB⁵. In this thesis, all children and adolescents with a CDPD, independent of their diagnosis or level of mobility (except those using a powered wheelchair) were included. The Functional Mobility Scale (FMS)⁶ was used to classify participants, based on their level of mobility. To specify the functional mobility in more detail in youngsters with neuromuscular disorders (CP and SB), the Gross Motor Function Classification System (GMFCS)^{7,8} was used for participants with CP and the adapted Hoffer classification⁹ for participants with SB. Youngsters with CP or SB in this thesis were all rated with a FMS score of 6 (able to walk independently on all surfaces) or 5 (able to walk independently on level surfaces but requiring a rail for stairs). From the perspective of functional mobility, individuals with these scores are the least restricted in daily life due to their neuromuscular disorder. All other participants with a chronic disease or disability were rated with a FMS score of 6.

We recruited participants via schools for special education, pediatric physical therapy practices, (adapted) sports organizations and via the outpatient clinic of the Wilhelmina Children's Hospital (Utrecht, the Netherlands). As described in the general introduction, next to the HAYS study, the Sport-2-Stay-Fit (S2SF) study¹⁰ was performed concurrently by our research group. The S2SF study was executed within schools for special

education, at both primary and secondary level. Some participants of the S2SF study also participated in the HAYS study. This selective approach of recruitment may have resulted in a population that is more in line with the population of pediatric rehabilitation medicine, rather than with the total population of youngsters with CDPD in the Netherlands². Interestingly, it was easier to recruit sports participants with CDPD for participation in this study than non-sporting peers. It is likely that youngsters who are physically active through sports participation are more attracted to a study that focuses on sports which are familiar to them, than their peers who do not have (positive) athletic experience. Moreover, it was easier to approach the sporting group for participation, contacting sport organizations being a simple and effective way of recruiting potential sporting participants.

Sports participation, health-related fitness and physical activity

Youngsters with CDPD participating in organized sports at least twice weekly benefit optimally from the positive effects of sports¹¹. Youngsters with CDPD participating at least twice weekly in sports were more physically active during school days (> 30 minutes per day) and during the weekend (25 minutes per day) than their non-sporting peers and those who participated in sports once a week. Physical activity was a strong predictor of aerobic fitness, whereas total sedentary time had no effect on peak oxygen uptake. In particular, youngsters with CP or SB with muscle weakness showed in our study that they can achieve VO_{2peak} values within the normal range for healthy Dutch peers. In addition, anaerobic fitness, agility and strength measurement outcomes were significant higher in sports participants compared to their non-sporting peers and those who participated in sports once a week; organized sports contributes to better muscular function (Panel 9.1: health-related fitness and muscular function).

Parallel to the HAYS study, the S2SF study was performed by our research group, measuring similar outcome measures. The main objective of the S2SF study was to investigate the effects of a short-term high-intensity interval training (HIIT) followed by a school-based sports program once a week for six months in youngsters with CDPD. The HIIT program (8 weeks) resulted in improvements in agility, anaerobic and aerobic fitness, while VO_{2peak} levels remained stable. These results indicates that sports participation once a week (at school) is sufficient to maintain, but insufficient to improve health-related fitness. These outcome, indicate that a frequency of at least twice a week is needed to achieve improvement on VO_{2peak} .

Panel 9.1: health-related fitness and muscular function

Joris is a 14-year old boy diagnosed with spastic, hemilateral cerebral palsy (CP). According to the GMFCS, he is classified as Level I. He walks and runs independently at home and at school (FMS 6). He is a very talented soccer player. At the time he participated in this research, he was playing in the Dutch CP Football talent team. He began training more intensively and more frequently, from 2 to at least 5 times per week in the past year. He showed progression in his football skills and he was also less restricted by his condition. He became stronger, both in muscle function and fitness level, which seemed like positive (side) effects.

As CP Football is a team sport, classification aims at ensuring fairness with regard to the impact of impairment between both teams. To accomplish this, the players are allocated to sports classes depending on how much their impairment impacts performance in their sport. CP Football has 3 classes; FT1, FT2 and FT3. For example, in FT1, the condition limits sport performance more than FT2 and FT3. To ensure a fair game between teams, each team has to have one FT1 player on the field at all times and is not allowed to have more than one FT3 player on the field.

Unfortunately, Joris' progression resulted in a less restricted classification for CP Football (from FT2 to FT3), which meant that, due to his positive progression in terms of aerobic and anaerobic performance, he became 'too good'. There was no place for another FT3 player in the team. Consequently, he was not selected for the CP Football talented team for the next year.

It is well known that being overweight or obese in childhood is associated with a higher risk of cardiovascular diseases, various types of cancer and premature death in adulthood¹²⁻¹⁴. The percentage of typically-developing youngsters aged 12 to 18 with overweight (13.8%) and obesity (2.3%) increased significantly between 1990 and 2017, among both Dutch boys and girls^{15,16}. Of Dutch youngsters with CDPD, 14% of the children aged 4 to 12 years and 19% of the youngsters aged 12 to 18 years are overweight or obese¹⁷. The increasing trend and high percentages is alarming for their future health, for both typically developing youngsters and their peers with CDPD. As a result of the S2SF study, youngsters with CDPD who participated in sports once a week for 45 minutes had lower percentages of fat mass after six months¹⁰. In our study, youngsters with CDPD participating in sports at least twice weekly had a significant lower waist-hip circumference ratio score and a lower percentage of fat mass than their non-sporting peers and those who participated in sports once a week, while those with a pathological gait pattern had a significantly higher percentage of fat mass and a higher waist-hip circumference ratio compared to peers with typically gait pattern.

These results indicates that sports participation once per week already contributes to a

lower percentage of fat mass, as the S2SF study shows. However, sports participation of at least twice a week is even better as it contributes to multiple health-related outcomes (aerobic and anaerobic fitness, physical activity level, agility and strength), especially for those youngsters with a pathological gait pattern.

Psychosocial health

Besides the positive associations with health-related fitness and PA, organized sports participation at least twice weekly resulted also in better psychosocial health outcomes; 1) a better health-related quality of life, 2) feelings of athletic competence, 3) greater feelings of social acceptance and 4) greater exercise self-efficacy¹⁸.

Panel 9.2: Psychosocial health

"Participation in sports gives me more self-confidence, self-compassion, and I am more confident in life." Ellis van Loenhout, World Champion Paralympic road cyclist, 2014.

In the S2SF study, no improvements were observed within the psychosocial domain from a school-based sports program once a week for six months¹⁹, which indicates that school-based sports may have different impacts than sports club participation. In the S2SF study, a school-based sports program was chosen, to remove possible barriers to sports participation. The underlying thought was that a school setting offers a familiar environment with supportive and well-educated trainers, acceptance, and no additional transportation to and from a sports club. In addition, school-based interventions are thought to be the most universally applicable and effective way to counteract low PA and fitness, since children and adolescents spend at least half of their waking hours in this setting²⁰. Improvements in anaerobic fitness and reduced fat mass were observed in the S2SF study, while no improvements were observed within the psychosocial domain. There is a possibility that, despite the benefits of school-based sport, sports participation at a club might possess components that engage children in a different way than do school-based sports²¹. For example, children playing in sports teams may have more intrinsic motivation than do youngsters who play at school for rewards or recognition from others²². Besides, children who play sports in school settings may be more prone to discontinue in sports participation after graduation because they have more barriers in adapting to PA participation to a new setting²³, so school-based sports may not be the

solution to overcoming all barriers. This outcome emphasizes even more the importance of finding other ways to enable organized sports participation for youngsters with CDPD. One possibility may be practicing individual sports which are less hindered by barriers and are therefore more easy to participate in than team sports for youngsters with CDPD. In addition, the advantage of participation in individual adapted forms of sports is that youngsters can better focus on their abilities rather than their inabilities²⁴. However, involvement in a team sport at a sports club, and interaction with others in this context, allows youngsters to develop their self-concept, especially related to the physical and social domains. Sports club and team-based sports participation may therefore have different impacts on psychosocial health outcomes than do individual-based sports¹⁸. It is my recommendation to investigate the influence of team versus individual sports participation on psychosocial health.

Cardiovascular health

In our study, sports participation was positively associated with all health-related fitness outcomes, with the exception of cardiovascular health (arterial stiffness)^{11,25}. This was the only outcome parameter as a sub-domain of health-related fitness where sports participation had no direct association (Figure 1). Poor cardiorespiratory fitness (a low peak oxygen uptake) and higher waist circumference were associated with increased arterial stiffness (higher pulse wave velocity (PWV)) among youngsters with CDPD. Better cardiorespiratory fitness is related to more compliant arteries, whereas lower levels of body adiposity could not explain the relationship in our study²⁵. Presumably, hereditary factors, social environment and lifestyle behavior (nutritional intake) had an impact on this relationship. These (sub)domains, according to the model of Shephard & Bouchard, were not investigated in this study. A systematic review on dietary and dietary measures nutrients as monotherapy for arterial stiffness showed that adding omega-3 to the diet resulted in a decrease in PWV²⁶. At the same time, salt restriction, soy-rich food, moderate alcohol consumption and smoking cessation were also associated with a decreased PWV²⁷. It should be noted, however, that alcohol use or smoking behavior was almost non-existent in youngsters in our research. Furthermore, social and personal environmental and hereditary factors such as: 1) secondhand smoking, 2) paternal hypertension, and 3) maternal obesity turned out to be additional, independent risk factors for higher PWV. The aforementioned hereditary factors, personal and environmental variables, not investigated in this study, are recommended for future research.

Knowledge of the impact of environmental variables and heredity will provide insight and help support the development of specific (behavioral) interventions for both children and their parents to prevent early arterial stiffness. In addition, it is important to determine the optimal training regime in terms of frequency, duration, and intensity (dose-response relationships) of sport activities to elicit favorable effects on arterial stiffness. Currently, the results of this dissertation and other studies indicate that interventions aimed at reducing body adiposity (i.e. diet and facilitating healthy nutrition habits) and improving cardiorespiratory fitness (exercise) may improve arterial stiffness among youngsters with CDPD.

Physical health

From the literature, we know that an unfavorable body composition, low levels of PA and/or physical fitness levels are related to poor fundamental movement skills²⁸⁻³⁰ and increased injury levels³¹. Sports participants in the HAYS study scored better on all anaerobic tests than their non-sporting peers and those who participated in sports once a week¹¹. Fundamental movement skills are required to achieve optimal performance on anaerobic tests³². In our study, sports participation at least twice weekly does not pose a higher risk of (sports)-related injuries in youngsters with CDPD³³, which probably indicates that fundamental movement skills are well trained and developed in youngsters who participate in sports at least twice weekly. In addition, poor fundamental movement skills negatively influence the degree of physical activity³⁴. The sporting participants in our study were more active overall during the weekend and schooldays than their non-sporting peers or those who participated once a week, which contributes to a healthy lifestyle.

The fear of (sports-related) injuries in youngsters with CDPD and their parents is baseless, while even youngsters who do not participate in sports get injured. Athletes who participate in sports at least twice weekly get injured mostly during their sporting activities, while peers who participate in sports once a week or not at all get injured during less intense physical activities, such as during physical education lessons, ADL or non-organized sports and play in leisure time. Fortunately, the social impact (severity) of (sports-related) injuries was limited overall, but must still be an important focus for further research, with focus on the prevention of (sports-related) injuries. It would be interesting to investigate the association between incidence of injury and health-related fitness and PA outcomes in order to gain insights into the exact mechanisms and risk of

injuries. Follow-up research with the HAYS dataset is therefore recommended. With that knowledge, sports injury prevention training programs could be developed, including strategies that target all known risk factors for (sports-related) injury in youngsters with CDPD.

METHODOLOGICAL CONSIDERATIONS

Study design

The design of the HAYS study was a cross-sectional one³⁵, which fitted the aim and allowed us to include a large and diverse group within an age range of 10 to 18 years and with varying medical diagnoses. However, the cross-sectional design limited conclusions on causality. It may be that those who are physically fit and athletically competent experience a better quality of life, or those who feel more socially accepted are more likely to participate in (adapted) organized sports. A longitudinal design would have allowed analyses focusing on underlying pathways.

The crudeness of the measure of sports participation can be seen as a study limitation. In our study, no association between sports participation and arterial stiffness was found while all other outcome parameters of health-related fitness were positively correlated¹¹. It may be that sports participation as a dichotomous outcome measure has insufficient distinctive capacity (statistically) to identify beneficial effects on arterial stiffness. Therefore it seems worthwhile to investigate the relationship between the objectively measured physical activity and arterial stiffness.

The number of years of sports participation, as well as the duration, intensity and type of the sports activities, has not been measured. The positive associations that we have found between sports participation and psychosocial health parameters¹⁸ could be further explored with this sports-specific additional information. It may be that team sports have different impacts on psycho-social health parameters than do individual sports, for example³⁴. Furthermore, the duration of sports participation could influence the athletic skills level of an individual which could be preventive of getting injured. However, pre-puberal children often change their type of sports activity, which makes it more complex to assemble a complete picture and develop insight into the long-term effects of sports.

Outcome measures

We used an extensive set of measurements to gather data in different domains of health,

according to the model of Shephard & Bouchard¹. We used physical examination, measurement instruments to objectify PA levels and health-related fitness, digital questionnaires and telephone-based interviews to identify injuries and illnesses, during one calendar year. All measurement instruments and performance tests used in this study are feasible, easy to use, and proven valid and reliable in youngsters with CDPD. In addition, the measurement outcomes of all tests from the current study can be used as a reference, which makes it possible to put the test outcome of our young patients or sports participants with CDPD in perspective with their sporting and non-sporting peers. PWV is a good marker of vascular wall stiffness and an important predictor of cardiovascular disease³⁶. From this perspective, it can be concluded that measurement of arterial stiffness has the potential to be used for additional screening of early signs of vascular dysfunction or arterial stiffness, especially in children and adolescents at risk (overweight and obesity) of developing cardiovascular diseases³⁷. The measurement of the PWV by use of the Arteriograph is non-invasive, safe, inexpensive, feasible in children and adolescents and quick to implement in preventive health examination^{36,38}.

Large-scale observational studies have used objective monitoring of PA by accelerometry. This technical innovation has substantially increased our knowledge of PA and how PA is associated with health outcomes in the young. However, interpreting PA intensity data from accelerometry during childhood and adolescence is challenging. Methodological issues include the definition of PA intensity thresholds (predictive equations for metabolic equivalent of tasks (METs) with an algorithm specified for children and adolescents with CDPD), and there is no consensus on the most appropriate PA intensity thresholds to use when measuring children's PA by accelerometry^{39,40}. For these reasons, in our research we measured the type (lying, sitting, standing, walking, running, cycling) and duration of PA with accelerometry, instead of PA intensity. Accelerometers to measure the type of PA were available and validated in healthy adults, in diverse patient groups and in adults who use wheelchairs for daily mobility⁴¹. No valid accelerometer for measuring the type of PA was available for use in youngsters with or without motor disability at the start of this study. To measure the type of PA in daily life, we validated the Activ8 for use in youngsters with and without motor impairment⁴². It is a major strength of this dissertation that there is now a validated device for use in a broad pediatric population, with and without motor impairment.

With reference to the model introduced at the beginning of this discussion, the impact of the social and physical environment, lifestyle behaviors and personal attributes

have not been investigated in this thesis. These are defined as 'other factors' in the model of Shephard & Bouchard¹. Each individual is born with a certain set of genes, grows up in a certain environment and develops a certain behavior pattern^{43,44}. Every individual is part of different systems (parents, family, friends, peers, classmates, sports mates, (PE) teachers, trainers or coaches, health-care professionals etc.) and interacts in various environments (home, playground, school, sports club, rehabilitation center etc.)^{43,44}. Every system or environment evokes certain behaviors, and each individual will respond in their own manner. Sports participation (from recreational to elite level) has positive and negative consequences, with different experiences and beliefs, for youngsters and their parents. In the HAYS study, discrepancies in beliefs between youngsters and their parents were evident (see Panel 9.4). Negative experiences or beliefs of the child itself, their parents or social environment can limit support or participation in sports. Optimal understanding of the impact and influence of personal and environmental factors by quantitative research is needed for optimal support and guidance of PA and sports participation.

PRACTICAL IMPLEMENTATION

We were able to use and analyze objectively PA data measured with the Activ8 in 75% of the participants. Missing data were mainly due to technical problems with Activ8. Participants wore the Activ8 24/7, even during swimming and bathing activities. However, the Activ8 is not waterproof and, although we secured it to avoid water damage (waterproof and skinfold tape), this may have resulted in technical problems. Furthermore, the study of Kotte et al. shows that the feasibility of using the Activ8 in pediatric physical therapy practice is limited⁴⁵, due to 1) difficulties applying an accelerometer, 2) reluctance of children and their parents to wear an accelerometer and 3) technical difficulties in reading and charging the accelerometer, which were considered complicated by therapists. The study of Kotte et al.⁴⁵ and Pettee Gabriel et al.⁴⁴ emphasizes the gap between feasibility of accelerometry and technology in general for research purposes and their implementation in rehabilitation to achieve therapeutic goals.

Within sports, from recreational to elite level, portable technology, like fitness trackers, heart rate monitors, GPS devices and smart watches, for optimizing sports performance is usual⁴⁶. In addition, big data are increasingly being used for health care purposes⁴⁷. New technological innovations, if applied correctly, can provide insights into an

individual's performance, both in research and in practice, that we were previously unable to gain. In addition, technology, such as the use of digital resources, can contribute to more effective collaboration between different disciplines. As an example, the systematic measurement of injuries and illnesses during three years was complicated, inefficient and time-consuming in this thesis³³, which was partly due to the limited digital resources available for us to use. At athlete level, injuries and illnesses are often registered in an online system by the athlete, to which the trainer or coach has access. If such online registration systems were accessible for recreational sports participants or patients, for health-care professionals and for researchers working in the sports domain, the collected data could contribute to improving care and sports performance, and could be used for epidemiological research purposes. Unfortunately, due to obstacles in data protection and funding, too many opportunities are still being missed to realize innovation and to implement improvements in health care more widely⁴⁸.

FUTURE PERSPECTIVES

Sports specific barriers and opportunities

In the Netherlands, we have many different organizations committed to making sports participation possible for youngsters with CDPD. There is an increasing focus on sports performance of athletes with disabilities up to and including Paralympics. More and more people with disabilities can participate in local sports events and competitions that are being organized in municipalities. Adapted and inclusive sports participation is growing; running events are now also accessible to hand cyclists, for example. However, each municipality has its own local policies, so there are local differences in how much attention and financial help there is to support organized sports participation for individuals with a disability. To enable organized sports participation for youngsters with CDPD in their own home environment, a number of developments are needed at sports clubs (i.e. facilities, guidance and support), and there is a need for acceptance by society (i.e. peers, parents and trainers) and for family support (i.e. attitude, active involvement and transportation)⁴⁹. To indicate specifically what parents of children with a disability encounter in society, Panel 9.3 provides an overview of a number of frequently voiced experiences by parents of children who participated in the HAYS study. It is important to mention that it was not an official part of the HAYS study to investigate barriers and opportunities with regard to sports participation. Nevertheless, the statements

correspond to the results of recent qualitative research studying facilitators and barriers to performing activities and participation in children with cerebral palsy from the caregiver's perspective⁴⁹.

Panel 9.3 emphasizes the importance of a multidisciplinary approach, with professionals from the medical, social, sport and educational domains working together for youngsters with CDPD to overcome barriers and discover the possibilities of inclusive sports participation.

Panel 9.3: an overview of a number of sport-specific experienced barriers with brief recommendations

- 1) Adapted sports aids, prostheses, wheelchairs and hand bikes are sometimes required for participation in sports activities safely and without additional risk of injury. Access to sports aids is limited particularly for children and adolescents, because they are still growing: a tailor-made prosthesis does not last long and is therefore quite expensive. Financial arrangements for sports aids, prosthesis, wheelchairs and hand bikes differ per municipality, may often change and may be ambiguous. It often happens that a device for sports is not reimbursed, because another similar device has already been purchased for use in daily life.
- 2) There are a number of obstacles that restrict the accessibility of (adapted) sports. The accessibility of sports locations and forms of sport can be limited, but can sometimes be increased by simple adjustments, like an extra handrail or wheelchair-friendly pavement.
- 3) Information about suitable sports is often hard to find, and the right guidance for both accessibility and suitable sports can sometimes be decisive. Neighborhood teams and sports coaches can raise awareness of the range of sports available locally. Sports providers could be more open to youngsters with a chronic disease or disability and search together with them and their parents for opportunities to solve existing problems.
- 4) The significance of sports for health should receive more, and sometimes earlier, attention in treatment and/or rehabilitation. Professionals working in rehabilitation medicine should create awareness of the benefits of sports participation for health from the first stage of the rehabilitation.
- 5) Insufficient knowledge and expertise of sports coaches and trainers in the field of guiding children with a chronic disorder, especially at the recreational level of sports participation, limits access. It is therefore important to implement evidence-based information through workshops within a sports organization.

Regular and adapted organized sports participation

Fortunately, steps are being taken at policy level to improve, support and facilitate inclusive sports participation for this vulnerable group of children and adolescents. Recently, there have been three main developments in the Netherlands.

Firstly, a lifestyle intervention is included in the health insurance⁵⁰. Proven effective lifestyle interventions focusing on healthier food and eating habits, more exercise and, if necessary, individual psychological treatment to support behavioral change are included. Secondly, the Dutch coalition agreement stipulates that there must be a national prevention strategy that focuses on reducing smoking and preventing smoking at a young age, obesity in childhood and adolescence and excessive alcohol consumption^{51,52}. There were already a number of youth projects; 'Jongeren op Gezond Gewicht', 'Gezonde school' and 'Gezonde schoolkantine', which are being strengthened and/or accelerated by this prevention accord⁵³. Thirdly, based on the coalition agreement 'Trust in the future', the sports accord was established in 2018⁵⁴. The Ministry of Health, Welfare and Sport (VWS), Association of Sports and Municipalities (VSG), Association of Dutch Municipalities (VNG) and Dutch Olympic Committee/Dutch Sports Federation (NOC*NSF) are the strategic partners of this national sports accord. Attention is paid to diversity in the composition of sports associations, development of open sports clubs and accessible access to sports for children. More than 150 parties are involved in successfully implementing this sports accord in the Netherlands. To achieve this, the strategic partners have formulated six ambitions, for which objectives and interventions have been worked out. The ambitions are: 1) integration of sports and exercise, 2) establishing sustainable sports infrastructure, 3) vital sports and exercise organizations, 4) developing a positive sports culture, 5) teaching movement skills from an early age, and 6) elite athlete that inspires. One part of the ambition is to have more children comply with exercise guidelines and to reverse the downward spiral of children's competence in fundamental movement skills. The ambition is targeted at all children between the ages of 0 and 12 years. In terms of taking sufficient exercise, there is attention to the drop-out from sports and exercise of youngsters between 12 and 18 years of age. The interventions described fit in perfectly with the findings of this thesis and subsequent recommendations for practice. In the recommendations section, two points have been highlighted and worked out specifically for youngsters with CDPD, based on the results of our HAYS study and other research.

With these political developments, and especially from a financial point of view, there are more opportunities for professionals to promote healthy and active behavior, such as participation in organized sports, in the field of rehabilitation medicine and in the social and sports domains. The next step is the adequate implementation of proven effective intervention programs targeting an active and healthy lifestyle among youngsters with CDPD in the field of rehabilitation medicine and in the social and sports domains, and this requires a specific approach. To be effective in both the health and sports domains, health care professionals, educational program makers, researchers, neighborhood teams and sports coaches should work together⁵⁵. They should bring their knowledge (scientific and experience-based) together into behavior lifestyle programs⁵⁶, while neighborhood coaches can raise awareness of the range of sports available in the surroundings⁵⁵.

Furthermore, educating future professionals (students) in the preventive health and sports domain could be an effective and efficient strategy for implementing innovative evidence-based tools or interventions. As an example, the research project ("What moves you ?!") which recently started focuses on the development of toolboxes for improving participation in physical activity in children with disabilities⁵⁷. With that perspective, collaboration between researchers, educational program makers and future professionals from different backgrounds is important to successfully implement research outcomes into educational programs and society. RAAK-PRO and the National Scientific Agenda (NWA) promotes the quality of applied research at universities of applied sciences and strengthens the research capacity of the research groups involved. This is in close cooperation with the national knowledge network and professional practice⁵⁸.

RECOMMENDATIONS

Development of programs targeting PA and sports participation

Youngsters with and without CDPD often do not meet the guidelines for healthy PA^{59,60} but increasing physical activity is very complex⁶¹. An important finding in our study is the association between PA and age in children and adolescents with CDPD. PA levels are declining with increasing age, also observed in typically-developing youngsters. In the latter, patterns of increased physical inactivity and sedentary behavior may even track into adulthood^{62,63}. This trend is still being researched in the population with CDPD. Low levels of PA will also result in poor fundamental movement skills and increased risk of

overweight and of developing chronic conditions^{64,65}. In the Netherlands, inspectors of education have found that the movement skills of primary school pupils had decreased over the past 10 years⁵⁴. In addition, children with poor fundamental movement skills experience less pleasure in sports, and more frequently stand on the sidelines because they cannot catch up with their peers when playing or doing gymnastics. Consequently, the chance of quitting sport altogether is greater³⁴. Early fostering of broad fundamental movement skills and enjoyment of sport ensure that children can discover and develop their talents, but also that youngsters continue being physically active throughout their lives, i.e. long-term athletic development³⁴. It is crucial that professionals adopt a systematic approach to long-term athletic development for youngsters of all ages, abilities, and aspirations. It is therefore important in the first place that children and adolescents with CDPD meet the guidelines for physical activity (Table 1).

Table 1: Physical activity guidelines for youngsters with chronic diseases or physical disabilities.

Physical activity guidelines for youngsters with chronic diseases or physical disabilities.	
<p>Key guidelines</p> <p>Children and adolescents with CDPD ages 6 through 17 years should do 60 minutes (1 hour) or more of moderate-to-vigorous physical activity daily:</p> <ol style="list-style-type: none"> Aerobic: Most of the 60 minutes or more per day should be either moderate- or vigorous-intensity aerobic physical activity and should include vigorous-intensity physical activity on at least 3 days a week. Muscle-strengthening: As part of their 60 minutes or more of daily physical activity, children and adolescents should include muscle-strengthening physical activity on at least 3 days a week. Bone-strengthening: As part of their 60 minutes or more of daily physical activity, children and adolescents should include bone-strengthening physical activity on at least 3 days a week. 	
2.	It is important to provide children and adolescents with CDPD opportunities and encouragement to participate in physical activities that are appropriate for their age, medical status, motor skills level, and that are enjoyable and offer variety.
3.	When possible, children and adolescents with CDPD should meet the key guidelines. When youngsters with CDPD are not able to participate in the appropriate types or amounts of physical activities needed to meet the key guidelines, they should be as active as possible and avoid being inactive.
4.	Youngsters with CDPD and their parents and caregivers should work with a health care professional or physical activity specialist (pediatric/sports physical therapist) to understand the types, frequency, intensity and amounts of physical activity or sports activity appropriate for them.

Adapted from the American guideline for children and adolescents (3-17 years) and the Dutch guideline specifically for children and adolescents with a chronic disease or physical disability⁶⁰.

Participation in sport can be a means of achieving the goal of being intensively physically active at least 3 times a week (aerobic, muscle-strengthening and bone-strengthening). Coaches, physical educators, and trainers should develop programming to improve fundamental movement skills, as these are a predictor of PA levels. Engagement in sports can enhance fundamental movement skills and participation in sports is recommended for that purpose, but may need to take place with a different coaching approach in youngsters with CDPD (e.g., a more mastery-oriented approach with the greatest focus on task competency)²². Table 2 gives ten pillars for successful long-term athletic development in youngsters with CDPD. It might be advisable to encourage initial participation in less competitive or individual sports when movement skills are poor to start with. It is possible that low levels of some fundamental movement skills among obese children or those with more severe disabilities result in avoidance of competitive sports, since doing poorly may lead to negative psychological outcomes (i.e., low levels of enjoyment and confidence). During puberty and among girls, dropout from sports participation is high and support of sports participation is then especially important. Supporting strategies should target the youngsters, their parents and caregivers and all other persons in the social environment of youngsters with CDPD.

Table 2: Ten pillars for successful long-term athletic development in youngsters with CDPD.

Ten pillars for successful long-term athletic development in youngsters with CDPD	
1.	For long-term athletic development, youngsters of all ages with CDPD should engage in organized sports activities that promote physical fitness, physical activity and psychosocial wellbeing at least twice weekly.
2.	Sports-specific training programs should accommodate the highly-individualized and non-linear nature of the growth, maturation and development of youngsters with CDPD.
3.	Youngsters with CDPD should be encouraged to enhance their physical fitness and physical activity from early childhood, with a primary focus on a broad range of fundamental movement skill development and on the fun aspects of sports.
4.	Health and wellbeing of the child or adolescent should always be the central focus of long-term athletic development programs.
5.	From adolescence, there should be a shift from fundamental movement skills-oriented development to sport-specific development in training, where movement skills are trained more selectively in accordance with the sport.
6.	Youngsters with CDPD should participate in (organized) sports at least twice weekly, helping reduce the risk of injury and ensure their ongoing participation in long-term athletic development programs.
7.	Long-term athletic development programs should provide all youngsters with CDPD a range of training modes to enhance both health- and movement skill-related components of fitness.
8.	Practitioners should use relevant monitoring and assessment tools specifically developed and validated for youngsters with CDPD as part of an effective long-term athletic development strategy.
9.	Practitioners working with youngsters with CDPD should systematically progress and individualize training programs for successful long-term athletic development.
10.	Qualified professionals and sound pedagogical approaches are fundamental to the success of long-term athletic development programs in youngsters with CDPD.

Adapted from Lloyd et al.³⁴.

Panel 9.4 gives a selection of statements by parents of children who participated in the HAYS study.

Panel 9.4: statements of parents about their children participating in the HAYS study

'I am not sure if my child is able to pursue all these sport specific tests, whereas the child reacts: 'Mom, I am familiar with some of the tests, I do them during physical education lessons at school! I can do this!'

'I am very surprised, my child is able to do all these exercises, I did not know that he is capable like that'.

'Normally, at home, the only thing he likes to do is online gaming. Surprised that my child is so enthusiastic about performing all these sports specific tests. Maybe we have to look for a suitable sport'.

Exercise guidelines

There is insufficient knowledge and expertise among sports coaches and trainers in guiding youngsters with CDPD, especially at the recreational level of sports participation. There are exercise guidelines to optimize sports performance in typically-developing youngsters³⁴. In rehabilitation medicine, improving fitness levels is an important goal and youngsters with CDPD have been trained with success⁶⁶: an eight-week High Intensity Interval Training (HIIT) was proven feasible and safe when applied in children and adolescents with CDPD in the S2SF study⁶⁷. Professionals (trainers, coaches, PE teachers, sports physical therapists) working in the sports domain can use the same training guidelines as for typically-developing youngsters for improving sport performance in youngsters with CDPD. There is no reason so far to believe that exercise guidelines for healthy children cannot be used for peers with CDPD. There is no need to be reluctant. It may take the trainer more time in the beginning to get to know the child with a specific health problem, and it takes some effort to find out what works and what does not. Trainers may facilitate the child to explore their own capabilities and some creativity and perseverance is then required from the trainers. Children and adolescents often know very well what works and what doesn't for them, and coordination and contact with the parents is important when there are uncertainties. Individuals with a neuromuscular disease in particular often experience reluctance of their trainer to engage⁶⁸. Trainers, sports coaches and PE

teachers should be trained so that they can better understand and guide youngsters with CDPD and their parents towards a specific sport for optimal sports performance. Collaboration between professionals working in rehabilitation medicine and education and sports domain is important for that goal.

CONCLUSIONS

Support for sports participation

According to the main findings of this thesis, (organized) sports participation at least twice weekly is an excellent way to improve or maintain physical activity levels, health-related fitness and psychosocial health. In addition, sports participation does not pose an increased risk of injuries and illnesses. Sports organizations play a crucial role in the realization of the global recommendations for youngsters with CDPD to accumulate at least 60 minutes per day of moderate-to-vigorous aerobic PA, of which at least three times per week should be vigorous PA, muscle and bone strengthening, in addition to the activity they undertake as part of everyday living^{60,69}. As pointed out in the introduction, 1 in 4 youngsters grows up with a chronic condition². It is recommended that sports organizations, health care professionals and physical education teachers strengthen their role in the promotion of regular physical activity and sport for healthy active living. Inclusion of youngsters with CDPD in community sports, organized regular or adapted sports requires extra effort for optimal guidance and support because several sports-specific barriers exist.

Mission

Enabling organized sports participation (regular or adapted) for every child or adolescent with a chronic disease or physical disability in their own living environment.

REFERENCES

1. Shephard RJ, Bouchard C. Principal components of fitness: Relationship to physical activity and lifestyle. *Can J Appl Physiol* 1994;19(2):200-214.
2. Hal van L, Tierolf B, Rooijen van M, Hoff van der M. Een actueel perspectief op kinderen en jongeren met een chronische aandoening in Nederland: Omvang, samenstelling en participatie. April 2019;ISBN 978-90-5830-936-5.
3. Schoenmakers MA, de Groot JF, Gorter JW, Hillaert JL, Helders PJ, Takken T. Muscle strength, aerobic capacity and physical activity in independent ambulating children with lumbosacral spina bifida. *Disabil Rehabil* 2009;31(4):259-266. doi: 10.1080/09638280801923235.
4. Verschuren O, Maltais DB, Douma-van Riet D, Kruitwagen C, Ketelaar M. Anaerobic performance in children with cerebral palsy compared to children with typical development. *Pediatr Phys Ther* 2013;25(4):409-413. doi: 10.1097/PEP.0b013e3182a47022.
5. Kohleis K, Storck M, Geissler-Preuss S, Hirsch A, Kuhn FD, Ortfeld S, Rapp M, Bode H. Risk factors for mental health problems in children with cerebral palsy and spina bifida. *Klin Pediatr* 2019;231(1):28-34. Accessed 5/14/2019 10:40:02 AM. doi: 10.1055/a-0664-0832.
6. Graham HK, Harvey A, Rodda J, Nattrass GR, Pirpiris M. The functional mobility scale (FMS). *J Pediatr Orthop* 2004;24(5):514-520. doi: 00004694-200409000-00011.
7. Wood E, Rosenbaum P. The gross motor function classification system for cerebral palsy: A study of reliability and stability over time. *Dev Med Child Neurol* 2000;42(5):292-296.
8. Palisano RJ, Rosenbaum P, Bartlett D, Livingston MH. Content validity of the expanded and revised gross motor function classification system. *Dev Med Child Neurol* 2008;50(10):744-750. doi: 10.1111/j.1469-8749.2008.03089.x.
9. Schoenmakers MA, Gulmans VA, Gooskens RH, Pruijs JE, Helders PJ. Spinal fusion in children with spina bifida: Influence on ambulation level and functional abilities. *Eur Spine J* 2005;14(4):415-422. doi: 10.1007/s00586-004-0768-3.
10. Zwinkels M. From exercise training to school-based sports, the effects on fitness and health in youth with physical disabilities. [Dissertation] Utrecht University; 2018.
11. Lankhorst K, Takken T, Zwinkels M, et al. Sports participation, physical activity, and health-related fitness in youth with chronic diseases or physical disabilities: The Health in Adapted Youth Sports study. *J Strength Cond Res* 2019. doi: 10.1519/JSC.0000000000003098.
12. Dencker M, Andersen LB. Health-related aspects of objectively measured daily physical activity in children. *Clin Physiol Funct Imaging* 2008;28(3):133-144. doi: 10.1111/j.1475-097X.2008.00788.x.

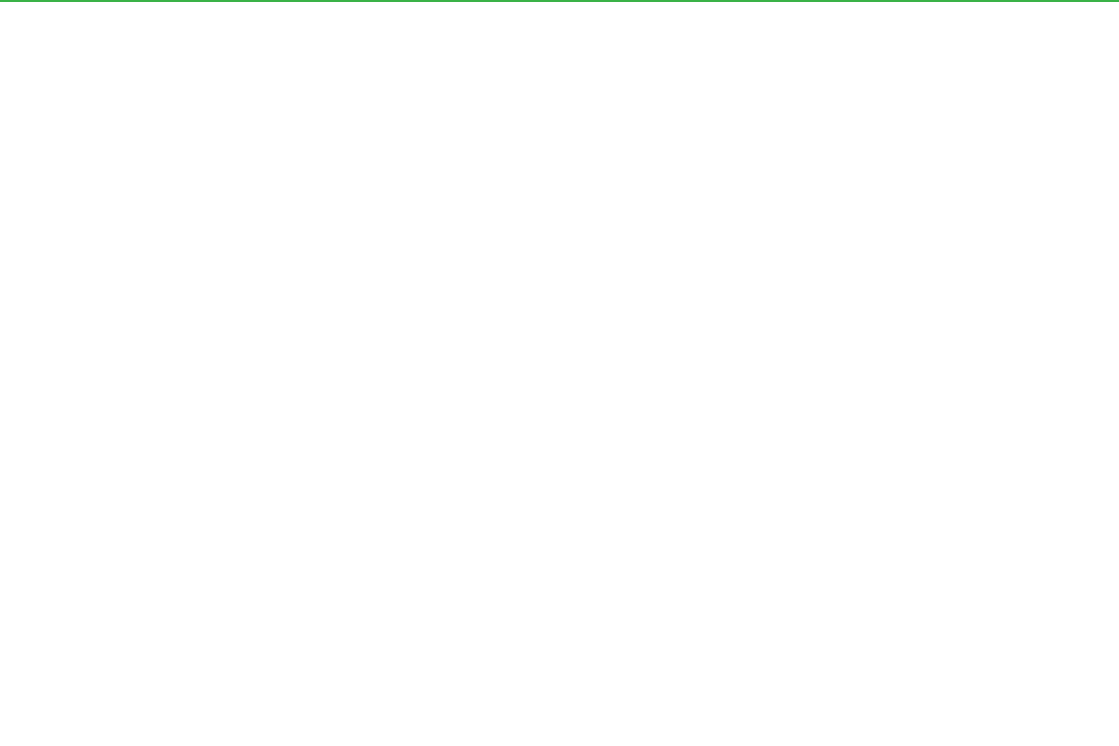
13. Jimenez-Pavon D, Kelly J, Reilly JJ. Associations between objectively measured habitual physical activity and adiposity in children and adolescents: Systematic review. *Int J Pediatr Obes* 2010;5(1):3-18. Accessed 7/7/2017 9:28:46 AM. doi: 10.3109/17477160903067601.
14. LaMonte MJ, Blair SN. Physical activity, cardiorespiratory fitness, and adiposity: Contributions to disease risk. *Curr Opin Clin Nutr Metab Care* 2006;9(5):540-546. doi: 10.1097/01.mco.0000241662.92642.08.
15. Stevens G, van Dorsselaer S, Boer M, de Roos S, Duinhof E, ter Bogt T, van den Eijnden R, Kuyper L, Visser D, Vollebbergh W, de Looze M. HBSC 2017. Gezondheid en welzijn van jongeren in Nederland. Utrecht: Universiteit Utrecht Faculteit Sociale Wetenschappen, Algemene Sociale Wetenschappen. 2018.
16. Ministry of Public Health. Gezondheidsenquête/leefstijlmonitor CBS, overgewicht. <https://www.volksgezondheidenzorg.info/onderwerp/overgewicht/cijfers-context/trends#node-trend-overgewicht-kinderen>. Updated 2017.
17. Jong de NB, Takken T, Berg van de SW, Vos GCW. Active healthy kids the Netherlands. http://www.super-lab.nl/wp-content/uploads/Report_Card-2018-ENG-website.pdf. 2018.
18. Te Velde SJ, Lankhorst K, Zwinkels M, et al. Associations of sports participation with self-perception, exercise self-efficacy and quality of life among children and adolescents with a physical disability or chronic disease - a cross-sectional study. *Sports Med Open* 2018;4(1):38-018-0152-1. doi: 10.1186/s40798-018-0152-1.
19. Zwinkels M, Verschuren O, Balemans A, et al. 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. *Front Pediatr* 2018;6:75. doi: 10.3389/fped.2018.00075.
20. Kriemler S, Meyer U, Martin E, van Sluijs EM, Andersen LB, Martin BW. Effect of school-based interventions on physical activity and fitness in children and adolescents: A review of reviews and systematic update. *Br J Sports Med* 2011;45(11):923-930. doi: 10.1136/bjsports-2011-090186.
21. Merkel DL. Youth sport: Positive and negative impact on young athletes. *Open Access J Sports Med* 2013;4:151-160. doi: 10.2147/OAJSM.S33556.
22. Geidne S, Quennerstedt M, Eriksson C. The youth sports club as a health-promoting setting: An integrative review of research. *Scand J Public Health* 2013;41(3):269-283. doi: 10.1177/1403494812473204.
23. Eime RM, Payne WR. Linking participants in school-based sport programs to community clubs. *J Sci Med Sport* 2009;12(2):293-299. doi: 10.1016/j.jsams.2007.11.003.

24. Yazicioglu K, Yavuz F, Goktepe AS, Tan AK. Influence of adapted sports on quality of life and life satisfaction in sport participants and non-sport participants with physical disabilities. *Disabil Health J* 2012;5(4):249-253. doi: 10.1016/j.dhjo.2012.05.003.
25. Haapala EA, Lankhorst K, de Groot J, Zwinkels M, Verschuren O, Wittinnk 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(10):1102-1111. doi: 10.1177/2047487317702792.
26. Pase MP, Grima NA, Sarris J. The effects of dietary and nutrient interventions on arterial stiffness: A systematic review. *Am J Clin Nutr* 2011;93(2):446-454. doi: 10.3945/ajcn.110.002725.
27. Steppan J, Barodka V, Berkowitz DE, Nyhan D. Vascular stiffness and increased pulse pressure in the aging cardiovascular system. *Cardiol Res Pract* 2011;2011:263585. doi: 10.4061/2011/263585.
28. Lubans DR, Morgan PJ, Cliff DP, Barnett LM, Okely AD. Fundamental movement skills in children and adolescents: Review of associated health benefits. *Sports Med* 2010;40(12):1019-1035. doi: 10.2165/11536850-000000000-00000
29. Webster EK, Martin CK, Staiano AE. Fundamental motor skills, screen-time, and physical activity in preschoolers. *J Sport Health Sci* 2019;8(2):114-121. doi: 10.1016/j.jshs.2018.11.006.
30. Bolger LE, Bolger LA, O'Neill C, Coughlan E, O'Brein W, Lacey S, Burns C. Accuracy of children's perceived skill competence and its association with physical activity. *J Phys Act Health* 2018:1-8. doi: 10.1123/jpah.2017-0371.
31. Nauta J, Martin-Diener E, Martin BW, van Mechelen W, Verhagen E. Injury risk during different physical activity behaviours in children: A systematic review with bias assessment. *Sports Med* 2015;45(3):327-336. doi: 10.1007/s40279-014-0289-0.
32. Hrysomallis C. Balance ability and athletic performance. *Sports Med* 2011;41(3):221-232. doi: 10.2165/11538560-000000000-00000.
33. Lankhorst, K., de Groot, J., Tim Takken, Backx, F., on behalf of the HAYS study group. Sports participation related to injuries and illnesses among youth with chronic diseases: Results of the Health in Adapted Youth Sports study. *BMC Sports Sci Med Rehab* (under review)
34. Lloyd RS, Cronin JB, Faigenbaum AD, et al. National Strength and Conditioning Association position statement on long-term athletic development. *J Strength Cond Res* 2016;30(6):1491-1509. doi: 10.1519/JSC.0000000000001387.

35. Lankhorst K, van der Ende-Kastelijin K, de Groot J, Zwinkels M, Verschuren O, Backx F, Visser-Meily A, 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;4:796-015-1589-z. eCollection 2015. doi: 10.1186/s40064-015-1589-z.
36. Townsend RR. Arterial stiffness: Recommendations and standardization. *Pulse (Basel)* 2017;4(Suppl 1):3-7. doi: 10.1159/000448454.
37. Thurn D, Doyon A, Sozeri B, Bayazit A, Canpolat N, Duzova A, Querfeld U, Schmidt B, Schaefer F, Wuhl E, Melk A. Aortic pulse wave velocity in healthy children and adolescents: Reference values for the Vicorder device and modifying factors. *Am J Hypertens* 2015;28(12):1480-1488. doi: 10.1093/ajh/hpv048.
38. Noortman LCM, Haapala EA, Takken T. Arterial stiffness and its relationship to cardiorespiratory fitness in children and young adults with a Fontan circulation. *Pediatr Cardiol* 2019;40(4):784-791. doi: 10.1007/s00246-019-02065-8.
39. Bussmann JB, van den Berg-Emons RJ. To total amount of activity..... and beyond: Perspectives on measuring physical behavior. *Front Psychol* 2013;4:463. doi: 10.3389/fpsyg.2013.00463.
40. Chinapaw M, Altenburg T, Brug J. Sedentary behaviour and health in children - evaluating the evidence. *Prev Med* 2015;70:1-2. doi: 10.1016/j.ypmed.2014.10.029.
41. Lankhorst K, Oerbekke M, Berg-Emons R, Takken T, Groot J de. Instruments measuring physical activity in individuals who use wheelchairs: A systematic review of measurement properties using the COSMIN criteria. *Arch Phys Med Rehab* (under review)
42. Lankhorst K, van den Berg-Emons RJ, Bussmann JBJ, Horemans HLD, de Groot JF. A novel tool for quantifying and promoting physical activity in youths with typical development and youths who are ambulatory and have motor disability. *Phys Ther* 2019;99(3):354-363. doi: 10.1093/ptj/pzy152.
43. Flores FS, Rodrigues LP, Copetti F, Lopes F, Cordovil R. Affordances for motor skill development in home, school, and sport environments: A narrative review. *Percept Mot Skills* 2019;126(3):366-388. doi: 10.1177/0031512519829271.
44. Pettee Gabriel KK, Morrow JR,Jr, Woolsey AL. Framework for physical activity as a complex and multidimensional behavior. *J Phys Act Health* 2012;9 Suppl 1:S11-8.
45. Kotte E, Veenhof C, Winkler A, Horemans H, Takken T, Groot de J. Monitoring physical activity in primary pediatric physical therapy practice: Difficulties, opportunities and a case study illustration. Submitted, *Pediatr Phys Ther* 2019.

46. Schaars D. The top fitness trends in 2019: Draagbare technologie, groepstraining en HIIT. <https://www.allesoversport.nl/artikel/de-top-fitness-trends-in-2019-draagbare-technologie-groepstraining-en-hiit/>. Updated November 13, 2018.
47. Kools F. Big data in healthcare. <https://www.maastrichtuniversity.nl/news/big-data-healthcare>. Updated April 25th, 2019.
48. ZonMw. Verspreiden van implementatiekennis, kennissyntheses. <https://www.zonmw.nl/nl/over-zonmw/maatschappelijke-impact/implementatie-overzicht/implementatiebeleid/verspreiden-van-implementatiekennis/kennissyntheses/>.
49. Earde PT, Praipruk A, Rodpradit P, Seanjumla P. Facilitators and barriers to performing activities and participation in children with cerebral palsy: Caregivers' perspective. *Pediatr Phys Ther* 2018;30(1):27-32. doi: 10.1097/PEP.0000000000000459.
50. Schaars D. Gecombineerde leefstijl interventies (GLI) in de basiszorgverzekering. <https://www.allesoversport.nl/artikel/gecombineerde-leefstijl-interventies-gli-in-de-basiszorgverzekering/>. Updated February 19, 2019.
51. Leemrijse C, Kappen H, Boeije H. Sport en bewegen door mensen met een lichamelijke beperking belemmeringen en mogelijkheden. Nivel. 2019, Utrecht, Netherlands (ISBN 978-94-6122-541-2).
52. Rijksinstituut voor Volksgezondheid en Milieu. Loket gezond leven, erkende interventies/jeugd. ministerie van volksgezondheid, welzijn en sport. <https://interventies.loketgezondleven.nl/interventieoverzicht1/>. Updated 2019.
53. Rijksinstituut voor Volksgezondheid en Milieu. Gezondheid en preventie; maatregelen in het nationaal preventieakkoord. <https://www.rijksoverheid.nl/onderwerpen/gezondheid-en-preventie/nationaal-preventieakkoord>. Updated 2019.
54. Bruins B, Bolsius L, van Zanen J, Bolhuis A. Nationaal sportakkoord, sport verenigt Nederland. Ministerie van Volksgezondheid, Welzijn en Sport. 2018.
55. Wagemakers A, Molleman G. Buurtsportcoaches slaan brug tussen zorg en sport. ZonMw. <https://mediator.zonmw.nl/mediator-33/buurtsportcoaches-slaan-brug-tussen-zorg-en-sport/>. Updated June, 2019.
56. Ministerie van volksgezondheid, welzijn en sport. Alles over erkende effectieve gecombineerde leefstijlinterventies. kenniscentrum sport, ede. <https://www.kenniscentrumsport.nl/sportinterventies-en-beweeginterventies/alles-over-erkende-effectieve-gecombineerde-leefstijlinterventies/>. Updated 2019.
57. Wittink H, Lugt van der R, Renes R, et al. Wat beweegt jou?! <https://www.onderzoek.hu.nl/Projecten/Wat-beweegt-jou/>. Updated 2018.

58. Nationaal Regieorgaan Praktijkgericht Onderzoek SIA. Nationaal regieorgaan praktijkgericht onderzoek SIA zet zich in voor meer en nog beter praktijkgericht onderzoek door hogescholen. <http://www.regieorgaan-sia.nl/>. Updated 2019.
59. Burghard M, Knitel K, van Oost I, Tremblay MS, Takken T, Dutch Physical Activity Report Card Study Group. Is our youth cycling to health? results from the Netherlands' 2016 report card on physical activity for children and youth. *J Phys Act Health* 2016;13(11 Suppl 2):S218-S224. doi: 10.1123/jpah.2016-0299.
60. Burghard M, de Jong NB, Vlieger S, Takken T. 2017 Dutch report card+: Results from the first physical activity report card plus for Dutch youth with a chronic disease or disability. *Front Pediatr* 2018;6:122. doi: 10.3389/fped.2018.00122.
61. Verschuren O, Peterson MD, Balemans AC, Hurvitz EA. Exercise and physical activity recommendations for people with cerebral palsy. *Dev Med Child Neurol* 2016;58(8):798-808. Accessed 12/19/2016 11:09:27 AM. doi: 10.1111/dmcn.13053.
62. Biddle SJ, Gorely T, Stensel DJ. Health-enhancing physical activity and sedentary behaviour in children and adolescents. *J Sports Sci* 2004;22(8):679-701. Accessed 12/19/2016 11:21:40 AM. doi: 10.1080/02640410410001712412.
63. Ortega FB, Konstabel K, Pasquali E, et al. Objectively measured physical activity and sedentary time during childhood, adolescence and young adulthood: A cohort study. *PLoS One* 2013;8(4):e60871. doi: 10.1371/journal.pone.0060871.
64. Hills AP, King NA, Armstrong TP. The contribution of physical activity and sedentary behaviours to the growth and development of children and adolescents: Implications for overweight and obesity. *Sports Med* 2007;37(6):533-545.
65. Tompsett C, Sanders R, Taylor C, Cobley S. Pedagogical approaches to and effects of fundamental movement skill interventions on health outcomes: A systematic review. *Sports Med* 2017;47(9):1795-1819. doi: 10.1007/s40279-017-0697-z.
66. Zwinkels M, Takken T, Ruyten T, Visser-Meily A, 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;71:70-76. doi: <https://doi.org/10.1016/j.ridd.2017.09.021>.
67. Zwinkels M, Verschuren O, de Groot JF, et al. Effects of high-intensity interval training on fitness and health in youth with physical disabilities. *Pediatr Phys Ther* 2019;31(1):84-93. doi: 10.1097/PEP.0000000000000560.
68. Hollander dEL, Milder IE, Proper KI. Beweeg- en sportgedrag van mensen met een chronische aandoening of lichamelijke beperking. Bilthoven; RIVM rapport 2015-0064.
69. Takken T. Nederlandse jeugd zevende op mondiale beweegranglijst. <https://www.allesoversport.nl/artikel/nederlandse-jeugd-zevende-op-mondiale-beweegranglijst/>. Updated May 24, 2019.



Nederlandse samenvatting

Kristel Lankhorst

Sportparticipatie laat bij kinderen en jongeren een positieve relatie zien met zowel de fysieke als ook de psychosociale gezondheid. Regelmatig sporten is dus belangrijk. Echter, jongeren met een fysieke beperking of een chronische ziekte sporten minder in vergelijking met hun leeftijdsgenoten. Slechts 26% van de jongeren met een chronische aandoening sport één keer per week bij een sportvereniging, vergeleken met 71% van hun niet-chronisch zieke leeftijdsgenoten. Terwijl voor deze populatie een actieve levensstijl zeer belangrijk is voor het bereiken van een optimaal niveau van fysieke en psychosociale gezondheid zowel op de korte als de lange termijn. Er zijn verschillende manieren om gezondheidsproblemen te voorkomen en een actieve leefstijl te bevorderen. Geregeld sporten bij een sportvereniging is daarvan een voorbeeld. Echter, voor jongeren met een chronische ziekte of een lichamelijke beperking is het niet vanzelfsprekend om te sporten bij een sportvereniging. Het doel van de Health in Adapted Youth Sports (HAYS) studie is daarom ook, om zowel de negatieve als positieve relaties van sport voor kinderen en jongeren met een chronische ziekte of lichamelijke beperking te onderzoeken. De resultaten zullen inzichtelijk maken of sportparticipatie een positieve invloed heeft op het bereiken van een optimaal niveau van gezondheid en lichamelijke activiteit bij bovengenoemde doelgroep kinderen en jongeren. Het onderzoek kan resulteren in het formuleren van adviezen voor professionals die binnen de gezondheidszorg met deze kinderen en jongeren werken, waardoor zij deze doelgroep beter kunnen begeleiden.

Het protocol van de HAYS studie wordt beschreven in **hoofdstuk 2**. In deze studie zijn er twee groepen kinderen en jongeren met een chronische ziekte of lichamelijke beperking met elkaar vergeleken. De eerste groep sport tenminste twee keer per week bij een sportvereniging en de andere groep sport één keer bij een sportvereniging of helemaal niet. Er is bij beide groepen kinderen gekeken naar de verschillen met betrekking tot fitheid, dagelijkse lichamelijke activiteit, psychosociale gezondheid en risico op (sportgerelateerde) blessures en ziektes. In deze studie zijn kinderen en jongeren in de leeftijd van 10-19 jaar met een lichamelijke beperking of chronische ziekte meegenomen die in staat zijn om zelfstandig te lopen. "Sport" is gedefinieerd als deelname aan een georganiseerde sport, tenminste twee keer per week gedurende een periode van 3 maanden of meer.

Doelgroep van de HAYS studie

Aan dit onderzoek hebben jongeren deelgenomen met een chronische ziekte of een fysieke beperking. De ziekten en aandoeningen die de kinderen en jongeren hebben bleken zeer divers. We hebben kinderen gerekruteerd via scholen voor speciaal onderwijs, kinderfysiotherapiepraktijken, (aangepaste) sportorganisaties en via de polikliniek van het Wilhelmina Kinderziekenhuis. Ook hebben kinderen en jongeren die aan het Sport-2-Stay-Fit (S2SF) onderzoek hebben deelgenomen meegedaan. Het S2SF onderzoek liep parallel aan het HAYS onderzoek, waarbij dezelfde uitkomstparameters werden gemeten. Het S2SF onderzoek richtte zich op het meten van de effectiviteit van een naschools-sportprogramma bij kinderen en jongeren met een chronische ziekte of een fysieke beperking. De ziekten en aandoeningen die voorkwamen in het HAYS onderzoek waren: hart- en vaatziekten (aangeboren hartziekten), longaandoeningen (astma), metabole ziekten (diabetes mellitus), spier- of skeletaandoeningen, niet-progressieve spieraandoeningen, aandoeningen als gevolg van een hersenbeschadiging (cerebrale parese, spina bifida), immunologische en hematologische aandoeningen, kanker en epilepsie.

Het meten van lichamelijk activiteit

In de HAYS studie hebben kinderen en jongeren deelgenomen met onder andere Spina Bifida of Cerebrale Parese. Deze groep jongeren ervaart als gevolg van hun aandoening spasticiteit en / of verlamming van de spieren, wat het lopen beperkt. Tot op heden was er nog geen goed meetinstrument om de dagelijkse lichamelijke activiteit te meten van kinderen en jongeren met een loopbeperking. Om de dagelijkse lichamelijke activiteit te kunnen meten bij de kinderen en jongeren die deelnemen aan de HAYS studie is in **hoofdstuk 3** een fysieke activiteitenmonitor (Activ8) gevalideerd voor gebruik bij kinderen en jongeren met en zonder een beperking van het looppatroon. Dit hoofdstuk laat zien dat de Activ8 de dagelijkse activiteiten nauwkeurig kan meten - zoals liggen, zitten, staan, lopen, fietsen en rennen - , van zowel kinderen en jongeren mét als zonder een loopbeperking. Daarnaast kan het meetinstrument gebruikt worden om interventies gericht op fysieke activiteit te monitoren en te evalueren. De Activ8 is op basis van de uitkomsten van dit onderzoek ingezet om de lichamelijk activiteit te meten van alle deelnemers in de HAYS studie.

De impact van georganiseerd (aangepast) sporten

In de **hoofdstukken 4, 5, 6 en 7** zijn de resultaten van de HAYS studie te lezen. In **hoofdstuk 4** staat beschreven wat de impact van sportdeelname is op fitheid (zuurstofopname, spierkracht, sprintsnelheid, lichaamsgewicht, vetpercentage) en lichamelijke activiteit. Het blijkt dat kinderen en jongeren die minstens twee keer in de week sporten bij een sportclub beter presteren op alle fysieke uitkomstparameters in vergelijking met hun leeftijdsgenootjes die niet of één keer per week sporten. De meer frequente sportgroep heeft 1) een significant hogere zuurstofopname (VO_{2peak}), 2) bereikte een hoger maximaal vermogen op de muscle power sprint test, 3) is sneller op de 10 x 5 meter sprinttest, 4) heeft gemiddeld een hogere handknijpkracht en 5) was in staat om verder te springen. De kinderen en jongeren die minstens twee keer per week aan sport deden, bleken een gezonder gewicht, een lagere body mass index (het gewicht in verhouding tot lichaamslengte) en gemiddeld een lager vetpercentage te hebben in vergelijking met hun leeftijdsgenootjes die niet of één keer per week sporten. Ook bleken zij actiever in het dagelijks leven; gedurende een schoolweek zijn zij gemiddeld meer dan 30 minuten per dag langer actief en gedurende het weekend is dat gemiddeld 25 minuten per dag langer vergeleken met hun niet sportende of één keer per week sportende leeftijdsgenootjes. De positieve relatie tussen sportdeelname en de zuurstofopname (VO_{2peak}) werd voor 31% beïnvloed door de dagelijkse lichamelijke activiteit. De mate van inactiviteit (liggen en zitten) had geen invloed op de positieve relatie tussen sportdeelname en de zuurstofopname (VO_{2peak}).

Hoofdstuk 5 beschrijft de impact van sportdeelname op de psychosociale gezondheid bij kinderen en jongeren met een chronische ziekte of fysieke beperking. Degenen die minstens twee keer per week aan sport doen hebben een hogere kwaliteit van leven en voelen zich atletisch kompetenter in vergelijking met hun niet of één keer per week sportende leeftijdsgenootjes. Kinderen (10-12 jaar), maar niet de jongeren (13-18 jaar), die minstens twee keer per week aan sport deelnemen ervaren een hoger gevoel van sociale acceptatie. Daarnaast blijkt dat wanneer men tenminste twee keer per week sport, dit sterk geassocieerd is met een hogere mate van beweging gerelateerde zelfredzaamheid.

Sportdeelname van tenminste twee keer in de week laat dus een positieve relatie zien met fitheid, lichamelijk activiteit en psychosociale gezondheid bij kinderen en jongeren met een chronische ziekte of fysieke beperking. Echter betekent dit niet dat we deze positieve relatie terug zien bij de cardiorespiratoire fitheid en de stijfheid van de arteriën.

Het onderzoek in **hoofdstuk 6** beschrijft dat een lage fitheid en een grotere buikomtrek beide zijn gerelateerd aan een hogere slagaderlijke stijfheid. Een verhoogde slagaderlijke stijfheid verhoogd het risico op het ontwikkelen van hart- en vaatziekten. Echter, de relatie tussen de fitheid en de slagaderlijke stijfheid werd deels verklaard door de buikomtrek. Na een uitgebreide analyse konden we concluderen dat kinderen en jongeren met een chronische ziekte of fysieke beperking met een buikomtrek van >73 cm en een zuurstofopname van <35 ml/kg/min zeer waarschijnlijk een hogere stijfheid van de slagaderen hebben. Deze resultaten suggereren dat interventies die gericht zijn op het faciliteren van een gezond gewicht en lichaamssamenstelling en het verbeteren van de fitheid mogelijkwerijs bijdragen aan het verbeteren van de slagaderlijke stijfheid. Interventieonderzoek zal echter moeten uitwijzen of deze aannames terecht zijn.

Naast de positieve invloed van sportdeelname op de gezondheid, bestaan er ook veronderstellingen dat sport resulteert in het oplopen van blessures of het verergeren van de bestaande chronische ziekte of aandoening. Deze veronderstellingen kunnen aanleiding zijn om niet deel te nemen aan georganiseerd sporten. Hoofdstuk 7 beschrijft het risico op het krijgen van (sportgerelateerde) blessures of ziekten. In dit onderzoek is een vergelijking gemaakt tussen kinderen en jongeren met een chronische ziekte of fysieke beperking die minstens twee keer per week deelnemen aan sport in vergelijking met leeftijdsgenootjes die niet of één keer per week sporten. Resultaten uit dit onderzoek tonen aan dat sportdeelname van tenminste twee keer in de week niet een significant hoger risico op de incidentie van een blessure of ziekte per 1000 uur lichamelijke activiteit met zich meebrengt in vergelijking met leeftijdsgenootjes die niet of eenmaal per week deelnemen aan sport. Elk kind of elke jongere loopt een even groot risico op, op het krijgen van een blessure of een ziekte. Degene die minstens twee keer per week deelnemen aan sport lopen voornamelijk een blessure op door hun, terwijl de kinderen en jongeren die niet of één keer per week sporten, blessures oplopen in het dagelijks leven. Dat gebeurt voornamelijk tijdens gymles op school, activiteiten in het dagelijks leven en niet-georganiseerde sport- en spelactiviteiten in de vrije tijd.

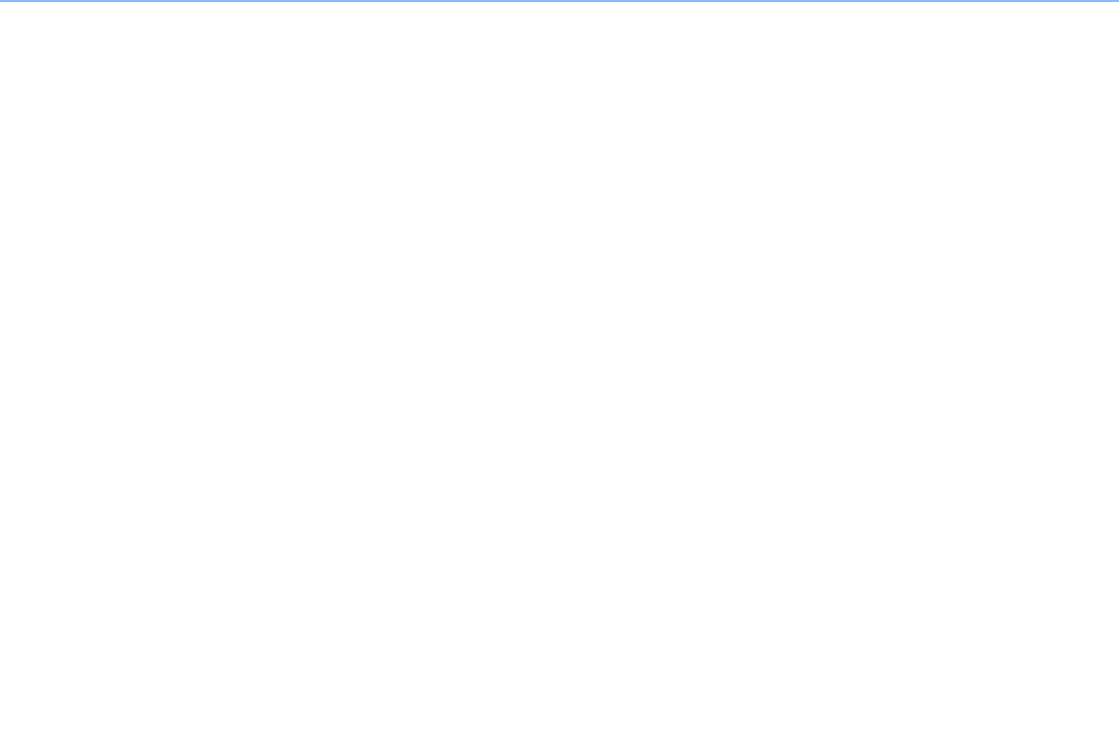
Op basis van de resultaten van de **hoofdstukken 4, 5, 6 en 7** kan aan kinderen en jongeren met een chronische ziekte of fysieke beperking geadviseerd worden om tenminste twee keer in de week deel te nemen aan (aangepast) georganiseerd sporten. Het is nu zeer wenselijk dat sportdeelname bij een sportclub voor iedereen uit deze doelgroep ook daadwerkelijk mogelijk wordt gemaakt. Er bestaan immers nog vele barrières waardoor het nog niet vanzelfsprekend is dat deze kwetsbare groep kinderen en jongeren onbezorgd kan sporten bij een sportclub bij hen in de buurt. Door sportdeelname bij een sportclub in hun eigen leefomgeving mogelijk te maken kunnen deze kinderen en jongeren zowel op schooldagen als ook in het weekend minstens twee keer per week lichamelijk actief zijn.

Het meten van lichamelijke activiteit bij mensen in een rolstoel

Nu we de impact van georganiseerde sportdeelname kennen voor de ambulante populatie (zij die niet gebonden zijn aan een rolstoel) kinderen en jongeren met een chronische ziekte of fysieke aandoening, ontstaat de volgende vraag wat de impact van sportparticipatie is voor hen die gebruik maken van een rolstoel in het dagelijks leven. Hiervoor is het belangrijk om ook de mate van lichamelijke activiteit te kunnen meten. Een meetinstrument dat in de ambulante populatie kinderen en jongeren wordt gebruikt kan niet per definitie gebruikt worden voor de jeugd in een rolstoel. In **hoofdstuk 8** bestuderen we op basis van bestaande literatuur welk meetinstrument er op dit moment voorhanden is voor het meten van de lichamelijke activiteit bij personen die gebruik maken van een rolstoel in het dagelijks leven. De twee vragenlijsten die veelbelovend lijken, zijn de Physical Activity Scale for Individuals with Physical Disabilities (PASIPD) en de Physical Activity Recall Assessment for People with Spinal Cord Injury (PARA-SCI). Daarnaast zijn er vier objectieve meetinstrumenten (activiteitenmonitoren) die op een valide manier de intensiteit van lichamelijke activiteit in het dagelijks leven meten. Dit zijn de GENEactive, Actigraph GT3X+, Actiheart en de PAMS. Om het type lichamelijk activiteit te meten zijn zowel de PAMS als ook de VitaMove valide instrumenten. De praktische toepasbaarheid verschilt per instrument. Ook hebben we uit de bestaande literatuur kunnen concluderen dat een hartslagmeter op basis van individuele kalibratie (het instellen van hartslagzones) een makkelijke en goedkope vervanger is, wanneer de hierboven genoemde activiteitenmonitoren voor volwassenen niet voorhanden zijn.

De bevindingen van dit proefschrift zijn in **hoofdstuk 9** in perspectief geplaatst binnen de huidige wetenschappelijk kennis en het (aangepast) georganiseerd sportklimaat in Nederland. Nu we weten dat sportdeelname van minstens twee keer in de week een positieve impact heeft op de gezondheid van kinderen en jongeren met een chronische ziekte of fysieke beperking, is het noodzakelijk dat sportdeelname ook mogelijk wordt gemaakt.

Gelukkig is er toenemende aandacht voor (aangepast) sporten voor kinderen en jongeren met een chronische ziekte of fysieke beperking. Het huidige regeerakkoord is daar een goed voorbeeld van. Om georganiseerde sportdeelname mogelijk te maken voor elk kind en jongere met een chronische ziekte of fysieke beperking in hun eigen woonomgeving, staan we nog wel voor een aantal uitdagingen. Denk bijvoorbeeld aan het toegankelijk maken van sportfaciliteiten, het bieden van de juiste begeleiding en ondersteuning, acceptatie door leeftijdsgenoten, ouders en trainers. Maar ook ondersteuning van familie (transport, actieve participatie en een positieve houding) is nodig om sportdeelname bij een sportclub mogelijk te maken voor onderhavige doelgroep. Inclusie van kinderen en jongeren met een chronische aandoening of fysieke beperking in het reguliere sportaanbod, aangepast sporten en/of sporten binnen een eigen gemeenschap vereist extra aandacht. Om optimale begeleiding en ondersteuning mogelijk te maken zullen eerst verschillende sportspecifieke barrières moeten worden weggenomen. Het is belangrijk dat sportorganisaties, gezondheidszorgprofessionals en gymdocenten hun krachten bundelen en lichamelijke activiteit en sportdeelname promoten voor een gezonde levensstijl bij deze jeugdigen.



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Kristel Lankhorst

Het is zover, het moment dat ik mijn dankwoord schrijf. Het moment dat ik afscheid neem van mijn reis als PhD student. Op reis gaan is spannend, een nieuw avontuur met nieuwe kansen, mogelijkheden en uitdagingen. Deze reis ga ik niet vergeten, het heeft mooie en leerzame momenten opgeleverd, waardevolle samenwerkingen en vrienden voor het leven. Op deze momenten blik ik terug in dit dankwoord. Deze reis heb ik niet alleen afgelegd, ik ben veel inspirerende mensen tegengekomen die allemaal op hun eigen manier een bijdrage hebben geleverd aan de totstandkoming van dit prachtige proefschrift, waar ik enorm trots op ben.

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About the author

Kristel Lankhorst

CURRICULUM VITAE



Kristel Lankhorst was born in Wijhe, the Netherlands, on March 11th, 1984. After graduating from the University of Applied Sciences in Enschede as Physical Therapist (PT) in 2006, where she was an active member of the study association for physiotherapy and podiatry (S.V. Tabula Rasa). After working one year of work experience as PT, she moved from Enschede to Amsterdam, to study Human Movement Sciences at the VU University in Amsterdam. She wanted to explore and learn more about the scientific background of physical therapy, rehabilitation and sports.

During her study she worked as a PT in a private practice for back & neck problems and treated work-related physical problems of employees at a law firm. In her free time she was active member of the study association of the faculty of Human Movement Sciences (V.I.B.) and of the sports association for cycling and ice-speed skating (SKITS). She successfully finished her Master internship on the impact of combined skin sparing mastectomy and immediate subpectoral prosthetic reconstruction on the pectoralis major muscle function. The Master internship was in close collaboration with the department of plastic surgery at the Antoni van Leeuwenhoek Hospital. Parallel to her Master program she has obtained her teaching qualification for higher education. In 2009, she received her Master of Science degree.

Two months of traveling in New Zealand and Australia followed before she started working as a PT and junior researcher at the department of physiotherapy and rehabilitation medicine at the VU Medical Center in Amsterdam. Her work at the hospital resulted in a Dutch publication; Is physical exercise during chemotherapy feasible for cancer patients? During one year she combined her work at the VU Medical Center with working as a lecturer at the Master Program Physical Therapy, specialization Sports Physical Therapy, of the HU University of Applied Sciences Utrecht. In 2013 she got the opportunity to start a PhD program in the Research group Lifestyle and Health of the HU University of Applied Sciences Utrecht. She participated within two research projects 'Fit for the Future!' and 'Health in Adapted Youth Sports'. She investigated the impact

of (adapted) organized sports participation on health-related fitness, physical activity, psychosocial functioning and risk of sport-related injuries and illnesses in youth with a chronic disease or physical disability. During her PhD, she combined research with lecturing activities by integrating research into the education program. She developed two modules for the Master Program Physical Therapy; 'Sports Physical Therapy and the Adapted Athlete' and 'Sports Physical Therapy and Youth Sports'.

Kristel lives in Amsterdam. Quality time with friends and family is important for her. She loves backpacking around the world and is a sports fanatic. She likes to go for a run in her free time, go cycling with friends, playing indoor soccer with FC Dubbelfrisss, is a member of the gym for crossfit, circuit training, yoga, and interval-running classes. During wintertime she likes speed skating at the ice-arena but preferably on frozen lakes.

PUBLICATIONS

International publications

Lankhorst K, Takken T, Zwinkels M, van Gaalen L, Velde ST, Backx F, Verschuren O, Wittink H, de Groot J. Sports Participation, Physical Activity, and Health-Related Fitness in Youth With Chronic Diseases or Physical Disabilities: The Health in Adapted Youth Sports Study. *J Strength Cond Res*. 2019 Mar 13.

Lankhorst K, van den Berg-Emons RJ, Bussmann JBJ, Horemans HLD, de Groot JF. A Novel Tool for Quantifying and Promoting Physical Activity in Youths With Typical Development and Youths Who Are Ambulatory and Have Motor Disability. *Phys Ther*. 2019 Mar 1;99(3):354-363.

Te Velde SJ, **Lankhorst K**, Zwinkels M, Verschuren O, Takken T, de Groot J; Associations of sport participation with self-perception, exercise self-efficacy and quality of life among children and adolescents with a physical disability or chronic disease-a cross-sectional study. *Sports Med Open*. 2018 Aug 15;4(1):38. doi: 10.1186/s40798-018-0152-1.

Zwinkels M, Verschuren O, Balemans A, **Lankhorst K**, Te Velde S, van Gaalen L, de Groot J, Visser-Meily A, 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. *Front Pediatr*. 2018 Mar 26;6:75.

Lankhorst K, Haapala EA*, 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 Jul;24(10):1102-1111.

Lankhorst K, van der Ende-Kastelijin K*, de Groot J, Zwinkels M, Verschuren O, Backx F, Visser-Meily A, 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 Dec 22;4:796.

Zwinkels M, Verschuren O, **Lankhorst K**, van der Ende-Kastelijin K, de Groot J, Backx F, Visser-Meily A, Takken T; Sport-2-Stay-Fit study group. 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 Oct 6;7:22.

Hage JJ, van der Heeden JF, **Lankhorst K**, Romviel SM, Vlutters ME, Woerdeman LA, Visser B, Veeger HE. Impact of combined skin sparing mastectomy and immediate subpectoral prosthetic reconstruction on the pectoralis major muscle function: a preoperative and postoperative comparative study. *Ann Plast Surg*. 2014;72(6):631-7.

Submitted publications

Lankhorst K, Oerbekke M, van den Berg-Emons R, Takken T, de Groot J. Instruments measuring physical activity in persons who use a wheelchair: a systematic review of measurement properties. *Archives of Physical Medicine and Rehabilitation*, accepted for publication.

Lankhorst K, de Groot J, Takken T, Backx FJ. Sports participation related to injuries and illnesses among youth with chronic diseases: results of the health in adapted youth sports study. *BMC Sports Science, Medicine and Rehabilitation*, under review.

National publications

Duijf M, **Lankhorst K**, Zwinkels M. Sporten is gezond, ook voor kinderen en jongeren met een chronische aandoening of lichamelijke beperking! <https://www.allesoversport.nl/> 13 januari 2018.

Lankhorst K, Geleijn E, Huijsmans R, Dekker J, Boven E. Is fysieke training tijdens chemotherapie voor patiënten met kanker haalbaar? *Ned Tijdschr Oncol* 2017;14:90-9.

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

Zwaan W, **Lankhorst K**, Rehorst J. Pillen slikken, de rem erop! Pijnstillers en ontstekingsremmers zijn funest voor sporters. *Sportgericht* 2016;5:34.

Lankhorst K, Zwinkels M, De Groot J, Verschuren O, Takken T. Is sport gezond voor chronisch zieke jongeren? *Sportgericht* 2016;3:25.

Van der Pijl K, **Lankhorst K**. Revaliderende sporter met diabetes Type 1, beperkingen of uitdagingen? *Sportgericht* 2016;2:26.

Van den Berg I, Daane K, Gereke K, Den Hartog B, Hilgersom H, Westerhof A, **Lankhorst K**. Behandelprotocol voor het patello-femoraal pijnsyndroom. Sportgericht 2014;1:18.

PRESENTATIONS

International scientific conferences and presentations

Lankhorst K. The impact of (adapted) organized sports participation on health in youth with a chronic disease or physical disability, Health in Adapted Youth Sports Study. Presentation. Summer School Pediatric Sport and Exercise Medicine, July 15th 2019, Utrecht, the Netherlands.

Lankhorst K. The impact of (adapted) organized sports participation on health in youth with a chronic disease or physical disability, Health in Adapted Youth Sports Study. Presentation. 6th International Rehabmove State-of-the-art congress, December 12-14th 2018, Groningen, the Netherlands.

Lankhorst K. Sports participation, physical activity, and health-related fitness in youth with chronic diseases or physical disabilities: Health in Adapted Youth Sports Study. Presentation. Summer School Pediatric Sport and Exercise Medicine, August 15th 2017, Utrecht, the Netherlands.

Lankhorst K. Sports participation, physical activity, and health-related fitness in youth with chronic diseases or physical disabilities: Health in Adapted Youth Sports Study. Presentation. VvBN PhD-day Human Movement Sciences. December 1th 2017, Rotterdam, The Netherlands.

Lankhorst K, van den Berg-Emons R, Bussmanns J, Horemans H, de Groot J. A novel tool to quantify and promote physical activity in youth and ambulatory youth with a motor disability. Presentation. Pediatric Work Physiology meeting XXX. October 3-8th 2017, Thessaloniki, Greece.

De Groot J, on behalf of **Lankhorst K**, Haapala EA, 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. Presentation. World Confederation for Physical Therapy, 2-4th July 2017, Cape Town, South Africa.

De Groot J, on behalf of **Lankhorst K**, van den Berg-Emons R, Bussmanns J, Horemans H, Stam H, de Groot J. Criterion validity of a novel feedback device to assess physical behavior in youth. Poster presentation. World Confederation for Physical Therapy, 2-4th July 2017, Cape Town, South Africa.

Lankhorst K, van den Berg-Emons R, Bussmann J, Horemans H, Stam H, Takken T, de Groot J. Measuring physical behavior in youth. Poster Pitch and Presentation. 3rd European Workshop on Clinical Pediatric Exercise Testing. November 11-12th 2016, Zeist, the Netherlands.

Lankhorst K, van den Berg S, Zwinkels M, van der Ende-Kastelijnn K, Verschuren O, Takken T, Backx F, de Groot J. Injuries in children and adolescents with a physical disability or chronic disease: a cross-sectional retrospective study: preliminary results. Poster presentation. Pediatric Work Physiology meeting XXIX, September 9-13th 2015, Utrecht, the Netherlands.

Lankhorst K, van der Ende-Kastelijnn K, Zwinkels M, Muselaers E, Verschuren O, Takken T, Backx F, de Groot J. Sport participation reduces sedentary time, but does it improve physical fitness in youth with a physical disability or chronic disease? Poster presentation. Pediatric Work Physiology meeting XXIX, September 9-13th 2015, Utrecht, the Netherlands.

Lankhorst K, Oerbekke M, van den Berg-Emons R, Takken T, de Groot J. Instruments measuring physical behavior in wheelchair users: a systematic review of measurement properties. Poster presentation. European Workshop on Clinical Pediatric Exercise Testing. November 7-8th 2014, Zeist, the Netherlands.

Lankhorst K, van der Ende-Kastelijnn K, de Groot J, Zwinkels M, Verschuren O, Backx F, Visser-Meily A, Takken T. Health in Adapted Youth Sports study: health effects of sports participation in children and adolescents with a chronic disease or physical disability. Utrecht Summer School, April 14th 2014, Utrecht, the Netherlands.

National scientific conferences and presentations

Lankhorst K, Zwinkels M. Gezondheidseffecten van sport voor kinderen en jongeren met een chronische ziekte. Presentatie. Symposium Kind en Bewegen - innovaties in de zorg voor kinderen. Hogeschool Utrecht, Kinderbewegingscentrum WKZ, kenniscentrum revalidatiegeneeskunde Utrecht, 3 november 2017, Utrecht.

Lankhorst K, Te Velde SJ, Zwinkels M, Verschuren O, Takken T, de Groot J. Associations of sport participation with self-perception, exercise self-efficacy and quality of life among children and adolescents with a physical disability or chronic disease-a cross-sectional study. Poster Presentatie. Symposium Kind en Bewegen - innovaties in de zorg voor kinderen. Hogeschool Utrecht, Kinderbewegingscentrum WKZ, kenniscentrum revalidatiegeneeskunde Utrecht, 3 november 2017, Utrecht.

Lankhorst K, de Groot J, Takken T, Backx FJ. Blessures bij kinderen en jongeren met een fysieke beperking of chronische ziekte. Hebben sportende kinderen meer blessures? Presentatie. Najaarscongres Nederlandse Vereniging voor Kinderfysiotherapie (NVFK) congres, 6 november 2015, Maarssen.

Lankhorst K. Health in Adapted Youth Sports (HAYS): De relatie tussen sportparticipatie en gezondheid van kinderen met een chronische ziekte of lichamelijke beperking. Presentatie. Dag van het Sportonderzoek, 29 oktober 2015, Zwolle.

Lankhorst K, van der Ende-Kastelijn K, Zwinkels M, Muselaers E, Verschuren O, Takken T, van den Berg-Emons R, de Groot J. Sport participatie vermindert sedentaire tijd, maar verbetert het ook de fysieke fitheid bij jeugd met een fysieke beperking of chronische ziekte? Presentatie. Dag van het Sportonderzoek, 29 oktober 2015, Zwolle.

Lankhorst K, van der Ende-Kastelijn K, Zwinkels M, van den Berg S, Verschuren O, Takken T, Backx F, de Groot J. Blessures bij kinderen en jongeren met een fysieke beperking of chronische ziekte: Een retrospectieve studie: eerste resultaten. Presentatie. Dag van het Sportonderzoek, 29 oktober 2015, Zwolle.

Lankhorst K, van der Ende-Kastelijn K, de Groot J, Zwinkels M, Verschuren O, Backx F, Visser-Meily A, Takken T. Health in Adapted Youth Sports (HAYS): De relatie tussen sportparticipatie en gezondheid van kinderen met een chronische ziekte of lichamelijke beperking. Presentatie. Dag van het Sportonderzoek, 30 oktober 2014, Nijmegen.

Presentations at faculties, for (medical) professionals and other activities

Lankhorst K. De impact van sport deelname op de gezondheid van jongeren met een chronische ziekte of fysieke beperking. Presentatie. Landelijke trainers dag, Esther Vergeer Foundation, 5 juli 2019, Arnhem.

Lankhorst K. Gezondheid en sport voor kinderen en jongeren met een chronische ziekte. Presentatie. Landelijke Fitkids dag, 8 juni 2018, Zeist.

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

Lankhorst K, van den Berg-Emons RJ, Bussmann JBJ, Horemans HLD, de Groot JF. De validiteit van de Activ8 bij rolstoel rijdende kinderen. Presentatie. Fit for the Future! Faculteit Revalidatiegeneeskunde, Erasmus Universiteit, 9 oktober 2017, Rotterdam.

Lankhorst K, Zwinkels M, de Groot J, Takken T. Blessures bij kinderen en jongeren met een fysieke beperking of chronische ziekte. Hebben sportende kinderen meer blessures? Presentatie. Terugkomdag ouders en kinderen op de Supportbeurs, 28 mei 2016, Utrecht.

Lankhorst K, van den Berg-Emons RJ, Bussmann JBJ, Horemans HLD, de Groot JF. Validiteit studie Activ8. Presentatie. Werkveld- / scholingsdag Fit for the Future!, Faculteit Gezondheidszorg, Hogeschool Utrecht, 16 januari 2016, Utrecht.

Lankhorst K, van den Berg-Emons RJ, Bussmann JBJ, Horemans HLD, de Groot JF. De validiteit van de Activ8 bij jongeren met en zonder een beperking. Presentatie. Fit for the Future! Faculteit Revalidatiegeneeskunde, Erasmus Universiteit, 22 september 2015, Rotterdam.

Lankhorst K, Zwinkels M, Takken T, de Groot J, Verschuren O, MVOTV. Kennisclip HAYS en S2SF studies. 6 november 2017. <https://youtu.be/VuWS63avfsM>.

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