PHYSICAL FITNESS AND PHYSICAL BEHAVIOR IN (WHEELCHAIR-USING) YOUTH WITH SPINA BIFIDA

Fysieke fitheid en fysiek gedrag
bij (rolstoel-rijdende) kinderen en adolescenten
met spina bifida

MANON BLOEMEN

PHYSICAL FITNESS AND PHYSICAL BEHAVIOR IN (WHEELCHAIR-USING) YOUTH WITH SPINA BIFIDA

FYSIEKE FITHEID EN FYSIEK GEDRAG
BIJ (ROLSTOEL-RIJDENDE) KINDEREN EN ADOLESCENTEN
MET SPINA BIFIDA

(met een samenvatting in het Nederlands)

COVER Bart Heideman

LAYOUT Bart Heideman

PRINTED BY Proefschriftmaken - www.proefschriftmaken.nl

ISBN 978-94-6295-639-1

© 2017 M.A.T. Bloemen.
All rights reserved. No part of this publication may be reproduced, stored in a retrieval system, or transmitted, in any form or by any means, electronically, mechanically, by photocopying, recording or otherwise, without the prior written permission of the author.

PROEFSCHRIFT

ter verkrijging van de graad van doctor aan de Universiteit Utrecht op gezag van de rector magnificus, prof. dr. G.J. van der Zwaan, ingevolge het besluit van het college voor promoties in het openbaar te verdedigen op woensdag 7 juni 2017 des ochtends te 10.30 uur
door

MARIA ANNE TRUDE BLOEMEN
geboren op 24 februari 1976
te Oldenzaal

Promotor	Prof. dr. F.J.G. Backx
Copromotoren	Dr. J.F. de Groot
	Dr. T. Takken

Dit proefschrift werd (mede) mogelijk gemaakt met financiële steun van Stichting Steunfonds Gezondheidszorgopleidingen van de Hogeschool Utrecht, K-J Projects, Wetenschappelijk College Fysiotherapie (WCF) van het Koninklijk Nederlands Genootschap Fysiotherapie (KNGF), Nederlandse Vereniging voor Kinderfysiotherapie (NVFK), ProCare B.V. en Lode B.V., Double Performance B.V. en de Hogeschool Utrecht.

De onderzoeken zijn gesubsidieerd door Foundation Innovation Alliance – Regional Attention and Action for Knowledge circulation (SIA RAAK) project nummers 2011-13-35P en SIA RAAK PRO-4-03 en een promotievoucher van de Hogeschool Utrecht.

CONTENTS

1

General Introduction

GENERAL INTRODUCTION

Spina bifida (SB) is the most frequently seen congenital deformity of the neural tube with an incidence ranging from 3 to 12.8 new cases per 10.000 births.[1] The malformation of the spinal cord and often the brain can result in both motor and sensor impairment, incontinence for bowel and bladder and cognitive impairment.[2] There are several types of SB, with varying impact on daily life: SB occulta (closed type), meningocele and myelomeningocele (both open types of SB), with the latter being the most common and involved type of SB.[2] Depending on the type of SB and the height of the lesion level of the spinal cord, children with SB experience difficulties with ambulation. The ambulation level is classified according to the Hoffer classification adjusted by Schoenmakers et al. (table 1) and ranges from normal ambulatory (level 1) to non-ambulatory (Level 5).[3]

Table 1. *Hoffer classification adjusted to Schoenmakers et al.*[3]

LEVEL	DESCRIPTION
Level 1	Normal ambulatory
Level 2	Community ambulatory
Level 3	Household ambulatory
Level 4	Therapeutic ambulatory
Level 5	Non-ambulatory

Looking at prognosis and development of SB, a 25-year cohort study found a decline in ambulation as a main mode of locomotion from 95% at age 0-5 to 46% at age 20-25.[4] This shows that part of ambulating children with SB will use a wheelchair for their main mobility when they grow older. Other evidence shows that about 50% of children with myelomeningocele uses a wheelchair for daily mobility.[5] Of the remaining 50% that are functional ambulatory (Hoffer 1-3), a large part will use a manual wheelchair for long distances or for sports participation. In this thesis, “wheelchair-using” is defined as using a wheelchair for either daily activities but also as using a wheelchair for solely long distances or sports participation. This means that children can be classified as Hoffer level 1 through level 5.

Due to advances in the medical approach, mortality rates of children with SB have decreased over the last years and 75%-80% of children with SB can now be expected to live to be adults.[4, 6] This requires a different approach in management of these patients from childhood through adolescence and adulthood, not only focusing on pathological aspects, but also at preventable medical and social consequences of the congenital disorder.[7] In general, adults with SB have an inactive lifestyle and thus show unfavorable physical behavior.[8] Evidence reports that they have lower physical fitness, higher prevalence of obesity, and lower health-related quality of life compared to peers.[9] Moreover, adults classified as Hoffer 4 and 5 show even more unfavorable physical behavior and lower levels of fitness compared to adults with SB classified as Hoffer 1-3.[10]

RATIONALE PHYSICAL FITNESS & PHYSICAL BEHAVIOR

Physical fitness consists of health-related fitness and skill-related fitness.[11] An important component of health-related fitness is cardiorespiratory endurance, with peak oxygen uptake (VO_{2peak}) as the single best indicator of the cardio-respiratory system. Skill-related fitness consists of power, speed, agility, coordination, balance and reaction time and is reflected in activities such as playing outside or playing wheelchair sports.[11] Physical behavior consists of sedentary activity and physical activity and is performed in a specific context with a certain motivation.[12] Sedentary activity is defined as "sitting or lying during waking hours with an energy expenditure lower than 1.5 metabolic equivalent task (MET)" whereas physical activity has been defined as "any bodily movement that results in energy expenditure".[11, 13]

Many studies have demonstrated the associations between low physical fitness, unfavorable physical behavior and higher cardiovascular and overall mortality.[14, 15] Recent Lancet series have shown that population rates of physical activity participation are low which has a major health impact worldwide. The authors concluded that decrease of this unhealthy behavior could improve health substantially.[14, 16, 17] Even though several interventions to improve this global unfavorable physical behavior have been developed and implemented in recent years, it remains a challenging international problem as physical activity has not increased.[18, 19]

While general population rates for participation in physical activity are already low, youth seems to do even worse than adults when looking at public health guidelines for recommended levels of physical activity.[16] Even though there are differences between countries, a large proportion of typically developing youth does not comply with these guidelines.[16, 20] For example, only an average of 29% of Dutch 12-17 year olds meet the Dutch guideline of physical activity (moderate to vigorous physically active for at least 60 minutes per day).[21] While typically developing children already are increasingly inactive, children with a physical disability like SB are even at higher risk of developing decreased levels of physical fitness and unfavorable physical behavior due to several psychosocial factors (Figure 1).[22] Moreover, children with SB seem to be prone to a vicious circle of decreased mobility and functional impairment; an inactive lifestyle, resulting in obesity, reduced health related quality of life and lower levels of physical fitness, in its turn influencing activities and participation in patients with SB.[23-26] Several studies have shown that physical fitness and physical behavior in youth with other physical disabilities than SB is low compared to typically developing peers.[27-29] Regarding youth with SB, evidence shows that ambulatory children also have reduced levels of physical fitness and are less physically active compared to typically developing peers.[30] A study with ambulating and wheelchair-dependent adolescents and young adults with SB showed that 39% of the participants were classified as inactive, with 37% as extremely inactive.[10] The wheelchair dependent participants had lower levels of physical fitness and showed unfavorable physical behavior compared to the ambulating participants. In the wheelchair dependent participants, physical fitness was significantly related to physical behavior.[10]

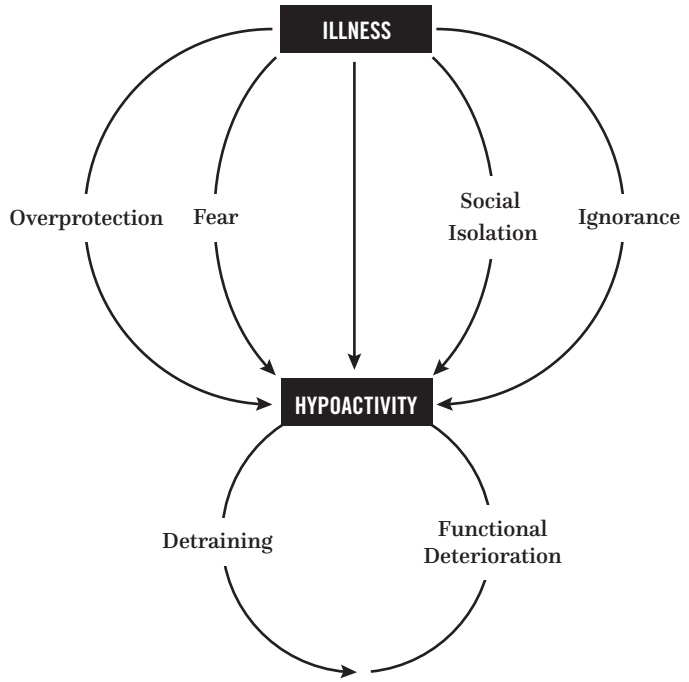

Figure 1. *Direct and indirect links between illness and hypoactivity according to Bar Or[22]*

PHYSICAL FITNESS TESTING

The knowledge-base regarding physical fitness testing for wheelchair-using youth is still very small. Even though assessment and optimizing physical fitness in youth with chronic conditions like spina bifida (SB) are important goals in pediatric rehabilitation, there are no valid and reliable tests available for clinicians to measure physical fitness in wheelchair-using children with spina bifida.[31] Valid and reliable tests will contribute to evaluation of interventions and the clinical reasoning process of clinicians. It is important for clinicians that tests do not require expensive equipment, are easy to implement in practice and that they are task specific. Moreover, it is of great importance that children can use their own wheelchair, as it takes into account the wheelchair-user interface integration.[32, 33] These considerations underline the importance of the development of valid and reliable field-based tests for measuring physical fitness in wheelchair-using youth with SB.

In wheelchair-using adults, arm cranking protocols are often used to assess VO_{2peak}. However, arm cranking protocols lack specificity to wheelchair propulsion and therefore the validity of these types of protocols are questioned.[33] Consequently,

wheelchair propulsion might be a more appropriate way of testing VO_{2peak} in wheelchair-using youth with SB.[34] In **chapter 2** the best test performance and feasibility is analyzed using a Graded Arm Cranking Protocol versus a Graded Wheelchair Propulsion Test in wheelchair-using youth with SB after which the reliability of the best test is determined.[35] After determining the best laboratory test to measure VO_{2peak}, **chapter 3** analyzes the validity and reliability of the Shuttle Ride Test (SRiT) in wheelchair-using youth with SB, a field-based test to measure cardiorespiratory endurance.[36] **Chapter 4** analyzes the validity and reliability of the field-based tests to assess skill-related fitness in wheelchair-using youth with SB.[37]

PHYSICAL BEHAVIOR

There is no evidence in the literature that presents an overview of physical behavior in wheelchair-using youth with SB. Also relations with VO_{2peak} or other determinants such as age, gender and ambulatory status are lacking. Knowing the level of physical behavior in wheelchair-using youth with SB and understanding its relations with certain determinants will help us to tailor and optimize interventions specific for this population. **Chapter 5** describes the physical behavior of wheelchair-using youth with SB and **chapter 6** analyzes the relations between physical behavior and VO_{2peak}, age, gender, and Hoffer classification in wheelchair-using youth with SB.[38, 39]

Unfavorable physical behavior in children with physical disabilities is a complex problem and pediatric physical therapists have a unique position in implementing interventions to achieve healthy active lifestyles in youth with physical disabilities like SB.[31] Qualitative research offers insight in the children's and parental perspectives regarding the problems they experience when trying to improve physical behavior. **Chapter 7** analyzes these children's and parental perspectives and describes the personal and environmental factors to consider when aiming to improve participation in physical activity in children with SB.[40] Furthermore, it is important to analyze all available evidence about factors that influence physical behavior to be able to develop an intervention in the future that may improve physical behavior in children with physical disabilities like SB. So **chapter 8** presents an overview of this available evidence and analyzes which factors are associated with physical activity in youth with a physical disability.[41] **Chapter 9** shows evidence already present in the literature for interventions aiming to increase physical activity in children with physical disabilities.[42] This will help us to understand which aspects of interventions that are already used show effectiveness and which aspects not. Finally, **chapter 10** presents the theoretical and clinical implications, methodological considerations, directions for future research and the overall conclusion.

The results presented in chapter 2,3,4,5 and 6 are part of the *Let's Ride... Study*, analyzing physical fitness and physical behavior in wheelchair-using youth with SB.

REFERENCES

1. Castilla EE, Botto LD, Bakker MK. (2011)
International Clearinghouse for Birth Defects Surveillance and Research.
Annual Report 2011 with Data for 2009.
http://www.icbdsr.org/filebank/documents/ar2005/Report2011.pdf
Accessed December 03, 2012.

2. Singh DK.
Families of Children with Spina Bifida : A Review.
Journal of developmental and physical disabilities 2003;15: 37-54.

3. Schoenmakers MA, Gulmans VA, Gooskens RH, Helders PJ.
Spina bifida at the sacral level: more than minor gait disturbances.
Clin Rehabil 2004;18: 178-185.

4. Bowman RM, McLone DG, Grant JA, Tomita T, Ito JA.
Spina bifida outcome: a 25-year prospective.
Pediatr Neurosurg 2001;34: 114-120.

5. Schoenmakers MA, Uiterwaal CS, Gulmans VA, Gooskens RH, Helders PJ.
Determinants of functional independence and quality of life in children with spina bifida.
Clin Rehabil 2005;19: 677-685.

6. Mitchell LE, Adzick NS, Melchionne J, Pasquariello PS, Sutton LN, Whitehead AS.
Spina bifida.
Lancet 2004;364: 1885-1895.

7. Simeonsson RJ, McMillen JS, Huntington GS.
Secondary conditions in children with disabilities: spina bifida as a case example.
Ment Retard Dev Disabil Res Rev 2002;8: 198-205.

8. Van den Berg-Emons HJ, Bussmann JB, Brobbel AS, Roebroeck ME, van Meeteren J, Stam HJ.
Everyday physical activity in adolescents and young adults with meningomyelocele as measured with a novel activity monitor.
J Pediatr 2001;139: 880-886.

9. Crytzer TM, Dicianno BE, Kapoor R.
Physical activity, exercise, and health-related measures of fitness in adults with spina bifida: a review of the literature.
PM R 2013;5: 1051-1062.

10. Buffart LM, Roebroeck ME, Rol M, Stam HJ, van den Berg-Emons RJ, Transition Research Group South-West Netherlands.
Triad of physical activity, aerobic fitness and obesity in adolescents and young adults with myelomeningocele.
J Rehabil Med 2008;40: 70-75.

11. Caspersen CJ, Powell KE, Christenson GM.
Physical activity, exercise, and physicaL fitness: definitions and distinctions for health-related research.
Public Health Rep 1985;100: 126-131.

12. Bussmann JB, van den Berg-Emons RJ.
To total amount of activity... and beyond: perspectives on measuring physical behavior.
Front Psychol 2013;4: 463.

13. Sedentary Behaviour Research Network.
Letter to the editor: standardized use of the terms "sedentary" and "sedentary behaviours".
Appl Physiol Nutr Metab 2012;37: 540-542.

14. Lee IM, Shiroma EJ, Lobelo F, Puska P, Blair SN, Katzmarzyk PT, et al.
Effect of physical inactivity on major non-communicable diseases worldwide: an analysis of burden of disease and life expectancy.
Lancet 2012;380: 219-229.

15. Kohl HW,3rd, Craig CL, Lambert EV, Inoue S, Alkandari JR, Leetongin G, et al.
The pandemic of physical inactivity: global action for public health.
Lancet 2012;380: 294-305.

16. Hallal PC, Andersen LB, Bull FC, Guthold R, Haskell W, Ekelund U, et al.
Global physical activity levels: surveillance progress, pitfalls, and prospects.
Lancet 2012;380: 247-257.

17. Bauman AE, Reis RS, Sallis JF, Wells JC, Loos RJ, Martin BW, et al.
Correlates of physical activity: why are some people physically active and others not?
Lancet 2012;380: 258-271.

18. Sallis JF, Bull F, Guthold R, Heath GW, Inoue S, Kelly P, et al.
Progress in physical activity over the Olympic quadrennium.
Lancet 2016;388: 1325-1336.

19. Reis RS, Salvo D, Ogilvie D, Lambert EV, Goenka S, Brownson RC, et al.
Scaling up physical activity interventions worldwide: stepping up to larger and smarter approaches to get people moving.
Lancet 2016;388: 1337-1348.

20. Van Hecke L, Loyen A, Verloigne M, van der Ploeg HP, Lakerveld J, Brug J, et al.
Variation in population levels of physical activity in European children and adolescents according to cross-European studies: a systematic literature review within DEDIPAC.
Int J Behav Nutr Phys Act 2016;13: 70-016-0396-4.

21. 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: S218-S224.

22. Bar-Or O.
Role of exercise in the assessment and management of neuromuscular disease in children.
Med Sci Sports Exerc 1996;28: 421-427.

23. Van den Berg-Emons HJ, Bussmann JB, Meyerink HJ, Roebroeck ME, Stam HJ.
Body fat, fitness and level of everyday physical activity in adolescents and young adults with meningomyelocele.
J Rehabil Med 2003;35: 271-275.

24. Rimmer JA, Rowland JL.
Physical activity for youth with disabilities: a critical need in an underserved population.
Dev Neurorehabil 2008;11: 141-148.

25. Rimmer JH, Marques AC.
Physical activity for people with disabilities.
Lancet 2012;380: 193-195.

26. Durstine JL, Painter P, Franklin BA, Morgan D, Pitetti KH, Roberts SO.
Physical activity for the chronically ill and disabled.
Sports Med 2000;30: 207-219.

27. Balemans AC, van Wely L, Middelweerd A, van den Noort J, Becher JG, Dallmeijer AJ.
Daily stride rate activity and heart rate response in children with cerebral palsy.
J Rehabil Med 2014;46: 45-50.

28. Obeid J, Balemans AC, Noorduyn SG, Gorter JW, Timmons BW.
Objectively measured sedentary time in youth with cerebral palsy compared with age-, sex-, and season-matched youth who are developing typically: an explorative study.
Phys Ther 2014;94: 1163-1167.

29. Ryan JM, Forde C, Hussey JM, Gormley J.
Comparison of Patterns of Physical Activity and Sedentary Behavior Between Children With Cerebral Palsy and Children With Typical Development.
Phys Ther 2015;95: 1609-1616.

30. 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.

31. Rowland JL, Fragala-Pinkham M, Miles C, O'Neil ME.
The scope of pediatric physical therapy practice in health promotion and fitness for youth with disabilities.
Pediatr Phys Ther 2015;27: 2-15.

32. Mason BS, van der Woude LH, Goosey-Tolfrey VL.
The ergonomics of wheelchair configuration for optimal performance in the wheelchair court sports.
Sports Med 2013;43: 23-38.

33. Goosey-Tolfrey VL, Leicht CA.
Field-based physiological testing of wheelchair athletes.
Sports Med 2013;43: 77-91.

34. Verschuren O, Zwinkels M, Ketelaar M, Reijnders-van Son F, Takken T.
Reproducibility and validity of the 10-meter shuttle ride test in wheelchair-using children and adolescents with cerebral palsy.
Phys Ther 2013;93: 967-974.

35. 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: 432-437.

36. Bloemen MAT, de Groot JF, Backx FJG, Benner J, Kruitwagen CLJJ, Takken T.
The wheelchair shuttle test for assessing aerobic fitness in youth with spina bifida; a validity and reliability study.
Phys Ther Accepted.

37. Bloemen MA, Takken T, Backx FJ, Vos M, Kruitwagen CL, de Groot JF.
Validity and Reliability of Skill-Related Fitness Tests for Wheelchair-Using Youth With Spina Bifida.
Arch Phys Med Rehabil 2016.

38. Bloemen MAT, van den Berg-Emons RHJG, Tuijt M, Nooijen CF, Takken T, Backx FJG, et al. Physical behavior in wheelchair-using children and adolescents with spina bifida. Submitted.

39. Bloemen MAT, Takken T, de Groot JF, Kruitwagen CLJJ, Rook RA, van den Berg-Emons RHJG, et al.
Determinants of physical behavior in wheelchair-using youth with spina bifida. Submitted.

40. 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;15: 11-015-0265-9.

41. 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;57: 137-148.

42. Bloemen MAT, Van Wely L, Mollema J, Dallmeijer A, de Groot JF.
Increasing physical activity in children with physical disability: what is the evidence? A systematic review.
Dev Med Child Neurol Accepted.

2

Arm cranking versus wheelchair propulsion for testing aerobic fitness in children with spina bifida who are wheelchair dependent.

Manon A.T. Bloemen[1,2], MSc; Janke F. de Groot[1,2], PhD; Prof. Frank J.G. Backx[3]; Rosalyne A. Westerveld[1], BA; Tim Takken[2], PhD

1 Research Group Lifestyle and Health, HU University of Applied Sciences Utrecht, Utrecht, The Netherlands
2 Child Development and Exercise Center, Wilhelmina Children's Hospital, University Medical Center Utrecht, The Netherlands
3 Department of Rehabilitation, Physical Therapy Science and Sports, University Medical Center Utrecht, The Netherlands

J Rehabil Med 2015 May; 47(5): 432-7

ABSTRACT

Objective
To determine the best test performance and feasibility using a Graded Arm Cranking Test vs a Graded Wheelchair Propulsion Test in young people with spina bifida who use a wheelchair, and to determine the reliability of the best test.

Design
Validity and reliability study.

Subjects
Young people with spina bifida who use a wheelchair.

Methods
Physiological responses were measured during a Graded Arm Cranking Test and a Graded Wheelchair Propulsion Test using a heart rate monitor and calibrated mobile gas analysis system (Cortex Metamax). For validity, peak oxygen uptake (VO_{2peak}) and peak heart rate (HR_{peak}) were compared using paired t-tests.
For reliability, the intraclass correlation coefficients, standard error of measurement, and standard detectable change were calculated.

Results
VO_{2peak} and HR_{peak} were higher during wheelchair propulsion compared with arm cranking (23.1 vs 19.5 ml/kg/min, $p = 0.11$; 165 vs 150 beats/min, $p < 0.05$). Reliability of wheelchair propulsion showed high intraclass correlation coefficients (ICCs) for both VO_{2peak} (ICC=0.93) and HR_{peak} (ICC=0.90).

Conclusion
This pilot study shows higher HR_{peak} and a tendency to higher VO_{2peak} in young people with spina bifida who are using a wheelchair when tested during wheelchair propulsion compared with arm cranking. Wheelchair propulsion showed good reliability.
We recommend performing a wheelchair propulsion test for aerobic fitness testing in this population.

INTRODUCTION

Aerobic fitness is related to clustered cardiovascular disease risk factors in children and adolescents.[1] Several studies have shown young people with disabilities to be less active and less fit compared with their peers.[2–4] Furthermore, studies have reported that adolescents and young adults who use a wheelchair are more inactive and less fit than their peers who walk.[5, 6] For example, in a study of young adults with spina bifida (SB) who use a wheelchair, 39% were classified as inactive, and 37% as extremely inactive.[6] Thus, aerobic fitness testing in children and adolescents with disabilities has become an important issue in both research and clinical practice.[2, 7–10]

Several studies have examined methods of testing aerobic fitness in children with disabilities who are ambulatory.[2] However, the knowledge-base regarding aerobic fitness testing for children and adolescents who use a wheelchair is small. In aerobic fitness testing peak oxygen uptake (Vo_{2peak}) is considered to be the single best indicator of the cardio-respiratory system. The gold standard for measuring VO_{2peak} is an incremental ergometer test with gas exchange until volitional exhaustion.

In wheelchair ergometry, arm cranking protocols are often used in aerobic fitness testing. However, a recent review of wheelchair testing in adults suggests that arm cranking protocols lack specificity to wheelchair propulsion and therefore questions the validity of these types of protocols.[11] A recent study in young people with cerebral palsy observed that a field test using wheelchair propulsion yielded higher cardiorespiratory parameters compared with arm cranking. Therefore wheelchair propulsion might be a more appropriate way of testing children and adolescents who use a wheelchair.[12] To test aerobic fitness in children with neuromuscular disease, Bar-or stated that "testing in the laboratory has the advantage (over field conditions) of better standardization". Bar-or further emphasized that, in children with neuromuscular disease, assessment of the oxygen transport system, reduced muscle function (strength and endurance) and other limitations should be taken into account when developing a test to measure VO_{2peak}.[13]

In preparation for a larger intervention study, using VO_{2peak} as 1 of the outcome measures, the aims of this pilot study were: (I) to investigate the best test performance and feasibility using a Graded Arm Cranking Test (GACT) vs a Graded Wheelchair Propulsion Test (GWPT) in the laboratory to measure VO_{2peak} in children and adolescents with SB who use a wheelchair, and (II) to determine the reliability of the best test in young people with SB who use a wheelchair.

METHODS

This study is part of the larger "*Let's Ride… Study*", looking at fitness and physical activity in young people with SB. The present study comprised 2 parts: "a best test performance and feasibility study (study 1; study population 13 children)" and "a reliability study (study 2; study population 24 children)".

Subjects

Children and adolescents were recruited through the BOSK (the association for people with a physical disability and their parents in the Netherlands), pediatric physical therapists working with these children, and several rehabilitation centres and SB outpatient services in the Netherlands. Inclusion criteria were: age range 6–18 years at enrolment; a diagnosis of SB; using a manual wheelchair for daily life and/or sports; and able to follow test instructions. Parents, and children who were aged 12 years and over, had to sign informed consent. Children and adolescents were excluded if they had any (medical) events that might interfere with testing (e.g. a change of wheelchair during the testing period or any acute medical events). The study was approved by the medical ethics committee of the University Medical Center Utrecht.

Demographics and morphological parameters

A questionnaire was used to record age, gender, type of SB, lesion level, sport activities before testing, health status, use of wheelchair and type of wheelchair. Body mass was measured using an electronic wheelchair scale (KERN MWS-300K100M, KERN & SOHN GmbH, Balingen, Germany). Height was measured using a non-stretchable tape while seated using the arm-span length (middle finger-tip to middle finger-tip) as recommended in wheelchair-dependent children, due to the presence of contractures when lying supine.[14] Fat-free mass was determined with a bioelectrical impedance analysis system (BIA; the Bodystat® QuadScan 4000 System; Bodystat Ltd, Isle of Man, UK). BIA is a non-invasive simple test to distinguish lean body mass and fat by comparing conductivity and resistance in the body.[15]

Exercise testing

Two graded exercise tests were used in this study; the GACT and the GWPT (see below). During these tests, physiological responses, including breath-by-breath gas analysis, were measured using a heart rate (HR) monitor (miniCardio, Hosand Technologies Srl, Verbania, Italy) and calibrated mobile gas analysis system (Cortex Metamax B3, Cortex Medical GmbH, Leipzig, Germany). The Cortex Metamax is a valid and reliable system for measuring gas-exchange parameters during exercise.[16]

Graded Arm Cranking Test

During the GACT a modified McMaster all-out progressive continuous arm cranking protocol was performed[17] on an electro-magnetically braked arm cranking ergometer (Lode Angio, Procare BV, Groningen, the Netherlands). The participant used his/her own wheelchair in order to ensure adequate support and a stable position during the GACT. After an initial warm-up phase at 0 Watts, the resistance was increased every minute by 8 Watts. The participant was encouraged to maintain the recommended cadence of 60–80 rpm.

Graded Wheelchair Propulsion Test

The GWPT was performed on custom-made rolling bars (wheelchair ergometer) for wheelchairs (based on the Cateye Ergociser 3600, Osaka, Japan). The participant was seated in his/her own wheelchair and was secured to the rolling bars. Resistance was increased by 0.1 torque increments every minute, while participants were encouraged to maintain wheelchair propulsion at the same speed throughout the test. The speed was a self-selected comfortable speed of between 60 and 120 rpm for the first few minutes of testing.

Both protocols were continued until the participant stopped due to exhaustion, despite verbal encouragement from the test leader. After a 5-min rest period, participants were tested for a maximum of 5 min at 110% of the maximum resistance they reached. This type of supra-maximal testing has been described previously in healthy adults by Rossiter et al., and is explained below under "Exercise testing parameters".[18] For the "best test performance and feasibility study" (study 1) the children and adolescents visited the laboratory twice; once for the GACT and once for the GWPT, with 1–2 weeks between testing. For the "reliability study" (study 2) the children and adolescents visited the laboratory twice, both for the GWPT, with 1–2 weeks between testing. Conditions during testing were identical during visits 1 and 2.

Data analysis

Exercise testing parameters

Both peak and supra-maximal exercise parameters were calculated as the mean value over the highest 30 s during the exercise test. Normalized VO_{2peak} was calculated as VO_{2peak}/kg or $VO_{2supramaximal}$/kg and expressed as ml/kg/min. Aerobic testing usually includes physiological responses and criteria for maximal exercise testing as set out by Rowland.[19] These criteria are subdivided into subjective and objective criteria, where every child has to meet the first and at least 1 of the latter to confirm true maximal aerobic fitness testing. The subjective criteria include signs of intense effort (sweating; facial flushing; clear unwillingness to continue despite encouragement), whereas the objective criteria for aerobic fitness testing include an evaluation of HR_{peak} (≥ 180 beats/min) and peak respiratory exchange ratio (RER_{peak}) (> 0.99).

Because these objective criteria may not be applicable for wheelchair ergometry, a supra-maximal protocol, as described by Rossiter et al., was used to confirm $VO_{2peak.}$[18] In earlier studies in our laboratory, this type of supra-maximal testing has been proven useful and feasible in children with disabilities and chronic conditions.[20, 21]

Best test performance and feasibility study (study 1)

- GACT and GWPT were compared by calculating descriptives and differences, both at the individual level and at group level. Two- tailed t-tests were used to test differences between the GACT and GWPT, after testing for normal distribution and equality of means. The significance level was set at $\alpha<0.05$.
- Maximal effort was defined as the presence of subjective criteria for intense effort, such as sweating, facial flushing and clear unwillingness to continue despite encouragement.
- Acceptability was defined as the willingness to perform the test again in the future, based on experienced burden. Children and adolescents were asked which test they preferred and why.
- Adverse events following exercise testing were monitored by asking the children, adolescents and parents during their next visit.

Table 1. *Study population*

	STUDY1 BEST TEST PERFORMANCE AND FEASIBILITY (N = 13)	**STUDY2** RELIABILITY (N = 24)
LEVEL OF LESION	**FREQUENCY**	**FREQUENCY**
Thoracic	2	5
Lumbar	11	19
ANTHROPOMETRICS	**MEAN (SD)**	**MEAN (SD)**
Age (years)	13.4 (3.5)	14.8 (3.0)
Arm span (cm)	157 (23)	160 (16)
Weight (kg)	46.2 (18.7)	54.5 (16.2)
BMI	17.9 (3.2)	22 (7.1)
Fat free mass (%kg)	69.3 (7.3)	69 (14)
HOFFER AMBULATION LEVEL[25]	**FREQUENCY**	**FREQUENCY**
Community ambulatory	1	2
Household ambulatory	1	3
Therapeutic ambulatory	2	2
Non ambulatory	9	17

CM = Centimeter | KG = Kilogram | SD = Standard Deviation

Table 2. *Outcome data of GACT and GWPT in youth with SB who are wheelchair dependent.*

	GACT MEAN (SD) (N=12)	**GWPT** MEAN (SD) (N=12)
HR_{PEAK} (BPM)	150 (28)	165 (25)*
VO_{2PEAK} (ML/KG/MIN)	19.5 (4.4)#	23.1 (7.3)$$
RER_{PEAK}	1.19 (0.32)#	1.20 (0.21)$$
DIFFERENCE $VO_{2SUPRAMAX}$ (ML/KG/MIN)	1.4 (4.6)#	0.7 (2.2)$$
DURATION OF TESTING (MIN SEC)	7min 11sec (4min 22sec)	8min 27sec (2min 14sec)
MAXIMAL EFFORT	8	8
ADVERSE EVENTS	0	0

* = p<0.05 for differences between graded Graded Arm Cranking Test (GACT) and Graded Wheelchair Propulsion Test (GWPT) | HR = Heart Rate | BPM = Beats Per Minute | VO_{2peak} = peak oxygen uptake RER = Respiratory Exchange Ratio | SD = Standard Deviation | MIN = Minutes | SEC = Seconds
#n = 11 because child did not want to wear Metamax Cortex | $$ n = 10 because Cortex Metamax did not function properly.

Reliability study (study 2)

- Reliability was tested with the intra-class correlation coefficient (ICC) Shrout and Fleiss model 2.1.A.[22, 23]
- Measurement error was analysed using the standard error of measurement$_{agreement}$ ($SEM_{agreement}$) and the smallest detectable change (SDC), calculated using the following equations:
  - $SEM_{agreement} = \sqrt{\sigma^2_m + \sigma^2_{residual}}$, where σ^2_m accounts for the systematic errors between both measurements and $\sigma^2_{residual}$ accounts for the random error.[22,23]
  - $SDC = 1.96 * \sqrt{2} * SEM_{agreement.}$[22]

RESULTS

The study population for the "best test performance and feasibility study" (study 1) comprised 13 children (9 boys, 4 girls), mean age 13.4 (age range 8–17) years. The study population for the "reliability study" (study 2) comprised of 24 children (13 boys, 11 girls), mean age 14.8 (age range 8–19) years. The children in study 1 were different children from those in study 2. Children's age, height, weight, body mass index (BMI), fat-free mass, lesion level (classified according to American Spinal Injury Association (ASIA) guidelines[24]) and ambulation level according to Hoffer adapted by Schoenmakers et al.[25] are shown in table I.

Best test performance and feasibility (study 1)

Comparison between GACT and GWPT

One child (#8) could not be tested using the GACT because his arms were too short to reach the pedals. However, all children were able to perform the GWPT. One child (#12) reported experiencing pain in his forearms in the early stage of the GWPT, and therefore this test was discontinued. Outcome data for both the GACT and the GWPT at the group level are shown in table II and at the individual level in table III.

At the group level peak heart rate (HR_{peak}) was significantly higher during the GWPT compared with the GACT (165 ± 25 vs 150 ± 28 beats per min (bpm), $p < 0.05$). VO_{2peak} was not significantly different ($p = 0.11$); however, the difference in favour of the GWPT (23.1 vs 19.5 for GACT: >15%) was clinically relevant. Other exercise testing parameters, mean duration time and maximal effort were similar.

Examining the individual results, VO_{2peak} and HR_{peak} were higher in, respectively, 6/8 and 9/11 cases in GWPT, while RER_{peak} was comparable. HR_{peak} was below 180 bpm in 10/12 cases in GACT and in 8/12 cases in GWPT. However, RER_{peak} was higher than 0.99 in 8/11 cases in GACT and in 9/10 cases in GWPT. The minimum duration time was better during GWPT compared with GACT (4 min 27 s vs 1 min 12 s, respectively). In addition, only 3 children had a duration time of less than 6 min during the GWPT compared with 6 children during the GACT. Both the GACT and the GWPT showed good results during supramaximal testing, as only very small or even negative differences were found between VO_{2peak} and $VO_{2supramaximal}$ (table III).

Table 3. *Outcome data graded exercise testing at the individual level.*

	GACT					GWPT				
	VO_{2PEAK} (ML/KG/MIN)	$VO_{2SUPRAMAX}$ (ML/KG/MIN)	HR_{PEAK} (BPM)	RER_{PEAK}	TIME (MIN:SEC)	VO_{2PEAK} (ML/KG/MIN)	$VO_{2SUPRAMAX}$ (ML/KG/MIN)	HR_{PEAK} (BPM)	RER_{PEAK}	TIME (MIN:SEC)
1	27.5	28.1	198	1.18	13:30	MD[$$]	MD[$$]	200	MD[$$]	11:00
2	18.7	23.0	172	1.27	11:00	33.7	31.7	189	1.24	11:45
3	15.3	15.3	114	1.25	4:56	13.3	13.3	157	1.59	8:20
4	20.7	10.7	163	1.19	8:13	22.0	22.7	192	1.21	11:05
5	MD[#]	MD[#]	117	MD[#]	4:22	11.7	10.7	119	0.96	6:15
6	17.0	17.7	175	1.83	13:48	24.8	19.2	162	1.43	10:00
7	26.3	25.0	140	0.77	1:12	30.7	32.0	185	1.10	5:00
8	MD[$]	MD[$]	MD[$]	MD[$]	MD[$]	28.3	30.3	172	1.06	4:27
9	21.0	23.0	142	1.04	5:30	21.0	20.7	164	1.01	7:30
10	13.3	7.3	121	0.90	3:00	13.5	14.0	131	1.04	7:10
11	21.7	22.7	132	0.96	3:26	MD[$$]	MD[$$]	154	MD[$$]	5:16
12	28.7	27.3	180	1.04	9:24	MD[##]	MD[##]	MD[##]	MD[##]	MD[##]
13	24.0	23.3	175	1.25	10:07	25.7	25.3	165	1.01	9:30

GACT = Graded Arm Cranking Test | GWPT = Graded Wheelchair Propulsion Test | HR = Heart Rate | BPM = Beats Per Minute | VO_{2peak} = peak oxygen uptake | RER = Respiratory Exchange Ratio | SD = Standard Deviation | MIN = Minutes | SEC = Seconds | [$] = MD because arm crank ergometer was to large | [$$] = MD because Cortex Metamax did not function properly | [#] = MD because child did not want to wear the Cortex Metamax | [##] = MD because child had to stop early in the test because of pain in his lower arms.

Table 4. *Outcome aerobic fitness parameters GWPT in youth with SB.*

	GWPT (N=24) MEAN (SD)	
	TEST	RETEST
HR_{PEAK} (BPM)	185 (18)	180 (20)
VO_{2PEAK} (ML/KG/MIN)[$]	23.5 (7.4)	22.8 (6.6)
RER_{PEAK} [$]	1.23 (0.14)	1.20 (0.15)
DURATION OF TESTING (MIN SEC)(RANGE)	6:43 (1:36)	6:37 (1:40)
ADVERSE EVENTS	0	0

GWPT = Graded Wheelchair Propulsion Test | SB = Spina Bifida | HR = Heart Rate | BPM = Beats Per Minute VO_{2peak} = peak oxygen uptake | RER = Respiratory Exchange Ratio | SD = Standard Deviation | MIN = Minutes SEC = Seconds | $ n = 19 due to failure of the Cortex Metamax

Maximal effort, acceptability and adverse events

In both tests 8 children met the subjective criteria for maximal aerobic fitness testing. Most children and adolescents preferred the GWPT compared with the GACT (5/9); mostly because of familiarity with wheelchair propulsion and because of muscle fatigue in the neck, shoulders and arms during the GACT. However, 4 children did not have a preference. No adverse events occurred during testing.

Reliability (study 2)

The reliability of the GWPT was high, with excellent intra-class correlation coefficients (ICCs) for VO_{2peak} and high ICCs for HR_{peak}. The SEM was, respectively, 1.87 for VO_{2peak} and 6 for HR_{peak}. The SDC was, respectively, 5.18 for VO_{2peak} and 17 for HR_{peak}. The results are shown in tables IV and V.

Table 5. *Outcome reliability data.*

	RELIABILITY GWPT (N=24 HR_{PEAK}, N=23 VO_{2PEAK})		
	ICC 2.1.A (95% CI)	$SEM_{AGREEMENT}$	SDC
VO_{2PEAK} (ML/KG/MIN)	0.93 (0.83-0.97)	1.87	5.18
HR_{PEAK} (BPM)	0.90 (0.73-0.96)	6	17

GWPT = Graded Wheelchair Propulsion Test | HR = Heart Rate | VO_{2peak} = peak oxygen uptake | $SEM_{agreement}$ = Standard Error of Measurement | ICC = Intra Class Correlation Coefficient | 95% CI = 95% Confidence Interval | SDC = Smallest Detectable Change | BPM = Beats Per Minute.

DISCUSSION

The primary aim of this pilot study was to investigate best test performance and feasibility (study 1) using GACT vs GWPT in the laboratory setting to measure VO_{2peak} in children with SB who use a wheelchair. A secondary aim was to examine the reliability of the best test (study 2). Significantly higher HR_{peak} and clinically relevant higher VO_{2peak} values were found during the GWPT compared with the GACT; other exercise parameters, maximal effort and acceptability were similar in both tests. Because of this, reliability was determined for the GWPT. The reliability was high, with excellent ICCs for VO_{2peak} and high ICCs for HR_{peak}. The SEMs were acceptable and SDCs of 5.2 for VO_{2peak} and 17 for HR_{peak} were found at the individual level.

Best test performance and feasibility (study 1)

The preference for wheelchair propulsion compared with arm cranking in this study is similar to the results of a study by Verschuren et al.[12], which reported a higher VO_{2peak} (26.0 vs 25.3 ml/kg/min) and a significantly higher HR_{peak} (172 vs 161 bpm) during a wheelchair propulsion field test compared with a maximal GACT in children with cerebral palsy. Findings for adults using a wheelchair are equivocal, with studies showing no significant differences in VO_{2peak} when comparing arm cranking with wheelchair propulsion[26–28] and results indicating higher VO_{2peak} during functional wheelchair propulsion[29, 30]. Results for HR_{peak} also remain equivocal, both higher HR_{peak} during wheelchair propulsion[27, 31] and higher HR_{peak} during arm cranking[32] have been reported. The results of this study combined with the literature about children and adolescents support a change in functional propelling protocols, as suggested by Bar-or.[13]

Regarding feasibility, 1 child was unable to perform the GACT because of his limited arm span. Using the Cortex Metamax during the GACT was also complicated for older children and adolescents, due to the large dimensions of the arm crank ergometer, the flow sensor and face mask, which would probably have limited their maximum effort. Since we wanted to include children aged 6 years and over, this aspect supported our preference for the GWPT. However, 1 adolescent had to stop the GWPT prematurely, due to pain in his forearms. This individual was community ambulatory; he used his wheelchair only for long distances, which may explain the pain he experienced during the GWPT.

In this study VO_{2peak} and HR_{peak} were the main outcome parameters. Power output is also an important outcome parameter often used during aerobic fitness testing. However, it was not possible to report power output for the GWPT on the wheelchair ergometer. Measuring the resistance of the wheelchair on the ergometer is difficult, resulting in problems with measuring power output on the wheelchair ergometer. This problem could have been solved by using a wheelchair propulsion test on a treadmill, as is often used in adults with spinal cord injury who use a wheelchair.[30] In our opinion this was not feasible in our population, as children and adolescents with SB are often anxious,[33] which is likely to limit their maximum effort when tested on a treadmill.

Wheelchair propulsion can also be measured through field tests, such as the multistage field test and the Shuttle Ride Test.[12, 34] The benefits of these field tests are the absence of expensive equipment, the specificity of the task and the possibility of testing several participants at the same time. We decided, however, that we first had to determine the best laboratory test for measuring VO_{2peak} in children and adolescents with SB who use a wheelchair, as field testing may be influenced by, for example, wheelchair skills or anaerobic performance.[11] The validity and reliability of field-testing will be examined in a future study.

Validity for VO_{2peak} usually includes criteria for "maximal effort", subdivided into subjective and objective criteria.[19] These criteria apply to children who are developing typically; however, there are no criteria for maximal aerobic fitness testing in children and adolescents who use a wheelchair. Also, for adults who use a wheelchair the criteria for maximal aerobic fitness testing are unclear. Therefore, Goosey-Tolfrey & Leicht[11] recommended performing a verification protocol for measuring VO_{2peak}. In this study the protocol according to Rossiter et al.[18] was used. No differences were found between VO_{2peak} and $VO_{2supramaximal}$, assuming that maximal effort was achieved in both tests. However, 2 participants (#7 and #10) achieved both relatively low HR_{peak} (140 and 121 bpm, respectively) and low RER_{peak} values (0.77 and 0.90, respectively) during GACT assuming peripheral limitation instead of cardiovascular limitation. They did achieve higher HR_{peak} and RER_{peak} values during GWPT, again supporting our preference for using GWPT during exercise testing. When examining the criteria for maximal aerobic fitness testing for HR_{peak} and RER_{peak}, more children achieved the criterion for RER_{peak} of > 0.99 compared with the criterion for HR_{peak} of > 180 bpm. Future research should determine the criteria for maximal aerobic fitness testing in children and adolescents who use a wheelchair, so that these criteria can be used in both research and care.

Regarding acceptability, we asked the children and adolescents about their preference (GACT vs GWPT). Most children were able to explain why they preferred either the GACT or the GWPT. We also tried to apply the OMNI scale of perceived exertion[35] because research indicated a relationship between the rate of perceived exertion and VO_{2peak}.[36] However, using the OMNI scale of perceived exertion appeared to be questionable in this population. Children and adolescents often stated they were "not tired at all", even though they were visibly flushing and sweating and both HR_{peak} and RER_{peak} were high. Most children and adolescents with SB have lower IQ scores, and they may have difficulty interpreting the OMNI scale of perceived exertion.[37, 38]

Reliability (study 2)

The mean values of VO_{2peak} in our population were 23.5 (SD 7.4) and 22.8 (SD 6.6) ml/kg/min, with a total range of 12–36.7 ml/kg/min. The SDC was 5.2 ml/kg/min, equivalent to 22% of the mean VO_{2peak}. No literature is available about intervention studies regarding VO_{2peak} in children and adolescents with SB who use a wheelchair. Differences of 4% were found after training in children and adolescents with SB who were ambulatory.[39] However, a recent systematic review of exercise training programmes and wheelchair propulsion capacity in adults showed significant improvements in VO_{2peak} of 14–36%

after mixed training and 10–94% after endurance training.[40] The interpretation of an SDC of 5.1 ml/kg/min remains unclear in this population, in particular because of the low levels of fitness and the known inactivity of these children and adolescents.[2, 3] When participating in a training programme, they may experience a steep increase in VO_{2peak} due to their low starting point. Future research may provide information about progression in VO_{2peak} after training in children and adolescents with SB and, consequently, about interpretation of the SDC.

Study limitations

This study has several limitations. The first part of this pilot study involved only 13 participants, which may have resulted in clinical, yet not statistically significant, differences. However, when combining the results with those of the reliability study, the outcomes for VO_{2peak} appear to be consistent and even higher for HR_{peak}. Therefore, and supported by the best available evidence, we consider the choice in favour of GWPT to be justified. Another possible limitation is the use of fixed protocols for both the GACT and the GWPT for all participants, as this did not take into account differences in lesion level, age, height and physical activity level. This may have influenced the duration of the tests, and therefore also VO_{2peak} and HR_{peak}. It is important to expand our knowledge and experience regarding aerobic fitness testing in children who use a wheelchair, so that guidelines for more individual protocols may be developed in the future, comparable to the Godfrey protocols for children on a cycle ergometer. Furthermore, other clinimetric properties of the GWPT remain unclear, such as the minimal clinically important difference and responsiveness; these aspects may be the focus of future research.

In conclusion, this pilot study shows higher HR_{peak} and VO_{2peak} in children and adolescents with SB who are using a wheelchair when tested during wheelchair propulsion compared with arm cranking. The GWPT showed good reliability. We recommend performing a wheelchair propulsion test for aerobic fitness testing in children and adolescents who use a wheelchair.

ACKNOWLEDGEMENTS

The study was founded by Foundation Innovation Alliance – regional Attention and Action for knowledge circulation (SIA RAAK), project number 2011-13-35P and by personal PhD grant HU University of Applied Sciences Utrecht.

REFERENCES

1. Anderssen SA, Cooper AR, Riddoch C, Sardinha lB, Harro M, Brage S, et al. Low cardiorespiratory fitness is a strong predictor for clustering of cardiovas cular disease risk factors in children independent of country, age and sex. Eur J Cardiovasc Prev Rehabil 2007; 14: 526–531.

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: 2–14.

3. Van den Berg-Emons RJ, Bussmann JB, Stam HJ. Accelerometry based activity spectrum in persons with chronic physical conditions. Arch Phys Med Rehabil 2010; 91: 1856–1861.

4. Nooijen C, Slaman J, van der Slot W, Stam H, Roebroeck M, van den Berg-Emons R, et al. Health-related physical fitness of ambulatory adolescents and young adults with spastic cerebral palsy. J Rehabil Med 2014; 46: 642–647.

5. Van den Berg-Emons HJ, Bussmann JB, Brobbel AS, Roebroeck ME, van Meeteren J, Stam HJ. Everyday physical activity in adolescents and young adults with meningomyelocele as measured with a novel activity monitor. J Pediatr 2001; 139: 880–886.

6. Buffart LM, Roebroeck ME, Rol M, Stam HJ, van den Berg-Emons RJ, Transition research Group South-West Netherlands. Triad of physical activity, aerobic fitness and obesity in adolescents and young adults with myelomeningocele. J Rehabil Med 2008; 40: 70–75.

7. De Groot JF, Takken T, Gooskens RH, Schoenmakers MA, Wubbels M, van Hees l, et al. Reproducibility of maximal and submaximal exercise testing in “normal ambulatory” and “community ambulatory” children and adolescents with spina bifida: which is best for the evaluation and application of exercise training? Phys Ther 2011; 91: 267–276.

8. Van Brussel M, Lelieveld OT, van der Net J, Engelbert RH, Helders PJ, Takken T. Aerobic and anaerobic exercise capacity in children with juvenile idiopathic arthritis. Arthritis Rheum 2007; 57: 891–897.

9. Verschuren O, Ketelaar M, Keefer D, Wright V, Butler J, Ada l, et al. Identification of a core set of exercise tests for children and adolescents with cerebral palsy: A Delphi survey of researchers and clinicians. Dev Med Child Neurol 2011; 53: 449–456.

10. 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: 1107–1117.

11. Goosey-Tolfrey VL, Leicht CA. Field-based physiological testing of wheelchair athletes. Sports Med 2013; 43: 77–91.

12. Verschuren O, Zwinkels M, Ketelaar M, Reijnder-van Son F, Takken T. Reproducibility and Validity of the 10-meter shuttle ride test in wheelchair-bound children and adolescents with cerebral palsy. Phys Ther 2013; 93: 967–974.

13. Bar-or o. Role of exercise in the assessment and management of neuromuscular disease in children. Med Sci Sports Exerc 1996; 28: 421–427.

14. Dosa NP, Foley JT, Eckrich M, Woodall-ruff D, Liptak GS. Obesity across the lifespan among persons with spina bifida. Disabil Rehabil 2009; 31: 914–920.

15. Mok E, Letellier G, Cuisset JM, Denjean A, Gottrand F, Hankard R. Assessing change in body composition in children with duchenne muscular dystrophy: anthropometry and bioelectrical impedance analysis versus dual-energy X-ray absorptiometry. Clin Nutr 2010; 29: 633–638.

16. Medbo JL, Mamen A, Welde B, von Heimburg E, Stokke R. Examination of the Metamax I and II oxygen analysers during exercise studies in the laboratory. Scand J Clin Lab Invest 2002; 62: 585–598.

17. Bar-or o, Rowland T. Pediatric Exercise Medicine. From physiologic principles to healthcare application. Champaign, Il: Human kinetics; 2004.

18. Rossiter HB, Kowalchuk JM, Whipp BJ. A test to establish maximumO2 uptake despite no plateau in the O2 uptake response to ramp incremental exercise. J Appl Physiol (1985) 2006; 100: 764–770.

19. Rowland T. Developmental exercise physiology. Champaign, Il: Human kinetics; 1996.

20. Werkman MS, Hulzebos HJ, van de Weert-van Leeuwen PB, Arets HG, Helders PJ, Takken T. Supramaximal verification of peak oxygen uptake in adolescents with cystic fibrosis. Pediatr Phys Ther 2011; 23: 15–21.

21. De Groot JF, Takken T, de Graaff S, Cooskens RHJM, Helders PJM, Vanhees L. Treadmill testing of children who have spina bifida and are ambulatory: does peak oxygen uptake reflect maximum oxygen uptake? Phys Ther 2009; 89: 679–687.

22. De Vet H, Terwee C, Mokkink l, Knol D. Measurement in medicine. Practical guides to biostatistics and epidemiology. New York: Cambridge University Press; 2013.

23. De Vet HC, Terwee CB, Knol D, Bouter lM. When to use agreement versus reliability measures. J Clin Epidemiol 2006; 59: 1033–1039.

24. Maynard FM Jr, Bracken MB, Creasey G, Ditunno JF Jr, Donovan WH, Ducker TB, et al. International standards for neurological and functional classification of spinal cord injury. American Spinal Injury Association. Spinal Cord 1997; 35: 266–274.

25. Schoenmakers MA, Gulmans VA, Gooskens RH, Helders PJ. Spina bifida at the sacral level: more than minor gait disturbances. Clin Rehabil 2004; 18: 178–185.

26. Pitetti KH, Snell PG, Stray-Gundersen J. Maximal response of wheelchair-confined subjects to four types of arm exercise. Arch Phys Med Rehabil 1987; 68: 10–13.

27. Arabi H, Vandewalle H, Pitor P, de Lattre J, Monod H. Relationship between maximal oxygen uptake on different ergometers, lean arm volume and strength in paraplegic subjects. Eur J Appl Physiol Occup Physiol 1997; 76: 122–127.

28. Bhambhani Y. Physiology of wheelchair racing in athletes with spinal cord injury. Sports Med 2002; 32: 23–51.

29. Gass GC, Camp EM. The maximum physiological responses during incremental wheelchair and arm cranking exercise in male paraplegics. Med Sci Sports Exerc 1984; 16: 355–359.

30. Haisma JA, van der Woude LH, Stam HJ, Bergen MP, Sluis TA, Bussmann JB. Physical capacity in wheelchair-dependent persons with a spinal cord injury: a critical review of the literature. Spinal Cord 2006; 44: 642–652.

31. McConnell TJ, Horvat MA, Beutel-Horvat TA, Golding LA. Arm crank versus wheelchair treadmill ergometry to evaluate the performance of paraplegics. Paraplegia 1989; 27: 307–313.

32. Gayle GW, Pohlman Rl, Glaser RM, Davis GM. Cardiorespiratory and perceptual responses to arm crank and wheelchair exercise using various handrims in male paraplegics.
Res Q Exerc Sport 1990; 61: 224–232.

33. Flanagan A, Kelly EH, Vogel lC. Psychosocial outcomes of children and adolescents with early-onset spinal cord injury and those with spina bifida.
Pediatr Phys ther 2013; 25: 452–459.

34. Goosey-Tolfrey VL, Tolfrey K. The multi-stage fitness test as a predictor of endurance fitness in wheelchair athletes.
J Sports Sci 2008; 26: 511–517.

35. Robertson RJ, Goss FL, Boer NF, Peoples JA, Foreman AJ, Da-bayebeh IM, et al. Children's OMNI scale of perceived exertion: mixed gender and race validation.
Med Sci Sports Exerc 2000; 32: 452–458.

36. Goosey-tolfrey VL, Paulson TA, Tolfrey K, Eston RG. Prediction of peak oxygen uptake from differentiated ratings of perceived exertion during wheelchair propulsion in trained wheelchair sportspersons.
Eur J Appl Physiol 2014; 114: 1251–1258.

37. Dennis M, Barnes MA. The cognitive phenotype of spina bifida meningomyelocele.
Dev Disabil Res Rev 2010; 16: 31–39.

38. Kelly NC, Ammerman RT, Rausch JR, Ris MD, Yeates KO, Oppenheimer SG, et al. Executive functioning and psychological adjustment in children and youth with spina bifida.
Child Neuropsychol 2012; 18: 417–431.

39. 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; 25: 597–606.

40. Zwinkels M, Verschuren O, Janssen TW, Ketelaar M, Takken T, on behalf of the Sport-2-Stay-Fit study group. Exercise training programs to improve hand rim wheelchair propulsion capacity: a systematic review.
Clin Rehabil 2014; 28: 847–861.

3

The wheelchair shuttle test for assessing aerobic fitness in youth with spina bifida; a validity and reliability study

Manon AT Bloemen MSc[1,2,3], Janke F de Groot PhD[1,2], prof. Frank JG Backx[3] MD PhD, Joyce Benner MSc[1], Cas LJJ Kruitwagen MSc[4], Tim Takken PhD[2]

1 Research Group Lifestyle and Health, HU University of Applied Sciences Utrecht, Utrecht, The Netherlands
2 Child Development and Exercise Center, Wilhelmina Children's Hospital, University Medical Center Utrecht, The Netherlands
3 Department of Rehabilitation, Nursing Science and Sports, University Medical Center Utrecht, The Netherlands
4 Department of Biostatistics and Research Support, Julius Center, University Medical Center Utrecht, The Netherlands

Phys Ther Accepted

ABSTRACT

Background
Testing aerobic fitness in youth is important because of expected relationships with health.

Objective
To estimate validity and reliability of the Shuttle Ride Test in youth with spina bifida who use a wheelchair for mobility and sport.

Design
Validity and reliability study.

Methods
The Shuttle Ride Test, Graded Wheelchair Propulsion Test and skill related fitness tests were administered. N=33 for the validity study (14.5 ± 3.1 years) and N=28 for the reliability study (14.7 ± 3.3 years).

Results
No significant differences were found between the Graded Wheelchair Propulsion Test and Shuttle Ride Test for most cardiorespiratory responses. Correlations between the Graded Wheelchair Propulsion Test and Shuttle Ride Test were moderate to high (r=0.55 – 0.97). The variance of VO_{2peak} could be predicted for 77% by height, number of shuttles completed and weight with large prediction intervals. High correlations were found between number of shuttles completed and skill related fitness tests (0.73 ; -0.92). ICCs were high (0.77 – 0.98), with a smallest detectable change of 1.5 for number of shuttles completed and coefficient of variations of 6.2% and 6.4% for absolute and relative VO_{2peak} respectively.

Conclusion
When measuring VO_{2peak} directly by using a mobile gas analyses system, the Shuttle Ride Test is highly valid for testing VO_{2peak} in youth with spina bifida who use a wheelchair for mobility and sport. The outcome measure shuttles represents aerobic fitness, while also being highly correlated with both anaerobic performance and agility. It is not possible to predict VO_{2peak} accurately using the number of shuttles completed. Moreover, the Shuttle Ride Test is highly reliable in youth with spina bifida with a good smallest detectable change for the number of shuttles completed.

INTRODUCTION

Testing aerobic fitness in youth with a physical disability like spina bifida (SB) is currently an important aspect of pediatric physical therapy because of the expected relationships between fitness and health.[1, 2] The aerobic fitness of youth with SB is low compared to typically developing peers but also compared to youth with other chronic diseases.[3-5] A study with adolescents and young adults with SB showed that the average aerobic fitness was 42% lower than their typically developing peers, with the lowest scores in participants who are wheelchair-using.[6]

We analyzed the measurement properties of the Graded Wheelchair Propulsion Test (GWPT), a highly valid and highly reliable laboratory test to measure peak oxygen uptake (VO_{2peak}) in youth with SB who use a wheelchair for mobility and sport.[7] Despite the benefits of a laboratory test, field tests are more applicable for pediatric physical therapists as these tests can be performed in their own setting without the investment of expensive and sophisticated equipment. Valid and reliable field-based tests will contribute to evaluation of interventions and the clinical reasoning process of pediatric physical therapists concerning aerobic fitness.

The Shuttle Ride Test (SRiT), derived from the well-known and frequently used Shuttle Run Test in youth who are ambulatory, has been used in other clinical populations and seems to be the most appropriate maximal aerobic field test for measuring aerobic fitness in youth with SB who use a wheelchair for mobility and sport.[8, 9] During the SRiT, which is a stepwise incremental maximal exercise test, children propel their wheelchair back and forth to exhaustion with a standardized increasing speed between two lines that are 10 meters apart. The main outcome measure is the number of shuttles they achieved, with one shuttle corresponding to approximately one minute of wheelchair propulsion. A shuttle is a stage with a constant speed and the speed is increased approximately every 1 minute. This principle has been used over decades in field exercise tests in children.[10] The SRiT is a highly valid test for measuring VO_{2peak} when using a mobile gas analysis system in youth with Cerebral Palsy (CP) Gross Motor Function Classification System III and IV who use a wheelchair for mobility and sport. As disorder-related characteristics differ between youth with CP and SB, the validity should also be estimated for youth with SB who use a wheelchair for mobility and sport, comparing VO_{2peak} of the SRiT to the GWPT using a mobile gas analyses system.

While criterion validity concerns comparing VO_{2peak} measures between the SRiT and the GWPT, most pediatric physical therapists and other clinicians do not have the access to a mobile gas analysis system when assessing the SRiT. They have to use the metric shuttle during their clinical reasoning process. Evidence in athletes who are wheelchair-using, showed that it is difficult to predict VO_{2peak} using the distance travelled during the incremental SRiT.[11, 12] In children with Osteogenesis Imperfecta (OI), poor correlations were found between VO_{2peak} and the outcome measure shuttles.[9] Furthermore, the importance of anaerobic performance and agility during the SRiT has been hypothesized, as turning, deceleration and acceleration seem to be important skills during the SRiT.[8, 12] At the same time both personal aspects (e.g. age) and wheelchair features may play

an important role in the number of shuttles that a child achieves during the SRiT.[12, 13] So even though literature suggests that the SRiT is a highly valid, inexpensive field test for measuring aerobic fitness in children with CP and OI who use a wheelchair for mobility and sport, the construct of the metric shuttles is still unclear. Therefore, it is interesting to evaluate the possibility to predict VO_{2peak} achieved during the SRiT by using the outcome measure shuttles and to test the hypotheses of moderate to high correlations between the shuttles and anaerobic performance, agility, personal factors and wheelchair features in children with SB who use a wheelchair for mobility and sport. This information will help pediatric physical therapists to interpret the outcome measure shuttles during their clinical reasoning process.

Besides validity, reliability is an important measurement property and highly relevant when evaluating the effects of training. The SRiT is highly reliable in youth with CP and OI who use a wheelchair for mobility and sport, with ICCs > 0.95.[8, 9] The reliability of the SRiT has yet to be established in youth with SB who use a wheelchair for mobility and sport. Therefore the aims of this study are (1) to estimate the criterion validity of the SRiT by comparing cardiorespiratory responses of the SRiT to the GWPT using a mobile gas analysis system, (2) to estimate the construct validity of the metric shuttles by predicting VO_{2peak} and correlating the shuttles with anaerobic performance, agility, personal factors and wheelchair features and (3) to estimate the reliability of the SRiT in youth with SB who use a wheelchair for mobility and sport.

METHODS

Participants

This study is part of the larger "*Let's Ride... Study*", looking at fitness and physical activity in youth with SB who use a wheelchair for mobility and sport.[7, 14] The recruitment of participants took place in the Netherlands by pediatric physical therapists, rehabilitation centers, the BOSK (Association of an by parents of youth and adults with a disability) and SB outpatient services. Participants were included if they were diagnosed with SB, 5-18 years of age during enrollment, and were able to follow instructions of the measurements. They had to self-propel a wheelchair during daily life, long distances or sports participation, meaning that they had to be experienced wheelchair-users. Participants were excluded if any (medical) event was present that intervened with testing outcome.

Procedures

The study procedures were approved by the Medical Ethics Committee of the University Medical Center Utrecht, the Netherlands. Children aged 12 years and over and all parents had to sign informed consent, as this is required by the Dutch law and regulations.

The participants of the validity study were measured twice (one day for the GWPT and one day for the SRiT), those who also participated in the reliability study were measured three times (a third day for the second SRiT), with three days to one week between testing. The laboratory test GWPT was measured in the laboratory of the HU University of Applied Sciences (Utrecht, the Netherlands). All field tests were measured either in the gymnasium of the HU University of Applied Sciences or in a gymnasium nearby the participants home. The participants were always tested in similar conditions: in their own wheelchair, with the same tire pressure (maximum that was allowed) and on the same floor. The testing order differed between the participants, because of practical aspects like availability of the gymnasium or laboratory. Only one maximal aerobic exercise test was performed on a single day and the maximal aerobic exercise test was always the last test of that day. Between the other short duration tests, a resting period of at least five minutes was scheduled.

A standard questionnaire recorded age, gender, type of SB, lesion level, use of wheelchair and type of wheelchair. Before testing, body mass and wheelchair mass were determined by an electronic wheelchair scale (Kern MWS-300K100M, KERN & SOHN GmbH, Balingen, Germany). Arm span while seated as proxy for height was measured through the use of a non-stretchable tape (middle finger-tip to middle finger-tip) as recommended in children who are wheelchair-using, because of possible contractures in hips and knees when lying supine.[15] The body mass index (BMI, body mass divided by the square of length) was adjusted with x 0.95 for mid-lumbar lesions and x 0.90 for high lumbar/ thoracic lesions.[15]

Maximal aerobic exercise testing

Two graded exercise tests, the GWPT and the SRiT, were performed by all participants. A heart rate (HR) monitor (miniCardio, Hosand Technologies Srl, Verbania, Italia) measured HR during the tests and a calibrated mobile gas analysis system (Cortex Metamax B3, Cortex Medical GmbH, Leipzig, Germany) was used to measure cardio-respiratory responses during both the GWPT and the SRiT. The Cortex Metamax has been used in multiple studies regarding exercise testing in youth who use a wheelchair for mobility and sport and is valid and reliable for measuring gas-exchange parameters during exercise.[7, 9, 16]

Both the GWPT and the SRiT were continued until the participants stopped due to exhaustion, despite verbal encouragement from the test leader (MB), who was an experienced pediatric physical therapist.

Graded Wheelchair Propulsion Test (GWPT)

The incremental protocol of the GWPT is described in an earlier study, it is a highly valid and reliable test to assess VO_{2peak} (ICCs > 0.9) in youth with SB.[7] The participants were sitting in their own wheelchair and were secured to a wheelchair ergometer (custom-made, based on the Cateye ergociser 3600, Osaka, Japan). The participants had to maintain their self-selected comfortable wheelchair propulsion speed (between 60-120 rpm), while the resistance was increased by 0.1 torque increments every minute[7].

Shuttle Ride Test (SRiT)

The protocol of the SRiT, as described earlier by Verschuren et al. in youth with CP and by Bongers et al. in youth with OI, was used.[8, 9] The participants had to propel their wheelchair back and forth from one line to another line, 10 meters apart. Participants

were instructed to cross the lines with their front wheels and then turn 180 degrees and proceed without stopping. The starting speed was 2.0 km/h and the speed increased with 0.25 km/h every minute. This incremental pace was dictated by an auditory cue ("beep") played by a standard CD player, so the children knew when to start and in what pace they had to propel their wheelchair between the lines. The main performance outcome measure of the SRiT is the total number of achieved shuttles (ranging from 0.5 to 23 shuttles), with one shuttle corresponding to approximately one minute of wheelchair propulsion. The children had to continue until they failed to reach the line within 1.5 meter, on two consecutive beeps, despite verbal encouragements. All participants were accompanied by the test leader to help pace themselves and to encourage them to achieve maximal effort.[8, 9] Next to cardiorespiratory responses, the total number of achieved shuttles was recorded.

Exercise testing parameters – cardiorespiratory responses

Objective criteria for maximal aerobic exercise testing for ambulating children were defined as a HR_{peak} > 180/min, peak Respiratory Exchange Ratio (RER_{peak}) >0.99 or the presence of a VO2 plateau.[17] The applicability of these objective criteria for youth who use a wheelchair is unclear, so in this study, data of both the GWPT and the SRiT were included for analysis if the subjective criteria (signs of intense effort such as sweating, facial flushing, clear unwillingness to continue despite encouragement) were met.[17]

Original data from the Cortex Metamax were prepared for analyses using a 10 seconds moving average interval.[18] Absolute VO_{2peak}, peak carbon dioxide production (VCO_{2peak}), RER_{peak}, peak minute ventilation (VE_{peak}), peak tidal volume (TV_{peak}) and peak breathing frequency (BF_{peak}) were calculated as the average value over the highest 30 seconds during the GWPT and the SRiT (prior to termination of the test). HR_{peak} was defined as the highest value during the tests. VO_{2peak} was normalized (ml/kg/min) by dividing absolute VO_{2peak} by body mass. The ventilator anaerobic threshold (VAT) was determined by the ventilatory equivalents method. When results were uncertain, the V-slope method was used to verify the VAT.[19-22]

Field-based skill related fitness testing

Three field-based skill related fitness tests were used in order to estimate the construct validity of the outcome measure shuttles from the SRiT.

Anaerobic performance - Muscle Power Sprint Test (MPST)[14]

Anaerobic performance was measured by the MPST, a valid (r > 0.72 arm-cranking Wingate Anaerobic test) and reliable field test (ICCs > 0.95) in youth with SB.[14] Participants had to sprint 15 meters, marked by two lines, a total of four times. They had 10 seconds to turn and prepare for the next sprint, between every sprint. The time of a 15 meter sprint was manually recorded with a stopwatch to one hundredth of a second. Power output for each of the four sprints was calculated using the following formula: Power = (total mass x distance^2) / time^{23}, where total mass included mass of the wheelchair and the child. Mean power (MP) was defined as the average power of the four sprints and was used as the outcome measure.

Agility - 10x5 Meter Sprint Test (10x5MST) and slalom test[14]

Agility was measured by the 10x5MST and the slalom test. Both tests are valid (r=0.93) and reliable (ICCs>0.95) field tests in youth with SB.[14] For the 10x5MST, participants had to sprint and turn 10 times continuously between 2 lines that were 5 meter apart as fast as possible. During the slalom test, participants had to slalom between four cones that were 1.5 meter apart, turn at the end and sprint back as fast as possible and repeat the same procedure. Time taken to perform the tests was manually recorded with a stopwatch to one hundredth of a second and was used as the outcome measure.

Statistical analysis

Power analysis

We estimated the range of sample values given a hypothesized population Intraclass Correlation Coefficient (ICC) and sample size by a general method, prior to data collection.[24] Assuming an ICC of 0.85 a sample size of 21 was needed for the lower limit of a one-sided 95% confidence interval to exceed a value of 0.70.[25]

Criterion validity SRiT and GWPT

Criterion validity between the SRiT and GWPT was evaluated by analyzing the cardiopulmonary responses (absolute and relative VO_{2peak}, VCO_{2peak}, HR_{peak}, VE_{peak}, RER_{peak}, VT_{peak}, BF_{peak} and VO_2 at VT) using two-tailed t-tests and Pearson correlation coefficients. First, normality of the data was checked with histograms and Q-Q plots. If there was uncertainty about the normality of the data, bootstrapping with a bias corrected accelerated (BCa) confidence interval was used to confirm the results. Also, linearity of relationships between the cardiopulmonary responses of the SRiT and the GWPT was assessed with scatterplots. Weak correlations were defined as the lower bound of the 95% CI r<0.5, moderate correlations were defined as the lower bound of the 95% CI r=0.5 – 0.7, high correlations as the lower bound of the 95% CI r=0.7 – 0.9 and excellent correlations as the lower bound of the 95% CI r=0.9 – 1.0.[26]

Construct validity outcome measure shuttles of the SRiT

For construct validity, a stepwise regression was used to predict VO_{2peak} based on the number of shuttles achieved on the SRiT. Possible independent variables were personal factors (gender, age, weight, height, BMI), wheelchair features (wheelchair mass, tire pressure) and factors obtained during the SRiT (HR_{peak}, shuttles). First, linearity of relationships between VO_{2peak} and the independent variables was assessed with scatterplots and quantified with Pearson correlation coefficients. Subsequently, a weighted stepwise forward multiple regression analysis was performed to identify the independent variables that contributed to the prediction of VO_{2peak} during the SRiT. Variables were included with a p-value <0.05 and excluded with a p-value >0.1. Multicollinearity was checked for by assuring a tolerance of > 0.1.[27] To present the accurateness of the regression on the individual level, we calculated prediction intervals for all participants.

The correlations between the number of achieved shuttles of the SRiT and the anaerobic performance (Mean Power of the MPST) and agility (seconds of the 10x5MST and seconds of the slalom test) were established by Pearson correlation coefficients. Additionally, possible relations between the number of achieved shuttles during the SRiT and

Table 1. *Participants characteristics.*

	MEAN (SD) N=51*	MEAN (SD) N=33 VALIDITY**	MEAN (SD) N=28 RELIABILITY***
AGE (YEARS;MONTHS)	13.6 (3.7)	14.5 (3.1)	14.7 (3.3)
BODY MASS (KG)	48.8 (18.8)	53.5 (16.3)	51.3 (15.2)
HEIGHT (CM)	154.7 (21.3)	158.7 (16.7)	157.7 (17.2)
BODY MASS INDEX (KG/M²)	22.9 (6.5)	24.6 (7.1)	24.2 (7.4)
WEIGHT WHEELCHAIR (KG)	19.2 (6.2)	18.3 (5.5)	20.0 (5.9)
	N	**N**	**N**
GENDER (BOYS/GIRLS)	30/21	16/17	16/12
TYPE (OPEN/CLOSED)	47/4	32/1	27/1
LEVEL OF LESION[39]			
• THORACIC	11	5	7
• LUMBAR	39	28	21
• SACRAL	1	0	0
AMBULATION LEVEL[40]			
• COMMUNITY AMBULATOR	5	1	0
• HOUSEHOLD AMBULATOR	6	4	4
• THERAPEUTIC AMBULATOR	4	4	1
• NON AMBULATOR	36	24	23

KG = Kilogram | CM = Centimeter | M = Meter | N = Number | SD = Standard Deviation | * = Characteristics from all participants | ** = Characteristics from participants who were included in the validity part of this study | *** = Characteristics from participants from participants who were included in the reliability part of this study.

personal factors (age, weight, height, and BMI) and wheelchair features (wheelchair mass, tire pressure) were analyzed.

Reliability of the SRiT

Reliability was tested using the ICC Shrout and Fleiss model 2.1.A,[28, 29] after checking for normality. ICCs were defined as excellent if the lower bound of the 95% CI > 0.80, ICCs were defined as high if the lower bound of the 95% CI ranged between 0.7 – 0.8 and ICCs were defined as moderate if the lower bound of the 95% CI ranged between 0.5 – 0.7.[30]

For agreement, the measurement error was analyzed using the Standard Error of Measurement$_{agreement}$ (SEM$_{agreement}$) and the Smallest Detectable Change (SDC). The SEM$_{agreement}$ was calculated by SEM$_{agreement}$= $\sqrt{(\sigma^2_m + \sigma^2_{residual})}$, in which σ^2_m accounts for the systematic errors between both measurements and $\sigma^2_{residual}$ accounts for the random error.[28, 29] The SDC was calculated by SDC = 1.96 * √2 * SEM$_{agreement}$.[29]

In case of heteroscedasticity, checked by using a Bland-Altman plot,[31] the coefficient of variation (CV) was calculated as a measure of agreement.[32, 33] The spreadsheet calculations by Hopkins were used with log transformation of the data.[34] Data were anti-logged to obtain the 95% confidence interval of the CV.[34] A low CV presents a more reliable measurement than a high CV with CVs between 5% and 10% defined as acceptable.[35-37] For practical interpretation, we also created a Bland and Altman plot where we used the log transformed data for creating the limits of agreement.[33, 38] After anti-logging the data, the limits of agreement were placed in the Bland and Altman plot. These limits of agreement give an indication of the absolute agreement between the two measurements and can be interpreted as a true change, comparable to the SDC.[33]

RESULTS

A total of 51 children (30 boys / 21 girls) were recruited, with a mean age of 13.6 (SD 3.7) years. For the validity part, 33 participants successfully completed both the GWPT and the SRiT. For the reliability part, 28 participants completed both the SRiT and the retest. Participants' characteristics, including level of lesion (classified according to the American Spinal Injury Association guidelines[39]) and modified Hoffer classification[40] are depicted in Table I. A small number of participants were community or household ambulatory and thus self-propelling a wheelchair for long distances or sports participation. The majority were therapeutic or non-ambulatory, meaning that they were self-propelling a wheelchair during daily life. Table 2 represents an overview of the data collection for the GWPT, SRiT and retest SRiT, including information regarding missing data.
No adverse events occurred during testing.

Criterion Validity SRiT and GWPT

A total of 11 participants (mean age 11.3 years, SD 4.1; 7 boys and 4 girls) were excluded because they did not achieve maximal effort on the GWPT (n=2) or the SRiT (n=2) or on

Table 2. *Overview data collection of the GWPT and the SRiT in youth with SB who use a wheelchair for mobility and sport.*

TEST	N	COMPLETED	REASON MISSING DATA
GWPT	51	33 (65%)	6 (12%) not able to come to university 2 (4%) too heavy for child 2 (4%) shoulder pain 8 (16%) did not meet subjective criteria
SRiT1	51	41 (80%)	1 (2%) too difficult for child 9 (18%) did not meet subjective criteria
SRiT2	32	28 (88%)	4 (13%) did not meet subjective criteria

GWPT = Graded Wheelchair Propulsion Test | SRiT = Shuttle Ride Test | N = Number

Table 3. *Results of the Graded Wheelchair Propulsion Test (GWPT) and the Shuttle Ride Test(SRiT).*

	GWPT MEAN (SD)	SRiT MEAN (SD)	MEAN DIFFERENCE (95% CI)	CORRELATION COEFFICIENT (95% CI)
ABSOLUTE VO_{2PEAK} (L/MIN, N=29[$])	1.140 (0.400)	1.179 (0.302)	-0.039 (-0.121;0.042)	r=0.848 (0.699;0.926)
RELATIVE VO_{2PEAK} (ML/KG/MIN, N=29[$])	22.7 (6.9)	23.6 (6.5)	-0.9 (-2.4;0.5)	r=0.841 (0.686;0.923)
VCO_{2PEAK} (L/MIN, N=29[$])	1.399 (0.565)	1.467 (0.513)	-0.068 (-0.153;0.017)	r=0.918 (0.832;0.961)
HR_{PEAK} (BPM, N=33)	185 (16)	188 (14)	-3 (-8;1)	r=0.683 (0.423;0.839)
VE_{PEAK} (22 L/MIN, N=29[$])	53.21 (22.13)	54.31 (18.15)	-1.09 (-4.64;2.46)	r=0.911 (0.818;0.957)
RER_{PEAK} (N=29[$])	1.22 (0.17)	1.25 (0.20)	-0.03 (-0.09;0.04)	r=0.592 (0.288;0.787)
VT_{PEAK} (L, N=29[$])	1.012 (0.346)	0.965 (0.348)	0.047 (0.013;0.081)	r=0.967 (0.931;0.984)
BF_{PEAK} (N=28[#])	56.50 (16.14)	61.15 (13.25)	-4.65 (-7.37;-1.93)	r=0.904 (0.802;0.954)
VO_2 AT VT (L/MIN, N=28[#])	0.757 (0.290)	0.730 (0.262)	0.027 (-0.075;0.128)	r=0.552 (0.226;0.767)

VO_{2peak} = Peak Oxygen Uptake | VCO_{2peak} = Peak Carbon Dioxide Production | HR_{peak} = Peak Heart Rate VE_{peak} = Peak Minute Ventilation | RER_{peak} = Peak Respiratory Exchange Ratio | VT_{peak} = Peak Tidal Volume BF_{peak} = Peak Breathing Frequency | VO_2 at VT = Oxygen Uptake at Ventilatory Threshold | L/MIN = Liters Per Minute | ML/KG/MIN = Milliliter per Kilogram per Minute | BPM = Beats Per Minute | L = Liter | [$] = Missing data of respiratory gas analysis measurements in 4 participants | [#] = Missing data of respiratory gas analysis measurements in 5 participants.

both (n=7). Two participants for which the GWPT was too heavy and the SRiT was too difficult were 5 years and 9 months and 6 years old respectively.

No significant differences were found for HR_{peak}, absolute VO_{2peak}, relative VO_{2peak}, VCO_{2peak} and VE_{peak} between the GWPT and SRiT. TV_{peak} was significantly higher during the GWPT, while BF_{peak} was significantly higher during the SRiT. High correlations between the GWPT and the SRiT were found for absolute VO_{2peak}, relative VO_{2peak}, VCO_{2peak}, VE_{peak}, TV_{peak} and BF_{peak} (lower bound 95% CI r>0.7), while weak correlations were found for HR_{peak}, RER_{peak} and VO_2 at VT (lower bound 95% CI r=0.2 – 0.4) (table 3).

Construct Validity

A total of 38 participants achieved the subjective criteria for maximal effort during the first SRiT and data of these participants were used for predicting absolute VO_{2peak}.[17] Of the variables that concerned individual features (age, weight, height, BMI), wheelchair-user features (wheelchair mass, tire pressure) and outcomes obtained during the SRiT (shuttles, HR_{peak}), only age (r=0.47), weight (r=0.54), height (r=0.72) and shuttles (r=0.58) correlated significantly with absolute VO_{2peak}. Subsequently, height, weight and shuttles were used as independent variables in the weighted multiple linear regression analysis. We used a weighted regression, because of heteroscedasticity during a normal multiple linear regression. The unstandardized predicted values were divided into three groups, using "1 / variance" as the weighing factor. It was possible to predict VO_{2Peak} for 77% by height, shuttles and weight. The Standard Error of Estimates (SEE) were 0.12, 0.14 and 0.26 respectively. Looking at individual prediction intervals, we saw a mean range of 0.734 liters / minute, with the smallest range being 0.51 L/min and the largest range being 1.10 L/min. The results of the weighted regression are presented in table 4.

Regarding the construct of the outcome measure shuttles, high correlations were found with the 10x5MST and the slalom test, both measuring agility (lower bound of the 95% CI r> -0.8). The MPST, measuring anaerobic power, correlated moderately (lower bound of the 95% CI r = 0.5) with the number of shuttles. Looking at the personal variables, both height and BMI showed weak correlations with the number of shuttles (lower bound 95% CI r=0.2 and -0.2). Finally wheelchair mass (lower bound 95% CI r=-0.2) correlated weakly with the number of shuttles as well (table 5).

Reliability

The reliability of the SRiT was high, with excellent ICCs for absolute VO_{2peak}, VCO_{2peak}, TV_{peak}, VE_{peak} and the number of achieved shuttles (lower bound of 95% CI ICC > 0.9), high ICCs for BF_{peak} and relative VO_{2peak} (lower bound of 95% CI ICC = 0.8) and moderate ICCs for HR_{peak}, RER_{peak} and VO_2 at VT (lower bound of 95% CI ICC = 0.5 – 0.6).

For agreement, the SDC for the number of achieved shuttles is 1.5. The CV for absolute and relative VO_{2peak} is 6.2% (95% CI 5.0 ; 8.2) and 6.4% (95% CI 5.2 ; 8.6) respectively. The limits of agreement are for both absolute and relative VO_{2peak} + 0.20 × the mean of 1st – 2nd measurement (table 6, figures 1 and 2).

Table 4. *Regression models for predicting VO_{2peak}.*

	B	95% CI	BETA	SIG.	ADJUSTED R SQUARE
INTERCEPT	-0.960	-1.618 ; -0.302		0.005	0.509
HEIGHT	0.013	0.009 ; 0.017	0.723	0.000	
INTERCEPT	-1.068	-1.694 ; -0.442		0.001	0.566
HEIGHT	0.012	0.007 ; 0.016	0.640	0.000	
NO. OF SHUTTLES	0.025	0.004 ; 0.047	0.272	0.022	
INTERCEPT	-0.853	-1.313 ; -0.393		0.001	0.773
HEIGHT	0.004	0.000 ; 0.008	0.207	0.073	
NO. OF SHUTTLES	0.056	0.037 ; 0.075	0.607	0.000	
WEIGHT	0.012	0.008 ; 0.016	0.643	0.000	

CI = Confidence Interval | Sig. = Significance | N=38, missing data of respiratory gas analysis measurements in 3 participants.

Table 5. *Correlation of number of achieved shuttles during the SRiT with skill-related fitness tests, personal factors and wheelchair features.*

	MPST (95% CI)	10X5MST (95% CI)	SLALOM TEST (95% CI)	AGE (95% CI)	HEIGHT (95% CI)
SHUTTLES	0.73 (0.54;0.85)	-0.92 (-0.96;-0.86)	-0.89 (-0.94;-0.80)	0.11 (-0.20;0.41)	0.45 (0.16;0.66)
	WEIGHT (95% CI)	**BMI** (95% CI)	**WHEELCHAIR MASS** (95% CI)		**TIRE PRESSURE** (95% CI)
SHUTTLES	-0.12 (-0.19;0.41) (0.16;0.66)	-0.47 (-0.70;-0.19)	-0.54 (-0.72;-0.18)		-0.15 (-0.44;0.26)

BMI = Body Mass Index | MPST = Muscle Power Sprint Test
10x5MST = 10 x 5 Meter Sprint Test | 95% CI = 95% Confidence Interval

Table 6. *Outcome reliability data SRiT*

	RELIABILITY SRiT				
	SRiT 1 MEAN (SD)	**SRiT 2 MEAN** (SD)	**ICC 2.1.A** (95% CI)	**$SEM_{AGREEMENT}$**	**SDC**
HR_{PEAK} (BPM) N=27$	189 (13)	188 (15)	0.77 (0.56 – 0.90)	7	18
VCO_{2PEAK} (L/MIN) N=25#	1.436 (0.482)	1.505 (0.549)	0.97 (0.94 – 0.99)	0.083	0.232
RER_{PEAK} N=25#	1.22 (0.13)	1.21 (0.18)	0.78 (0.52 – 0.91)	0.06	0.18
BF_{PEAK} N=26@	59.50 (14.27)	57.82 (14.90)	0.92 (0.83 – 0.96)	4.195	11.629
TV_{PEAK} (l) N=27$	0.981 (0.333)	1.024 (0.364)	0.95 (0.89 – 0.98)	0.083	0.232
VE_{PEAK} (L/MIN) N=25#	53.99 (18.07)	56.95 (21.42)	0.96 (0.91 – 0.98)	9.314	25.818
VO_2 AT VT (L/MIN) N=24^	0.714 (0.257)	0.742 (0.273)	0.773 (0.55 – 0.90)	0.126	0.351
SHUTTLES N=28	14 (4)	14 (4)	0.98 (0.96 – 0.99)	0.5	1.5
	SRiT 1 MEAN (SD)	**SRiT 2 MEAN** (SD)	**ICC 2.1.A** (95% CI)	**CV** (95% CI)	
VO_{2PEAK} (L/MIN) N=25#	1.193 (0.362)	1.245 (0.387)	0.96 (0.92 – 0.98)	6.2% (5.0 – 8.2)	
VO_{2PEAK} (ML/KG/MIN) N=25#	24.1 (6.0)	25.1 (6.8)	0.93 (0.84 – 0.97)	6.4% (5.2 – 8.6)	

SRiT = Shuttle Ride Test | HR = Heart Rate | VO_{2peak} = Peak Oxygen Uptake | VCO_{2peak} = Peak Carbon Dioxide Production | RER_{peak} = Peak Respiratory Exchange Ratio | BF_{peak} = Peak Breathing Frequency | TV_{peak} = Peak Tidal Volume | VE_{peak} = Peak Minute Ventilation | VO2 at VT = Oxygen Uptake at Ventilatory Threshold ICC = Intra Class Correlation | SEMagreement = Standard Error of Measurement | SDCagreement = Smalles Detectable Change | BPM = Beats Per Minute | L/MIN = Liters Per Minute | L = Liters | ML/KG/MIN = Milliliter Per Kilogram Per Minute | CV = Coefficient of Variation | CI = Confidence Interval | $ = Missing data of respiratory gas analysis measurements in 1 participant | @ = Missing data of respiratory gas analysis measurements in 2 participants | # = Missing data of respiratory gas analysis measurements in 3 participants | ^ = Missing data of respiratory gas analysis measurements in 4 participants.

Figure 1. *Limits of Agreement for absolute VO_{2peak}*

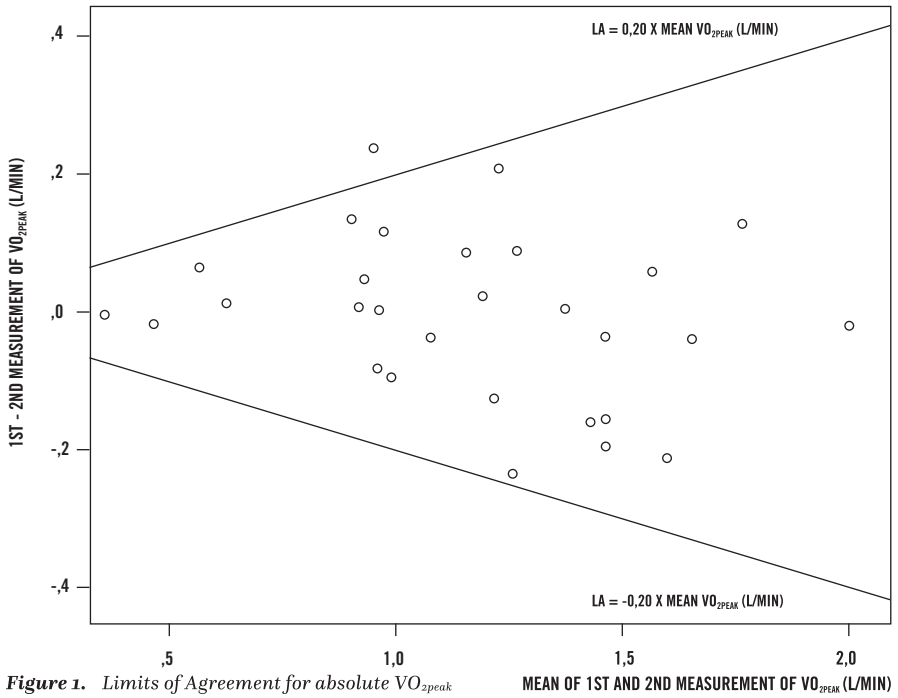

Figure 1. *Limits of Agreement for absolute VO_{2peak}*

LA = Limits of Agreement | L/MIN = Liter Per Minute

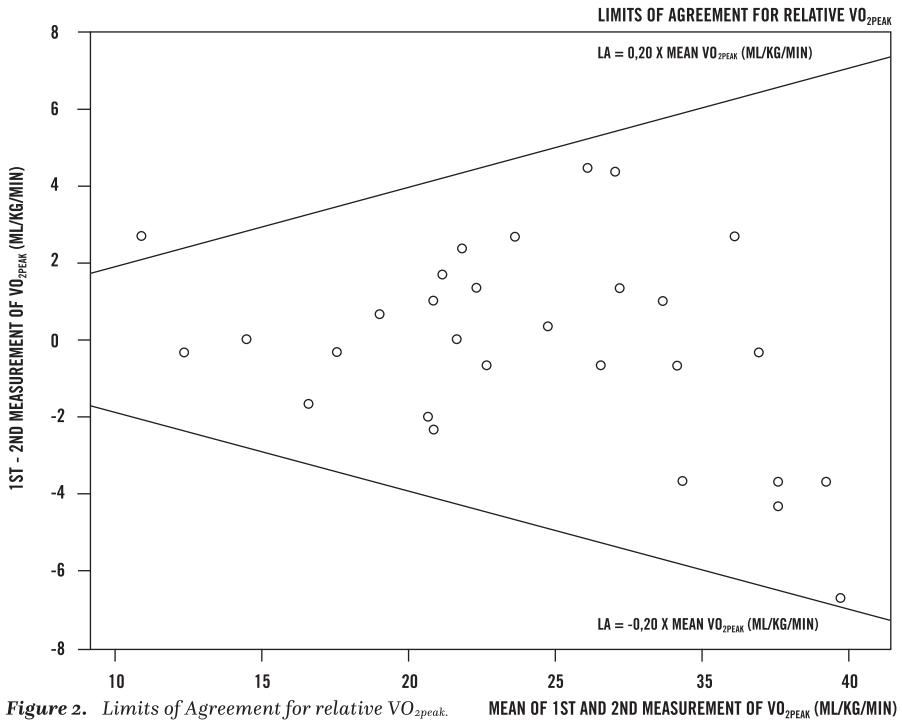

Figure 2. *Limits of Agreement for relative VO_{2peak}.*

LA = Limits of Agreement | ML/KG/MIN = Milliliter Per Kilogram Per Minute

DISCUSSION

This study focused on the validity and reliability of the SRiT in youth with SB who use a wheelchair for mobility and sport. To our knowledge, only one other study investigated the validity of the SRiT in youth who are wheelchair using so far. Verschuren et al. compared the SRiT with the graded arm exercise test (GAET) in youth with CP.[8] They found a significantly higher HR_{peak} and VE_{peak} during the SRiT and hence questioned the GAET as a gold standard to measure cardiorespiratory demands in children who are wheelchair using.[8] We therefore considered the wheelchair propulsion test (GWPT) as our gold standard to ensure specificity of testing as we found a higher HR_{peak} and VO_{2peak} during the GWPT compared to the GAET in an earlier study.[7] The differences we found in TV_{peak} and BF_{peak} between the SRiT and GWPT may be explained by the differences in test performance. During the GWPT, the child has to propel with a continuous speed and increasing load while during the SRiT, the child has to increase his or her speed. Our hypothesis is that this difference in test performance may affect the breathing pattern and thus explaining the increase of BF during the SRiT and increase of TV during the GWPT. Future research may clarify these different physiologic responses during incremental exercise testing protocols in youth who are wheelchair using.

Regarding cardiopulmonary responses of youth with SB, we observed higher HR_{peak}, RER_{peak} and VE_{peak} compared to youth with CP, with similar VO_{2peak} results.[8] When comparing our cardiopulmonary responses with youth with OI, we observed lower HR_{peak}, RER_{peak} and higher VE_{peak}, again with comparable VO_{2peak} values.[9] It would be interesting to further analyze these cardiopulmonary responses in youth with different diagnosis, to gain a deeper insight in exercise physiology of youth who use a wheelchair for mobility and sport so these findings can be interpreted more adequately. Moreover, this would also help to understand which objective criteria for maximal aerobic exercise testing should be used in this population. Until now, the applicability of the existing objective criteria for maximal aerobic exercise testing (HR_{peak} > 180/min and RER_{peak} >0.99)[17] is unclear for youth who use a wheelchair for mobility and sport because of the smaller muscle mass in the arms compared to the legs. In this study we used subjective criteria for maximal exercise testing17 to conclude if a child performed maximal at either the GWPT or the SRiT. We only included data in the analyses, if the subjective criteria were met. There were no specific characteristics regarding participants who did not meet the subjective criteria, so unfortunately we were not able to conclude in which children the SRiT cannot be used for maximal cardiorespiratory exercise testing. We also tried to use the OMNI-scale of perceived exertion,[41] unfortunately, these results were unreliable due to the cognitive impairments often present in youth with SB.

To our knowledge, no study in youth who use a wheelchair for mobility and sport tried to predict VO_{2peak} using the number of achieved shuttles during the SRiT so far. A recent meta-analysis concerning the original 20-meter Shuttle Run Test for typically developing children showed a moderate to high criterion-related validity for estimating VO_{2peak}.[42] However, Castro-Pinero et al. stated that existing equations for estimating VO_{2peak} should not be used at an individual level in typically developing children.[43]

A study in adults was only able to predict VO_{2peak} for 59% of the variance using the number of exercise stages during an incremental SRiT[44] and another study in adults concluded strong reservations about predicting VO_{2peak} in adults who are wheelchair-using.[12] Even though we were able to explain 77% of the variance, relatively large prediction intervals were present, indicating large errors when using the prediction equation at an individual level. Of course, our relatively small sample size of 38 should be taken into account, so our results should be interpreted as tentative.[45] This is why, for now, we recommend not to use this prediction equation and advise to use a mobile gas analysis system to measure VO_{2peak} in children with SB when interested in aerobic fitness.

We then tried to clarify the construct of the outcome measure shuttle, mostly used by pediatric physical therapists because they do not have the availability of a mobile gas analyses system. Unfortunately, it was not possible to explain which independent variables contribute to the number of achieved shuttles using a multiple linear regression, due to multicollinearity between the skill-related fitness tests. The moderate to high correlations between the number of achieved shuttles and both anaerobic performance and agility confirm the hypothesis generated by Verschuren et al. and Vanlandewijck et al. about the importance of anaerobic performance and agility during the SRiT.[8, 12] It underlines the importance of mastering wheelchair skills as deceleration, turning and acceleration, next to optimizing VO_{2peak}. Another interesting subject would be to analyze whether the increase in VO2 is equal during every incremental shuttle of the SRiT. This might help to analyze which part of the SRiT is explained by VO_2-uptake and which part may be explained by, for example, anaerobic performance or agility.

It was interesting to see the significant negative correlation between wheelchair mass and number of achieved shuttles, indicating the importance of light weight wheelchairs. In our previous study about skill related fitness tests in youth with SB,[14] we also found that a lighter wheelchair mass contributed to the distance traveled during one push. Literature about adults confirm the relevance of light weight wheelchairs.[46] Interestingly, some participants had relatively heavy wheelchairs, so wheelchair mass should be considered more carefully when providing a wheelchair to a child. Unfortunately, we were not able to take wheelchair features as rolling resistance, internal resistance and the wheelchair configuration into account.[47-49] These aspects are difficult to measure in this population, as all children have individually adjusted wheelchairs. Future research may take these wheelchair features into account.

The results regarding the reliability of the achieved shuttles during the SRiT are comparable with wheelchair using youth with CP and OI.[8, 9] The SDC for the number of achieved shuttles (SDC=1.5) are again similar to wheelchair using youth with CP (SDC=1.4) and slightly better than in wheelchair using youth with OI (SDC=1.9).[8, 9]

In conclusion, The SRiT is a highly valid field test for measuring VO_{2peak} in youth with SB who use a wheelchair for mobility and sport, when applying a mobile gas analysis system during testing. If one has the availability of a mobile gas analyses system, both the SRiT and the GWPT can be used to measure VO_{2peak}. It is not possible to predict VO_{2peak} accurately using the number of achieved shuttles. For pediatric physical therapists using the metric shuttles, the number of achieved shuttles represents aerobic fitness and is moderately correlated with anaerobic performance and highly correlated with agility. Because the SRiT is highly reliable and has a good SDC for the number of achieved shuttles, the SRiT can be used to monitor effectiveness of interventions to improve aerobic performance in youth with SB who use a wheelchair for mobility and sport.

ACKNOWLEDGEMENTS

We would like to thank all children, adolescents and their parents for their enthusiastic participation. Also all students who participated in this study, as part of their thesis, are being acknowledged.

FUNDING

Funded by Foundation Innovation Alliance - Regional Attention and Action for Knowledge circulation (SIA RAAK), project number 2011-13-35P and by personal PhD grant HU University of Applied Sciences Utrecht.

DECLARATION OF CONFLICTING INTERESTS

The author(s) declare that they have no competing interests.

REFERENCES

1. Bouchard C, Shephard RJ, Stephens T. (1994) Physical activity, fitness and health the model and key concepts. In: Anonymous Physical activity, fitness, and health consensus statement.
Champaign, IL: Human Kinetics.

2. Rowland JL, Fragala-Pinkham M, Miles C, O'Neil ME. The scope of pediatric physical therapy practice in health promotion and fitness for youth with disabilities.
Pediatr Phys Ther 2015;27: 2-15.

3. Bongers BC, van Brussel M, Hulzebos EHJ, Takken T. (2014) Pediatric norms for carciopulmonary exercise testing in relation to se and age.
's-Hertogenbosch: BOXPress.

4. 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.

5. De Groot JF, Takken T, Schoenmakers MA, Vanhees L, Helders PJ. Limiting factors in peak oxygen uptake and the relationship with functional ambulation in ambulating children with spina bifida.
Eur J Appl Physiol 2008;104: 657-665.

6. Buffart LM, Roebroeck ME, Rol M, Stam HJ, van den Berg-Emons RJ, Transition Research Group South-West Netherlands. Triad of physical activity, aerobic fitness and obesity in adolescents and young adults with myelomeningocele.
J Rehabil Med 2008;40: 70-75.

7. 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: 432-437.

8. Verschuren O, Zwinkels M, Ketelaar M, Reijnders-van Son F, Takken T. Reproducibility and validity of the 10-meter shuttle ride test in wheelchair-using children and adolescents with cerebral palsy.
Phys Ther 2013;93: 967-974.

9. Bongers BC, Rijks EB, Harsevoort AG, Takken T, van Brussel M. 10-m Shuttle Ride Test in Youth With Osteogenesis Imperfecta Who Use Wheelchairs: Feasibility, Reproducibility, and Physiological Responses.
Phys Ther 2016;96: 679-686.

10. Leger LA, Mercier D, Gadoury C, Lambert J. The multistage 20 metre shuttle run test for aerobic fitness.
J Sports Sci 1988;6: 93-101.

11. Goosey-Tolfrey VL, Tolfrey K. The multi-stage fitness test as a predictor of endurance fitness in wheelchair athletes.
J Sports Sci 2008;26: 511-517.

12. Vanlandewijck Y, van de Vliet P, Verellen J, Theisen D. Determinants of shuttle run performance in the prediction of peak VO2 in wheelchair users.
Disabil Rehabil 2006;28: 1259-1266.

13. Goosey-Tolfrey VL, Leicht CA. Field-based physiological testing of wheelchair athletes.
Sports Med 2013;43: 77-91.

14. Bloemen MA, Takken T, Backx FJ, Vos M, Kruitwagen CL, de Groot JF. Validity and Reliability of Skill-Related Fitness Tests for Wheelchair-Using Youth With Spina Bifida.
Arch Phys Med Rehabil 2016.

15. Dosa NP, Foley JT, Eckrich M, Woodall-Ruff D, Liptak GS. Obesity across the lifespan among persons with spina bifida.
Disabil Rehabil 2009;31: 914-920.

16. Medbo JI, Mamen A, Welde B, von Heimburg E, Stokke R. Examination of the Metamax I and II oxygen analysers during exercise studies in the laboratory.
Scand J Clin Lab Invest 2002;62: 585-598.

17. Rowland T. (1996) Developmental exercise physiology.
Champaign, IL: Human Kinetics.

18. Paap D, Takken T. Reference values for cardiopulmonary exercise testing in healthy adults: a systematic review.
Expert Rev Cardiovasc Ther 2014;12: 1439-1453.

19. Visschers NC, Hulzebos EH, van Brussel M, Takken T. Comparing four non-invasive methods to determine the ventilatory anaerobic threshold during cardiopulmonary exercise testing in children with congenital heart or lung disease.
Clin Physiol Funct Imaging 2015;35: 451-459.

20. American Thoracic Society, American College of Chest Physicians. ATS/ACCP Statement on cardiopulmonary exercise testing.
Am J Respir Crit Care Med 2003;167: 211-277.

21. Ohuchi H, Nakajima T, Kawade M, Matsuda M, Kamiya T. Measurement and validity of the ventilatory threshold in patients with congenital heart disease. Pediatr Cardiol 1996;17: 7-14.

22. Beaver WL, Wasserman K, Whipp BJ. A new method for detecting anaerobic threshold by gas exchange. J Appl Physiol (1985) 1986;60: 2020-2027.

23. 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: 108-115.

24. Shrout PE, Fleiss JL. Intraclass correlations: uses in assessing rater reliability. Psychol Bull 1979;86: 420-428.

25. 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: 1107-1117.

26. Cohen J. (1988) Statistical Power Analysis For The Behavioural Sciences, 2nd edition. Hillsdale, NJ, UK: Lawrence Erlbaum Associates.

27. Field A. (2006) Discovering Statistics Using SPSS. London, United Kingdom: SAGA Publications Ltd.

28. de Vet H, Terwee C, Mokkink L, Knol D. (2013) Measurement in Medicine. Practical Guide to Biostatistics and Epidemiology. New York, United States of America: Cambridge University Press.

29. de Vet HC, Terwee CB, Knol DL, Bouter LM. When to use agreement versus reliability measures. J Clin Epidemiol 2006;59: 1033-1039.

30. Portney LG, Watkins MP. (2000) Foundations of Clinical Research: Applications to Practice. Norwalk, CT: Appleton and Lange.

31. Bland JM, Altman DG. Statistical methods for assessing agreement between two methods of clinical measurement. Lancet 1986;1: 307-310.

32. Atkinson G, Nevill AM. Statistical methods for assessing measurement error (reliability) in variables relevant to sports medicine. Sports Med 1998;26: 217-238.

33. Stekelenburg CM, van der Wal MB, Knol DL, de Vet HC, van Zuijlen PP. Three-dimensional digital stereophotogrammetry: a reliable and valid technique for measuring scar surface area. Plast Reconstr Surg 2013;132: 204-211.

34. Hopkins WG. (2013) A new view of statistics. http://sportsci.org/resource/stats/ Accessed February, 2016.

35. Figueroa-Colon R, Hunter GR, Mayo MS, Aldridge RA, Goran MI, Weinsier RL. Reliability of treadmill measures and criteria to determine VO2max in prepubertal girls. Med Sci Sports Exerc 2000;32: 865-869.

36. Johnston KN, Jenkins SC, Stick SM. Repeatability of peak oxygen uptake in children who are healthy. Pediatr Phys Ther 2005;17: 11-17.

37. de Groot JF, Takken T, Gooskens RH, Schoenmakers MA, Wubbels M, Vanhees L, et al. Reproducibility of maximal and submaximal exercise testing in “normal ambulatory” and “community ambulatory” children and adolescents with spina bifida: which is best for the evaluation and application of exercise training? Phys Ther 2011;91: 267-276.

38. Euser AM, Dekker FW, le Cessie S. A practical approach to Bland-Altman plots and variation coefficients for log transformed variables. J Clin Epidemiol 2008;61: 978-982.

39. Maynard FM,Jr, Bracken MB, Creasey G, Ditunno JF,Jr, Donovan WH, Ducker TB, et al. International Standards for Neurological and Functional Classification of S pinal Cord Injury. American Spinal Injury Association. Spinal Cord 1997;35: 266-274.

40. Schoenmakers MA, Gulmans VA, Gooskens RH, Helders PJ. Spina bifida at the sacral level: more than minor gait disturbances. Clin Rehabil 2004;18: 178-185.

41. Robertson RJ, Goss FL, Boer NF, Peoples JA, Foreman AJ, Dabayebeh IM, et al. Children’s OMNI scale of perceived exertion: mixed gender and race validation. Med Sci Sports Exerc 2000;32: 452-458.

42. Mayorga-Vega D, Aguilar-Soto P, Viciana J. Criterion-Related Validity of the 20-M Shuttle Run Test for Estimating Cardiorespiratory Fitness: A Meta-Analysis. J Sports Sci Med 2015;14: 536-547.

43. Castro-Pinero J, Artero EG, Espana-Romero V, Ortega FB, Sjostrom M, Suni J, et al. Criterion-related validity of field-based fitness tests in youth: a systematic review. Br J Sports Med 2010;44: 934-943.

44. Vanderthommen M, Francaux M, Colinet C, Lehance C, Lhermerout C, Crielaard JM, et al. A multistage field test of wheelchair users for evaluation of fitness and prediction of peak oxygen consumption. J Rehabil Res Dev 2002;39: 685-692.

45. Babyak MA. What you see may not be what you get: a brief, nontechnical introduction to overfitting in regression-type models. Psychosom Med 2004;66: 411-421.

46. 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.

47. Mason BS, van der Woude LH, Goosey-Tolfrey VL. The ergonomics of wheelchair configuration for optimal performance in the wheelchair court sports. Sports Med 2013;43: 23-38.

48. van der Woude LH, Veeger HE, Dallmeijer AJ, Janssen TW, Rozendaal LA. Biomechanics and physiology in active manual wheelchair propulsion. Med Eng Phys 2001;23: 713-733.

49. van der Woude LH, de Groot S, Janssen TW. Manual wheelchairs: Research and innovation in rehabilitation, sports, daily life and health. Med Eng Phys 2006;28: 905-915.

4

Validity and reliability of skill-related fitness tests for wheelchair-using youth with spina bifida

Manon A. Bloemen, MSc,[1,2,3] Tim Takken, PhD,[2] Frank J. Backx, PhD,[4] Marleen Vos, MSc,[1,3] Cas L. Kruitwagen, MSc,[5] Janke F. de Groot, PhD,[1,4]

1 Research Group Lifestyle and Health, HU University of Applied Sciences Utrecht, Utrecht, The Netherlands
2 Child Development and Exercise Center, Wilhelmina Children's Hospital, University Medical Center Utrecht, The Netherlands
3 Master Program Pediatric Physical Therapy, HU University of Applied Sciences Utrecht, Utrecht, The Netherlands
4 Department of Rehabilitation, Physical Therapy Science and Sports, University Medical Center Utrecht, The Netherlands
5 Department of Biostatistics and Research Support, Julius Center, University Medical Center Utrecht, The Netherlands

Arch Phys Med Rehabil 2016 Sep 12. Epub ahead of print

ABSTRACT

Objectives
To determine content validity of the Muscle Power Sprint Test (MPST), and construct validity and reliability of the MPST, 10x5 Meter Sprint Test (10x5MST), slalom test, and One Stroke Push Test (1SPT) in wheelchair-using youth with spina bifida (SB).

Design
Clinimetric study.

Setting
Rehabilitation centers, SB outpatient services, and private practices.

Participants
A convenience sample of children and adolescents (N=53; 32 boys, 21 girls; age range, 5-19y) with SB who use a manual wheelchair. Participants were recruited through rehabilitation centers, SB outpatient services, pediatric physical therapists, and the BOSK (Association of Physically Disabled Persons and their Parents).

Interventions
Not applicable.

Main Outcome Measures
Construct validity of the MPST was determined by comparing results with the arm-cranking Wingate Anaerobic Test (WAnT) using paired t tests and Pearson correlation coefficients, while content validity was assessed using time- based criteria for anaerobic testing. Construct validity of the 10x5MST, slalom test, and 1SPT was analyzed by hypothesis testing using Pearson correlation coefficients and multiple regression. For reliability, intraclass correlation coefficients (ICCs) and smallest detectable changes (SDCs) were calculated.

Results
For the MPST, the mean ± SD exercise time of 4 sprints was 28.1±6.6 seconds. Correlations between the MPST and arm-cranking WAnT were high (r>.72, *P*<.01). Excellent correlations were found between the 10x5MSTand slalom test (r=.93, *P*<.01), while correlations between the 10x5MST or slalom test and MPST and 1SPT were moderate (r=-.56 to -.70; r=.56, *P*<.01). The variation of the 1SPTwas explained for 38% by wheelchair mass (ß=-.489) and total upper muscle strength (ß=.420). All ICCs were excellent (ICCs>.95), but the SDCs varied widely.

Conclusion
The MPST is a valid and reliable test in wheelchair-using youth with SB for measuring anaerobic performance. The 10x5MST and slalom test are valid and reliable for measuring agility. For the 1SPT, both validity and reliability are questionable.

INTRODUCTION

Assessment and optimizing physical fitness in youth with chronic conditions such as spina bifida (SB) are important goals in pediatric rehabilitation.[1] About 50% of children with SB use a wheelchair as their main mobility, and a large number of ambulatory children use a wheelchair for community mobility or sports.[2,3] While several physical fitness tests have been developed for ambulatory youth with disabilities, evidence for wheelchair-using youth is lacking.[4,5] Skill-related fitness is part of physical fitness as defined by Caspersen et al,[6] and consists of power, speed, agility, coordination, balance, and reaction time. In daily life of wheelchair-using youth, skill-related fitness is reflected in activities such as playing outside or playing wheelchair sports.[7] Since participation in outside play and sports is an essential goal in pediatric rehabilitation, assessment of skill-related fitness is important. This assessment enhances clinical reasoning and supports evaluation of training programs.

Field-based testing does not require expensive equipment, is task specific, and children use their own wheelchair, which is of great importance because it takes into account the wheelchair-user interface integration.[4,8-12] For wheelchair-using people, several field-based tests have been developed in which aspects of skill-related fitness, such as power, speed, agility, and coordination, play an important role. The Muscle Power Sprint Test (MPST), combining both power and speed, measures anaerobic performance during 15-m sprints.[5,9,11,13] Content and construct validity of the MPST have been established for children with cerebral palsy (CP).[9,11,13] Content validity is defined as "the degree to which the content of a measurement instrument is an adequate reflection of the construct to be measured."[14(p743)] Anaerobic performance contains short-term high-intensity exercise, with adenosine triphosphate, phosphocreatine, and glycogen being the dominant fuel sources.[13,15] Therefore, high-intensity exercise should be performed for a maximum of 30 seconds. In ambulatory youth with CP this results in 6 sprints, while for wheelchair-using youth with CP the total number of sprints is 3.[9,11,13] Construct validity is "the degree to which the scores of a measurement instrument are consistent with hypotheses, for instance relationships to scores of other instruments."[14(p743)] The arm-cranking Wingate Anaerobic Test (WAnT) is the criterion standard laboratory assessment for anaerobic capacity in wheelchair-using people and is thus suitable to determine the construct validity of the MPST.[15]

Agility refers to acceleration, deceleration, and turning and is reflected by the 10x5 Meter Sprint Test (10x5MST) and slalom test.[5,11,16] The One Stroke Push Test (1SPT) measures aspects of coordination (propelling technique) and also wheelchair features and physical factors (eg, strength).[10,17] No criterion standards are available for the 10x5MST, slalom test, and 1SPT. However, identifying the relationships between these different skill-related fitness tests contributes to clarification of the underlying constructs.

Reliability concerns "the degree to which the measurement is free from measurement error" and consists of both reliability and measurement error.[14(p743),18] While there is some evidence for validity and reliability of the MPST, 10x5MST, and 1SPT, evidence is lacking for wheelchair-using youth with SB. Therefore, the aims of this study were to

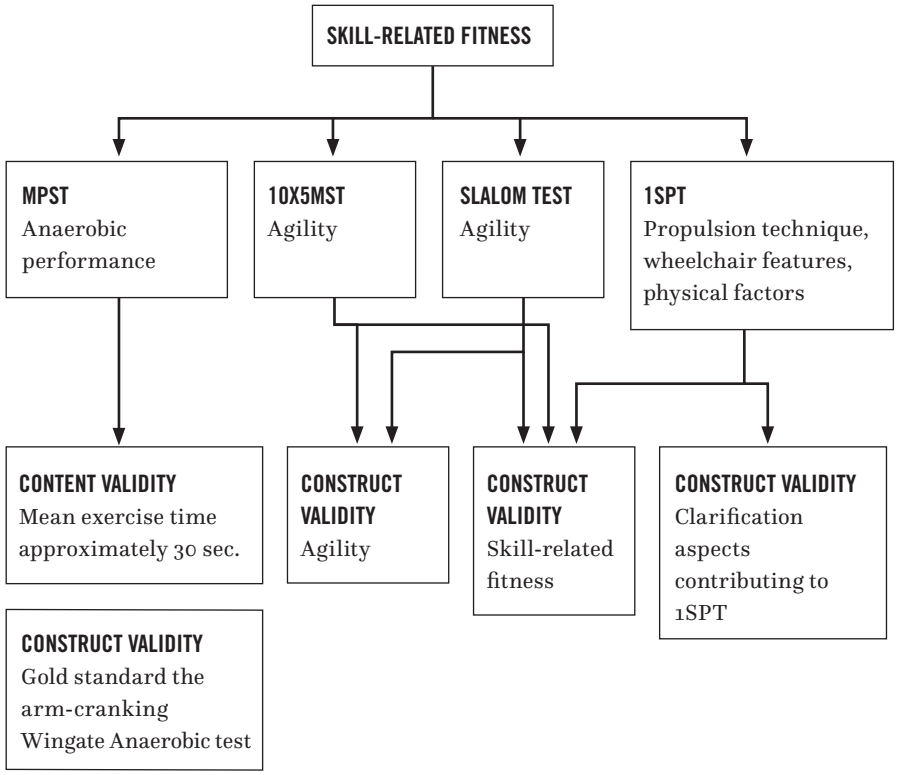

Figure 1. *Overview of testing for field-based skill-related fitness tests in wheelchair-using youth with SB.*

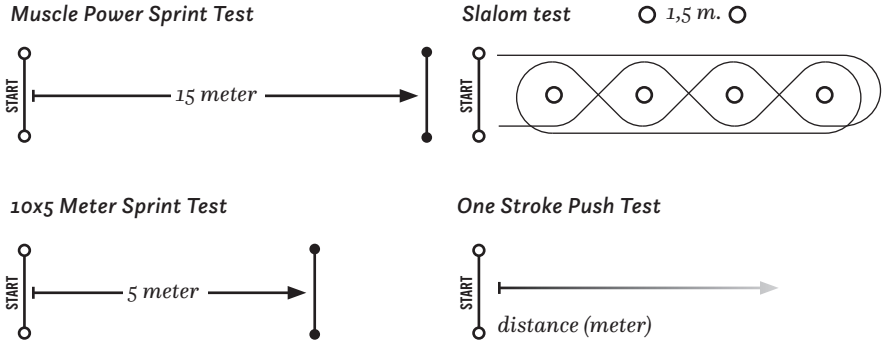

Figure 2. *Overview of the field based skill-related fitness tests.*

determine (1) the content and construct validity of the MPST; (2) the construct validity of the 10x5MST, slalom test, and 1SPT; and (3) the reliability of the MPST, 10x5MST, slalom test, and 1SPT in wheelchair-using youth with SB. Concerning content validity, we hypothesized that the total number of sprints of the original ambulatory version of the MPST (6 sprints) should be adjusted to a lower number. For construct validity, we hypothesized high correlations between the MPST and the criterion standard laboratory assessment for anaerobic power, the arm-cranking WAnT. In addition, we hypothesized high to excellent correlations between the 10x5MST and slalom test, as both tests measure agility. Moderate correlations were expected between the 10x5MST or slalom test and the MPST and 1 SPT, since they all measure different yet related aspects of skill-related fitness. Moreover, we hypothesized that wheelchair features such as wheelchair mass and physical factors such as muscle strength contribute to the 1SPT.

METHODS

The Medical Ethics Committee of the University Medical Center Utrecht, the Netherlands, approved the study procedures (no. 11- 557). Parents, and the children aged ≥12 years signed informed consent.

Participants

This study is part of the larger "*Let's Ride… Study*", focusing on fitness and physical activity in wheelchair-using youth with SB.[19] Recruitment and inclusion and exclusion criteria of the participants are described earlier in our validity and reliability study regarding aerobic fitness testing in the lab environment in wheelchair-using youth with SB.[19] Participants were recruited in the Netherlands and included if they had received a diagnosis of SB, were aged 5 to 18 years during enrollment, used a wheelchair, and were able to follow instructions.

Procedures

Figure 1 presents the clinimetric properties evaluated in this study. Participants were assessed twice (validity part) or 3 times (validity and reliability part), with 3 days to 1 week between testing moments. The tester was a pediatric physical therapist, and both the tester and the participants were unaware of previous results. Age, sex, type of SB, lesion level, use of wheelchair, and type of wheelchair were recorded through a standard questionnaire. An electronic wheelchair scalea was used to register body mass and wheelchair mass. Arm span length (middle fingertip to middle fingertip) was used as an indicator for height as recommended in wheelchair-using people, using nonstretchable tape.[20] Body mass index was calculated as body mass divided by the square of height, with an adjustment x.95 for midlumbar lesions and x.90 for high lumbar/thoracic lesions.[20]

Exercise testing

Both verbal instructions and demonstrations were provided using a standardized protocol, and included verbal encouragements throughout all tests to ensure maximal

effort. Every test started with a habituation period during which participants were familiarized with the test, with 5 minutes of resting before starting the actual measurement. *Figure* 2 presents an overview of the skill-related fitness tests.

Muscle Power Sprint Test

Participants were instructed to propel a distance of 15m marked by 2 lines as fast as possible. This was repeated 6 times. Between every sprint, participants had 10 seconds to turn and prepare. The main outcome measure was the manually recorded time per 15-m sprint (to .01s). Power output for each sprint was determined as follows: Power = Total mass (Body mass + Wheelchair mass) x Distance^2/Time. The highest power is presented as peak power (PP), while the average power over the sprints is presented as mean power (MP).[11]

Arm-cranking WAnT

We used an electromagnetically braked arm ergometer[b] to perform the arm-cranking WAnT, while participants sat in their own wheelchair that was fixated to the floor. During the first 2 minutes (warmup phase), no breaking force was applied and participants had to crank at a comfortable speed. During the last 10 seconds of the warmup, a countdown was given to allow them to maximize their pace, after which a braking force of .26Nm/kg was immediately applied, and participants had to crank as fast as possible for 30 seconds.[11] Both PP (highest mechanical power) and MP (average power over 30s) were recorded with the fully computerized Lode Ergometry Manager Software.[11,15,c]

10x5 Meter Sprint Test

Participants were instructed to sprint and turn 10 times continuously as fast as possible, between 2 lines that were 5m apart. The main outcome measure was the manually recorded time (to .01s).[11]

Slalom Test

Participants were instructed to slalom as fast as possible between 4 cones placed 1.5m apart. Participants had to turn at the end, sprint back, and repeat the same procedure once. The main outcome measure was the manually recorded time (to .01s).[16]

One Stroke Push Test

Participants had to cover as much distance as possible by using 1 push. The main outcome measure was the distance (in centimeters) measured from the starting line to the most anterior point of the front wheel furthest away. The mean distance of 3 trials was calculated.[10]

Muscle strength

Muscle strength of the upper extremities (shoulder abductors, elbow flexors and extensors, wrist dorsal flexors) was measured by the CITEC handheld dynamometer[d] using the break method according to Beenakker et al.[21] It is a reliable method for measuring muscle strength in youth with SB.[22] Total upper muscle strength was defined as the summed score of these 4 muscle groups.[21]

Statistical analysis

Before the data collection, a sample size estimation was performed. With the use of the method described by Shrout and Fleiss,[23] a sample size of 25 will, with 95% probability, result in a sample intraclass correlation coefficient (ICC) of >.75 (considered to be good) when the true ICC is as high as .85. This sample size estimation was based on the reliability part of the study. Data were analyzed for normality using quantile-quantile plots, histograms, and scatterplots.

Content and construct validity of MPST

For content validity of the MPST, the number of sprints with a mean duration time close to 30 seconds was determined. Consequently, this number of sprints was used for calculating the MP and PP. Construct validity between the MPST and the arm-cranking WAnT was evaluated by Pearson correlation coefficients and paired t tests.

Construct validity of 10x5MST, slalom test, and 1SPT

Pearson correlation coefficients were used to determine construct validity between the MPST, 10x5MST, slalom test, and 1SPT. In addition, we analyzed the contribution of wheelchair features and physical factors to the distance covered during the 1SPT. First, linearity of relationships between the 1SPT and the independent variables "tire pressure," "wheelchair mass," "wheelchair mass + body mass," "body mass," "Body Mass Index," "age," and "total muscle strength" were assessed with scatterplots. Second, univariate analyses were quantified with Pearson correlation coefficients to select a maximum of 4 independent variables in the multiple regression analyses, to ensure stability of the parameter estimates given the sample size. Subsequently, a forward stepwise multiple regression analysis was performed. Variables were included with a P value <.05 and excluded with a P value >.1.

Reliability

Reliability was analyzed by the ICC Shrout and Fleiss model 2.1.A.[18,24] The standard error of measurement agreement and the smallest detectable change (SDC) were determined for the measurement error. The standard error of measurement agreement was calculated by $\sqrt{\sigma^2_m + \sigma^2_{residual}}$, in which σ^2_m represents the systematic errors between both measurements, and $\sigma^2_{residual}$ represents the random error.[18,24] The SDC was calculated by 1.96 * √2 * standard error of measurement agreement.[24] For interpretation, both the standard errors of measurement and SDCs were calculated as percentages of mean scores.

Data interpretation

Moderate correlations were defined as r=0.5 to 0.7, high correlations as r=0.7 to 0.9, and excellent correlations as r=0.9 to 1.0.[25] High correlations ($r \geq 0.7$) were required for establishing construct validity of the MPST compared with the arm-cranking WAnT. Moderate correlations were required for establishing construct validity of the 10x5MST, slalom test, and 1SPT. ICCs of 0.7 to 0.9 were defined as good, and ICCs >.90 were defined as excellent.[25]

RESULTS

The total study population consisted of 53 participants (32 boys, 21 girls), with a mean age ± SD of 13.6 ± 3.11 years. The total number of participants was much higher than the minimum of 25 participants as estimated, because this study was part of the larger "*Let's Ride… Study*." In this larger study, all participants were assessed with several tests measuring fitness and physical activity, but only some of them participated in the reliability study of the skill-related fitness tests. Participants' age, sex, height, weight, body mass index,[20] wheelchair mass, type of lesion, level of lesion,[26] and ambulation level[27] are presented in table 1. Table 2 lists the reasons for missing data.

Table 1. *Participants characteristics (N=53)*

CHARACTERISTICS	MEAN (SD)
Age (years; months)	13;6 (3;11)
Body mass (kg)	47.9 (18.9)
Arm span length (m)	1.54. (0.22)
Body Mass Index (kg/m²)	22.6 (6.6)
Wheelchair mass (kg)	19.6 (7.0)
	N (%)
Sex (male/female)	32/21 (60/40)
Type (open/closed)	49/4 (92/8)
Level of lesion[26] • Thoracic • Lumbar • Sacral	 7 (13) 41 (77) 5 (10)
Ambulation level[27] • Community ambulatory • Household ambulatory • Therapeutic ambulatory • Non ambulator	 5 (9) 6 (11) 4 (8) 38 (72)

KG = Kilogram | M = Meter | SD = Standard Deviation

Content and construct validity of MPST

Concerning content validity, the mean ± SD exercise time for 6 sprints was 42.5 ± 10.3 seconds. The cutoff point for 30 seconds was 4 sprints, with a mean ± SD of 28.1 ± 6.6 seconds. Therefore, the calculations of MP and PP were based on 4 sprints.

For construct validity, significant high correlations were found between the arm-cranking WAnT and the MPST for both PP and MP (r>.74, P<.01). Moreover, the PP and MP were significantly lower in the MPST (mean PP, 59.2W; mean MP, 54.0W) compared with the arm-cranking WAnT (mean PP, 176.6W; mean MP, 100.8W; P<.01) (table 3).

Construct validity of 10x5MST, slalom test, and 1SPT

A significant excellent correlation (r=.93, P<.01) was found between the 10x5MST and slalom test. Significant (P<.01) moderate correlations were found between the 10x5MST and MPST (r=-.70), 10x5MST and 1SPT (r=-.56), slalom test and MPST (r=-.67), slalom test and 1SPT (r=-.60), and 1SPT and MPST (r=.56).

For explaining the variation in the 1SPT, significant (P<.01) moderate correlations between the 1SPT and wheelchair mass (r=.48) and total upper muscle strength (r=.41) were found. Relations with all other variables (tire pressure, wheelchair mass + body mass, body mass, Body Mass Index, age) showed P>.05. Subsequently, sex, wheelchair mass, and total upper muscle strength were used as independent variables in the regression analyses. Wheelchair mass (ß=-.489) and total upper muscle strength (ß=.420) explained 38% of the variation in 1SPT distance (table 4). Heteroscedasticity and multicollinearity assumptions were not violated.

Reliability of MPST, 10x5MST, slalom test, and 1SPT

Reliability of the MPST, 10x5MST, slalom test, and 1SPT was high, with ICCs >.95. The SEMs varied from 3.7% (10x5MST) to 14.5% (1SPT) of the mean, with SDCs varying from 10.1% (10x5MST) to 40.6% (1SPT) of the mean (table 5).

Table 2. *Number of participants of the skill-related fitness tests in wheelchair-using youth with SB.*

	TEST			RETEST		
	N	COMPLETED	REASON MD	N	COMPLETED	REASON MD
WANT	53	42 (79%)	5 (9%) not able to come to university 6 (11%) limitations ergometer*	n.a.	n.a.	n.a.
MPST	53	53 (100%)	n.a.	38	38 (100%)	n.a.
10X5MST	53	48 (91%)	5 (9%) too difficult	37	32 (86%)	5 (14%) too difficult
SLALOM TEST	53	51 (96%)	2 (4%) too difficult	38	34 (89%)	4 (11%) too difficult
1SPT	53	48 (91%)	1 (2%) too difficult 4 (8%) lack of space	33	28 (85%)	1 (3%) too difficult 4 (12%) lack of space

WAnT = The arm-cranking Wingate Anaerobic Test | MPST = Muscle Power Sprint Test | 10x5MST = 10 x 5 Meter Sprint Test | 1SPT = One Stroke Push TesT | N.A. = Not Applicable | MD = Missing Data | * = Ergometer proportions did not fit the participant

Table 3. *Test results (paired t-tests and Pearons correlation coefficients) of the WAnT and MPST (construct validity)*

	WAnT (N=42)		MPST (N=53)		WAnT - MPST	
	MEAN TEST (SD)	RANGE TEST	MEAN TEST (SD)	RANGE TEST	DIFF. MEAN	r
PP (W)	176.6 (90.7)	35.9–436.6	59.2 (39.1)	5.0 - 143.4	117.4*	0.74*
MP (W)	100.8 (56.6)	18.0–243.3	59.2 (39.1)	4.1 - 127.0	46.8*	0.88*

WAnT = The arm-cranking Wingate Anaerobic Test | MPST = Muscle Power Sprint Test | SD = Standard deviation | Diff. = Difference | r = Pearson Correlation Coefficient | * p< 0.01

Table 4. *Regression models for explained variance in distance covered during*

	B	95% CI	ß	SIG.	ADJUSTED R^2
CONSTANT	16.639	12.416 – 20.861		0.000	0.210
WHEELCHAIR MASS	-0.360	-0.559 – -0.161	-0.477	0.001	
CONSTANT	11.566	6.862 – 16.270		0.000	0.376
WHEELCHAIR MASS	-0.370	-0.547 – -0.161	-0.489	0.000	
UPPER MUSCLE STRENGTH	0.010	0.004 – 0.015	0.420	0.001	

1SPT = One Stroke Push Test | 95% CI = 95% Confidence Interval | SIG. = Significance

Table 5. *Outcome reliability data.*

TEST	MEAN TEST (SD) RANGE	MEAN RETEST (SD) RANGE	ICC AGREEMENT	95% CI	SEM AGREEMENT	SEM % OF MEAN	SDC	SDC % OF MEAN
MPST PP (W) N=38	59.2 (39.05) 5.0-143.4	60.6 (48.1) 4.4-156.4	0.98	0.96 - 0.99	6.8	11%	18.7	31.6%
MPST MP (W) N=38	54.0 (36.05) 4.1-127.0	55.1 (43.8) 3.5-141.2	0.98	0.97 - 0.99	5.4	10%	15.0	27.8%
10X5MST (SEC.) N=32	43.4 (8.9) 32.8-72.1	43.0 (8.4) 32.8-66.9	0.97	0.93 - 0.98	1.6	3.7%	4.4	10.1%
SLALOM TEST (SEC.) N=34	22.3 (5.7) 16.1-39.9	22.2 (5.9) 15.7-42.1	0.97	0.94 - 0.98	1.0	4.5%	2.7	12.1%
1SPT (METERS) N=28	9.6 (5.6) 1.76-26.6	9.9 (6.4) 1.78-26.44	0.95	0.95 - 0.99	1.4	14.5%	3.9	40.6%

MPST = Muscle Power Sprint Test | MP = Mean Power | PP = Peak Power | W = Watt | 10x5MST = 10x5 Meter Sprint Test | SEC= Seconds | 1SPT = One Stroke Push Test | N = Number | SD = Standard Deviation ICC = Intra Class Correlation | CI = Confidence Interval | SEM = Standard Error of Measurement | SDC = Smallest Detectable Change.

DISCUSSION

Validity

Content validity of the MPST as an outcome measure for anaerobic fitness (<30s) resulted in a total of 4 sprints as opposed to 3 sprints in wheelchair-using children with CP. Therefore, when the MPST is used for wheelchair-using youth with SB, it should be adapted to 4 sprints. High correlations between the arm-cranking WAnT and the MPST supported evidence for good construct validity of the MPST, in line with data in youth with CP. At the same time, also in line with data in youth with CP, the MPST yielded significantly lower PP and MP than did the arm-cranking WAnT.[11] These differences might be explained by the differences in performance during both tests: continuous hand cycling during the arm-cranking WAnT versus intermittent propelling during the MPST. Furthermore, 6 participants from our study were not able to perform the arm-cranking WAnT because the ergometer proportions did not fit the participants, while all participants were able to perform the MPST. Moreover, the MPST is inexpensive and easy to administer, and therefore a good field-based alternative for the lab-based arm-cranking WAnT when measuring anaerobic performance in wheelchair-using youth with SB.

For construct validity, the excellent correlation between the 10x5MST and slalom test supports the hypothesis that both tests measure agility. In addition, the moderate correlations between the 10x5MST or slalom test and 1SPT and MPST support the hypothesis that all tests measure skill-related fitness. The negative correlations we found were as expected, as higher scores on the MPST and 1SPT and lower scores on the slalom test and 10x5MST indicate better performance. Since it was hypothesized that the 1SPT measures propelling technique, wheelchair features, and physical factors, we analyzed the contribution of various variables in relation to the distance measured. Wheelchair mass (wheelchair feature) explained 21% of the variation and seemed to be most important. Subsequently, total upper muscle strength (physical factor) also seemed to play an important role. However, both variables only explained about 38% of the variation. A limitation was the inability to measure propulsion technique in biomechanical terms and the friction between the wheel and the floor; these variables appear to be important aspects contributing to the distance covered during the 1SPT.[28,29] We are, however (to our knowledge), the first to try to understand what the 1SPT truly measures in wheelchair-using youth. Future research may be able to take these biomechanical aspects into account and provide more insight into the different factors that contribute to the distance covered in 1 stroke. For now, clinicians, parents/patients, and manufacturers should realize the importance of lightweight wheelchairs, besides upper muscle strength, since this seems to affect performance in skill-related fitness tests positively and thus in daily life activities.

Reliability

We found excellent ICCs, comparable with the ICCs found in wheelchair-using youth with CP.[5,11] However, the observed SDCs varied widely. These SDCs are important for clinicians, because they provide information about the true change of an individual patient.[14] We expressed them as percentages of the mean scores found in our study because outcomes from intervention studies are lacking. SDCs ranged from acceptable for the 10x5MST and slalom test, to questionable for the MPST and relatively high for the 1SPT. For the MPST and 10x5MST, they seem to be comparable or slightly lower compared with those for wheelchair-using youth with CP.[11] However, the SDC of the 1SPT measured in this study was slightly higher compared with that for wheelchair-using youth with CP.[10] Future research should clarify the minimal clinically important change and responsiveness of all tests, to give more insight into the interpretation of the SDCs.

Study limitations

Certain limitations should be considered when interpreting the results of this study. First, no objective criteria were available to determine whether participants performed maximally during all tests. Second, the time taken to execute the MPST, 10x5MST, and slalom test was recorded manually, which can be a source of error. However, this manual recording of time is highly representative of clinical practice. In addition, test and retest were performed by the same tester, so only intrarater reliability can be interpreted. Clinics or rehabilitation centers are advised to determine the interrater reliability between therapists working at their clinic.

Conclusions

Regarding content validity, the MPST should be adapted to 4 sprints when used in wheelchair-using youth with SB. It shows good construct validity with the arm-cranking WAnT for measuring anaerobic performance. Even though reliability of the MPST is high, its clinical use is questionable because of large measurement errors. The construct validity of the 10x5MST and slalom test is good. The reliability of the 10x5MST and slalom test is high, and both tests have an acceptable measurement error. Depending on individual patient goals, clinicians can choose which test to use for measuring agility. The clinical use of the 1SPT is still questionable because the construct is unclear and measurement error seems quite large.

Suppliers

a. Kern MWS-300K100M; Kern & Sohn GmbH.
b. Lode Angio; Procare BV.
c. Lode Ergometry Manager Software; Procare BV.
d. CITEC handheld dynamometer; C.I.T. Technics-Center for Innovative Technics.

REFERENCES

1. Rowland JL, Fragala-Pinkham M, Miles C, O'Neil ME.
The scope of pediatric physical therapy practice in health promotion and fitness for youth with disabilities.
Pediatr Phys Ther 2015;27:2-15.

2. Bowman RM, McLone DG, Grant JA, Tomita T, Ito JA.
Spina bifida outcome: a 25-year prospective.
Pediatr Neurosurg 2001;34:114-20.

3. Schoenmakers MA, Uiterwaal CS, Gulmans VA, Gooskens RH, Helders PJ.
Determinants of functional independence and quality of life in children with spina bifida.
Clin Rehabil 2005;19:677-85.

4. de Groot JF, Takken T, Gooskens RH, Schoenmakers MA, Wubbels M, Vanhees L, et al.
Reproducibility of maximal and submaximal exercise testing in "normal ambulatory" and "community ambulatory" children and adolescents with spina bifida: which is best for the evaluation and application of exercise training?
Phys Ther 2011;91:267-76.

5. 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;19:108-15.

6. Caspersen CJ, Powell KE, Christenson GM.
Physical activity, exercise, and physical fitness: definitions and distinctions for health-related research.
Public Health Rep 1985;100:126-31.

7. Sonenblum SE, Sprigle S, Lopez RA.
Manual wheelchair use: bouts of mobility in everyday life.
Rehabil Res Pract 2012;2012:753165.

8. Balemans AC, Fragala-Pinkham MA, Lennon N, Thorpe D, Boyd RN, O'Neil ME, et al.
Systematic review of the clinimetric properties of laboratory- and field-based aerobic and anaerobic fitness measures in children with cerebral palsy.
Arch Phys Med Rehabil 2013;94:287-301.

9. 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;25:25-8.

10. Verschuren O, Ketelaar M, De Groot J, Vila Nova F, Takken T. Reproducibility of two functional field exercise tests for children with cerebral palsy who self-propel a manual wheelchair.
Dev Med Child Neurol 2013;55:185-90.

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

12. Goosey-Tolfrey VL, Leicht CA. Field-based physiological testing of wheelchair athletes.
Sports Med 2013;43:77-91.

13. Verschuren O, Takken T.
The muscle power sprint test.
J Physiother 2014;60:239.

14. Mokkink LB, Terwee CB, Patrick DL, Alonso J, Stratford PW, Knol DL, et al.
The COSMIN study reached international consensus on taxonomy, terminology, and definitions of measurement properties for health-related patient-reported outcomes.
J Clin Epidemiol 2010;63:737-45.

15. Bar-Or O. The Wingate Anaerobic Test: an update on methodology, reliability and validity.
Sports Med 1987;4:381-94.

16. Hutzler Y. Physical performance of elite wheelchair basketball players in armcranking ergometry and in selected wheeling tasks.
Paraplegia 1993;31:255-61.

17. May LA, Butt C, Minor L, Kolbinson K, Tulloch K.
Measurement reliability of functional tasks for persons who self-propel a manual wheelchair.
Arch Phys Med Rehabil 2003;84:578-83.

18. de Vet HC, Terwee CB, Knol DL, Bouter LM.
When to use agreement versus reliability measures.
J Clin Epidemiol 2006;59: 1033-9.

19. 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;47:432-7.

20. Dosa NP, Foley JT, Eckrich M, Woodall-Ruff D, Liptak GS.
Obesity across the lifespan among persons with spina bifida.
Disabil Rehabil 2009;31:914-20.

21. 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;11:441-6.

22. Mahony K, Hunt A, Daley D, Sims S, Adams R.
Inter-tester reliability and precision of manual muscle testing and hand-held dynamometry in lower limb muscles of children with spina bifida.
Phys Occup Ther Pediatr 2009;29:44-59.

23. Shrout PE, Fleiss JL.
Intraclass correlations: uses in assessing rater reliability.
Psychol Bull 1979;86:420-8.

24. de Vet H, Terwee C, Mokkink L, Knol D.
Measurement in medicine: practical guide to biostatistics and epidemiology.
New York: Cambridge Univ Pr; 2013.

25. Cohen J.
Statistical power analysis for the behavioural sciences.
2nd ed. Hillsdale: Lawrence Erlbaum Associates; 1988.

26. Maynard FM,Jr, Bracken MB, Creasey G, Ditunno JF,Jr, Donovan WH, Ducker TB, et al.
International Standards for Neurological and Functional Classification of Spinal Cord Injury.
American Spinal Injury Association. Spinal Cord 1997;35: 266-74.

27. Schoenmakers MA, Gulmans VA, Gooskens RH, Helders PJ.
Spina bifida at the sacral level: more than minor gait disturbances.
Clin Rehabil 2004;18:178-85.

28. de Groot S, Vegter R, Vuijk C, et al.
WHEEL-I: development of a wheelchair propulsion laboratory for rehabilitation.
J Rehabil Med 2014;46:493-503.

29. van der Woude LH, Veeger HE, Dallmeijer AJ, Janssen TW, Rozendaal LA.
Biomechanics and physiology in active manual wheelchair propulsion.
Med Eng Phys 2001;23:713-33.

5

Physical behavior in wheelchair-using youth with spina bifida: an observational study

Manon AT. Bloemen,[1] Rita HJG. vd Berg-Emons,[2] Matthijs Tuijt,[3] Carla F.J. Nooijen,[2,4] Tim Takken,[5] Frank JG. Backx,[6] Marleen Vos,[1] Janke F. de Groot[1,5]

1 Research Group Lifestyle and Health, HU University of Applied Sciences Utrecht, Utrecht, The Netherlands
2 Department of Rehabilitation Medicine, Erasmus MC University Medical Center Rotterdam, Rotterdam, the Netherlands
3 Research Group Human Movement and Adaptation, HU University of Applied Sciences Utrecht, Utrecht, The Netherlands
4 Department of Public Health, Karolinska Institutet, Stockholm, Sweden
5 Child Development and Exercise Center, Wilhelmina Children's Hospital, University Medical Center Utrecht, The Netherlands
6 Department of Rehabilitation, Physical Therapy Science and Sports, University Medical Center Utrecht, The Netherlands

Submitted

ABSTRACT

Aim

To quantify physical behavior in wheelchair-using youth with spina bifida (SB) and evaluate the intensity of activities

Method

VitaMove data of 34 and Actiheart data of 36 wheelchair-using (for daily life, long distances or sports) youth (5-18 years) with SB were collected to assess type of activity and intensity. Type of activity was presented as time spent in sedentary activities and physical activities and compared to reference data. Intensity was analyzed according to the percentage of heartrate reserve. Data of 25 participants could be used to combine type of activity and intensity.

Results

Participants spent more time in sedentary activities (94.3% per 24 hours versus 78.0% per 24 hours, p<0.00) and less time in physical activities (5.0% per 24 hours versus 12.2% per 24 hours, p<0.00) compared to typically developing peers. Physical behavior during weekend days was significantly unfavorable compared to school days; 19% met the Guidelines of Physical Activity during school days and 8% during weekend days. The intensities per activity varied extensively between participants.

Interpretation

Wheelchair-using youth shows unfavorable physical behavior, with weekend days being even more unfavorable compared to school days. The different intensities during activities indicate the importance of individually tailored assessments and interventions.

What this paper adds

- Quantification of physical behavior in wheelchair-using youth with spina bifida.
- Quantification of intensity of activities in wheelchair-using youth with spina bifida.

INTRODUCTION

Physical behavior consists of both sedentary activities and physical activities and is performed in a specific context with a certain motivation.[1] Sedentary activities are defined as sitting or lying during waking hours whereas physical activity has been defined as “any bodily movement that results in energy expenditure”.[2, 3] International recommendations somewhat vary, but agree that both limiting sedentary activities and optimizing physical activity levels are important.[4-6] Systematic reviews concluded that there is an association between increased sedentary time and unfavorable physical and psychological health outcomes in youth. For example, sedentary time was associated with unfavorable body composition, decreased fitness, higher clustered cardio metabolic risk scores, and decreased academic achievement.[4, 7] Moreover, independent of sedentary time, higher levels of physical activity are positively associated with these physical and psychological health outcomes and thus leading to greater health benefits.[8, 9] Therefore, time spent in sedentary activities and in physical activities are two independent risk factors for health outcomes in youth.

A recent study shows that an average of only 29% of typically developing Dutch 12- to 17 year olds meet the Dutch guideline of physical activity.[10] As a consequence of their reduced mobility or time spent in the wheelchair, youth with spina bifida (SB) may be even at higher risk of developing unfavorable physical behavior.[11, 11, 12] Accelerometry- based evidence in ambulatory and wheelchair-using adolescents and young adults (mean age 21 years) with SB showed decreased levels of physical activity.[13] Furthermore, in this study the small subgroup of wheelchair-using participants were found to be even less active than their ambulating peers.[13] To our knowledge, no evidence exists about objectively measured physical behavior of wheelchair-using youth with SB, even though the majority uses a wheelchair for their main mobility, for long distances, or for sports participation.

Different concepts can be considered when measuring physical behavior, such as type of activity (which can further be analyzed in terms of duration) and the physiologic response of the body to physical behavior, representing the intensity. All these dimensions require different equipment.[14] The VitaMove is an accelerometry-based activity monitor, which can distinguish sedentary activities such as lying and sitting as well as physical activities such as wheelchair propulsion and cycling.[15] Heartrate monitors can be used to measure heartrate, which can then be used to classify intensity of physical behavior, ranging from very light to maximal intensity according to the American College of Sports Medicine.[6]

Combining type of activity and intensity is interesting as it provides information at what intensity certain activities are performed. The intensity of activities might be different in clinical populations compared to typically developing peers because of the severity of the disability.[16, 17] This information is lacking for wheelchair-using youth with SB. While it could seem that wheelchair-using youth is less active as defined by time spent in certain types of activities, the intensity level could show other results. Understanding physical behavior (both type of activity and intensity) and the intensity

of different types of activities in wheelchair-using youth with SB will help us to tailor and optimize interventions specific for this population. Therefore, the aims of this study were to quantify in wheelchair-using youth with SB:

1. the physical behavior in both type of activity and intensity
2. the intensity of different types of activities

METHODS

Participants

This observational study is part of the "*Let's Ride… Study*", evaluating fitness and physical behavior in wheelchair-using youth with SB. Participants were recruited in the Netherlands and were included if they were diagnosed with SB, 5-18 years of age during enrollment, used a manual wheelchair during daily life, for long distances, or for sports participation and if they were able to follow test instructions. Participants were excluded if they had any events that might intervene with the outcomes of the testing. All parents and participants aged 12 years and older signed informed consent. The Medical Ethics Committee of the University Medical Center Utrecht approved the study procedures (number 11-557). [18-20]

Demographic and morphologic parameters

The participants visited our lab to record age, gender, type of SB, lesion level, sport activities and type of wheelchair by a standard questionnaire. Body mass was measured using an electronic wheelchair scale (Kern MWS-300K100M, KERN & SOHN GmbH, Balingen, Germany) and height was measured using a non-stretchable tape while seated using the arm span length (middle finger-tip to middle finger-tip) as recommended in wheelchair-using youth, due to the presence of contractures when lying supine.
The Body Mass Index (BMI, body mass divided by the square of height) was adjusted with x 0.95 for mid-lumbar lesions and x 0.90 for high lumbar/thoracic lesions.[21]

Equipment for measuring type of activity and intensity

The VitaMove (2M Engineering, Veldhoven, the Netherlands) was used for measuring the type, and consequently duration, of activities. The VitaMove is an ambulatory monitoring system with wireless body-fixed accelerometers (Freescale MMA7260Q, Denver, USA) which is highly valid for measuring mobility-related activities in wheelchair-using youth as well as in able-bodied people.[15, 22, 23] For non-ambulatory participants, the system consists of three recorders: one recorder on the sternum and one on each wrist. The following activities can be distinguished: lying, sitting, wheeling, handbiking and non-cyclic moving. Participants who were both walking and wheelchair-using wore two additional recorders, one on each thigh, to additionally distinguish standing, walking, running, and biking. The VitaMove signals were uploaded to a computer for kinematic analysis by the VitaScore Software (VitaScore BV, Gemert, the Netherlands) (ref Nooijen2014). Detailed descriptions of the configuration and analysis have been described elsewhere.[15, 22]

The Actiheart (CamNtech Ltd, Papworth Everard, United Kingdom) was used for measuring the intensity of physical behavior. It is a highly valid device for measuring heartrate (HR) and is easy to use in wheelchair-using children with SB.[24] The Actiheart was attached to the chest by electrocardiogram electrodes (H99SG, Kendall, Covidien, Ireland) and measured the HR every 30 seconds.

Protocol

Participants were asked to wear both devices simultaneously for two school days and one weekend day from the moment they got dressed until they went to bed, except during bathing and swimming. They also were asked to keep an activity diary so we could to correct for swimming and check for peculiarities in the data. To avoid measurement bias, we did not explain that we were measuring physical behavior beforehand and instructed the participants to continue their ordinary life.

Data analysis

A minimum duration of one day and a minimum wear time of 8 hours per day was required to be included in the analysis.[25] Data were excluded if participants were ill during recording days. Type and total duration of activities were obtained from the VitaMove. Sitting and lying were clustered and presented as sedentary activities. Walking, running, wheeling, (hand)biking and non-cyclic moving were clustered and presented as physical activities. Standing was separately analyzed. All activities were expressed as a percentage of wear time, to control for differences in total wear time between participants.

VitaMove data of the participants were compared to reference activity monitor data of 20 typically developing youths aged 8 – 20 years who had worn the VitaMove during two school days (48-hour measurement). These data were available from previous studies at the department of Rehabilitation Medicine at Erasmus University Medical Center Rotterdam.[26] For comparison between wheelchair-using youth with SB and typically developing youth, the data were expressed as a percentage of 24 hours.[26]

As a measure of intensity, the heart rate reserve (HRR) was determined from the registered HRs by the Actiheart, using the following formula:
- $HRR = (HR_{measured\ by\ actiheart} - HR_{rest})/(HR_{peak} - HR_{rest})\ x\ 100\%$[27]

Peak HR (HR_{peak}) was recorded during maximal exercise testing. For the "*Let's Ride… Study*", both the Shuttle Ride Test and the Graded Wheelchair Propulsion Test were performed; these are valid and reliable maximal exercise tests for wheelchair-using youth with SB.[18, 20] Before maximal exercise testing, HR_{rest} was measured while participants had to sit still for 10 minutes in their own wheelchair (while e.g. reading a book). If either a higher HR_{peak} or a lower HR_{rest} was measured by the Actiheart in daily life, these values were used.[28] The HRRs were classified into five different zones of activity intensity according to the American College of Sports Medicine with 0-30% HRR classified as very light , 30-40% as light , 40-60% as moderate, 60-90% as vigorous and >90% as near to maximal.[6] To control for differences in wear time, both total minutes per day and % of wear time were determined.

Statistical analyses

Histograms, QQ-plots and the Shapiro Wilk test showed that data of the Vitamove and Actiheart separately were not normally distributed. Wilcoxon Signed Rank tests showed no differences between the first and second school day, justifying the use of data when only one school day was available. When data of two school days were available, data were averaged. Differences between school days and weekend days were tested with the Wilcoxon Signed Rank test.

For type of activity, the durations were presented as median, interquartile range (IQR) and minimum and maximum. For comparing sedentary activities and physical activities between our participants and typically developing peers, differences were analyzed using linear regression correcting for gender and age.

For intensity, we presented median, interquartile range (IQR), minimum and maximum and also described how many of our participants met the physical activity guideline (at least 60 minutes per day on at least moderate intensity of which at least 30 minutes at vigorous intensity).[29]

For determining intensity of different activities, data of the VitaMove and data of the Actiheart were combined using MatLab (MatLab, R2014b, The MathWorks Inc., Natick, MA, USA). We upsampled the data of the Actiheart so the 30-second intervals of the Actiheart could be combined with the 1-second intervals of the VitaMove. The mean HRRs, standard deviation, minimum and maximum HRRs per activity were calculated. The time spent in at least moderate physical activity (>40% HRR) and the time spent in at least vigorous physical activity (>60% HRR) per type of activity was calculated.

RESULTS

A total of 53 wheelchair-using youths with SB participated in the *Let's Ride... Study*. VitaMove data of 34 participants could be used for the analysis of the type of activities and Actiheart data of 36 participants could be used for the analysis of the intensity of physical behavior. For intensity of different activities, data of 25 participants could be combined (VitaMove and Actiheart) (Table 1). Missing data were caused by not properly functioning of the devices, wear time less than 8 hours (f.e. because of irritation) or illness. Results for type of activities compared to their typically developing peers, expressed as a % of 24-hours, are presented in table 2. Wheelchair-using participants with SB spent a significantly higher amount of time in sedentary activities (94.3% per 24 hours versus 78.0% per 24 hours, p<0.00) and a significantly lower amount of time in physical activities (5.0% per 24 hours versus 12.2% per 24 hours, p<0.001) on a school day. This corresponds with approximately 72 minutes in physical activities on a school day for wheelchair-using youth with SB compared to 175 minutes for typically developing peers.

Table 1. *Characteristics of the participants.*

	VITAMOVE N = 34 MEAN (SD)	ACTEHEART N = 36 MEAN (SD)	VITAMOVE - ACTIHEART N = 25 MEAN (SD)
AGE (YEARS;MONTHS)	13.7 (3.2)	13.5 (3.6)	13.4 (3.3)
BODY MASS (KG)	52.8 (18.1)	49.7 (19.5)	53.4 (19.3)
HEIGHT (CM)	159.1 (19.5)	155.5 (21.4)	158.9 (21.2)
BODY MASS INDEX (KG/M2)	23.9 (6.3)	23.2 (7.4)	24.1 (7.2)
WHEELCHAIR MASS (KG)	19.6 (6.7)	19.1 (5.8)	19.5 (5.9)
HEARTRATE REST (BEATS PER MIN.)	na	76 (9)	na
HEARTRATE PEAK (BEATS PER MIN.)	na	189 (15)	na
HEARTRATE RESERVE (BEATS PER MIN.)	na	113 (17)	na
	N	**N**	**N**
GENDER (BOYS/GIRLS)	20/14	21/15	15/10
SPORTS (NR. OF TIMES A WEEK)	7/14/9/4	6/17/9/4	5/11/6/3
TYPE (APERTA/OCCULTA)	33/1	33/3	24/1
LEVEL OF LESION			
• THORACIC	5	6	2
• LUMBAR	29	29	23
• SACRAL	0	1	0
AMBULATION LEVEL			
• COMMUNITY AMBULATORY	2	4	2
• HOUSEHOLD AMBULATORY	3	6	3
• HERAPEUTIC AMBULATORY	3	2	1
• NON AMBULATORY	26	24	19

N = Number of Participants | SD = Standard Deviation | KG = Kilogram | CM = Centimeter | M = Meter
NA = Not Applicable

Results for type of activities compared to their typically developing peers, expressed as a % of 24-hours, are presented in table 2. Wheelchair-using participants with SB spent a significantly higher amount of time in sedentary activities (94.3% per 24 hours versus 78.0% per 24 hours, p<0.00) and a significantly lower amount of time in physical activities (5.0% per 24 hours versus 12.2% per 24 hours, p<0.001) on a school day. This corresponds with approximately 72 minutes in physical activities on a school day for wheelchair-using youth with SB compared to 175 minutes for typically developing peers.

Results for type of activities on a school day and a weekend day, expressed as a % of wear time, are presented in table 3. Wheelchair-using youth with SB spent 90% of the wear time (IQR 8%) sitting or lying during a school day compared to 96% (IQR 10%) during a weekend day (p<0.01). Furthermore, they spent significantly (p< 0.01) more time in physical activities during a school day (median 8.9 % of the wear time, IQR 7%) compared to a weekend day (median 4 % of the wear time, IQR 6%).

The results for intensity of physical behavior are presented in table 4. Overall, the intensity was significantly higher during a school day compared to a weekend day. Of all participants, 19% met the Guidelines of Physical Activity (> 60 minutes moderate to vigorous intensity of which 30 minutes > vigorous intensity) during a school day and 8% during a weekend day.[29]

The intensity of the different activities are presented in table 5. Overall, the intensity varied extensively per activity as can be seen by the broad ranges reported in table 5. An example of the strain of different activities for a wheelchair-using adolescent during a school day and weekend day is presented in figure 1 and 2. These figures illustrate the differences in type of activities and intensity during a school day and a weekend day.

Table 2. *Percentage of time spend in different types of activities on a school day, comparing wheelchair-using youth with SB to typically developing peers.*

CHARACTERISTICS	YOUTH WITH SB	YOUTH WHO IS TYPICALLY DEVELOPING	DIFFERENCE	P[3]
NUMBER OF PARTICIPANTS	34	20		
AGE (YEARS) **MEAN** (SD)	13.7 (3.2)	13.8 (2.9)	-0.1	0.939
GENDER (BOYS/GIRLS)	20 / 14	10 / 10		0.580
WEIGHT (KG) **MEAN** (SD)	52.8 (18.1)	45.7 (14.3) (n=10)	7.1	0.258
HEIGHT (CM) **MEAN** (SD)	159.1 (19.5)	158.5 (14.5) (n=10)	0.6	0.929
% PHYSICAL ACTIVITIES[1] **MEDIAN** (IQR)	5.0 (3.5)	12.2 (6.1)	-7.2	0.000
• WALKING	0.0 (0.0)	8.3 (6.2)		
• RUNNING	0.0 (0.0)	0.1 (0.2)		
• WHEELING	3.7 (2.5)	0.0 (0.0)		
• (HAND)BIKING	0.1 (2.1)	1.2 (3.0)		
• NON-CYCLING MOVEMENT	0.1 (0.2)	2.2 (1.5)		
% SEDENTARY ACTIVITIES[2] (SITTING AND LYING) **MEDIAN** (IQR)	94.3 (4.3)	78.3 (6.3)	16.3	0.000
% STANDING MEDIAN (IQR)	0.0 (0.7)	8.7 (2.4)	-8.7	

SB = Spina Bifida | SD = Standard Deviation | KG = Kilogram | CM = Centimeter | IQR = Interquartile Range

1 Physical activities are total duration of walking, running, wheeling, (hand)biking and non-cyclic moving, as a % of 24 hours.
2 Sedentary Activities are total duration of sitting and lying, as a % of 24 hours.
3 Difference in characteristics between participants with SB and typically developing children was tested with a two sample t-test (age, weight, height) and chi-square (gender). Differences in physical-active activities and sedentary activities were analyzed with regression analyses corrected for age and gender.

Table 3. *Duration of the types of activities in wheelchair-using youth with SB, separately presented for a school day and a weekend day.*

	SCHOOL DAY	WEEKEND DAY	P[4]
WEARTIME VITAMOVE[1]	13.2 (1.6)	10.9 (1.9)	0.000
PHYSICAL ACTIVITIES[2]	9 (7; 2-24)	4 (6; 0-24)	0.003
• WALKING	0 (0; 0-13)	0 (0; 0-2)	
• RUNNING	0 (0; 0-0)	0 (0; 0-0)	
• NON-CYCLIC MOVING	0 (0; 0-6)	0 (0; 0-5)	
• WHEELING	7 (5; 2-13)	3 (4; 0-16)	
• (HAND)BIKING	0 (3; 0-10)	0 (1; 0-10)	
SEDENTARY ACTIVITIES[3]	90 (7; 53-98)	96 (10; 50-100)	0.007
• SITTING	84 (11; 38-96)	85 (15; 23-98)	
• LYING	4 (6; 1-23)	6 (12; 0-73)	
STANDING	0 (1; 0-35)	0 (1; 0-28)	

Type of activities are presented as % of wear time (median and interquartile range).

1 Wear time is total wear time in hours, presented as mean (standard deviation).
2 Physical activities are total duration of walking, running, non-cyclic movement, wheeling and (hand)biking, presented as a % of wear time.
3 Sedentary activities are total duration of sitting and lying, presented as a % of wear time.
4 Differences between a school day and weekend day for wear time was tested with the paired samples t-test. Differences between a school day and weekend day for physical activities and sedentary activities were tested with the Wilcoxon Signed Rank test.

Table 4. *Intensity of physical behavior for wheelchair-youth with SB, separately presented for school days and weekend days.*

	MINUTES PER 24 HOURS MEDIAN (IQR)			% WEARTIME MEDIAN (IQR)		
	SCHOOL DAY	WEEKEND DAY	P	SCHOOL DAY	WEEKEND DAY	P
WEAR TIME	761 (117)	628 (140)	0.000			
VERY LIGHT (0-30%)	575 (180)	500 (145)	0.010	73.96 (14.37)	87.71 (25.90)	0.045
LIGHT (30-40%)	110 (57)	56 (100)	0.001	14.91 (8.29)	8.43 (15.50)	0.032
MODERATE (40-60%)	55 (55)	20 (76)	0.027	7.15 (7.83)	3.66 (9.40)	0.086
VIGOROUS (60-90%)	10 (25)	1 (7)	0.005	1.35 (3.43)	0.15 (0.90)	0.010
NEAR-MAX TO MAX (>90%)	0 (1)	0 (0)	0.001	0.03 (0.11)	0.00 (0.00)	0.002

WAnT = The arm-cranking Wingate Anaerobic Test | MPST = Muscle Power Sprint Test | 10x5MST = 10 x 5 Meter Sprint Test | 1SPT = One Stroke Push TesT | N.A. = Not Applicable | MD = Missing Data | * = Ergometer proportions did not fit the participant

Table 5. *Intensity of the different activities.*

	LYING	SITTING	STANDING	WALKING	WHEELING	(HAND) BIKING	NON-CYCLIC MOVING
%HRR[1]	22 (9;0-76)	22 (10;0-99)	36 (12; 0-86)	44 (12; 3-82)	33 (12; 0-98)	32 (9; 0-100)	29 (9; 5-71)
MINUTES >40% HRR[2]	0 (1; 0-7)	24 (27; 8-118)	19 (23; 9-37)	3 (25; 0-37)	6 (9; 0-64)	0 (7; 0-30)	0 (1; 0-19)
% OF WEAR TIME >40% HRR[2]	0 (0; 0-1)	4 (4; 1-15)	3 (3; 1-6)	1 (4; 0-6)	1 (1; 0-8)	0 (1; 0-4)	0 (0; 0-2)
MINUTES >60% HRR[2]	0 (0; 0-1)	2 (5; 0-14)	3 (9; 0-17)	1 (19; 0-23)	1 (2; 0-32)	0 (2; 0-16)	0 (0; 0-3)
% OF WEAR TIME >60% HRR2	0 (0; 0-0)	0 (1; 0-2)	0 (1; 0-3)	0 (2: 0-4)	0 (0; 0-4)	0 (0; 0-2)	0 (0; 0-0)

1. % HRR presented as mean (Standard Deviation; Minimum - Maximum)
2. Minutes / % of time >40% HRR and Minutes / % of time >60% HRR presented as median (Interquartile Range; Minimum – Maximum)

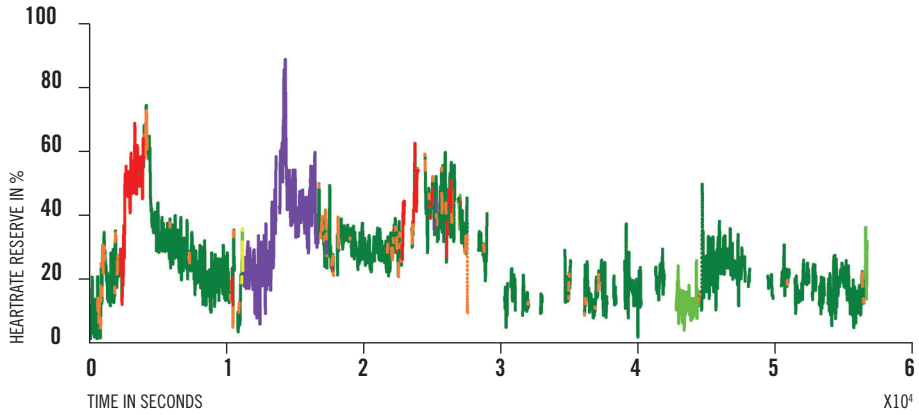

Figure 1. *The % of HRR for the several types of activity performed on a school day by a wheelchair-using adolescent with SB.*

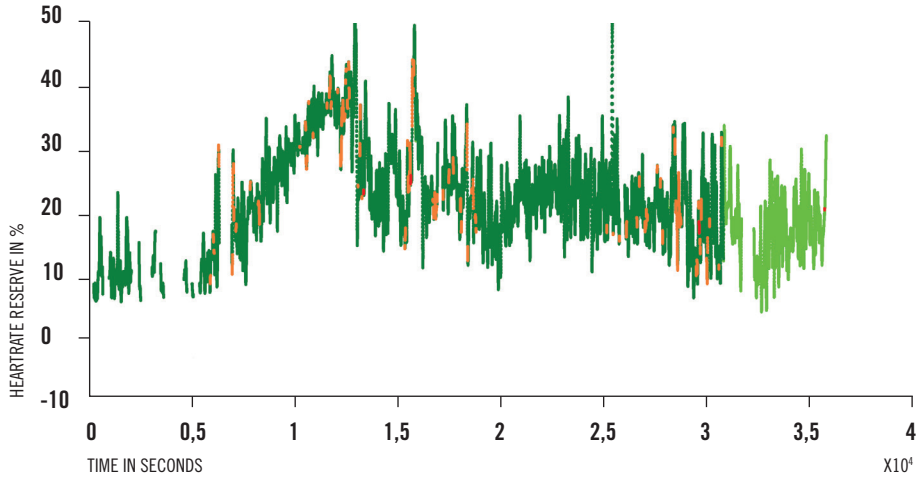

Figure 2. *The % of HRR for the several types of activity performed on a weekend day by a wheelchair-using adolescent with SB.*

DISCUSSION

This study showed results on physical behavior of wheelchair-using youth with SB combining information about types of activities and intensity. Wheelchair-using youth with SB spent more time in sedentary activities and less time in physical activities compared to typically developing peers. When comparing our results to wheelchair-using adolescents and young adults with SB, similar percentages of time in sedentary activities and physical activities were reported.[30] This may implicate that physical behavior does not change during transition into adulthood, but is already unfavorable during childhood in wheelchair-using youth with SB. Further longitudinal research may focus at the effect of age on physical behavior. When comparing our results to ambulatory youth and young adults with Cerebral Palsy (CP), these latter seem to be less sedentary and more physically active than our wheelchair-using participants with SB.[26, 31, 32]

As wheelchair-using youth with SB are only partial or not at all able to stand, this will of course result in more time spent sitting. A recent meta-analysis in adults showed that high levels of moderate physical activity intensity attenuates increased risk of death associated with high sitting time.[33] However, only 19% and 8 % of our participants met the physical activity guideline during respectively a school day and weekend day.[29] This indicates that, in general, the intensity of physical behavior of our participants was low and possibly too low to achieve health benefits. A recent study showed that about 50% of both typically developing and ambulating children with CP met the physical activity guideline.[28] Although the physical activity guideline has been described for typically developing youth and not for wheelchair-using youth, it does give us an implication of the unfavorable physical behavior of our participants. Even though our participants were wheelchair-using and diagnosed with SB, there was no medical reason why they would not be able to perform physical activities.

As evidence has shown that the activity levels during childhood track into adulthood, the challenge seems to be how to improve physical behavior during early childhood in wheelchair-using youth with SB.[34] There seems to be an opportunity during weekends, as the participants were more sedentary and less physically active on a weekend day. The participants might be fatigued after a whole school week and thus needing to rest during the weekend. It might also be, however, that there are not enough possibilities to be physically active during weekends or that there is not enough stimulation in the direct environment. Recent literature showed a variety of important facilitators and barriers when aiming to improve physical activity in youth with SB. The authors stated that we should focus on the individual possibilities and use individual facilitators for that specific child and context, so applying an individual approach and not an 'one-size fits all program'.[35, 36] Future research may give insight in possible effective interventions for improving physical behavior in wheelchair-using youth with SB.

Intensity of the activities varied extensively between the wheelchair-using participants, with for example wheeling and (hand)biking ranging from very light intensities to near to maximal intensities. This again underlines the individual approach needed when aiming to improve physical behavior in wheelchair-using youth with SB. It seems important to also measure the intensity of different activities when measuring physical behavior, besides measuring type of activity and overall intensity of physical behavior. In general, activities as wheeling and (hand)biking are activities that can be adequate in achieving higher intensities. Interestingly, the variability from very light to near to maximal intensity was also found for sitting. This might be due to the fact that HR responses during exercise are slightly delayed and thus not fully in line with the activity that is performed.[37] For example, when a participant starts wheeling, it takes some time for the HR to increase and to become in a steady state level. Similarly, if a participant stops wheeling (and thus sits according to the VitaMove), it takes time for the HR to recover and return to its resting rate.

A strength of this study was that physical behavior was measured with valid objective devices, using both the VitaMove and the Actiheart simultaneously. By doing so, we were able to measure sedentary activities and physical activities, as well as intensity. It offered the unique possibility of combining these results into intensity during several activities. Of course there are also limitations using these instruments. We missed data of about 35% of participants because of various reasons. In some cases the devices did not function properly but there were also some participants who did not want to wear these devices. Secondly, we used the HRR for the intensity, however, the variability of the HR is also related to other aspects such as emotional stress.[37] Finally, the analysis and interpretation of the data (Vitamove and Actiheart) are quite time consuming, making them not feasible for clinical practice yet. There is still a huge challenge in developing valid and reliable activity monitors that can be easily used in daily clinical practice in wheelchair-using youth. This is extremely important, so clinicians will be able to individually measure physical behavior in wheelchair-using youth with SB. This will support clinicians in developing individually tailored interventions and also evaluate these interventions.

In conclusion, wheelchair-using youth with SB are substantially more sedentary and less physically active (both in type of activity and intensity) compared to typically developing peers. Comparison between school days and weekend days showed that physical behavior on weekend days was less favorable. The intensity of the different activities varied extensively between the participants, indicating the importance of individually tailored assessments and interventions.

ACKNOWLEDGEMENTS

All children and adolescents and their parents are acknowledged for their participation in the "*Let's Ride... Study*".

Funded by Foundation Innovation Alliance - Regional Attention and Action for Knowledge circulation (SIA RAAK PRO), project number SIA RAAK PRO-4-03 and by personal PhD grant HU University of Applied Sciences Utrecht.

REFERENCES

1. Bussmann JB, van den Berg-Emons RJ.
To total amount of activity..... and beyond: perspectives on measuring physical behavior.
Front Psychol 2013;4: 463.

2. Sedentary Behaviour Research Network.
Letter to the editor: standardized use of the terms "sedentary" and "sedentary behaviours".
Appl Physiol Nutr Metab 2012;37: 540-542.

3. Caspersen CJ, Powell KE, Christenson GM.
Physical activity, exercise, and physical fitness: definitions and distinctions for health-related research.
Public Health Rep 1985;100: 126-131.

4. Tremblay MS, LeBlanc AG, Kho ME, Saunders TJ, Larouche R, Colley RC, et al.
Systematic review of sedentary behaviour and health indicators in school-aged children and youth.
Int J Behav Nutr Phys Act 2011;8: 98-5868-8-98.

5. Physical fitness and activity in schools. American Academy of Pediatrics.
Pediatrics 2000;105: 1156-1157.

6. Garber CE, Blissmer B, Deschenes MR, Franklin BA, Lamonte MJ, Lee IM, et al.
American College of Sports Medicine position stand. Quantity and quality of exercise for developing and maintaining cardiorespiratory, musculoskeletal, and neuromotor fitness in apparently healthy adults: guidance for prescribing exercise.
Med Sci Sports Exerc 2011;43: 1334-1359.

7. Carson V, Hunter S, Kuzik N, Gray CE, Poitras VJ, Chaput JP, et al.
Systematic review of sedentary behaviour and health indicators in school-aged children and youth: an update.
Appl Physiol Nutr Metab 2016;41: S240-65.

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;7: 40-5868-7-40.

9. Hallal PC, Andersen LB, Bull FC, Guthold R, Haskell W, Ekelund U, et al.
Global physical activity levels: surveillance progress, pitfalls, and prospects.
Lancet 2012;380: 247-257.

10. Burghard M, Knittel K, van Oost I, Tremblay MS, Takken T.
Is our Youth Cycling to Health? Results From the Netherlands' 2016 Report Card on Physical Activity for Children and Youth.
11, supplement 2 2016;13: S218-S224.

11. Rimmer JH, Marques AC.
Physical activity for people with disabilities.
Lancet 2012;380: 193-195.

12. Rimmer JA, Rowland JL.
Physical activity for youth with disabilities: a critical need in an underserved population.
Dev Neurorehabil 2008;11: 141-148.

13. Buffart LM, Roebroeck ME, Rol M, Stam HJ, van den Berg-Emons RJ, Transition Research Group South-West Netherlands.
Triad of physical activity, aerobic fitness and obesity in adolescents and young adults with myelomeningocele.
J Rehabil Med 2008;40: 70-75.

14. 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.

15. 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: 11-015-0004-x.

16. Brehm MA, Kempen JC, van der Kooi AJ, de Groot IJ, van den Bergen JC, Verschuuren JJ, et al.
Age-related longitudinal changes in metabolic energy expenditure during walking in boys with Duchenne muscular dystrophy.
PLoS One 2014;9: e115200.

17. De Groot JF, Takken T, Schoenmakers MA, Vanhees L, Helders PJ.
Limiting factors in peak oxygen uptake and the relationship with functional ambulation in ambulating children with spina bifida.
Eur J Appl Physiol 2008;104: 657-665.

18. 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: 432-437.

19. Bloemen MA, Takken T, Backx FJ, Vos M, Kruitwagen CL, de Groot JF.
Validity and Reliability of Skill-Related Fitness Tests for Wheelchair-Using Youth with Spina Bifida.
Arch Phys Med Rehabil 2016;.

20. Bloemen MAT, de Groot JF, Backx FJG, Benner J, Kruitwagen CLJJ, Takken T.
The Shuttle Ride Test for assessing aerobic performance in youth with spina bifida who are wheelchair-dependent; a validity and reliability study.
Physical Therapy Accepted;.

21. Dosa NP, Foley JT, Eckrich M, Woodall-Ruff D, Liptak GS.
Obesity across the lifespan among persons with spina bifida.
Disabil Rehabil 2009;31: 914-920.

22. Bussmann JB, Martens WL, Tulen JH, Schasfoort FC, van den Berg-Emons HJ, Stam HJ.
Measuring daily behavior using ambulatory accelerometry: the Activity Monitor.
Behav Res Methods Instrum Comput 2001;33: 349-356.

23. Postma K, van den Berg-Emons HJ, Bussmann JB, Sluis TA, Bergen MP, Stam HJ.
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.

24. Brage S, Brage N, Franks PW, Ekelund U, Wareham NJ.
Reliability and validity of the combined heart rate and movement sensor Actiheart.
Eur J Clin Nutr 2005;59: 561-570.

25. White DK, Wagenaar RC, Del Olmo ME, Ellis TD.
Test-retest reliability of 24 hours of activity monitoring in individuals with Parkinson's disease in home and community.
Neurorehabil Neural Repair 2007;21: 327-340.

26. Nooijen CF, Slaman J, Stam HJ, Roebroeck ME, Berg-Emons RJ, Learn2Move Research Group.
Inactive and sedentary lifestyles amongst ambulatory adolescents and young adults with cerebral palsy.
J Neuroeng Rehabil 2014;11: 49-0003-11-49.

27. Karvonen MJ, Kentala E, Mustala O.
The effects of training on heart rate; a longitudinal study.
Ann Med Exp Biol Fenn 1957;35: 307-315.

28. Balemans AC, van Wely L, Middelweerd A, van den Noort J, Becher JG, Dallmeijer AJ.
Daily stride rate activity and heart rate response in children with cerebral palsy.
J Rehabil Med 2014;46: 45-50.

29. American College of Sports Medicine.
Exercise prescription for healthy populations & special considerations. (2010)
Philadelphia: Lippincott Williams & Wilkins.

30. van den Berg-Emons HJ, Bussmann JB, Brobbel AS, Roebroeck ME, van Meeteren J, Stam HJ.
Everyday physical activity in adolescents and young adults with meningomyelocele as measured with a novel activity monitor.
J Pediatr 2001;139: 880-886.

31. Obeid J, Balemans AC, Noorduyn SG, Gorter JW, Timmons BW.
Objectively measured sedentary time in youth with cerebral palsy compared with age-, sex-, and season-matched youth who are developing typically: an explorative study.
Phys Ther 2014;94: 1163-1167.

32. Ryan JM, Forde C, Hussey JM, Gormley J.
Comparison of Patterns of Physical Activity and Sedentary Behavior Between Children With Cerebral Palsy and Children With Typical Development.
Phys Ther 2015;95: 1609-1616.

33. Ekelund U, Steene-Johannessen J, Brown WJ, Fagerland MW, Owen N, Powell KE, et al.
Does physical activity attenuate, or even eliminate, the detrimental association of sitting time with mortality? A harmonised meta-analysis of data from more than 1 million men and women.
Lancet 2016;388: 1302-1310.

34. Telama R, Yang X, Leskinen E, Kankaanpaa A, Hirvensalo M, Tammelin T, et al.
Tracking of physical activity from early childhood through youth into adulthood.
Med Sci Sports Exerc 2014;46: 955-962.

35. 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;57: 137-148.

36. 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;15: 11-015-0265-9.

37. Armstrong N, Welsman JR.
The physical activity patterns of European youth with reference to methods of assessment.
Sports Med 2006;36: 1067-1086.

6

Determinants of physical behavior in wheelchair-using youth with spina bifida

Manon AT Bloemen MSc[1,2,3], Tim Takken PhD[2], Janke F de Groot PhD[1,2], Cas LJJ Kruitwagen MSc[4], Rosanne A Rook MSc[1,3], Rita HJG. vd Berg-Emons[5], Frank JG Backx MD PhD[6]

1 Research Group Lifestyle and Health, HU University of Applied Sciences Utrecht, Utrecht, The Netherlands
2 Child Development and Exercise Center, Wilhelmina Children's Hospital, University Medical Center Utrecht, The Netherlands
3 Master Program Pediatric Physical Therapy, HU University of Applied Sciences Utrecht, Utrecht, The Netherlands
4 Department of Biostatistics and Research Support, Julius Center, University Medical Center Utrecht, The Netherlands
5 Department of Rehabilitation Medicine, Erasmus MC University Medical Center Rotterdam, Rotterdam, the Netherlands
6 Department of Rehabilitation, Physical Therapy Science and Sports, University Medical Center Utrecht, The Netherlands

Submitted

ABSTRACT

Aim

To explore associations between physical behavior and age, gender, peak oxygen uptake (VO_{2peak}) and Hoffer classification in wheelchair-using youth with spina bifida.

Method

VitaMove data of 34 and Actiheart data of 36 wheelchair-using youth with spina bifida (for daily life, long distances or sports) were used in this observational study to assess physical behavior. Time spent in sedentary activities, physical activities and Moderate to Vigorous Physical Activity (MVPA) was analyzed. The Shuttle Ride Test was used to measure VO2peak. Univariate and multivariate regression analyses were performed with physical behavior as the dependent variable. Independent variables were age, gender, VO_{2peak} and Hoffer classification.

Results

Sedentary and physical activities during a school day were influenced by both age (ß=0.326 / ß=-0.320) and Hoffer classification (ß=0.409 / ß=-0.534) and during a weekend day by Hoffer classification (ß=0.617 / ß=-0.428) alone. MVPA was influenced by Hoffer classification (ß=-0.527) during a school day and by age (ß=-0.600) during a weekend day.

Interpretation

Physical behavior is associated with age and Hoffer classification, with older age and the inability to walk influencing physical behavior negatively. Gender and VO_{2peak} seem not to be associated with physical behavior in wheelchair-using youth with spina bifida.

INTRODUCTION

Recent Lancet papers have recognized physical inactivity as a global pandemic and described the major negative health effects of physical inactivity on non-communicable diseases such as coronary heart disease, type 2 diabetes and cancer.[1-4] The authors concluded that decrease of this unhealthy behavior could improve health substantially.[2, 4, 5] A large proportion of typically developing youth does not comply with guidelines for physical behavior, that consists of both sedentary activities and physical activities and is performed in a certain context and with a specific goal.[2, 6-9] While it is already difficult for typically developing youth to develop and maintain favorable physical behavior, it is even harder for wheelchair-using youth with disabilities such as spina bifida (SB). They experience a wide variety of barriers such as lack of support from people and lack of suitable play and sport facilities.[10, 11] A recent study showed that wheelchair-using youth with SB were substantially more sedentary and less physically active than their typically developing peers.[12] These children were using their wheelchair for daily life or long distances or sports participation.[12]

Bouchard and Shephard have described a model in which relationships between physical activity, health related fitness and health are being presumed, while at the same time genetics and environmental conditions play an important role in this interaction.[13] Cardiorespiratory fitness is part of health-related fitness, with peak oxygen uptake (VO_{2peak}) as the gold standard outcome measure. Studies in typically developing youth have indeed shown low to moderate relationships (r=0.10 – r=0.45) between objectively measured physical activity and VO_{2peak}.[14] Evidence in youth with physical disabilities is unfortunately still scarce and shows conflicting findings for different clinical populations. Takken et al. analyzed the association between objectively measured physical activity and VO_{2peak} in ambulatory youth with juvenile idiopathic arthritis and found a significant moderate correlation (r=0.3, corrected for age) between physical activity and VO_{2peak}.[15] In contrast, Schoenmakers et al. found no correlation between subjectively measured physical activity and VO2peak in ambulatory children and adolescents with SB.[16]

Age and gender seem to be important personal factors that influence physical behavior in typically developing youth (4-18 years), with older age related to more time spent in sedentary activities and less time spent in physical activities, and boys being more active than girls.[5, 17, 18] A similar pattern is reported in ambulatory youth with Cerebral Palsy (CP), although the gender effect is not always found.[19-21]

In youth with CP, there is conflicting evidence regarding the association between physical behavior and the severity of the disability, as classified by the Gross Motor Function Classification System (GMFCS) ranging from GMFCS level I (walking with minor disability) to GMFCS level V (transported in a wheelchair).[22, 23] While evidence shows that a higher GMFCS level is associated with more sedentary time and lower physical activity[24], there are also studies that did not find a relation between GMFCS level and physical activity[19, 21, 24]. For children with SB, the Hoffer classification adjusted according to Schoenmakers et al. is used to classify the ambulatory status of the children ranging from normal ambulatory (Hoffer 1) to non-ambulatory (Hoffer 5) and provides information about the severity of the disability.[25]

It is important to explore and understand the presumed relationships between physical behavior and its determinants, in order to develop specific interventions for this population. To our knowledge, no evidence exists that evaluated the relationship between physical behavior and VO_{2peak}, age, gender and Hoffer classification in wheelchair-using youth with SB. So the aim of this study was to analyze the associations between physical behavior and VO_{2peak}, age, gender and Hoffer classification in wheelchair-using youth with SB.

METHODS

Participants
This study is part of the "*Let's Ride... Study*", analyzing fitness and physical behavior in wheelchair-using youth with SB.[26, 27] Participants were recruited nationwide in the Netherlands and included if they were diagnosed with SB, 5-18 years of age during enrollment, used a manual wheelchair during daily life, long distances or sports participation and if they were able to follow test instructions. Participants were excluded if they had any events that might intervene with the outcomes of the testing.[26, 27] The participants aged 12 years and over and all parents had to sign informed consent. The Medical Ethics Committee of the University Medical Center Utrecht approved the study procedures (number 11-557).

Demographic and morphologic parameters
Participants age, gender, type of SB, lesion level, sport activities, use of wheelchair, type of wheelchair and Hoffer classification[25] were registered through a standard questionnaire. The modified Hoffer classification categorized the ambulation level of people with SB in normal ambulatory (Hoffer 1), community ambulatory (Hoffer 2), house hold ambulatory (Hoffer 3), therapeutic ambulatory (Hoffer 4) and non-ambulatory (Hoffer 5).[25]

Body mass was measured using an electronic wheelchair scale (Kern MWS-300K100M, KERN & SOHN GmbH, Balingen, Germany) and height was measured using non-stretchable tape while seated using the arm span length (middle finger-tip to middle finger-tip) as recommended in wheelchair-using children, due to the presence of contractures when lying supine.[28] The body mass index (BMI, body mass divided by the square of height) was adjusted with x 0.95 for mid-lumbar lesions and x 0.90 for high lumbar/thoracic lesions.[29]

Physical Behavior equipment[12]
Physical behavior was measured using two objective monitors: the VitaMove (2M Engineering, Veldhoven, the Netherlands) and the Actiheart (CamNtech Ltd, Papworth Everard, United Kingdom) and has been described in detail elsewhere.[12] Participants had to wear both devices for two school days and one weekend day, from the moment they got dressed until they went to bed, except during bathing and swimming.

To avoid measurement bias, we explained that we wanted to know if these monitors were able to detect posture: we did not explain that we were measuring physical behavior and also provided instructions to continue their ordinary life.

In this study, the VitaMove measured the type of activities. It is a wireless monitoring system with body-fixed accelerometers (Freescale MMA7260Q, Denver, USA) and is valid for measuring mobility-related activities in wheelchair-using youth.[30] A detailed description of the configuration and analysis has been described elsewhere.[7, 12, 30, 31] The Actiheart was attached to the chest by electrocardiogram electrodes (H99SG, Kendall, Covidien, Ireland) and measures the physiological reaction of the body resulting in intensity. It is a valid device for measuring heart rate (HR) and is easy to use in wheelchair-using youth with SB.[32] A detailed description of the analysis has been given elsewhere.[12]

Physical behavior data analysis
A minimum duration of one normal day without any peculiarities such as illness and a minimum wear time of 8 hours per day was required to include the data in this study.[33] To correct for differences in wear time, all physical behavior data were calculated as % of wear time.

Regarding the type of activities, sitting and lying were clustered and presented as sedentary activities during awake hours. The activities walking, running, wheeling, (hand)biking and non-cyclic moving were clustered and presented as physical activities.

As a measure of intensity, the following formula was used to determine the percentage of Heart Rate Reserve (HRR):
$HRR = (HR_{measured\ by\ actiheart} - HR_{rest})/(HR_{peak} - HR_{rest}) \times 100\%$[34]

Heart rate peak (HR_{peak}) was recorded during the Shuttle Ride Test and the Graded Wheelchair Propulsion Test, both valid and reliable maximal exercise tests for wheelchair-using youth with SB.[26, 35] If a higher HR_{peak} was recorded in daily life by the Actiheart, this HR_{peak} was used.[36] Before maximal exercise testing, HR_{rest} was measured while participants had to sit still for 10 minutes in their own wheelchair (while e.g. watching tv or reading a book). If a lower HR_{rest} was measured in daily life by the Actiheart, this HR_{rest} was used.[36] The amount of time spent in moderate to vigorous physical activity (MVPA, >40% HRR, according to the American College of Sports Medicine) was used in the analyses.[37]

VO_{2peak}
VO_{2peak} was measured during the Shuttle Ride Test (SRiT), a highly valid and reliable maximal exercise test for assessing VO_{2peak} in wheelchair-using youth with SB.[35] Cardiorespiratory responses during the SRiT were measured by a calibrated mobile gas analysis system (Cortex Metamax B3, Cortex Medical GmbH, Leipzig, Germany) and a HR monitor (miniCardio, Hosand Technologies Srl, Verbania, Italia). Absolute VO_{2peak} was calculated as the average value over the highest 30 seconds during the SRiT and HR_{peak} was defined as the highest value during the tests. Data of the SRiT were included in the analysis if the subjective criteria for maximal exercise testing (signs of

intense effort such as sweating, facial flushing, clear unwillingness to continue despite encouragement) were met.[38]

Statistical Analyses

First, data was checked for normality using histograms, QQ-plots and the Shapiro Wilk Test. Because there were no significant differences between the first and second school day, data were averaged.[12, 33] If data of only one school day was obtained, we used this single school day. Due to significant differences between the school and weekend days, we analyzed determinants of physical behavior during a school day as well as during a weekend day.[12]

Linearity of relationships between physical behavior (time spent in sedentary activities, physical activities and MVPA) and age and VO_{2peak} were assessed with scatterplots. Thereafter, Spearman Rank Correlations were used to test the associations between physical behavior and age and VO_{2peak} separately. Mann-Whitney U tests were used to test for significant differences between boys and girls and between participants with Hoffer 1-3 and Hoffer 4-5 for physical behavior.[39] After the univariate analyses, multiple linear regressions were performed with physical behavior as the dependent variable. A separate analysis was performed for every outcome measure of physical behavior. The independent variables age, VO_{2peak}, gender, and Hoffer classification (Hoffer 1-3 versus Hoffer 4,5) were entered in the regression if the p-value $\leq$ 0.20 during univariate analysis.[19] Because we did not formulate a priori hypotheses about the order to include the independent variables, backward linear regressions were performed. Variables were excluded with a p-value > 0.1 and multicollinearity was checked for by assuring a tolerance of > 0.1. Normality and homoscedasticity of the residuals was visually checked by using histograms, QQ-plots and residual plots.

RESULTS

VitaMove data of 34 youngsters with SB were available for time spent in sedentary and physical activities and Actiheart data of 36 participants were available for the time spent in MVPA. Of these participants, 30 met the subjective criteria for maximal exercise testing during the SRiT, so only these data were included in the analyses regarding the associations between physical behavior and VO_{2peak}. The characteristics of the participants are presented in Table 1.

Spearman rank correlations for univariate analyses are presented in Table 2. We found correlations with a p-value < 0.20 between all outcome measures of physical behavior and age, except for the time spent in sedentary activities during a weekend day (p=0.251). Correlations between physical behavior and VO_{2peak} all showed p-values > 0.20.

Differences between boys and girls and between Hoffer 1-3 and Hoffer 4-5 are presented in Table 3. Gender differences with a p-value < 0.20 were found for MVPA during a school day. Hoffer 1-3 versus Hoffer 4-5 showed p-values < 0.20 for all outcome measures of physical behavior.

The independent variables with a p-value < 0.20 were used in the multiple regression analyses. Results are presented in Table 4, for school days and weekend days separately. Time spent in sedentary activities and physical activities during a school day were influenced by both age and Hoffer classification. During a weekend day, time spent in sedentary activities and physical activities was influenced by Hoffer classification alone. MVPA was influenced by Hoffer classification during a school day and by age during a weekend day. Overall, youth with SB with Hoffer 4-5 were performing worse than Hoffer 1-3 and older participants performing worse than younger participants.

Table 1. *Characteristics of the participants.*

	VITAMOVE N = 34	VITAMOVE AND MAXIMAL EXERCISE TESTING SRIT N = 30	ACTIHEART N = 36	ACTIHEART AND MAXIMAL EXERCISE TESTING SRIT N = 30
AGE (YRS;MTHS)	13.7 (3.2)	14.0 (3.0)	13.5 (3.6)	14.2 (3.1)
BODY MASS (KG)	52.8 (18.1)	53.5 (16.3)	49.7 (19.5)	51.8 (16.9)
HEIGHT (CM)	159.1 (19.5)	160.8 (18.0)	155.5 (21.4)	158.6 (18.1)
BODY MASS INDEX (KG/M2)	23.9 (6.3)	24.0 (6.4)	23.2 (7.4)	23.8 (7.6)
WHEELCHAIR MASS (KG)	19.6 (6.7)	18.8 (5.8)	19.1 (5.8)	19.4 (6.1)
PHYSICAL BEHAVIOR[1]				
• SEDENTARY ACTIVITIES SCHOOL DAY	90 (8)	91 (8)	Na	Na
• SEDENTARY ACTIVITIES WEEKEND DAY	96 (10)	96 (6)	Na	Na
• PHYSICAL ACTIVITIES SCHOOL DAY	8.9 (7)	8 (6)	Na	Na
• PHYSICAL ACTIVITIES WEEKEND DAY	4 (6)	3 (6)	Na	Na
• MVPA SCHOOL DAY	Na	Na	9 (9)	8 (8)
• MVPA WEEKEND DAY	Na	Na	4 (10)	4 (8)
VO_{2PEAK} (L/MIN)	Na	1.20 (0.36) *school day* 1.28 (0.40) *weekend day*	Na	1.19 (0.31) *school day* 1.21 (0.31) *weekend day*
	N	**N**	**N**	**N**
GENDER (BOYS/GIRLS)	20/14	18/12	21/15	*17/13*
SPORTS (NO / 1X / 2X / 3X A WEEK)	7/14/9/4	4/13/9/4	6/17/9/4	3/15/8/4
TYPE (OPEN/CLOSED)	33/1	30/0	33/3	29/1
LEVEL OF LESION				
• THORACIC	5	4	6	4
• LUMBAR	29	26	29	26
• SACRAL	0	0	1	0
AMBULATION LEVEL ACCORDING TO HOFFER				
• COMMUNITY AMBULATOR (HOFFER 2)	2	1	4	1
• HOUSEHOLD AMBULATOR (HOFFER 3)	3	2	6	5
• THERAPEUTIC AMBULATOR (HOFFER 4)	3	3	2	2
• NON AMBULATOR (HOFFER 5)	26	24	24	22

SRiT = Shuttle Ride Test | KG = Kilogram | CM = Centimeter | M = Meter | N = Number | SD = Standard Deviation
Na = Not Applicable | Age, body mass, height, body mass index, mass of the wheelchair and VO_{2peak} are presented as mean and standard deviations | [1] Physical behavior is depicted as a % of wear time. Median and interquartile ranges are presented.

Table 2. *Spearman Rank Correlations between physical behavior (sedentary activities, physical-active activities, MVPA) and age and VO_{2peak}.*

SPEARMAN RANK CORRELATIONS	SEDENTARY ACTIVITIES SCHOOL DAY	SEDENTARY ACTIVITIES WEEKEND DAY	PHYSICAL ACTIVITIES SCHOOL DAY	PHYSICAL ACTIVITIES WEEKEND DAY	MVPA SCHOOL DAY	MVPA WEEKEND DAY
AGE (YRS;MTHS)	**0.388 (p=0.028)* N=32**	0.229 (p=0.251) N=28	**-0.369 (p=0.037)* N=32**	**-0.286 (p=0.148) N=27**	**-0.311 (p=0.069) N=35**	**-0.512 (p=0.005)* N=29**
VO_{2PEAK} (L/MIN)	-0.042 (p=0.832) N=28	0.026 (p=0.872) N=23	-0.013 (p=0.946) N=28	-0.057 (p=0.795) N=23)	-0.091 (p=0.633) N=30	-0.259 (p=0.222) N=24

MVPA = Moderate to Vigorous Physical Activity | P = Significance Level | N = Number | L = Liters
Min = Minute | * = p<0.05 - **bold** = p<0.200

Table 3. *Differences between boys and girls and Hoffer 1-3 versus 4-5 for physical behavior (sedentary activities, physical activities, MVPA).*

MANN WHITNEY U TESTS	SEDENTARY ACTIVITIES SCHOOL DAY N=32	SEDENTARY ACTIVITIES WEEKEND DAY N=27	PHYSICAL ACTIVITIES SCHOOL DAY N=32	PHYSICAL ACTIVITIES WEEKEND DAY N=27	MVPA SCHOOL DAY N=35	MVPA WEEKEND DAY N=29
GENDER (BOYS/GIRLS)	91% / 88% (p=0.377)	96% / 93% (p=0.753)	8% / 12% (p=0.301)	4% / 6% (p=0.680)	**10% / 7% (p=0.158)**	4% / 5% (p=0.948)
HOFFER (1-3 / 4-5)	**67% / 92% (p=0.000)***	**86% / 96% (p=0.006)***	**18% / 8% (p=0.000)***	**7% / 3% (p=0.055)**	15% / 7% (p=0.002)*	**6% / 3% (p=0.199)**

MVPA = Moderate to Vigorous physical activity | P = Significance Level | N = Number | * = p<0.05, **bold** = p<0.200 | Sedentary Time, Dynamic-active behavior and MVPA are depicted as % of wear time. Medians are presented for boys / girls and Hoffer 1,2,3 / Hoffer 4,5.

Table 4. *Backward multiple regression analyses for physical behavior (sedentary activities, physical activities and MVPA), for school day and weekend day separately.*

	B	95% CI	SIGN	BETA	ADJUSTED R^2
SEDENTARY ACTIVITIES SCHOOL DAY (AGE AND HOFFER INCLUDED)					
CONSTANT	63.997	47.749 ; 80.246	0.000		
AGE	0.082	-0.004 ; 0.167	0.062	0.327	
HOFFER	12.197	1.908 ; 22.486	0.022	0.409	0.257
SEDENTARY ACTIVITIES WEEKEND DAY (HOFFER INCLUDED)					
CONSTANT	76.456	66.333 ; 86.579	0.000		
HOFFER	18.738	7.882 ; 29.594	0.002	0.617	0.351
PHYSICAL ACTIVITIES SCHOOL DAY (AGE AND HOFFER INCLUDED)					
CONSTANT	21.982	15.237 ; 28.726	0.000		
AGE	-0.037	-0.072 ; -0.001	0.062	-0.320	
HOFFER	-7.303	-11.574 ; -3.032	0.022	-0.534	0.391
PHYSICAL ACTIVITIES WEEKEND DAY (HOFFER INCLUDED)					
CONSTANT	12.113	5.344 ; 18.882	0.000		
HOFFER	-7.576	-14.835 ; -0.316	0.042	-0.428	0.144
MVPA SCHOOL DAY (AGE AND HOFFER INCLUDED)					
CONSTANT	17.979	13.242 ; 22.716	0.000		
HOFFER	-9.809	-15.414 ; -4.204	0.001	-0.527	0.256
MVPA WEEKEND DAY (HOFFER INCLUDED)					
CONSTANT	30.720	18.320 ; 43.120	0.000		
AGE	-0.138	-0.211 ; -0.065	0.001	-0.600	0.336

Sedentary activities, Physical activities and MVPA are % of wear time. | Age is presented in months. | Hoffer: Hoffer 1-3 is coded as 0 and Hoffer 4-5 as 1.

DISCUSSION

This study investigated associations between physical behavior and VO_{2peak}, age, gender and Hoffer classification in wheelchair-using youth with SB. Interestingly, only age and Hoffer classification influenced physical behavior in this population. However, we found different results between school days and weekend days and also between the different outcome measures of physical behavior. These results imply that there are different factors important regarding physical behavior during school days compared to weekend days but also between the type of activity and the intensity of activity. Hoffer classification seems to be the most important independent variable associated with physical behavior, with children who use a manual wheelchair during daily life (Hoffer 4-5) performing worse than children who use a manual wheelchair for sports or long distances (Hoffer1-3). Buffart et al. analyzed the associations between physical activity and Hoffer classification in adolescents and young adults (mean age 21 years) with SB.[39] They also found a significant association (beta -0.541) between physical activities and Hoffer classification, with wheelchair dependent participants (Hoffer 4-5) performing worse than the ambulatory participants.[39]

We did not find an effect of gender on physical behavior. These results are similar to the results from van Wely et al., who also found relations between physical behavior and age and GMFCS level in ambulatory children with CP, but no influence of gender on any outcome measure of physical behavior.[20] However, there are several studies in ambulatory youth with CP that reported that boys performed better than girls regarding physical behavior.[19, 21] We are not aware of evidence in youth with SB looking at the effect of gender on physical behavior. We believe that ambulatory status is a far more important determinant of physical behavior than gender.

Furthermore, we found no relationship between physical behavior and VO_{2peak} in wheelchair-using youth with SB. These results differ from evidence in typically developing youth, where low-to-moderate relationships were reported between physical behavior and $VO2_{peak}$.[14] Comparing our results with ambulatory children with SB, Schoenmakers et al. also found no correlations between subjectively measured physical activity and VO_{2peak}.[16] Buffart et al. analyzed also the associations between physical activity and VO_{2peak} in adolescents and young adults with SB (mean age 21 years) and did not find a relation between physical activities and VO_{2peak}.[39] When they separately analyzed the data of the participants classified as Hoffer 4- 5, they did find a significant association (beta 0.398) between physical activities and VO_{2peak}.[39] Secondary analyses with our data, including only Hoffer 4-5 (n=20 and n=25), still did not show any association between physical behavior and VO2peak. Our hypothesis is that a minimum level of VO_{2peak} is required in order to be physically active. All children and adolescents in our study probably have VO_{2peak} levels higher than this minimum required VO_{2peak} level, as all participants did participate in normal daily life (going to school, playing with friends, etc). Unfortunately, we do not know the exact level of VO_{2peak} that is required for wheelchair-using children, so we are not sure if this hypothesis is correct. Another aspect may be the fact that overall, the participants did not spend much time in MVPA and that the physical strain during different activities such as wheeling and (hand)

biking was rather low.[12] This may imply that the intensity of the physical behavior of wheelchair-using children with SB may be too low to have any effect on VO_{2peak}.

It seems that in wheelchair-using youth, other variables than VO_{2peak} or gender are important and influence physical behavior. Considering the explained variance, varying from 26% to 39% on a school day and 14% to 34% on a weekend day, a large percentage of the variance in physical behavior remained unexplained. There is an extensive variety of personal and environmental determinants that may be important in the amount of physical behavior, as reported in the qualitative study in youth with SB from Bloemen et al.[11] Moreover, several of these determinants will be individually based as the context differs tremendously between the individual children and adolescents.[10, 11] Future research may clarify the influence of other personal and environmental variables on physical behavior in wheelchair-using children and adolescents with SB.

Our study has several strengths and limitations. A strength is the use of objectively measured physical behavior by using valid monitors. In addition, we were able to measure VO_{2peak} directly using mobile gas analysis during a highly valid and reliable maximal incremental exercise test.[35] A limitation was that not all VO_{2peak} data could be included in the analysis as some participants did not reach the subjective criteria for maximal exercise testing. Unfortunately, we were not able to use objective criteria for maximal exercise testing, as it is still unclear if objective criteria used in ambulatory children (HR_{peak} > 180/min, peak respiratory exchange ratio >0.99 or the presence of a VO_2 plateau) are applicable for wheelchair exercise testing in youth with disabilities such as SB.[38] A second limitation was that we lost some physical behavior data due to technical problems or participants who refused to wear the physical behavior equipment. We therefore chose to first use univariate analysis to select the independent variables that were used during multiple regression analyses.[19] Finally, we used a cross sectional design, so no causal relationships could be established. Future research may be able to use longitudinal designs and also include other personal and environmental variables that potentially influence physical behavior in wheelchair-using children with SB. This will provide further insight in the different associations with physical behavior and will help to develop interventions to improve favorable physical behavior in wheelchair-using youth with SB.

In conclusion, physical behavior is associated with age and Hoffer classification in wheelchair-using youth with SB, with older age and the inability to walk influencing physical behavior negatively. Gender and VO_{2peak} were not associated with physical behavior in wheelchair-using youth with SB. Interestingly, still a large percentage of the variance in physical behavior remained unexplained, implicating that there are other important personal or environmental factors that should be explored for the improvement of physical behavior. We believe that focusing on a healthy active lifestyle should start as early as possible so it becomes routine behavior, especially in youth with SB classified as Hoffer 4 and 5. Clinicians should evaluate which personal and environmental factors might be important in an individual child concerning favorable physical behavior. Increasing cardiorespiratory fitness alone does not seem to be the proper intervention to improve physical behavior.

ACKNOWLEDGEMENTS

All children and adolescents and their parents are acknowledged for their participation in the "*Let's Ride... Study*".

Funded by Foundation Innovation Alliance - Regional Attention and Action for Knowledge circulation (SIA RAAK PRO), project number SIA RAAK PRO-4-03 and by personal PhD grant HU University of Applied Sciences Utrecht.

REFERENCES

1. Kohl HW,3rd, Craig CL, Lambert EV, Inoue S, Alkandari JR, Leetongin G, et al.
The pandemic of physical inactivity: global action for public health.
Lancet 2012;380: 294-305.

2. Hallal PC, Andersen LB, Bull FC, Guthold R, Haskell W, Ekelund U, et al.
Global physical activity levels: surveillance progress, pitfalls, and prospects.
Lancet 2012;380: 247-257.

3. Reis RS, Salvo D, Ogilvie D, Lambert EV, Goenka S, Brownson RC, et al.
Scaling up physical activity interventions worldwide: stepping up to larger and smarter approaches to get people moving.
Lancet 2016;388: 1337-1348.

4. Lee IM, Shiroma EJ, Lobelo F, Puska P, Blair SN, Katzmarzyk PT, et al.
Effect of physical inactivity on major non-communicable diseases worldwide: an analysis of burden of disease and life expectancy.
Lancet 2012;380: 219-229.

5. Bauman AE, Reis RS, Sallis JF, Wells JC, Loos RJ, Martin BW, et al.
Correlates of physical activity: why are some people physically active and others not?
Lancet 2012;380: 258-271.

6. Burghard M, Knittel K, van Oost I, Tremblay MS, Takken T.
Is our Youth Cycling to Health? Results From the Netherlands' 2016 Report Card on Physical Activity for Children and Youth.
11, supplement 2 2016;13: S218-S224.

7. Bussmann JB, Martens WL, Tulen JH, Schasfoort FC, van den Berg-Emons HJ, Stam HJ.
Measuring daily behavior using ambulatory accelerometry: the Activity Monitor.
Behav Res Methods Instrum Comput 2001;33: 349-356.

8. Van Hecke L, Loyen A, Verloigne M, van der Ploeg HP, Lakerveld J, Brug J, et al.
Variation in population levels of physical activity in European children and adolescents according to cross-European studies: a systematic literature review within DEDIPAC.
Int J Behav Nutr Phys Act 2016;13: 70-016-0396-4.

9. Verloigne M, Loyen A, Van Hecke L, Lakerveld J, Hendriksen I, De Bourdheaudhuij I, et al.
Variation in population levels of sedentary time in European children and adolescents according to cross-European studies: a systematic literature review within DEDIPAC.
Int J Behav Nutr Phys Act 2016;13: 69-016-0395-5.

10. 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;57: 137-148.

11. 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;15: 11-015-0265-9.

12. Bloemen MAT, van den Berg-Emons RHJG, Tuijt M, Nooijen CF, Takken T, Backx FJG, et al.
Physical behavior in wheelchair-using children and adolescents with spina bifida.
Submitted .

13. Bouchard C, Shephard RJ, Stephens T.
(1994) Physical activity, fitness and health the model and key concepts.
In: Anonymous Physical activity, fitness, and health consensus statement.
Champaign, IL: Human Kinetics.

14. Dencker M, Andersen LB.
Accelerometer-measured daily physical activity related to aerobic fitness in children and adolescents.
J Sports Sci 2011;29: 887-895.

15. Takken T, van der Net J, Kuis W, Helders PJ.
Physical activity and health related physical fitness in children with juvenile idiopathic arthritis.
Ann Rheum Dis 2003;62: 885-889.

16. 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.

17. Dumith SC, Gigante DP, Domingues MR, Kohl HW.
Physical activity change during adolescence: a systematic review and a pooled analysis.
Int J Epidemiol 2011;40: 685-698.

18. Stanley RM, Ridley K, Dollman J.
Correlates of children's time-specific physical activity: a review of the literature.
Int J Behav Nutr Phys Act 2012;9: 50-5868-9-50.

19. Mitchell LE, Ziviani J, Boyd RN. Characteristics associated with physical activity among independently ambulant children and adolescents with unilateral cerebral palsy.
Dev Med Child Neurol 2015;57: 167-174.

20. van Wely L, Becher JG, Balemans AC, Dallmeijer AJ.
Ambulatory activity of children with cerebral palsy: which characteristics are important?
Dev Med Child Neurol 2012;54: 436-442.

21. van Eck M, Dallmeijer AJ, Beckerman H, van den Hoven PA, Voorman JM, Becher JG.
Physical activity level and related factors in adolescents with cerebral palsy.
Pediatr Exerc Sci 2008;20: 95-106.

22. Bodkin AW, Robinson C, Perales FP.
Reliability and validity of the gross motor function classification system for cerebral palsy.
Pediatr Phys Ther 2003;15: 247-252.

23. 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.

24. Keawutan P, Bell K, Davies PS, Boyd RN.
Systematic review of the relationship between habitual physical activity and motor capacity in children with cerebral palsy.
Res Dev Disabil 2014;35: 1301-1309.

25. Schoenmakers MA, Gulmans VA, Gooskens RH, Helders PJ.
Spina bifida at the sacral level: more than minor gait disturbances.
Clin Rehabil 2004;18: 178-185.

26. 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: 432-437.

27. Bloemen MA, Takken T, Backx FJ, Vos M, Kruitwagen CL, de Groot JF.
Validity and Reliability of Skill-Related Fitness Tests for Wheelchair-Using Youth with Spina Bifida.
Arch Phys Med Rehabil (in press) 2016.

28. Dosa NP, Foley JT, Eckrich M, Woodall-Ruff D, Liptak GS.
Obesity across the lifespan among persons with spina bifida.
Disabil Rehabil 2009;31: 914-920.

29. Dosa NP, Foley JT, Eckrich M, Woodall-Ruff D, Liptak GS.
Obesity across the lifespan among persons with spina bifida.
Disabil Rehabil 2009;31: 914-920.

30. 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: 11-015-0004-x.

31. Postma K, van den Berg-Emons HJ, Bussmann JB, Sluis TA, Bergen MP, Stam HJ.
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.

32. Brage S, Brage N, Franks PW, Ekelund U, Wareham NJ.
Reliability and validity of the combined heart rate and movement sensor Actiheart.
Eur J Clin Nutr 2005;59: 561-570.

33. White DK, Wagenaar RC, Del Olmo ME, Ellis TD.
Test-retest reliability of 24 hours of activity monitoring in individuals with Parkinson's disease in home and community.
Neurorehabil Neural Repair 2007;21: 327-340.

34. Karvonen MJ, Kentale E, Mustalo O.
The effects of training on heart rate; a longitudinal study.
Ann Med Exp Biol Fenn 1957;35: 307-315.

35. Bloemen MAT, de Groot JF, Backx FJG, Benner J, Kruitwagen CLJJ, Takken T.
The Shuttle Ride Test for assessing aerobic performance in youth with spina bifida who are wheelchair-dependent; a validity and reliability study.
Physical Therapy (in press).

36. Balemans AC, van Wely L, Middelweerd A, van den Noort J, Becher JG, Dallmeijer AJ.
Daily stride rate activity and heart rate response in children with cerebral palsy.
J Rehabil Med 2014;46: 45-50.

37. Garber CE, Blissmer B, Deschenes MR, Franklin BA, Lamonte MJ, Lee IM, et al.
American College of Sports Medicine position stand. Quantity and quality of exercise for developing and maintaining cardiorespiratory, musculoskeletal, and neuromotor fitness in apparently healthy adults: guidance for prescribing exercise.
Med Sci Sports Exerc 2011;43: 1334-1359.

38. Rowland T. (1996)
Developmental exercise physiology.
Champaign, IL: Human Kinetics.

39. Buffart LM, Roebroeck ME, Rol M, Stam HJ, van den Berg-Emons RJ, Transition Research Group South-West Netherlands.
Triad of physical activity, aerobic fitness and obesity in adolescents and young adults with myelomeningocele.
J Rehabil Med 2008;40: 70-75.

7

Personal and environmental factors to consider when aiming to improve participation in physical activity in children with Spina Bifida: a qualitative study

Manon AT Bloemen[1,2], Olaf Verschuren[3], Claudia van Mechelen[1], Hanneke E Borst[1], Arina J de Leeuw[1], Marsha van der Hoef[1], Janke F de Groot[1,2]

1 Research Group Lifestyle and Health, HU University of Applied Sciences Utrecht, Utrecht, The Netherlands
2 Child Development and Exercise Center, Wilhelmina Children's Hospital, University Medical Center Utrecht, The Netherlands
3 Brain Center Rudolf Magnus and Center of Excellence for Rehabilitation Medicine, University Medical Center Utrecht and De Hoogstraat Rehabilitation, Utrecht, The Netherlands

BMC Neurol 2015 Feb 10;15:11

ABSTRACT

Background
Youth with spina bifida (SB) are less fit and active than other groups with childhood disability. While recent studies have shown benefits of exercise training, the increased fitness levels do not sustain or lead to increased levels of physical activity (PA) in these children. Therefore, it seems important to explore which factors are associated with participation in PA (or lack of) in youth with SB. The objective of this study is to describe both personal and environmental factors that are important for participation in physical activity as experienced by these children and their parents, in order to better develop intervention strategies to improve participation in PA in youth with SB.

Methods
Eleven semi-structured interviews with parents of children with SB aged 4–7 years, nine focus groups with youth with SB (n = 33, age 8–18 years) and eight focus groups with their parents (n = 31) were conducted, recorded and transcribed verbatim. Two independent researchers analyzed the data. Central themes for physical activity were constructed, using the model for Physical Activity for Persons with a Disability (PAD model) as a background scheme.

Results
Data showed that youth with SB encountered both personal and environmental factors associated with participation in PA on all levels of the PAD model. Bowel and bladder care, competence in skills, sufficient fitness, medical events and self-efficacy were important personal factors. Environmental factors that were associated with physical activity included the contact with and support from other people, the use of assistive devices for mobility and care, adequate information regarding possibilities for adapted sports and accessibility of playgrounds and sports facilities.

Conclusions
Our findings suggest that a variety of both personal and environmental factors were either positively or negatively associated with participation in PA. An individual approach, assessing possibilities rather than overcoming barriers within and surrounding the child may be a good starting point when setting up intervention programs to improve participation in PA. Therefore, assessment of both personal and environmental factors associated with physical activity should be standard care within multidisciplinary intervention programs aimed to encourage healthy active lifestyles in youth with SB.

BACKGROUND

Spina Bifida (SB) is the most frequently seen congenital deformity of the neural tube. The incidence ranges from 3–4 to 7–12.8 new cases per 10,000 births.[1] The malformation of the spinal cord and often the brain can result in both motor and sensory impairment, incontinence for bowel and bladder and cognitive impairment.[2] Due to advances in the medical approach, mortality rates have de- creased over the last years and 75%-80% of children with SB can now be expected to live to be adults.[3,4] This requires a different approach in management of these patients from childhood into adulthood, not only focusing on the pathological aspects, but also at the secondary prevention and healthy active living.[5] In optimizing health outcomes of youth with SB, like in other youth with chronic childhood conditions, physical activity (PA) is an increasingly important factor to consider. Not only be- cause of its presumed relation with fitness and health[6], but also because of increasing evidence suggesting that healthy and active children become healthy and active adults.[7]

The risk for reduced levels of activity and fitness has been confirmed in a recent study in ambulatory children with SB.[8] Additionally, in a study with adolescents and young adults with SB, 39% were classified as inactive, with 37% as extremely inactive. The average aerobic capacity was 42% lower than their typically developing peers, with obesity found in 35%.[9] Even more, youth with SB not only have low fitness and PA compared to their typically developing peers; they are also in the lowest range of fitness and PA when compared to other children and adolescents with physical disabilities.[10,11] The increased risk for components of metabolic syndrome due to the low PA has been described for youth, adolescents and young adults with SB.[12,13]

Training programs in children and adolescents with disabilities, including SB, have shown positive results in fitness.[14-17] At the same time, these studies have shown that the benefits of exercise training, e.g. the increased fitness levels do not sustain or lead to increased levels of physical activity in youth with SB, Given the benefits of PA in maintaining these gains in fitness and other health benefits, it seems important to explore which personal and environmental factors are associated with participation in PA (or lack off) in this specific population.

A recent review describes factors for PA in youth with a disability.[18] The authors concluded that most avail- able literature included several types of disabilities, making it difficult to understand whether the factors would differ between specific diagnoses.[18] Given the fact that youth with SB are even less active than other groups with childhood disability, it is important to see if there are factors that are specific for participation in PA (or lack off) in youth with SB. Shields et al. also stated that further research should not only examine negative factors or barriers but should also focus on positive factors or facilitators for PA, as these positive factors might be successful strategies to use in the development of interventions aimed at increasing participation in PA.[18] To date, no study has explored the specific factors for participation in PA in youth with SB. Knowing that parents play an important role in the lives of their children it is imperative to consider their perspectives as well on their child's potential to be physically active.[19,20] Therefore, the objective of this study is to describe both personal and environmental factors that are important for

participation in PA as experienced by both youth with SB and their parents, in order to better develop intervention strategies to improve participation in PA in this population.

METHODS

Design and data collection

This study employed a descriptive qualitative design, with a thematic analysis.[21] Social constructivism was the base for this research, as referred to by Cresswell et al. as being a "subjective meaning" of how people experience their world. In this approach questions remain broad to encourage the participants to construct their meaning, which is supported by interaction with others (the social part of social constructivism).[21,22]
A recent paper about qualitative data collection with children indicated that children are able to talk about and share their experiences and views.[23] Therefore, the data was collected using focus groups with children and adolescents 8 – 18 years of age and their parents. Separate focus groups for the children and adolescents and the parents were conducted, so that possible differences between these groups could become evident. Focus groups were chosen since the interaction during focus groups can be beneficial, as the members are encouraged to share and clarify individual and their shared ideas and opinions.[24] A number of six to ten participants is found ideal to create sufficient interaction between the participants with different views and experiences.[25,26] In this study however, the number of participants per focus group was decreased to three to six because of the frequent attention difficulties typical in youth with SB.[27,28] Because younger children (4–7 years of age) were not considered capable to specify factors associated with participation in PA, due to logistic reasons, semi-structured interviews and not focus groups were conducted with one or both parents within this group.
For this study PA consists of both PA in activities of daily life, such as (hand) biking to school or active play, and participation in (un)organized sports. It is defined as "any bodily movement, produced by skeletal muscles, that results in energy expenditure".[29]

Both the focus groups and the individual interviews were conducted in a rehabilitation center, a pediatric physical therapy institution or in the home situation. Experienced and trained interviewers with a (pediatric) physical therapy background conducted the interviews. Open- ended questions (see Appendix 1) were used to allow participants to express their feelings and opinions in their own words, where clarification could be provided when necessary. Participants were not directed towards any particular preconceived response. All focus groups and interviews were audio taped and filmed.

Prior to the focus groups and interviews general information regarding family composition, education level, ambulatory classification and PA patterns was gathered using a standardized questionnaire. The study was approved by the Internal Review Board from the HU University of Applied Sciences Utrecht.

Participants

In order to include the whole range of elementary and secondary school up to young adulthood, this study focused on children and adolescents with SB 4 – 18 years of age and their parents. A purposeful, maximum variation sampling was used so all possible factors would emerge.[30] The participants were recruited through the BOSK (Association from and by parents from children, adolescents and adults with a disability), pediatric physical therapists and several SB outpatient services in the Netherlands. In order to reduce the burden of travel and time, the groups were formed by convenience, rather than stratification by age, gender or level of PA. Youth with SB and/or their parents were included if they were 8 – 18 years of age or had children with SB aged 4–18 years. Written informed consent was signed by youth 12 years of age and older as well as by their parents prior to taking part in this research. For children < 12 years of age, only parents signed informed consent in line with Dutch law. The children, adolescents and the parents were excluded if they were insufficient in the Dutch language or if participation was not possible due to cognitive or behavioral problems.

Data analysis

All focus groups and interviews were transcribed verbatim based on the audio- and videotapes and transcriptions were checked (by CvM) independently to enhance dependability. After this step text that was determined as not relevant (such as "hmm", "aha") was deleted after consensus.[25] A thematic analysis was performed with an inductive strategy.[21] It was an iterative process in which fragments were coded, resulting in subthemes and finally themes were determined for every interview and focus group. Step one consisted of defining a text section as a PA, a positive or a negative determinant or a solution. Positive and negative determinants were aspects that were already present, whereas a solution was defined as an aspect that was not yet experienced in real life by the participants of that focus group or interview. During step two, the text was classified as a personal or an environmental determinant using the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY). The third step specified the detailed description of the PA, positive or negative determinant or solution.

The analyses for each focus group/interview were performed by two independent researchers with varying experiences in working with children and adolescents with SB (0 – 15 years). Consensus was reached after every step. In case of no consensus, a third researcher was consulted who had extensive experience in research in children and adolescents with SB. After analyzing all focus groups and interviews separately, central themes were constructed by two independent researchers (CvM, MB). The solutions from the separate focus groups and interviews were compared to the positive determinants; if a solution was already mentioned as a positive determinant in another focus group or interview, it was specified as a positive determinant theme. After construction of these central themes, they were discussed with the third re- searcher (JdG) and several experts working in the field of pediatric medicine. Member checking was performed by presenting the central themes to a different group of parents of children and adolescents with SB, asking if they agreed with the results and if there were any missing determinants. The final step consisted of categorizing the central themes in modifiable determinants, partly modifiable determinants and non-modifiable determinants by the two independent researchers (CvM, MB).

The Physical Activity for persons with Disability model (PAD model) was used as a background scheme.[31] In many studies looking at factors associated with participation in PA, the PAD model is being used to identify emerging themes.[18,32] This model combines the ICF with the model of Attitude, Social Influence and Self- Efficacy (ASE model).[31] This results in a model, enlarging the personal and environmental factors as part of the ICF model that either facilitate or hinder the intention to participate in physical activity. The personal factors consist of the levels of "Intention", "Attitude", "Self-efficacy", "Health condition" and "Facilitators and Barriers". "Intention" is the central determinant for participation in PA within the PAD model. Without intention to be active, a person is most likely not going to be active. At the same time though, a person may very well have the intention to be active, but this intention is influenced for better or worse by other contextual factors both at the per- sonal and environmental levels.[31] "Attitude" is defined as what an individual thinks and expresses about an active life- style for him- or herself and "Self-Efficacy" is the confidence that an individual has for performing PA. "Health condition" refers to specific aspects related with the diagno- sis, in this case SB. The environmental factors only consist of the level of "Social Influence", defined as what another person thinks about PA for that individual, and, like the personal factors, the level of "Facilitators and Barriers".[31]

To enhance the credibility and conformability, two independent researchers with varying experiences in pediatric physical therapy performed the analyses. In case of no consensus, a third researcher was consulted who had extensive experience in research in children and adolescents with SB.[33,34] Several experts working in pediatric medicine performed skeptical peer review to ensure dependability.[33,34] In addition member checking was performed by presenting the results to a different group of parents of children and adolescents with SB, leading to credibility.[33,34]

The data was analyzed through MaxQDA version 10 (VERBI, Berlin, Germany) to enhance standardization and transparency.[35]

RESULTS

Eleven semi-structured interviews with 13 parents from young children with SB, nine focus groups with youth (n = 33) with SB and eight focus groups with their parents (n = 31) were conducted. Participants did not discuss any new factors after the 7th focus group and 10th interview. Therefore, the researchers were confident that informational saturation was achieved. The children and adolescents attended both regular schools and schools for special education and their mobility varied from normal ambulatory to non-ambulatory.[36] Table 1 provides an overview of the characteristics of both the children and adolescents and the parents. In the Netherlands, children with special needs often attend special education schools, which are regionally distributed and are funded, like regular schools, by the Dutch government.

Table 1. *Characteristics of the children, adolescents and their parents (N=44).*

	PARENTS (N = 13: FROM CHILDREN 4-7 YEARS)	**PARENTS** (N = 31: FROM CHILDREN AND ADOLESCENTS 8-18 YEARS)
AGE	Mean 39 years (range 27-44)	Mean 47 years (range 34-64)
SEX (FEMALE/MALE)	11/2	25/6
NUMBER (%) ADHERING TO THE DUTCH GUIDELINES FOR HEALTHY PA	12 (92 %)	26 (84 %)
EDUCATION LEVEL (%) UNIVERSITY OR PROFESSIONAL LEVEL	35	30
	CHILDREN 4-7 YEARS (N= 11)	**CHILDREN AND ADOLESCENTS 8-18 YEARS** (N=33)
AGE	Mean 6 years (range 4-7)	Mean 13 years (range 8-18)
SEX (FEMALE/MALE)	4/7	15/18
DIAGNOSES	9 SB 2 SB with hydrocephalus	26 SB 7 SB with hydrocephalus
MOBILITY (HOFFER CLASSIFICATION) • NORMAL AMBULATORY • COMMUNITY AMBULATORY • HOUSEHOLD AMBULATORY • NON AMBULATORY	 1 2 1 7	 2 6 5 20
NUMBER (%) ADHERING TO THE DUTCH GUIDELINES FOR HEALTHY PA	6 (55 %)	17 (57 %)
NUMBER (%) WITH SIBLINGS	9 (90%)	28 (85%)
EDUCATION (REGULAR/SPECIAL)	6/5	9/24

SB = Spina Bifida | PA = Physical Activity

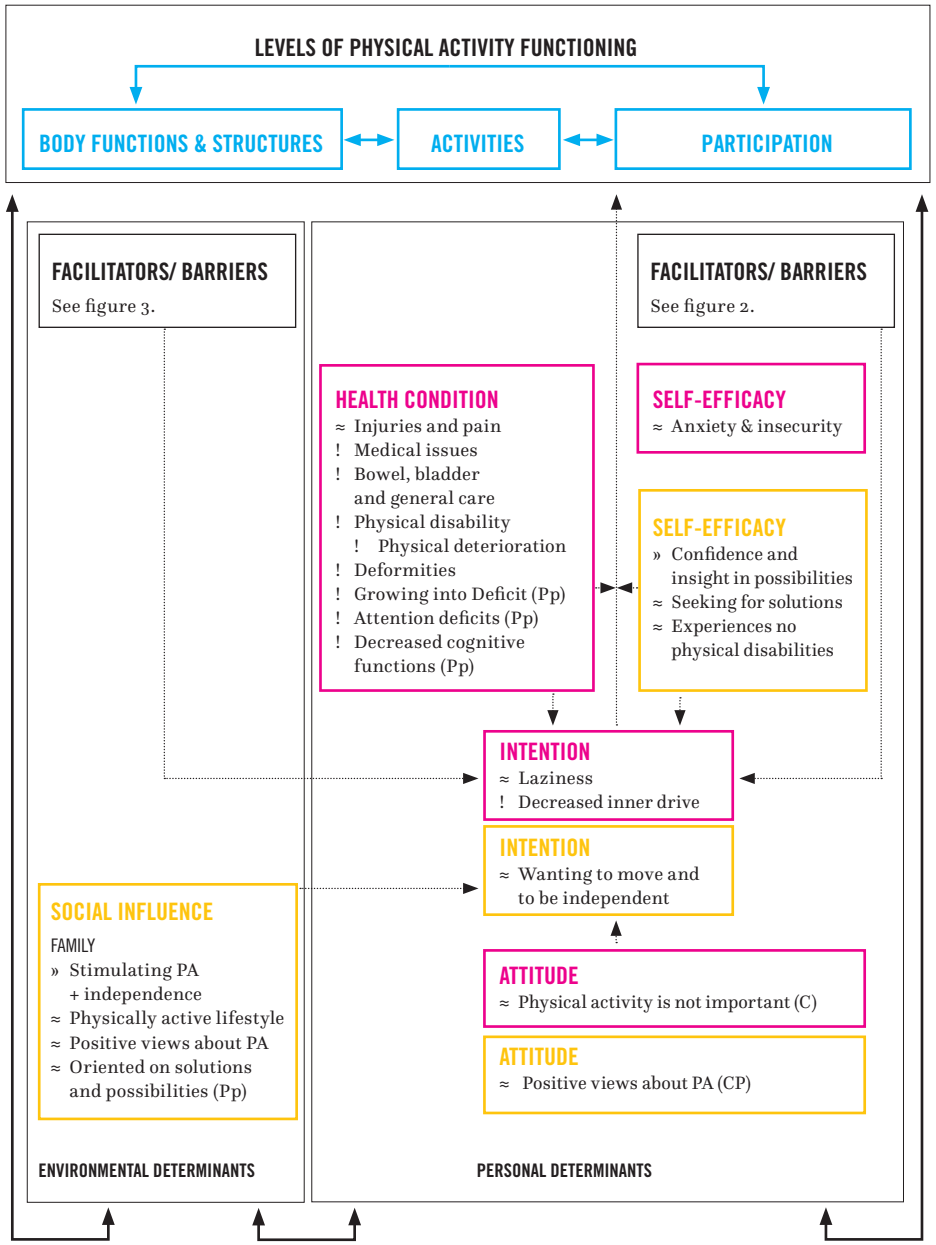

Figure 1. *Results of this study incorporated in the PAD-model* [31]

»	Modifiable Determinants
≈	Partly Modifiable Determinants
!	Not Modifiable Determinants
p	Parents (4 to 7 year old children)
P	Parents (8 to 18 year old children)
C	Children & Adolescents (8 to 18 years)

When there's no indicator behind a determinant, it means the determinant is mentioned in (P), (p) & (C).

PA Physical Activity
Negative Determinants
Positive Determinants

Data showed that youth with SB encountered a variety of both positive as negative personal and environmental factors for PA during childhood on all levels of the PAD model, with only minor differences between the children, adolescents and the parents. Individual differences were present and the factors varied in modifiability. Figures 1, 2 and 3 present overviews of the central themes on the different levels of the PAD model, the most important issues are discussed in the text. The quotes (P = Parents from children 8–18 years, p = parents from children 4–7 years, C = Children and adolescents 8–18 years) represent the literal translation of what the children, adolescents or parents said during the focus groups or the interviews.

Personal factors

Intention

Wanting to be physically active and to be independent seems to be a very strong positive theme; "(C) *I always self-propel my wheelchair, … at a certain point, … you have to do it yourself later on*". However, there also seems to be a large group of children and adolescents who lack an inner drive for PA, which seems to be difficult or sometimes even impossible to change; "(p) *the complication is, that the stimulation always has to come from us… what I experience from my healthy children, … is they ask us for help, … and say 'now you have to help me because … I want to do this and that', they ask. He doesn't ask, it always has to be stimulated by us*".

Attitude

Both the children, adolescents and the parents described a positive attitude towards PA in the children and adolescents, for example because of expected health benefits and social contacts; "(C) *if you play sports, you get energy, … you'll become fit and yes you'll notice*", "(C) b*ecause you're around people, you make contact with people, sometimes you make friends*". In the children and adolescents however, was also mentioned that PA was not important.

Self-efficacy

Both the children, adolescents and parents pointed out the importance of self-confidence; it seems to be crucial to have a notion and realization of own capacities and possibilities. Positive experiences were described of training programs or camps that also focus on developing self- efficacy. When this was explained by a girl "(C) *because I dare to do things now that others don't dare*", an adolescent reacted "(C) *yes, I would also like to do that, for my self-confidence because… if you fall, you know what to do*", the girl reacted "(C) *yes, that's what I mean.*"

Health condition

Medical problems, bowel and bladder care, injuries and pain, disabilities, deterioration and deformities were important physical negative factors of SB besides attention and cognitive dysfunctions. The bowel and bladder care influenced PA mostly when the child was incapable of self-catheterization; "(P) *really an obstacle, … every 3.5 hour it has to happen… so you always have to plan ahead, or you have to go back and forth, … you always have to say 'it's not possible to come directly after school because he has to go to the toilet first', it is even a bigger obstacle than the handicap, you always have to be there as a*

parent…" According to parents, both the physical and the cognitive dysfunctions may lead to growing into deficit when these children and adolescents grow older, meaning that the differences between the children and adolescents with SB and typically developing peers become more evident.

Facilitators and barriers within the child

The competence in both simple as complex skills are important facilitators and barriers; "(P) *Wheelchair training, that is very important I think, …. that they really learn to go up and down stairs… she can do much more now… a lot of places are not adjusted for wheel-chairs… and you can just go… your life becomes a lot more fun*". Social consequences are mentioned as an important facilitating motivational aspect. A requirement for PA seems to be a sufficient level of fitness, "(C) *being unfit*" was mentioned as a barrier because "(C) *you get tired more easily.*" Overweight or obesity is seen as a barrier for PA, because transfers are more difficult.

Environmental factors

Social influence

The importance of stimulating the child in a physically active and independent lifestyle was emphasized upon. All parents believed that PA is healthy because of multiple reasons such as positive effects on health, social relations and general development. Additionally, the parents reported a solution-orientated approach within the family, as a positive factor; not emphasizing on the difficulties, but focusing on solutions and possibilities. "(C) *I think partly maybe the way I was brought up, because my parents*

BARRIERS		
≈ Decreased motivation	≈ Insufficient control of skills	≈ Insufficient connection
» Decreased fitness	≈ Simple (Cp)	with peers (P)
	≈ Complex	! Interests for passive activities
	≈ Overweight (Pp)	

FACILITATORS		
» Sufficient fitness	≈ Motivation for PA	≈ Denies help (Cp)
≈ Control of skills	≈ Social element	! Mentality
≈ Simple	≈ Achievements	! Development by
≈ Complex	≈ Competition and play	growing older (p)
	≈ Goal (Cp)	
	≈ Asks help if necessary (p)	

Figure 2. *Personal Determinants. Barriers and facilitators.*

» Modifiable Determinants
≈ Partly Modifiable Determinants
! Not Modifiable Determinants
p Parents (4 to 7 year old children)
P Parents (8 to 18 year old children)
C Children & Adolescents (8 to 18 years)

When there's no indicator behind a determinant, it means the determinant is mentioned in (P), (p) & (C).

PA Physical Activity
Negative Determinants
Positive Determinants

they always say, you have to propel yourself as much as possible, because your fitness will increase … if I start to complain 'I'm tired, I want to go home' , well, they ignore it … I think it is ok, I think I will benefit later on. On the other hand, at that moment when I'm tired and they don't want to push me, I am mad (laughing)."

Facilitators and Barriers within the environment

- Other people

A major theme was the support from and contact with people in general. The protective attitude towards children and adolescents with a disability such as SB, was mentioned as an important barrier, but also the inability to be open-minded and flexible. A girl said "(C) *Sometimes I see handicapped children… older than I am, and they are treated like they are much younger and then I think, you just can't do that*", after which another boy remarked "(C) *well, I think it is like that, because they usually think that you're also mentally handicapped and that's why they think oh, he's not that smart.*" One of the adolescents trained in a local fitness centre and she stated: "(C) … *they easily think that activities are too hard … if I for example say 'I want to do this and that' he will say 'that is too hard for you … what if something happens … well. It is difficult to say otherwise…*" People in general seem to have a lack of knowledge about possibilities for PA in children with SB, but it varies widely how they cope with this. Certain people are willing and able to adjust activities, but also examples that were the opposite were mentioned. "(P) *We now have a teacher who absolutely doesn't want to make adjustments in the physical education class. They just say 'if he can't do it, he can't do it'. We had a huge discussion about his grade for physical education this year. She didn't want to give a higher grade than a C, because, well, that was just not possible.*"

- Possibilities to participate in sports

Sports possibilities were another major theme. In general, there were not enough suitable sports possibilities and the possibilities of participating in regular, local sport clubs were scarce. If they have to travel to sports clubs further away, transport problems will arise and it will also be more time consuming. "(C) *It has to be in the neighborhood … so you can go by yourself … so your parents don't have to take you. Yes, because when you grow older, it is annoying always having your parents around*". A tendency towards more sport possibilities for children and adolescents with a disability in local and regular clubs is noted by the parents, for example a regular local soccer club that set up a team for children with physical disabilities "(p) *There are a lot of enthusiastic people who said it really fits in our club, and we're going to take care of it!*" The support from the national sports associations for participation in local and regular sports clubs seems to increase and the necessity of this support was underlined, "(P) *They have a huge roll and they think it is important*", "(P) *You notice that they're working on it, but I think it should go faster*".

- Assistive devices

The importance of good assistive devices for optimal mobility and personal care was highlighted; "(C) *You can achieve the same things with an assistive device as an able bodied person, … a wheelchair is a replacement of your legs… but then you need good equipment… I should not have to adjust to my equipment… it should exactly be the other*

BARRIERS

FAMILY
» Necessary parental support (Pp)
≈ Over-protective and over-supportive
! Family composition and environment

SUPPORT FROM AND CONTACT WITH PEOPLE
» Insufficient support during care (P)
» Insufficient support for sports from health care professional (P)
≈ Over-protective and over-supportive
≈ Insufficient knowledge and inability to be open-minded
≈ Insufficient connection with peers

SPORTS
» Too few or inadequate sports possibilities
» Insufficient sports possibilities in regular sports club (Pp)
» Sports possibilities during difficult times

PLAY OPPORTUNITIES
» Not suitable

MOBILITY AND CARE AIDS
» Inadequate mobility and care aids
» Slow and complicated procedure

ENVIRONMENT
» Not or insufficient accessible
» Not safe enough (p)

SCHOOL
≈ Combination of distance and groups transportation in special education
» Insufficient support and vision in regular education (Pp)
» Insufficient adaptation in regular education (p)
» Decrease in PA (p)

! **WEATHER**

TRANSPORT
» Taxi transport is unreliable and takes time (Pp)
» Mobility aids are not allowed in the taxi (C)

INFORMATION TRANSFER
» Insufficient information regarding mobility aids (Cp)
» Insufficient information regarding sports possibilities (Pp)

FINANCIAL SUPPORT
≈ Expenses for adapted sports and the limited reimbursement
≈ Expenses care and mobility aids and the limited reimbursement
≈ Expenses adapted play materials and the limited reimbursements (p)
≈ Personal budget is limited utilizable (Pp)
! Financial cuts (P)

FACILITATORS

FAMILY
! Family composition and environment
≈ Family is able to support
≈ Assertiveness parents

SUPPORT FROM AND CONTACT WITH PEOPLE
» Presence of external stimuli
» Health care professional supports procedure care and mobility aids
» Support during PA, transport, care
≈ Presence of peers, taking possibilities into account
≈ Instructor and sportsmen are role models
≈ People are competent, open-minded and flexible

SPORTS
» Adapted sports possibilities
» Proper time (Cp)
» Possibility to try several sports (CP)
» Possibilities within regula clubs (Pp)
» Flexibility and support from National Sports Associations (Pp)

PLAY OPPORTUNITIES
» Suitable
» Several riding devices (Pp)
» WII (CP)

MOBILITY AND CARE AIDS
» Optimum mobility and care aids
» Adequate procedure for aids (Pp)

ENVIRONMENT
» Accessibility
» Safety (Pp)

SCHOOL
≈ Regular educational system is open-minded
» Possibilities for PA at special education
» Special education provides space after school hours (P)

TRANSPORT
≈ Individual transport (P)

INFORMATION TRANSFER
» Individual & practical support
» Contact with other parents (Pp)
» Information regarding sports possibilities

FINANCIAL SUPPORT
≈ Personal budget
≈ Sponsorship and reimbursement (P)

INTERVENTIONS REGARDING SKILLS AND SELF-EFFICACY
» Wheelchair training (CP)
» Sports camps (P)

» **INFORMATION FOR ENVIRONMENT (CP)**
! **SHORT DISTANCES SUPPORT PA (CP)**
≈ **BALANCE EFFORT AND REST (P)**
! **WEATHER**

Figure 3. *Environmental Determinants. Barriers and facilitators.*

way around". Even though all agreed upon the importance of optimal assistive devices, the reality is often otherwise unfortunately. "(P) *Since September, she can't handbike anymore, ... if everything is ok again, the summer is probably over, that's such a waist, ... her friend lives 3 kilometer away, she can easily bike it, but now we have to take her, ... she is very limited because of assistive mobility devices that do not work*".

- Accessibility
The environment, such as playgrounds and sports facilities, lacks accessibility. Playgrounds with sand or grass are difficult or impossible to enter and playgrounds often contain equipment that is not suitable for children with disabilities like SB. Inadequate use or the absence of care facilities are other examples that make it difficult to participate in PA.

- Information
Adequate information transfer seems essential, with a variety of informational aspects with several goals. Parents mentioned the scarce attention for and information regarding PA during hospital visits. "(P) *A lot of things you have to find out yourself... I do miss that... I think, if you're in a hospital, we visit the hospital regularly, that there should be... more information... and listening what the child wants and I do miss that... they ask for example 'how is it', 'yes everything goes well' he (the child) says, well he always says everything goes well... but I think... you should ask 'what else do you want, how is it going with playing sports, do you play sports', it is always about what school do you go to and that's that*". Information for parents about which sports possibilities are available, specified for what kind of disabilities (mental, physical or a combination) including the care facilities that are available, make it easier to find a suitable sport for the child or adolescent. Several rehabilitation centers and local authorities have sport counselors, which are greatly appreciated because of their individual and practical approach, "(p) *Especially one on one, somebody who says... that's all available, what kind of child do you have, what kind of situation, where do you live, what are you looking for, leading to something concrete*". Information for the environment is also emphasized upon, so people understand the possibilities of children and adolescents with SB. Parents themselves sometimes provide this information, but other possibilities are appreciated widely. "(P) *In third grade they spent a lesson on him, they have this book,... It is about a boy with SB... the teacher read it aloud and then they talked about it*", "(P) *We always had support from a regional expertise centre and a therapist comes in ones every so many times*".

» Modifiable Determinants
≈ Partly Modifiable Determinants
! Not Modifiable Determinants
p Parents (4 to 7 year old children)
P Parents (8 to 18 year old children)
C Children & Adolescents (8 to 18 years)

When there's no indicator behind a determinant, it means the determinant is mentioned in (P), (p) & (C).

PA Physical Activity
Negative Determinants
Positive Determinants

DISCUSSION

The goal of this study was to describe both personal and environmental factors that are important for participation in physical activity as experienced by both youth with SB and their parents, in order to better develop intervention strategies to improve participation in PA. Three recent reviews looking at PA in persons with SB conclude this is an important gap in current knowledge for this population. They all agree looking into factors that can influence the (maintenance of) PA level is important for the development of interventions to improve PA and fitness levels in a sustainable matter.[12,37,38] In our study, a variety of both positive and negative factors were found on all levels of the PAD-model, both personal and environmental and with varying modifiability. The views of the children and adolescents and the parents were predominantly similar, with only minor differences.

While results are comparable with results presented in two recent systematic reviews about perceived barriers and facilitators to PA for children with disability[18,39], specific factors were associated with the lack of PA in youth with SB. The items specific for youth with SB were mostly related to bowel and bladder care, including assistive devices for these issues, but also the need for privacy in bathrooms or adequate equipment in bathrooms (e.g. changing table) in public places. Another problem specific to children with SB seemed to be a lack of inner drive to initiate any type of behavior, as reported by parents. This means parents and other adults within the child's environment need to motivate the child to be active much more than in other children with a disability. Looking at another recent qualitative study, in ambulatory children and adolescents with cerebral palsy.[32] it shows that themes like 'contact with and support from people', 'mobility and care aids' and 'play opportunities' for example were not found in this study from Verschuren et al.[32] These differences may be present because our study not only focused on sports participation but on PA in general and also included both ambulating as non-ambulating participants. Another qualitative study by Buffart et al. has looked at factors associated with participation in sports in adolescents and young adults with SB.[40] Results are partly overlapping, while some, like personal goal attainment - e.g. wanting to maintain ambulatory skills (positive) - or having to wake up early (negative) seemed more specific for this older group of patients. Similar though were the lack of information, the limited number of adapted/ accessible sports facilities, SB related bowel and bladder complications, equipment issues, fatigue and more general lack of motivation.

Next to negative factors, a wide variety of important positive factors were found in this study. As we know, not all negative factors are modifiable, but may be overcome by using positive factors. The use of assistive devices for optimal mobility and self-care, the development of a sufficient level of fitness, the development of wheelchair skills and self-confidence and a solution-orientated approach were examples of positive factors (both environmental and personal) that contributed to participation in PA. It was very interesting though, to note individual differences that were apparent and the fact that most examples of positive factors were complemented with similar negative factors, meaning the existing positive factors may not be implemented and used enough in general. For example, when children go to regular schools, some physical education (PE) teachers seem to work according to the idea of inclusiveness and find ways to involve children with dis- ability in their lessons, while other teachers do not know how to deal with children with disability. A very active boy for example, walking and exercising with crutches received a fail mark for PE because he could not perform the standard list of required activities (somersault, hopping, etc.).

At the same time though, this study presents a variety of factors for PA and it is evident that the factors vary between the individual children and adolescents. In order to start looking at sustainable interventions to improve participation in PA, these results may serve as a start for developing a practical guide with possible factors contributing to PA that is applicable in individual children and adolescents with SB. Using the contextual factors as represented in the PAD model may help health care professionals to assess the most important factors for the individual child or adolescent in their practice. Currently, we are developing a conversation tool to discuss participation in PA and to identify the factors possibilities or barriers within the (environment) of the individual child. Interventions should be directed at trying to stimulate the positive factors and to deduct the existing negative factors for PA. Even more importantly, health care professionals can work on developing several positive factors. For example, children (and parents) in our study were very enthousiastic about a paralympic athlete, who was a great role model, teaching children to develop skills and confidence through wheelchair training, which was associated with PA lifestyle. If youth encounter positive experiences like this and know that they can perform activities, they will feel safer and be confident in performing these activities during daily life. This was also reported by the children. Feeling confident in their wheelchair and being able to negotiate obstacles in daily life (including going up and down the escalator!) gave these children the freedom to be more independent and active in their neighborhood. Considering the results of this study, interventions designed to improve PA should be individualized to the child and multidisciplinary in methods. Physical therapy may be initiated to work on some of the basic requirements to move (e.g. sufficient fitness, certain skills and knowledge regarding an active lifestyle), but they need to work together with other health care professionals if needed for this child, but also teachers at school and coaches at the local sports club. By doing so, the remarks about the scarce in- formation and scarce attention for PA during hospital visits may be overcome, so the future care of these children and adolescents will include concrete actions of preventable consequences of inactivity. It seems important to start early in childhood with promoting independence and the benefits of an active healthy lifestyle. Results of longitudinal studies support the idea that PA in youth is of great importance for the promotion of public health [7] and as the children stated themselves, "*you have to do it yourself later on*".

The environmental modifiable factors may be partly addressed by health care professionals through advocacy for children with disability and the importance of participation in PA. However, policy makers seem to have a much higher responsibility in dealing with these factors. Several negative environmental factors might be altered

by adjustments in policies about health care aspects, but also policies about accessibility and the presence of local sports facilities and possibilities, play possibilities and the attitude of people towards children and adolescents with SB and PA. As cultural context and organization of community based PA varies among countries, the identified factors may differ when interviewing families who live outside of the Netherlands. So it would also be of great interest, if the same research would be conducted in other countries and to see if other factors are being mentioned or if certain negative factors do not exist in other societies as other regulations and standards apply. In the United States for example, the Americans with Disabilities Act [41] is a wide-ranging civil rights law that prohibits discrimination based on disability [42]; this may for example, have an impact on the central theme "accessibility of the environment". Cultural differences in PA between the Netherlands and the United States have been reported for youth with Cerebral Palsy [43]. If different factors do exist between societies, this might provide insight in strategies that may be used by policy makers to overcome the mentioned negative factors.

Several weaknesses and strengths were present during this qualitative study. Selection bias may be present as only youth and parents might have participated who believed that PA is important. However, both active and inactive youth and their parents participated, as presented in Table 1. At the same time, only Dutch speaking people were included. This could underreport the barriers for PA in non-Dutch speaking ethnic minority groups, as it is known that low levels of health literacy are reported in this population.[44,45] We did not ask about socioeconomic status, which given the financial barriers, could have been an important factor. Because the cultural context and organization of community-based physical activity and sports varies among countries, the experiences of the children and parents who participated in this study may differ from those of children and parents who live outside of The Netherlands. Moreover, in this study, the majority of children who participated were non-ambulatory. Thus, it is important to consider that some of the barriers, facilitators, and solutions described in this study might not reflect the experiences of families with children with less severe dis- abilities. These limitations may of course have influenced the results and the generalizability. At the same time though, a heterogenic group of participants, both ambulatory as non-ambulatory, were included in this study, leading to a wide variety of determinants for PA. Despite this heterogeneity, data saturation was reached and the benefit of this approach was the overall view that could be presented for this population. The final step in our data analysis consisted of categorizing the central themes in modifiable determinants, partly modifiable determinants and non-modifiable determinants. This was done in an effort to reflect on the contextual or personal factors that are present, and may be either positive or negative, but looking at what is a given and not very easy to change or on the opposite factors that could be a goal for intervention. It is certainly true, one could question these labels of (partly) modifiable or not and argue everything is modifiable, but this classification was seen from the perspective of a healthcare provider, working with an individual child. While changes in society are definitely possible, they often require different types of actions, at a societal level. One of the strengths of this study was using the PAD model in presenting the central themes.[31] By doing so, it was possible to give insight into the different factors of "Intention", "Attitude", "Self-efficacy", "Health condition", "Social influence" and "Facilitators and Barriers". Using the PAD model allows specific and individually tailored interventions to be developed for becoming or maintaining a physically active lifestyle by looking at possibilities within the environmental and personal situation of the child with SB. Another strength was the general description of PA, not just participation in sports, but also in daily life. This is important, because for most people (non-athletes), PA in daily life is probably a much more important factor in attaining a physically active lifestyle than participation in sports alone. Finally, triangulation, member checking and skeptical peer review were used to meet several important methodological aspects of qualitative research, such as credibility, conformability and dependability.[33,34]

CONCLUSION

Our findings suggest that while negative factors should be addressed when setting up intervention programs, using positive factors within the individual child seems to be an important starting-point in improving physical activity in youth with SB. Therefore, individual assessment of both personal and environmental factors associated with PA should be standard care within multidisciplinary intervention programs aimed to aimed to encourage healthy active lifestyles in youth with SB.

APPENDIX 1

Main topics for the children and adolescents during the focus groups. The topics for the parents were similar, only rephrased.

1. What kind of physical activities do you perform?
2. Do you like physical activity and why?
3. What facilitators do you experience regarding physical activity?
4. What barriers do you experience regarding physical activity?
5. What solutions would there be for being more physically active?

COMPETING INTERESTS

The authors declare that they have no competing interests.

ACKNOWLEDGMENTS

This research is funded by SIA-RAAK, which had no role in the design, data collection, analysis and interpretation, reporting of this work, or the decision to submit the work for publication.

We thank all the participating children, adolescents and their parents for sharing their views and experiences with us! All participating rehabilitation centers, schools for special education, pediatric physical therapists and the BOSK are being acknowledged for handing out our information flyers and for using the facilities when conducting the focus groups. For advice and input regarding our qualitative research, we thank Dr. Henny Boeije, Janke Oosterhaven (PT, MSc), Marlies Welbie (PT, MSc), Jacqueline Outermans (PT, MSc) and Pieterbas Lalleman (MSc). We would also like to acknowledge all participating students for their enthusiastic participation in this research.

FUNDING

Funded by Foundation Innovation Alliance - Regional Attention and Action for Knowledge circulation (SIA RAAK), project number 2011-13-35P and by personal PhD grant HU University of Applied Sciences Utrecht.

REFERENCES

1. Annual Report 2011 with Data for 2009.
International Clearinghouse for Birth Defects Surveillance and Research.
http://www.icbdsr.org/filebank/documents/ar2005/Report2011.pdf

2. Singh DK.
Families of Children with Spina Bifida: A Review.
J Dev Phys Disabil. 2003;15(1):37–54.

3. Bowman RM, McLone DG, Grant JA, Tomita T, Ito J.
Spina bifida outcome: a 25-year prospective.
Pediatr Neurosurg. 2001;34(3):114–20.

4. Mitchell LE, Adzick NS, Melchionne J, Pasquariello PS, Sutton LN, Whitehead AS.
Spina bifida.
Lancet. 2004;364:1885–95.

5. Simeonsson RJ, McMillen JS, Huntington GS.
Secondary conditions in children with disabilities: spina bifida as a case example.
Ment Retard Dev Disabil Res Rev. 2002;8(3):198–205.

6. Bouchard C, Shephard RJ, Stephens T.
Physical activity, fitness and health the model and key concepts.
In: Anonymous, editor. Physical activity, fitness, and health consensus statement.
Champaign, IL: Human Kinetics; 1994.

7. Telama R.
Tracking of physical activity from childhood to adulthood: a review.
Obes Facts. 2009;2(3):187–95.

8. 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):259–66.

9. Buffart LM, Roebroeck ME, Rol M, Stam HJ, van den Berg-Emons RJ,
Transition Research Group South-West Netherlands.
Triad of physical activity, aerobic fitness and obesity in adolescents and young adults with myelomeningocele.
J Rehabil Med. 2008;40(1):70–5.

10. van den Berg-Emons RJ, Bussmann JB, Stam HJ.
Accelerometry-based activity spectrum in persons with chronic physical conditions.
Arch Phys Med Rehabil. 2010;91(12):1856–61.

11. 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.

12. Short KR, Frimberger D. A review of the potential for cardiometabolic dysfunction
in youth with spina bifida and the role for physical activity and structured
exercise.
Int J Pediatr. 2012;2012:11.

13. Buffart LM, van den Berg-Emons RJ, Burdorf A, Janssen WG, Stam HJ, Roebroeck ME.
Cardiovascular disease risk factors and the relationships with physical activity,
aerobic fitness, and body fat in adolescents and young adults with myelomeningocele.
Arch Phys Med Rehabil. 2008;89(11):2167–73.

14. 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;161(11):1075–81.

15. Liusuwan RA, Widman LM, Abresch RT, Johnson AJ, McDonald CM.
Behavioral intervention, exercise, and nutrition education to improve health and
fitness (BENEfit) in adolescents with mobility impairment due to spinal cord
dysfunction.
J Spinal Cord Med. 2007;30 Suppl 1:S119–26.

16. Andrade CK, Kramer J, Garber M, Longmuir P.
Changes in self-concept, cardiovascular endurance and muscular strength of
qchildren with spina bifida aged 8 to 13 years in response to a 10-week physical-
activity programme: a pilot study.
Child Care Health Dev. 1991;17(3):183–96.

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;25(7):597–606.

18. Shields N, Synnot AJ, Barr M.
Perceived barriers and facilitators to physical activity for children with
disability: a systematic review.
Br J Sports Med. 2012;46(14):989–97.

19. Trost SG, Sallis JF, Pate RR, Russel R, Freedson PS, Taylor WC, et al.
Evaluating a model of parental influence on youth physical activity.
Am J Prev Med. 2003;25(4):277–82.

20. Erkelenz N, Kobel S, Kettner S, Drenowatz C, Steinacker JM.
Parental Activity as Influence on Children's BMI Percentiles and Physical Activity.
J Sports Sci Med. 2014;13(3):645–50.

21. Braun V, Clarke V.
Using thematic analysis in psychology.
Qual Res Psychol. 2006;3(2):77–101.

22. Creswell J.
Research Design: Qualitative, Quantatative and Mixed Methods Approaches.
2nd ed. Thousand Oaks, California: Sage Publications; 2003.

23. Spratling R, Coke S, Minick P.
Qualitative data collection with children.
Appl Nurs Res. 2012;25(1):47–53.

24. Massey OT.
A proposed model for the analysis and interpretation of focus groups in
evaluation research.
Eval Program Plann. 2011;34(1):21–8.

25. Baarda DB, De-Goede MPP, Teunissen J.
Basisboek Kwalitatief Onderzoek. Handleiding voor het opzetten en uitvoeren
van kwalitatief onderzoek.
secondth ed. Groningen/Houten, the Netherlands: Wolters-Noordhoff; 2005.

26. Robson C.
Real World Research, A Resource for Social Scientists and
Practitioner-Researchers.
2nd ed. Oxford, UK: Blackwell Publishing; 2002.

27. Caspersen ID, Habekost T.
Selective and sustained attention in children with spina bifida myelomeningocele.
Child Neuropsychol. 2013;19(1):55–77.

28. Rose BM, Holmbeck GN. Attention and executive functions in adolescents
with spina bifida.
J Pediatr Psychol. 2007;32(8):983–94.

29. 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):126–31.

30. Patton M.
Designing Qualitative Studies, Purposeful Sampling. In: Anonymous, editor.
Qualitative evaluation and research methods.
Beverly Hills, CA: Sage; 1990. p. 169–86.

31. van der Ploeg HP, van der Beek AJ, van der Woude LH, van Mechelen W.
Physical activity for people with a disability: a conceptual model.
Sports Med. 2004;34(10):639–49.

32. 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):488–94.

33. Netwerk Kwalitatief Onderzoek AMC – UvA.
Richtlijnen voor kwaliteitsborging in gezondheids(zorg)onderzoek:
Kwalitatief Onderzoek. 2002.

34. Letts L, Wilkins S, Law M, Stewart D, Bosch J, Westmorland M.
Guidelines for critical review form - Qualitative studies (version 2.0),
McMaster University Occupational Therapy Evidence-Based Practice
Research Group. 2007.

35. Kuckartz U.
MAXQDA 2007 reference manual. (J. Poppe, Transl.).
Marburg: VERBI Software; 2007.

36. Schoenmakers MA, Gulmans VA, Gooskens RH, Helders PJ.
Spina bifida at the sacral level: more than minor gait disturbances.
Clin Rehabil. 2004;18(2):178–85.

37. Oliveira A, Jácome C, Marques A.
Physical fitness and exercise training on individuals with Spina Bifida:
a systematic review.
Res Dev Disabil. 2014;35(5):1119–36.

38. Crytzer T, Dicianno B, Kapoor R.
Physical activity, exercise, and health-related measures of fitness in adults with
spina bifida: a review of the literature.
PM R. 2013;5(12):1051–62.

39. Bloemen MA, Backx FJ, Takken T, Wittink H, Benner J, Mollema J, de Groot JF.
Factors associated with physical activity in children and adolescents with a
physical disability: a systematic review.
Dev Med Child Neurol 2015;57: 137-148.

40. Buffart L, Westendorp T, van den Berg-Emons R, Stam HJ, Roebroeck ME.
Perceived barriers to and facilitators of physical activity in young adults with c
hildhood-onset physical disabilities.
J Rehabil Med. 2009;41(11):881–5.

41. Americans with Disabilities Act.
http://www.ada.gov/

42. 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):1057–61.

43. Van Wely L, Dallmeijer AJ, Balemans AC, Zhou C, Becher JG, Bjornson KF.
Walking activity of children with cerebral palsy and children developing
typically: a comparison between the Netherlands and the United States.
Disabil Rehabil. 2014;36(25):2136–42.

44. Council on Community Pediatrics.
Providing care for immigrant, migrant, and border children.
Pediatrics. 2013;131(6):e2028–2034.

45. Hoopman R, Terwee CB, Muller MJ, Ory FG, Aaronson NK.
Methodological challenges in quality of life research among Turkish and Moroccan
ethnic minority cancer patients: translation, recruitment and ethical issues.
Ethn Health. 2009;14(3):237–53.

8

Factors associated with physical activity in children and adolescents with a physical disability: a systematic review

Manon AT Bloemen[1,2], Frank JG Backx[3], Tim Takken[2], Harriet Wittink[1], Joyce Benner[1], Jurgen Mollema[1], Janke F de Groot[1,2]

1 Research Group Lifestyle and Health, HU University of Applied Sciences Utrecht, Utrecht, The Netherlands
2 Child Development and Exercise Center, Wilhelmina Children's Hospital, University Medical Center Utrecht, The Netherlands
3 Department of Rehabilitation, Nursing Science and Sports, University Medical Center Utrecht, The Netherlands

Dev Med Child Neurol 2015 Feb;57(2):137-48

ABSTRACT

Aim

The aim of this review was to summarize the important factors associated with participation in physical activity in children and adolescents with physical disabilities.

Method

A systematic mixed-studies review was conducted using the databases Academic Search Elite, CINAHL, The Cochrane Library, EMBASE, PEDro, PsycINFO, PubMed, and SPORTDiscus, searching for studies conducted from January 2000 to May 2013. The studies were identified by two independent researchers following predetermined inclusion and exclusion criteria. The methodological quality was determined using the McMaster University critical review forms for qualitative or quantitative research and was numerically rated according to the criteria developed by Imms.

Results

The initial electronic search yielded 10 161 articles, of which six were qualitative and 12 were quantitative studies. These studies showed that a diverse range of positive and negative factors were associated with participation in physical activity, such as self-efficacy, physical fitness, increasing age, and the availability of equipment and local facilities.

Interpretation

Future intervention studies could use these results, within the context of an individual child and his or her environment, as the basis for increasing physical activity levels, starting in early childhood and continuing throughout adolescence and into adulthood. An increased awareness of and focus on providing appropriate equipment and adapted sports in the child's own environment by policy makers might increase physical activity levels.

WHAT THIS PAPER ADDS

- Positive factors, not only negative factors, might be the basis for individualized interventions for physical activity for children and adolescents with physical disabilities.
- It could be valuable to focus on increasing self-efficacy and to pay extra attention during adolescence.
- Adequately adapted equipment and the availability of local sports seem to be associated with increased physical activity

INTRODUCTION

A recent Lancet series[1] reported on the importance of being physically active in reducing the development and mortality of non-communicable diseases such as cancer, type II diabetes, and cardiovascular disease.[2] For children in particular, the benefits of physical activity have been consistently documented and it is recognized that encouraging a physically active lifestyle from an early age is important.[3]

Physical activity can increase the physical, emotional, and social wellbeing of children with physical disabilities, as well as increasing their functional independence, integration, and quality of life and positively impacting their future health.[4–6] Therefore, it is alarming to see a decline in the physical activity of young people.[7] Children with physical disabilities are even less physically active than their peers with typical development.[8–11] A systematic review recently showed that young people with cerebral palsy (CP) participated in habitual physical activity at a rate that was 13% to 53% lower than in those with typical development and 30% lower than the recommended guidelines.[12] A group of 85 Dutch children with several physical disabilities had significantly lower physical activity, with a prevalence of overweight and obesity three and six times higher respectively, than children with typical develop- ment.[13] Moreover, a large group of children with disabilities participating in a Fitkids exercise therapy programme showed both reduced aerobic fitness and a high prevalence of overweight and obesity before the programme.[14]

Another systematic review identified a range of personal, social, environmental, and policy- and programme-related factors that influence physical activity in children and adolescents with disabilities.[15] The available literature includes several types of disabilities, including both intellectual and behavioral disabilities, which makes it difficult to understand which factors could be associated with children with a specific physical disability. Recent intervention programmes aiming to improve physical activity in children with physical disability – including an internet-based intervention, counselling, home-based physical therapy, and motivational interviewing – have highlighted the difficulty of improving physical activity in children and adolescents with a physical disability, as no improvements in physical activity were found.[16–18] The improvement of physical activity behavior in this population requires the consideration and comprehension of many factors that play a role in obtaining and maintaining a physically active lifestyle. An overview of these factors can provide information that is useful for health professionals, teachers, policy makers, and sports clubs for developing new interventions to increase participation in physical activity among these children. A complete understanding of why children and adolescents with physical disabilities are or are not physically active is imperative in order to improve their physical activity levels. Therefore, the purpose of this systematic review was to identify the factors that both hinder and facilitate physical activity for children and adolescents with physical disabilities.

METHOD

A protocol regarding this systematic mixed-studies review[19] was developed a priori, including search strategy, inclusion and exclusion criteria, methodological quality assessment, data extraction, and data analysis, and can be accessed by contacting the corresponding author.

Inclusion and exclusion criteria

Studies were included if (1) the primary aim was to examine factors that hinder and/or facilitate physical activity in children with physical disabilities resulting in motor disorders; (2) the study participants included were children with a physical disability (age range 4–18y, mean age <18y) or parents and/or caregivers giving information about their children with physical disabilities; (3) they were full-text reports published after 1 January 2000; and (4) they were written in English or Dutch. Studies in which an intervention was examined or in which physical activity was not the main outcome were excluded from this review. Similarly, studies investigating sedentary leisure activities or functional capacities were excluded, as were studies in which only differences between groups were reported. Finally, studies were excluded if more than 50% of the participating children did not have a physical disability and the results were not presented separately; if the physical disability was of a progressive nature; or if conditions in which exacerbations could occur (such as juvenile idiopathic arthritis) were present. This review included only original, peer-reviewed published articles and dissertations and did not include any 'grey literature', defined as document types produced by all levels of government organizations, academics, business professionals, and other organizations in electronic and print formats in which the process was not controlled by commercial publishing, that is by organizations whose primary activity is not publishing.[20]

Search strategy and screening

A literature search was conducted from January 2000 up to and including May 2013 in the following electronic data- bases: Academic Search Elite, CINAHL, The Cochrane Library, EMBASE, PEDro, PsycINFO, PubMed, and SPORTDiscus. A comprehensive search strategy was developed in consultation with a medical information specialist with four major themes – children, disability, physical activity, and factors – with individual search terms for each database. The terms for 'children' were derived from the existing search strategy from Riphagen et al.[21] The key terms within the search strategy were mapped to medical subject headings in MEDLINE, and title and abstract search words and phrases were added. The complete search strategy of PubMed can be found in Appendix S1 (available online).

Initial screening of titles was performed by one of the reviewers to exclude obviously non-fitting titles. The titles, abstracts, and full text of these studies were then independently reviewed for eligibility by two reviewers. Any discrepancies in the agreement were discussed with a third reviewer until consensus was reached. Finally, the reference lists of included studies were manually searched to find additional studies.

Methodological quality assessment of the manuscripts

The McMaster Critical Review Forms for qualitative[22,23] or quantitative research[24,25] were used to assess methodological quality, providing a narrative assessment of methodological quality. The wide range of methodologies employed in qualitative studies makes it complex to rate quality numerically. Imms[26] developed criteria for both qualitative research and non-experimental quantitative research, such as that reviewed in this paper. Therefore, the numerical rating criteria developed by Imms[26] were applied to the McMaster Critical Review Forms to inter- pret methodological quality.

The qualitative studies were rated by evaluating four common quality procedure criteria: credibility, transferability, dependability, and confirmability.[26] These criteria are derived from more traditional quantitative criteria. The criterion 'credibility' refers to internal validity and contains triangulation, a search for disconfirming evidence, and member checking. The criterion 'transferability' resembles external validity and contains a 'thick' description, such as a detailed description of the study context, the investigator's role in the context, and clear delineation of how the context affects the study's ability to answer the research question. The criterion 'dependability' examines reliability and includes data archiving and the creation of an audit trail (e.g. whether or not there is consistency between the data and the findings). Finally, the criterion 'confirmability' refers to objectivity and contains sceptical peer review or audit, participant audit, and reflective journal keeping.[26,27]

The methodological quality of quantitative studies was rated by evaluating three key criteria: sample, measurement, and analyses.[26] The criterion 'sample' examined whether or not selection bias was reduced, the sample size was appropriate for the design and research question, and the participant characteristics were clearly described. The criterion 'measurement' examined whether or not measurement bias was reduced. The criterion 'analyses' examined whether or not the analyses were appropriate for the research question and outcome measure.[26]

For both research designs, each criterion was scored with one star (no evidence of the study meeting the criterion), two stars (some evidence of the study meeting the criterion or unclear reporting), or three stars (evidence of the study meeting the criterion).[26]

Two reviewers independently performed the methodological quality assessment. Any discrepancies between the two reviewers were discussed until consensus was reached. If consensus could not be reached, agreement was obtained through discussion with a third reviewer. The percentage of agreement between the reviewers was determined afterwards.

Data extraction

Two reviewers independently extracted relevant data, using a standardized form, such as the study design, study inclusion and exclusion criteria, demographic data, setting, methods of data collection, and identified factors associated with physical activity. Any discrepancies were resolved by discussion until consensus was reached.

Data analysis

The Physical Activity for People with a Disability (PAD) model was used to categorize the results during the analysis.[28] This model combines the International Classification of Functioning, Disability and Health, in which personal and environmental factors are defined, with the attitude, social influence, and self-efficacy (ASE) model.[29] This results in a model that defines several levels of both personal and environmental factors. Personal factors include levels such as intention, attitude, self-efficacy, health condition, and barriers and facilitators. 'Intention' is the central factor of physical activity within the PAD model, all other factors influence an individual's intention to become or stay physically active.[28] 'Attitude' is defined as what an individual thinks and expresses about an active lifestyle for him- or herself, whereas 'self-efficacy' is described as the confidence an individual has to engage in physical activity. 'Health condition' refers to aspects related to the diagnosis. Personal 'barriers and facilitators' include additional personal factors related to physical activity. 'Social influence' is defined as what another person thinks about physical activity for that individual and is grouped with environmental 'barriers and facilitators', which contain the additional environmental factors related to physical activity.[28]

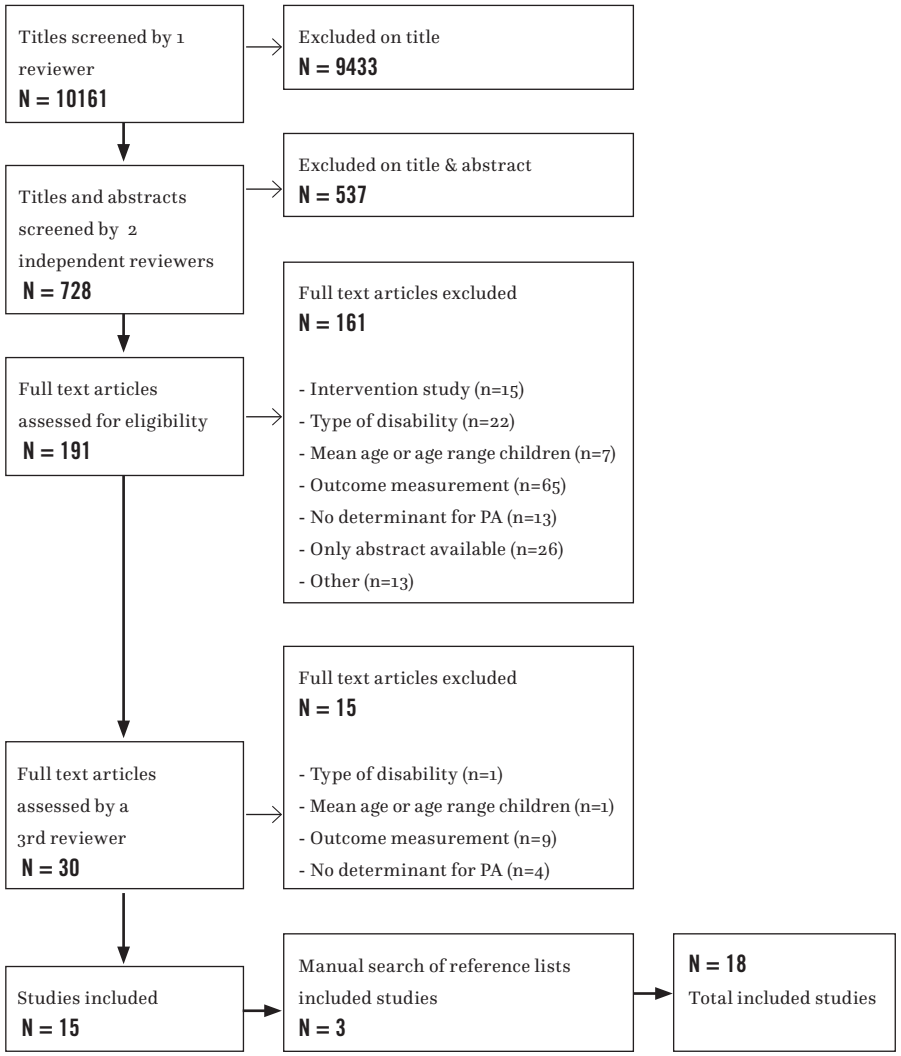

Figure 1. *Flow diagram detailing study selection process.*

RESULTS

Search results

The initial electronic search yielded 10.161 published articles (Fig. 1). After the titles were screened and the residual titles and abstracts were reviewed, full-text copies of 191 articles were retrieved. Following the predefined inclusion and exclusion criteria, 15 full-text articles were included in this systematic review.[10,30–43] A manual search in the reference lists of the articles identified three new studies that met the criteria;[44–46] therefore, a total of 18 studies were included. The studies were published between January 2000 and May 2013, employed either qualitative[30–35] or quantitative[10,36–46] study designs, and varied in the type of physical disability of the participants, sample size, participants' ages, and methods of data collection.

Two studies[39,40] used the same group of participants, but different factors were examined and different measures for physical activity were used, so both studies were included in this review. Two other studies[37,46] also used the same participants, but study results were only partly similar; the results from Matheri et al.[46] were used in this review and only the additional results from Frantz et al.[37] were presented.

Methodological quality assessment of the manuscripts

Agreement in the methodological quality assessment between the two reviewers was high, with agreement percentages ranging from 79 to 100% and a mean agreement of 86.8%. Tables I and II provide the methodological quality rating scores for the qualitative studies and quantitative studies respectively.

Qualitative studies

Six studies used a qualitative research design.[30–35] One study32 scored the maximum rating of three stars in all four criteria of the methodological quality assessment in accordance with the numerical rating scale of Imms.[26] In this study,[32] the researcher used triangulation, member checking, peer debriefing, and reflective journal keeping and provided a detailed description of the study context and her own role in it. The other five qualitative studies[30,31,33–35] scored the maximum of three stars[26] in at least two of the four criteria. The findings were always discussed with external researchers[30,31,33–35] and were sometimes checked by the participants.[35] However, in general, these studies lacked an adequate description of the study context and the investigator's role in it.[30,31,33–35]

Table 1. Summary of included qualitative studies.

FIRST AUTHOR	DESIGN	MEASURES	QUALITY CRITERIA	QUALITY RATING	PARTICIPANTS	PARTICIPANTS DETAILS				
						N	MEAN AGE (Y:MO)	AGE RANGE] (Y)	SEX	TYPE OF DISABILITY
Anderson (2005)[30]	Phenomeno-logical	Semi-structured interviews	Credibility Transferability Dependability Confirmability	** * *** ***	Children	14	13:7	10–16	14 female	Cerebral palsy n=6, spina bifida n=5, osteogenesis imperfecta n=2, other n=1
Barnett (2012)[31]	General Qualitative Research	Semi-structured interviews	Credibility Transferability Dependability Confirmability	** * *** ***	8 children 8 parents - 7 female - 1 male	8	14:1	13–15	8 male	Developmental coordination disorder (some also had mild learning difficulties n=4, diabetes n=1, scoliosis n=1, attention-deficit–hyperactivity disorder n=1)
Hunter (2009)[32]	Phenomeno-logical	Semi-structured interviews	Credibility Transferability Dependability Confirmability	*** *** *** ***	22 parents - 18 female - 4 male	23	13:7	9–17	10 female 13 male	Spina bifida n=16, cerebral palsy n=4, spinal cord injury n=1, caldar regression n=1, multiple spinal leaks n=1
Li (2012)[33]	General Qualitative Research	Semi-structured interviews	Credibility Transferability Dependability Confirmability	*** * * ***	Children	8	14:4 (SD 1:11)	11–16	5 female 3 male	Cerebral palsy
Luther (2010)[34]	General Qualitative Research	Semi-structured interviews	Credibility Transferability Dependability Confirmability	** * *** ***	12 children 12 parents - 9 female - 3 male	12	10:1	6–12	6 female 6 male	Spina bifida
Verschuren (2012)[35]	General Qualitative Research	Focus groups	Credibility Transferability Dependability Confirmability	*** *** * *	33 children 33 parents - 31 female - 2 male	33	–	7–17	15 female 18 male	Cerebral palsy

*No evidence of study meeting criteria | **some evidence of study meeting criteria or unclear reporting | ***evidence of study meeting criteria | – no data.

Quantitative studies

A quantitative research design was used by 12 studies,[10,36–46] No study scored the maximum rating of three stars[26] for all three criteria of the methodological quality assessment; however, one study[43] scored three stars[26] in two of the three criteria. The clinical importance of the results was not adequately addressed in all 12 studies[10,36–46] and, therefore, no study scored three stars for 'analyses'. How- ever, the methods used for the analyses seemed appropriate and statistical significance was reported in 12 studies.[10,36–46] All these studies[10,36–46] provided no or unclear evidence about their sample; three studies[36,38,42] did not describe their participants in detail and nine studies did not use a justified sample size.[10,37,39–41,43–46] Four studies[37,42,43,46] scored three stars for 'measurement', whereas eight studies scored only one[39,44,45] or two[10,36,38,40,41] stars. These studies did not report on the reliability[36,38–40,44,45] or validity[10,36,39,40,44,45] of all of their outcome measures, or they reported recall bias, since self-reported questionnaires were used.[10,38,41]

Data extraction

The results of the included studies are summarized in detail in Tables I and II.

Qualitative studies

Four of the six qualitative studies used a general qualitative research design[31,33–35] (Table I) and two qualitative studies used a phenomenological design.[30,32] Data were collected using semi-structured interviews[30–34] and focus groups.[35] Three studies included both children and parents,[31,34,35] two studies included only children,[30,33] and one study included only parents (the majority of which were mothers) reporting on their child.[32] The age of the children ranged from 6[34] to 17[32,35] years.

Two studies included children with CP,[33,35] one study included children with spina bifida,[34] one study included children with developmental coordination disorder,[32] and two studies included children with different physical disability diagnoses, such as CP, spina bifida, osteogenesis imperfecta, spinal cord injury, caldar regression, multiple spinal leaks, and other limited mobility.[30,32]

Quantitative studies

All quantitative studies used a cross-sectional design (Table II). Several physical activity monitors[36,39,40,43] were used to collect physical activity data, questionnaires[10,37,38,41–46] were used to collect objective physical activity data and factors related to physical activity, and measurements[36,39,40,44,45] were taken of factors related to physical activity. The age of participants ranged from 5[45] to 21[37,46] years, with the majority of children older than 10 years. Seven studies included children with CP[10,36,39,40,43–45] and four studies included children with different physical disability diagnoses, such as CP, spina bifida, muscular dystrophy, spinal cord injury, traumatic brain injury, paralysed limbs, and other limited mobility.[37,41,42,46] One study did not specify the physical disabilities.[38]

Data incorporated in the Physical Activity for People with a Disability model

The 18 included studies identified several factors associated with physical activity in children with physical disabilities, which were incorporated in the PAD model as shown in Figures 2 to 4. Thirteen studies reported on negative and positive factors,[30–37,40,41,43,44,46]

Table 2. *Summary of included quantitative studies.*

FIRST AUTHOR	DESIGN	MEASURES	QUALITY CRITERIA	QUALITY RATING	PARTICIPANTS	PARTICIPANTS DETAILS				
						N	MEAN AGE (Y:MO)	AGE RANGE] (Y)	SEX	TYPE OF DISABILITY
Capio (2012)[36]	Cross-sectional survey	Accelerometer, log diary, Test of Gross Motor Development – 2nd edition	Sample Methods Analysis	* ** **	Children	31	7:5 (SD 2:6)	–	–	Cerebral palsy
van Eck (2008)[44]	Cross-sectional survey	Question-naire, Self-Perception Profile for Children	Sample Methods Analysis	** * **	Parents	72	14:5 (SD 1:8)	12–16	26 female 46 male	Cerebral palsy
Kang (2007)[38]	Cross-sectional survey	46-item exercise barrier instrument	Sample Methods Analysis	* ** **	Children	145	<18	12–19	28 female 117 male	Physical (National Wheelchair Basketball Association classification I n=24, II n=53, III n=57)
Maher (2007)[10]	Cross-sectional survey	Physical Activity Question-naire for Adolescents	Sample Methods Analysis	** ** **	Children	112	13:11 (SD 1:11)	11–17	36 female 76 male	Cerebral palsy
Maltais (2005)[40]	Cross-sectional survey	Accelerome-ter, Biome-chanical Economy Quotient	Sample Methods Analysis	** ** **	Children	11	13:0 (SD 2:2)	10.6–16.3	4 female 7 male	Cerebral palsy
Maltais (2005)[39]	Cross-sectional survey	Heart rate monitor, peak oxygen uptake	Sample Methods Analysis	** * **	Children	11	13:0 (SD 2:2)	10.6–16.3	4 female 7 male	Cerebral palsy
Matheri (2009)[46] Frantz (2011)[37]	Cross-sectional descriptive survey	Physical Activity Scale for Individuals with Physical Disabilities	Sample Methods Analysis	** *** **	Children	234	17:1 (SD 1:11)	14–21	105 female 129 male	Paralysed limbs 38%, congenital malformations 15%, spinal injuries 14%, amputated limbs 9%, other 24%
Ortiz-Castillo (2011)[41]	Cross-sectional descriptive survey	Physical Activity Scale for Individu-als with Physical Disabilities, Physical Activity Determinants Scale	Sample Methods Analysis	** ** **	Children	93	–	12–18	37 female 56 male	Cerebral palsy n=28, spina bifida n=21, muscular dystrophy n=16, spinal cord injury n=9, other n=19
Shapiro (2010)[42]	Cross-sectional survey	Physical Self-Descrip-tion Ques-tionnaire	Sample Methods Analysis	* *** **	Children	36	16:0 (SD 2:10)	12–17	9 female 27 male	Cerebral palsy n=18, spina bifida n=9, traumatic brain injury n=3, muscular dystrophy n=2, other n=4
van Wely (2012)[43]	Cross-sectional survey	Step activity monitor, Self Perception Profile for Children, questionnaire	Sample Methods Analysis	*** *** **	Children	62	10:1 (SD 1:8)	7–13	23 female 39 male	Cerebral palsy
Zwier (2010)[45]	Cross-sectional survey	Questionnaire	Sample Methods Analysis	** * **	Parents	97	–	5–7	39 female 58 male	Cerebral palsy

*No evidence of study meeting criteria | **some evidence of study meeting criteria or unclear reporting | ***evidence of study meeting criteria | – no data.

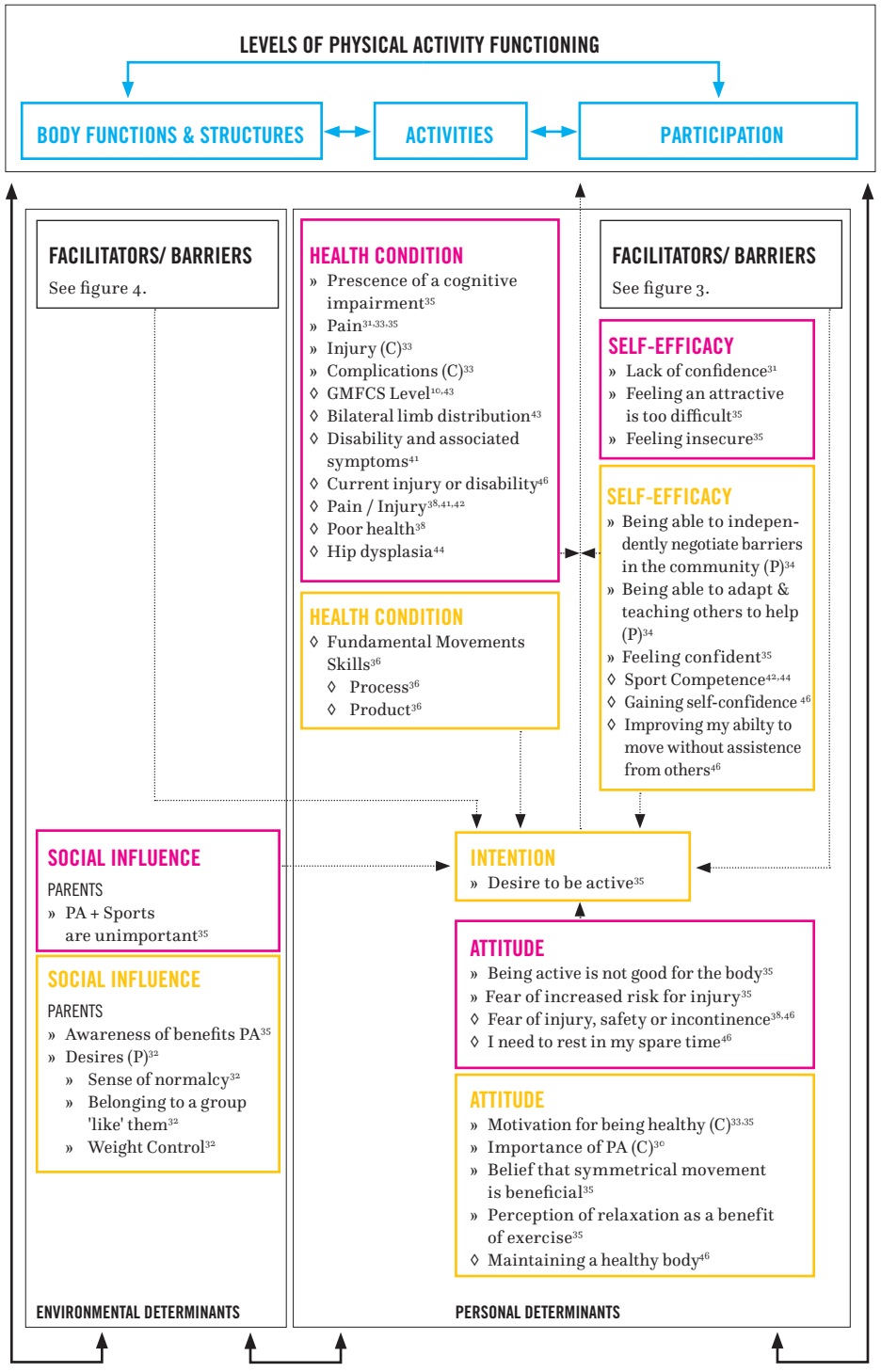

Figure 2. *Results of the included studies incorporated in the Physical Activity for People with a Disability model.*

four studies reported on only negative factors,[10,38,39,45] and one study reported on only positive factors.[42]

Personal Factors

The personal factors include 'intention', 'attitude', 'self- efficacy', 'health condition', and personal 'barriers and facilitators' (Fig. 2). Only one positive factor was identified at the level of intention.[35] The included studies identified mainly positive factors for attitude.[30,33,35,46] In general, children with physical disabilities think and express positive thoughts about an active lifestyle.[30,33,35,46] Two studies reported that having the opinions 'being active is not good for the body'[35] and 'I need to rest in my spare time'[46] negatively affected children's attitudes towards physical activity, and two studies reported on 'fear of safety, injury or incontinence'.[35,38] For self-efficacy, 'lack of confidence' or 'feeling insecure' were negative factors.[31,35] In contrast, 'feeling confident',[35] 'gaining self-confidence',[46] and 'sport competence'[42,44] were positive factors. Noteworthy positive factors were identified in the study by Luther.[34] Twelve children with spina bifida and their parents reported that 'being able to independently negotiate barriers in the community' and 'engaging others to help them negotiate their physical environment' helped them to participate in the physical activities they wanted to do,[34] which conformed with 'improving my ability to move without assistance from others'.[46] Finally, factors regarding health condition were identified. In particular, quantitative studies found several variables, such as the 'Gross Motor Function Classification System (GMFCS) level', that were significantly related to physical activity.[10,43]

At the level of personal 'barriers and facilitators', different factors were identified as related to the child's 'fitness',[31,33–35,38–40,42] 'motivation',[31,33–35,38,46] and 'abilities'[36] (Fig. 3). A striking negative factor was 'increasing age'.[10,34,43,44] Luther noted that, as children with spina bifida grew older, parents reported that 'their children became less active and rejected being involved in disability- adapted programs' and that 'their children were afraid to participate in physical activity'.[34] Maher[10] demonstrated an inverse association ($F=4.91$, $p=0.03$) between age and physical activity in children with CP; van Wely et al.[43] demonstrated that 'increasing age' ($p<0.001$), together with 'GMFCS level' and 'bilateral limb distribution', explained 52% of the variance in ambulatory activity in children with CP; and van Eck et al.[44] showed that older adolescents and females had significantly lower levels of physical activity, explaining 15% of the variance.

Environmental factors

In the PAD model, the environmental factors include 'social influence' and environmental 'barriers and facilitators' (Fig. 2). Social influence always concerned parental influence that was often positive: parents believe that physical activity is important for their child.[31,32,35]

» Qualitative Study Result
◊ Quantitative Study Result

PA Physical Activity
P Parents
C Children & Adolescents

When there is no indicator behind a factor, it means that the factor was mentioned by the parent or child in a qualitative study.

Barriers
Facilitators

At the level of environmental 'barriers and facilitators', factors were identified for different physical and social environments (Fig. 4). 'Family, teachers, peers, and other people can both hinder and promote physical activity',[30–35,37,38,41,46] 'lack of support from' or 'not being accepted or bullied by peers' were negative factors,[30,31,33–35,37,38] whereas 'getting support',[31,34,35,41] 'making friends',[34,46] or 'positive attitudes towards me by schoolmates, teachers and other people'[33,35,37] were reported as positive factors. The factor of 'teachers and instructors supporting you' seems to be a crucial positive factor associated with physical activity, as it was reported by both parents[31,34] and children.[33] In the study by Ortiz-Castillo,[41] 72% of adolescents with physical disabilities believed that 'having someone who can provide support' facilitates participation in physical activity.

BARRIERS

FITNESS
- » Fatigue[31,33,35]
- » Lack of energy and endurance[35,36]
- » Lack of motor skills[31]
- ◊ Oxygen cost of walking[39]
- ◊ Physical condition[38]
- ◊ Lack of skills[38]

MOTIVATION
- » Poor motivation[31,35]
- » Preferences for sedentary behavior (C)[33,34]
- » PA/sports not being 'fun'[35]
- ◊ Lack of interest, motivation or enjoyment[38]
- ◊ I'm not the sporty type[46]

AGE
- » Increasing age causes more fear[34]
- » Increasing age causes lack of motivation[34]
- ◊ Increasing age[10,43,45]
- ◊ Young age (<7 years)[45]

TIME
- » Lack of time[31,35]
- » Learning new skills is too time consuming[35]
- ◊ No / lack of time[38,46]
- ◊ Time taken to shower/change[38]

OTHER
- » Awareness of differences from peers[34]
- » Resisting asking for help[34]
- » Feeling like an outsider / being ashamed[35]
- » Not accepting (extent of) disability[35]
- ◊ How to use equipment[38]
- ◊ Not knowing how to exercise[38]
- ◊ Self-conscious/embarrassed[38]
- ◊ Inconvenience of sweat/combing[38]
- ◊ Female gender[44]

FACILITATORS

FITNESS
- ◊ Endurance[42]
- ◊ Biomechanical walking economy[40]

MOTIVATION
- » Enjoyment and happiness (C)[33,35]
- » Opportunity to 'clear the mind'[35]
- » Positive attitude towards being challenged[35]
- ◊ Enjoying and having fun[46]
- ◊ Learning new skills and experience[46]

SOCIALIZATION
- » Opportunity for social interaction[35]
- » Feeling accepted as part of a group[35]

ABILITIES
- ◊ Fundamental Movement Skills[36]

OTHER
- » Asking for help[34]
- » Accepting disability[35]
- » Having perseverance[35]
- » Activity gives sense of freedom (C)[30]

Figure 3. *Personal factors, barriers and facilitators of the included studies incorporated in the Physical Activity for People with a Disability model.*

» Qualitative Study Result
◊ Quantitative Study Result

PA Physical Activity
P Parents
C Children & Adolescents

When there is no indicator behind a factor, it means that the factor was mentioned by the parent or child in a qualitative study.

Barriers
Facilitators

'Adequately adapted activities,[31,33–35,38] programmes,[34,35,38] and equipment',[33,34,38,41,46] and 'access to transport'[31,34,35,38,41,46] were commonly reported as positive factors when they were available, and negative when they were not. It was also notable that the negative factors reported in this area were associated with sport facilities. After examining 33 ambulatory young people with CP and their parents, Verschuren et al.[35] noted a number of constraints in sport facilities, such as 'the teams being too big', 'a long waiting list', the child's 'abilities being underestimated', or the child 'not being allowed to play matches'. Other children with CP reported that 'physical education teachers only select students with better sport skills or performance', which hindered them from being physically active.[33] Another notable negative environmental factor was an 'uneven playground'. In the study by Matheri et al.,[46] 41% of the 234 adolescents with a disability reported that 'uneven playgrounds and compounds make it difficult to do exercise'.

DISCUSSION

The aim of this systematic review was to identify the factors that hinder or facilitate physical activity in children and adolescents with physical disabilities. A systematic search yielded 18 studies, including both qualitative[30–35] and quantitative[10,36–46] designs, in which a wide variety of factors were identified, with the majority originating from qualitative studies and being reported by children,[30,33] parents,[32] or a combination of the two.[31,34,35] Quantitative studies primarily examined the associations between factors and physical activity[36,38,40,42,46] or predictor variables for physical activity.[10,39,40,42–45] Three studies additionally or only reported descriptive statistics of factors.[37,41,46] The factors were distributed among all levels of the PAD model[28] and many could both hinder and promote physical activity, depending on the presence or absence of a certain factor.

The level of self-efficacy included some distinctive factors. A child's confidence seems to be an important positive factor associated with physical activity,[31,35,46] which may be influenced by factors related to health condition, such as motor skills or lack of them, as shown in the model. For example, if children are able to achieve the self-efficacy factors of 'being able to independently negotiate barriers in the community'[34] and 'teaching others to help',[34] they may overcome common environmental barriers; therefore, self-efficacy is imperative. At the same time, appropriate adaptive equipment for mobility and care[33,34,38,41,46] and appropriate opportunities at school,[46] at sport clubs,[31–33,35] and in the general community[30,31,34,35,38,41] seem to be important environmental requirements, whereas facilitating factors related to 'fitness'[40,42] and 'fundamental movement skills'[36] seem to be important personal requirements. Therefore, policy makers focusing their attention on appropriate adaptive equipment and opportunities may help to increase physical activity. Health care professionals can assist children in improving fitness and skills, but a focus on self-efficacy may also be beneficial, as it is more likely that children will continue physically active behavior if they are confident about their own abilities. Conversely, physical activity promotes self-efficacy, as adapted sports

programmes were reported to have a positive impact on children's confidence levels.[32] The results of this review are only partly comparable to results from the recent systematic review on perceived factors associated with physical activity for children with both cognitive and physical disability.[15] Only three of the included studies[30,32,38] in the review from Shields et al.[15] were also included in this review, because they included physical disabilities relevant to both reviews. We also included new studies in this review; the end date of our search was May 2013, compared with September 2010 in Shields et al.[15] Moreover, we included several quantitative studies reporting factors related to physical activity, which were excluded in the review from Shields et al.[15] Although this current review specifically sought factors associated with physical activity in children and adolescents with physical disabilities, we are aware that these children may also have some level of cognitive impairment. It was clear, however, that certain factors were identified more frequently or only for children with physical disabilities. Access to adequate equipment was reported only by children with physical disabilities[34,41] and positive factors related to attitude were more distinct.[30,35,46] In addition, it seems that children with intellectual or mental disabilities are more dependent on social influence to understand the importance of physical activity,[15] whereas children with physical disabilities generally have good personal attitudes towards physical activity.[31,33,35,46]

'Increasing age' was an important barrier specific to children and adolescents with physical disabilities.[10,34,43,44] Bearing in mind the likely long-term negative health consequences of physical inactivity in this population, we need to consider increasing age as a relevant factor associated with physical activity. As children and adolescents with physical disabilities progress into adulthood, they become more independent. Although young adults with physical disabilities still receive support from others, they more frequently reported personal factors as affecting their participation in physical activity, as described by Buffart et al.[47] In particular, the reasons why young adults

BARRIERS

FAMILY
- » Reliant on parents for transport and entrance to facilities (C)[31]
- » Lack of parental support (C)[33]
- » Lack of time of parent to assist (P)[34]
- » Parents not accepting extent of disability[35]
- » Parents concern for child's safety[34]
- » Parents feelings:
  - » Dissatisfaction with environment[35]
  - » Fear of child 'not fitting in'[35]
  - » Challenges with managing[35]
  - » Hesitating to ask for help from trainer(s)[35]
- ◊ People at home do not assist me[37]
- ◊ Low maternal level of education[45]

SUPPORT FROM AND CONTACT WITH PEOPLE
- » Not being accepted or being bullied by peers[31,33,35]
- » Not being accepted by other parents[35]
- » Teachers and instructors giving negative comments[31]
- » Lack of role models (C)[30]
- » Lack of PA partners to play with (C)[33]
- » Lack of professionals who know how to and can teach sport skills (C)[33]
- » Professionals are not assisting child in PA[34]
- ◊ Lack of trained staff/volunteer[38]
- ◊ People's misconception or unfriendly attitude[38]
- ◊ There's no one to do it with[46]

SPORTS
- » Activity type (competitive sports) (C)[31]
- » Activity not adequately adapted (P)[34,35]
- » Necessary equipment (C)[33]
- » Lack of opportunities (rules, regulation) (C)[30]
- » Sport facility:
  - » Teams too big[35]
  - » Waiting list[35]
  - » Being underestimated[35]
  - » Not allowed to play matches[35]
- ◊ Lack of clothes/(adapted) equipment[38,46]
- ◊ Where to exercise[38]
- ◊ Lack of LTE program[38]
- ◊ No space to exercise (with peers)[38]

ENVIRONMENT
- » Environment not adequately adapted (P)[34]
- ◊ Uneven playgrounds[46]
- ◊ Accessibility of facilities[38]
- ◊ Showering/changing facility[38]
- ◊ No suitable facilities[46]

SCHOOL
- » Lack of professional training in PE (C)[33]
- » PE teachers only select students with better sport skills or performance (C)[33]
- ◊ No suitable facilities at school[48]
- ◊ Having lot of homework[41]

TRANSPORT
- » Lack of (access to) transport[34,35]
- » Transport (living in more rural areas) (P)[31]
- ◊ Lack of transport[38,46]

FINANCIAL SUPPORT
- » Financial restrictions[35]
- » Financial commitment (P)[32]
- ◊ High cost[41]

OTHER
- » Consequences of stigma of disability (C)[30]
- » Time commitment (P)[32]
- ◊ Inappropriate weather[38,41]
- ◊ Too cold or hot[38]

FACILITATORS

FAMILY
- » Family resilience (P)[32]
- » Doing PA with parents[35]
- » Parental encouragement and motivation[31]
- » Parental support:[31,34,35]
- » Getting them to places[31]
- » Helping with transfers (P)[34]
- » Perseverance (in exploring options)[35]
- » Assertiveness (advocating for child)[35]
- » Having a positive attitude[35]
- ◊ Encouragement to be active[46]

SUPPORT FROM AND CONTACT WITH PEOPLE
- » Teachers + instructors supporting (P)[31]
- » Skilled helpers (P)[34]
- » Motivation from PE teacher + friends (C)[33]
- » Peer socialization (P)[34]
- » Being accepted by peers + other parents[35]
- ◊ Making friends[46]
- ◊ Positive attitude towards me by schoolmates, teachers and other people[37]
- ◊ Having someone who can provide support[41]
- ◊ Having someone with whom can do PA with[41]

SPORTS
- » Activity type (sports they enjoy doing)[31]
- » Chances to join competition (C)[33]
- » Opportunities for sport/PA[35]
- » Sport facility:[35]
  - » Having good trainer[35]
  - » Communication between trainers-coaches[35]
  - » Training in small groups[35]
- » Advantages of wheelchair sports (P):[32]
  - » Impact on child (confidence)[32]
  - » Community building[32]
  - » Friendship[32]
  - » Future aspirations[32]
- ◊ Having necessary equipment[41]
- ◊ Sportclub participation (on weekend days)[43]

ENVIRONMENT
- » Adaptive equipment (P)[34]
- » Access to sport/PA in c ommunity[31,35]
- ◊ Accessible community recreation facilities[41]
- ◊ Access to suitable facilities[46]

SCHOOL
- ◊ Participation in PE classes at school[46]

TRANSPORT
- » Transport (living close by the city) (P)[31]
- ◊ Having transportation[46]

OTHER
- » Appropriate group activities (P[34]
- » Disability-adopted programs (P)[34]
- » Opportunities away from home (C)[30]

Figure 4. *Environmental factors, barriers and facilitators of the included studies incorporated in the Physical Activity for People with a Disability model.*

» Qualitative Study Result
◊ Quantitative Study Result

PA Physical Activity
P Parents
C Children & Adolescents

When there is no indicator behind a factor, it means that the factor was mentioned by the parent or child in a qualitative study.

Barriers
Facilitators

with childhood-onset physical disabilities engaged in physical activity included motivational factors such as 'feelings of fulfillment and enjoyment', 'having a physical challenge', or 'wanting to achieve a goal', but there were also other factors such as 'maintaining a healthy body', 'functional independence', and 'physical appearance'.[47] Negative factors related to 'limited physical activity sports or facilities' and 'problems with transportation' were consistently reported among children and young adults, but young adults also reported that a 'lack of knowledge about where and how to exercise' and 'expensive equipment' or 'scarce second-hand assistive devices' kept them from being physically active.[47] This information seems to reaffirm the need for the focused attention of policy makers on adequate physical activity opportunities and appropriate adaptive equipment, and that motivation and confidence may be inevitable areas of focus for health professionals.

The current review used the PAD model to categorize and present the identified factors associated with physical activity.[28] The ASE model[29] incorporated herein is based on the theory of planned behavior,[48] which suggests that it is more likely that people will engage in physical activity when they have a positive attitude, perceive that there is social sup- port, and believe in their own ability to engage in physical activity.[28,48] The findings of this review seem to be consistent with this theory. By presenting the factors on different levels, it should be possible to develop interventions that may promote physical activity involving all these levels. However, a critical note has to be made when applying the PAD model. This model was originally developed for adults with a disability, and using this model for children and adolescents led to discussions about, for example, the factors of social influence. In young children, social influence mainly originates from the direct family,[31,32,35] whereas, as children get older, other people and the community seem to play a more important role.[47] Moreover, the results of this review demonstrated that, as age increases,[10,43,44] new negative factors such as 'more fear' and 'lack of motivation' are experienced.[34] As children progress into adulthood, the environment places greater demands that cannot always be fulfilled. Unfortunately, development in children and adolescents is difficult to categorize in the PAD model. In the future, the PAD model may be adjusted, or a new model may be developed, specifically for children and adolescents, with special emphasis on the developmental issues in paediatric research and care.

Future interventional research should aim to establish a practical guide based on these factors, which would help health care professionals realise individual approaches. It is obvious that no 'one-size-fits-all' concept is suitable for this population, but children with physical disabilities would experience greater benefit from individual approaches. As the reviewed literature originated from different countries, the factors and their strengths may differ; therefore, possible cultural differences should be taken into account when establishing such a guide.

The strengths of this review were that a sensitive search strategy was used, which produced an extensive yield of relevant literature, and 18 studies were located that met the selection criteria. Furthermore, every step in the study selection procedure, methodological quality assessment, data extraction, and data analysis was independently performed by at least two reviewers. The findings in the included studies were consistent with and applicable to the PAD model,[28] the use of which was also considered as a strength, since different levels could be addressed.

Some limitations should also be considered when interpreting the results of this review. Language and publication bias may be present because studies that were not written in English or Dutch were excluded, as were studies published before January 2000 and 'grey literature' (defined above). Only one[32] of the 18 included studies scored the maximum rating of three stars in the methodological quality assessment, which may have influenced the quality of the results of the studies. Although the McMaster Critical Review Forms[22–25] and the numerical rating criteria developed by Imms[26] are used in the literature, no information is available about their validity or reliability.[49] The relative importance of each factor should be considered, as the strength of the factors is mostly unclear and, therefore, it is uncertain which factors will be most important. Finally, there might be variability between the studies regarding the definition of physical activity, leading to different results.

CONCLUSION

The participation of children and adolescents with physical disabilities in physical activity is complex; a cluster of many factors exist that hinder or promote physical activity. An individualized approach in children and adolescents with physical disabilities, using the opportunities available to the child as a basis for increasing physical activity, may be an important element in future interventions. Increasing self-efficacy may also be of value, as it seems to support children in increasing physical activity behavior that may persevere throughout their life. Additional focus may be necessary during adolescence, as increasing age is an important negative factor that affects physical activity. Policy makers in schools, sport clubs, and the general community need to be aware of the importance of adequately adapted environments, and the availability of appropriate adaptive equipment and adapted sports in the child's own environment.

ACKNOWLEDGEMENTS

This research was funded by the Foundation Innovation Alliance – Regional Attention and Action for Knowledge circulation (SIA RAAK), project number 2011–13–35P, and by a personal PhD grant from the HU University of Applied Sciences Utrecht.

SUPPORTING INFORMATION

The following additional material may be found online:
Appendix SI: Search strategy PubMed.

REFERENCES

1. Physical activity, global health series.
Lancet. Available from: http://www.thelancet.com/series/physical-activity (accessed 18 July 2012).

2. Lee IM, Shiroma EJ, Lobelo F, et al.
Effect of physical inactivity on major non-communicable diseases worldwide: an analysis of burden of disease and life expectancy.
Lancet 2012; 380: 219–29.

3. Biddle SJ, Gorely T, Stensel DJ.
Health-enhancing physical activity and sedentary behaviour in children and adolescents.
J Sports Sci 2004; 22: 679–701.

4. 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: 1057–61.

5. Wilson PE, Clayton GH.
Sports and disability.
PM R 2010; 2: S46–54; quiz S55–6.

6. King G, Law M, King S, et al.
A conceptual model of the factors affecting the recreation and leisure participation of children with disabilities.
Phys Occup Ther Pediatr 2003; 23: 63–90.

7. 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: 533–45.

8. Rimmer JA, Rowland JL.
Physical activity for youth with disabilities: a critical need in an underserved population.
Dev Neurorehabil 2008; 11: 141–8.

9. Schoenmakers MA, de Groot JF, Gorter JW, et al.
Muscle strength, aerobic capacity and physical activity in independent ambulating children with lumbosacral spina bifida.
Disabil Rehabil 2009; 31: 259–66.

10. Maher CA, Williams MT, Olds T, et al.

Physical and sedentary activity in adolescents with cerebral palsy.
Dev Med Child Neurol 2007; 49: 450–7.

11. Buffart LM, Roebroeck ME, Rol M, et al.
Triad of physical activity, aerobic fitness and obesity in adoles- cents and young adults with myelomeningocele.
J Rehabil Med 2008; 40: 70–5.

12. Carlon SL, Taylor NF, Dodd KJ, et al.
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.

13. Neter JE, Schokker DF, de Jong E, et al.
The prevalence of overweight and obesity and its determinants in children with and without disabilities.
J Pediatr 2011; 158: 735–9.

14. Kotte EM, Winkler AM, Takken T.
Fitkids exercise therapy program in the Netherlands.
Pediatr Phys Ther 2013; 25: 7–13.

15. Shields N, Synnot AJ, Barr M.
Perceived barriers and facilitators to physical activity for children with disability: a systematic review.
Br J Sports Med 2012; 46: 989–97.

16. Maher CA, Williams MT, Olds T, et al.
An internet- based physical activity intervention for adolescents with cerebral palsy: a randomized controlled trial.
Dev Med Child Neurol 2010; 52: 448–55.

17. Van Wely L, Balemans AC, Becher JG, et al.
The effectiveness of a physical activity stimulation programme for children with cerebral palsy on social participation, self-perception and quality of life: a randomized controlled trial.
Clin Rehabil 2014; 28: 972–82.

18. Van Wely L, Balemans A, Becher J, et al.
Physical activity stimulation program for children with spastic cerebral palsy did not improve physical activity: a randomised controlled trial.
J Physiother 2014; 60: 40–9.

19. Grant MJ, Booth A.
A typology of reviews: an analysis of 14 review types and associated methodologies.
Health Info Libr J 2009; 26: 91–108.

20. GreyNet International.
Grey Literature Network Service.
Available from: http://www.greynet.org/ (accessed 9 July 2014).

21. Ket H, Riphagen I.
Pediatrics for PubMed, 2006.
Revised Ket H, 2009. http://www.medi- schebibliotheekznb.nl/Filters%20 PubMed.htm (accessed 16 April 2014).

22. Letts L, Wilkins S, Law M, et al. Guidelines for Critical Review Form – Qualitative Studies. Version 2.0. McMaster University, 2007.
Available from: http://www.srs- mcmaster.ca/Portals/20/pdf/ebp/ qualguidelines_version2. 0.pdf (accessed 9 July 2014).

23. Letts L, Wilkins S, Law M, et al.
Critical Review Form – Qualitative Studies. Version 2.0. McMaster University, 2007.
Available from: http://www.srs-mcmaster.ca/Portals/20/pdf/ebp/qualreview_ version2.0.pdf (accessed 9 July 2014).

24. Law M, Stewart D, Pollock N, et al.
Guidelines for Critical Review Form – Quantitative Studies. McMaster University, 1998.
Available from: http://www.srs-mcmas- ter.ca/Portals/20/pdf/ebp/ quanguidelines.pdf (accessed 9 July 2014).

25. Law M, Stewart D, Pollock N, et al. Critical Review Form – Quantative Studies. McMaster University, 1998. Available from: www.srs-mcmaster.ca/Portals/20/ pdf/ ebp/quanreview_form1.doc (accessed 9 July 2014).

26. Imms C.
Children with cerebral palsy participate: a review of the literature.
Disabil Rehabil 2008; 30: 1867– 84.

27. Netwerk Kwalitatief Onderzoek AMC – UvA.
Richtlijnen voor Kwaliteitsborging in Gezondheids(Zorg)Onderzoek: Kwalitatief Onderzoek. Amsterdam: Netwerk Kwalitatief Onderzoek AMC – UvA, 2002.

28. van der Ploeg HP, van der Beek AJ, van der Woude LH, et al.
Physical activity for people with a disability: a conceptual model.
Sports Med 2004; 34: 639–49.

29. de Vries H, Dijkstra M, Kuhlman P.
Self-efficacy: the third factor besides attitude and subjective norm as a predictor of behavioural intentions.
Health Educ Res 1988; 3: 273–82.

30. Anderson DM, Bedini LA, Moreland L.
Getting all girls into the game: physically active recreation for girls with disabilities.
J Park Recreat Admi 2005; 23: 78–103.

31. Barnett AL, Dawes H, Wilmut K.
Constraints and facilitators to participation in physical activity in teenagers with developmental co-ordination disorder: an exploratory interview study.
Child Care Health Dev 2013; 39: 393–403.

32. Hunter DE.
A phenomenological approach: the impact on families of sports participation for a child with a physical disability. (UMI Dissertations).
Ann Arbor, MI: ProQuest, 2010.

33. Li C, Chen S.
Exploring experiences of physical activity in special school students with cerebral palsy: a qualitative perspective.
Eur J Adapt Phys Activ 2012; 5: 7–17.

34. Luther BL.
Food and physical activity choices to prevent overweight in children with spina bifida. (UMI Dissertations).
Ann Arbor, MI: ProQuest, 2010.

35. Verschuren O, Wiart L, Hermans D, et al.
Identification of facilitators and barriers to physical activity in children and adolescents with cerebral palsy.
J Pediatr 2012; 161: 488–94.

36. Capio CM, Sit CHP, Abernethy B, et al.
Fundamental movement skills and physical activity among children with and without cerebral palsy.
Res Dev Disabil 2012; 33: 1235–41.

37. Frantz J, Phillips JS, Matheri JM, et al.
Physical activity and sport as a tool to include disabled children in Kenyan schools.
Sport Soc 2011; 14: 1227–36.

38. Kang M, Ragan BG, Zhu W, et al.
Exercise barrier severity and perseverance of active youth with physical disabilities.
Rehabil Psychol 2007; 52: 170–6.

39. Maltais DB, Pierrynowski MR, Galea VA, et al. P
hysical activity level is associated with the O2 cost of walking in cerebral palsy.
Med Sci Sports Exerc 2005; 37: 347–53.

40. Maltais DB, Pierrynowski MR, Galea VA, et al.
Habitual physical activity levels are associated with biomechanical walking economy in children with cerebral palsy.
Am J Phys Med Rehabil 2005; 84: 36–45.

41. Ortiz-Castillo E.
Physical activity patterns and factors influencing physical activity participation among adolescents with physical disabilities in urban communities.
(UMI Dissertations). Ann Arbor, MI, USA: ProQuest, 2012.

42. DR, Martin JJ.
Multidimensional physical self- concept of athletes with physical disabilities.
Adapt Phys Activ Q 2010; 27: 294–307.

43. van Wely L, Becher JG, Balemans AC, et al.
Ambulatory activity of children with cerebral palsy:
which characteristics are important?
Dev Med Child Neurol 2012; 54: 436–42.

44. van Eck M, Dallmeijer AJ, Beckerman H, et al.
Physical activity level and related factors in adolescents with cerebral palsy.
Pediatr Exerc Sci 2008; 20: 95–106.

45. Zwier JN, van Schie PE, Becher JG, et al.
Physical activity in young children with cerebral palsy.
Disabil Rehabil 2010; 32: 1501–8.

46. Matheri JM, Frantz JM.
Physical activity levels among young people with physical disabilities in selected high schools in Kenya and their perceived barriers and facilitators to participation.
J Commun Health Sci 2009; 4: 21–6.

47. Buffart LM, Westendorp T, van der Berg-Emons RJ, et al.
Perceived barriers to and facilitators of physical activity in young adults with childhood-onset physical disabilities.
J Rehabil Med 2009; 41: 881–5.

48. Ajzen I.
The theory of planned behavior.
Organ Behav Hum Dec 1991; 50: 179–211.

49. Crowe M, Sheppard L.
A review of critical appraisal tools show they lack rigor: alternative tool structure is proposed.
J Clin Epidemiol 2011; 64: 79–89.

9

Evidence for increasing physical activity in children with physical disability: a systematic review

Manon Bloemen[1,4], Leontien van Wely[2,4], Jurgen Mollema[1], Annet Dallmeijer[2,4], Janke de Groot[1,3,4]

1 Research Group Lifestyle and Health, HU University of Applied Sciences Utrecht, Utrecht, The Netherlands
2 Department of Rehabilitation Medicine and EMGO Institute for Health and Care Research, VU University Medical Center, Amsterdam, The Netherlands
3 Child Development and Exercise Center, Wilhelmina Children's Hospital, University Medical Center Utrecht, The Netherlands
4 On behalf of the Fit for the Future! Consortium

Dev Med Child Neurol *Accepted*

ABSTRACT

Aim

To summarize best evidence of interventions to increase physical activity (PA) in children with physical disabilities.

Method

A systematic review was conducted using an electronic search executed in in Academic Search Elite, Academic Search Premier, CINAHL, Embase, Medline, PeDro, PsychINFO and SPORTDiscus up to February 2016. Selection of articles has been performed independently by two researchers according to predetermined eligibility criteria. Data extraction, methodological quality and Levels of Evidence were independently assessed by two researchers using a data-collection form from the Cochrane Collaboration and according to the guidelines of the American Academy for Cerebral Palsy and Developmental Medicine.

Results

Seven studies were included. Five Randomized Controlled Trials ranged from strong Level-I to weak Level-II studies and two pre-post design studies were classified as Level-IV. There is Level-I evidence for no effect of physical training on objectively measured PA, conflicting Level-II evidence for interventions with a behavioral component on the increase of objectively measured PA directly after the intervention and Level-II evidence for no effect during follow-up. Results are limited to children with Cerebral Palsy as no other diagnoses were included.

Interpretation

Increasing PA in children with physical disabilities is very complex and demands further development and research.

WHAT THIS PAPER ADDS

- Physical training does not increase PA in youth with CP
- Behavioral interventions show conflicting evidence directly after the intervention
- Behavioral interventions show no increase in PA at follow-up

INTRODUCTION

Increasing physical activity (PA) in children and adolescents is a subject of great interest in pediatric research, with alarmingly growing rates of obesity and inactivity in youth. A physically active lifestyle is known to achieve fundamental health benefits for all individuals, including improved health-related quality of life, enhanced psychological well-being, improved physical functioning and prevention of non-communicable disease in both adults and children.[1-5] While being active is important for healthy children, children and adolescents with physical disabilities could benefit even more from regular PA in the prevention of not only comorbidity, but of functional decline and fatigue as well.[6-11] Despite these obvious benefits of PA, children with a physical disability are known to be at higher risk for an inactive lifestyle.[12-17]

Secondary to an inactive lifestyle, children with physical disabilities show reduced levels of cardiorespiratory fitness.[18, 19] Therefore, several studies have evaluated the effects of training in youth with physical disabilities. While these studies have shown positive results in measures of fitness, these studies also have shown that the increased levels of fitness do not sustain.[20, 21] This is not entirely surprising as these training programs were mostly aimed at increasing bodily functions such as fitness, and were not aimed at an increase in participation in PA. Given the benefits of PA in maintaining gains in fitness and other health benefits, however, it seems that sufficient levels of PA should also be a primary goal and outcome for pediatric practice to obtain durable effects. Increasing participation in PA requires behavioral change[22], as it is more than a skill as defined by the International Classification of Functioning, disability and health (ICF).[23] Factors associated with PA in youth with disability, such as self-efficacy and the presence of adaptive equipment, should ideally be incorporated into interventions aimed at increasing PA.[24-26] A recent paper describes the unique position of pediatric physical therapists in implementing interventions to achieve healthy active lifestyles in youth.[27]

While the evidence for effectiveness of interventions aiming to improve PA is still limited in healthy children[28, 29], the knowledge regarding children with physical disabilities seems even more limited and an overview of effective interventions is lacking. Therefore, the aim of this systematic review is to summarize best evidence of interventions to increase PA in children with physical disabilities. Results of this review will give more insight in effective ways to improve PA in pediatric rehabilitation practice.

METHODS

Search strategy

We conducted a systematic literature search up to and including February 2016 in the following electronic databases: MEDLINE, CINAHL, Academic Search Elite, Academic Search Premier, Embase, PEDro, PsycINFO and SPORTDiscus. In consultation with a medical information specialist (JM), a comprehensive search strategy was developed

around three major themes: 'children', 'disability' and 'physical activity'. Key terms within the search strategy were mapped to medical subject headings (MeSH) in Medline and expanded to include narrower terms. Also title and abstract search words were added to find the most recent, non-indexed literature. The MEDLINE search string was translated for the other databases. The complete search strategy for MEDLINE is presented in appendix 1. Finally, the reference lists of included studies were manually searched for additional studies.

Selection process and eligibility criteria

Titles, abstracts and full texts were independently reviewed for eligibility by two reviewers, with any discrepancies being discussed with a third reviewer until consensus was reached. Quantitative intervention studies were included if:

(1) the study participants were children (4-18 years, or a group mean age <18 years) with a physical disability;
(2) the studies were randomized controlled trials, clinical controlled trials or single-group designs;
(3) the interventions were part of pediatric physical therapy (such as physical training or exercising, task-oriented or functional training, interactive video gaming, promoting PA, coaching, motivational interviewing);
(4) one of the reported outcome measures was either objectively or subjectively assessed PA;
(5) it was a full-text peer reviewed article;
(6) it was written in English or Dutch.

Studies were excluded if:

- more than 50% of the participating children or adolescents did not have a physical disability and results were not presented separately,
- the physical disability was of a progressive nature or if conditions were present in which exacerbations could occur,
- it concerned single case reports or case series.

Data extraction

Relevant data were independently extracted by two reviewers, using an adapted data collection form from the Cochrane Library.30 Any discrepancies between reviewers were resolved by discussion until consensus was reached. The data were incorporated in a summary table according to the American Academy for Cerebral Palsy and Developmental Medicine (AACPDM) guidelines.31

Levels of evidence and methodological quality assessment of the manuscripts

The levels of evidence (LoE) and methodological quality according to the AACPDM were independently assessed by two researchers and any discrepancies between the reviewers were resolved by discussion until consensus was reached.[31] The LoE of the AACPDM are based on the LoE by Sacket[32] and range from level I (definite conclusions) to level V (no definite conclusions), as presented in table I. The studies that are classified as level I-III were assessed for methodological quality by seven criteria (see Table III). The ratings for methodological quality range from strong (6-7 criteria positive) to moderate (4-5 criteria positive) to weak (0-3 criteria positive).[31] The studies classified as LoE IV or V are not rated for methodological quality because of threats of internal validity due to the weak study design.[31]

Strength of the evidence

Due to heterogeneity of the studies a meta-analysis could not be performed. The outcomes, measures, and statistical results of the studies classified as LoE I-III are presented and analyzed as recommended by the AACPDM guidelines.[31] According to these guidelines the results should be classified according to the ICF into Body Structures / Body Functions, Activities and Participation, and Contextual Factors.[31]

Table 1. *Levels of Evidence for studies[31].*

LEVEL	INTERVENTION (GROUP) STUDIES
I	Systematic review of randomized controlled trials (RCTs) Large RCT (with narrow confidence intervals((n>100)
II	Smaller RCTs (with wider confidence intervals) (n<100) Systematic reviews of cohort studies "Outcome research" (very large ecologic studies)
III	Cohort studies (must have concurrent control group) Systematic reviews of case control studies
IV	Case series Cohort study without concurrent control group (e.g. with historical control group) Case-control study
V	Expert Opinion Case study or report Bench research Expert opinion based on theory or physiologic research Common sense / anecdotes

RESULTS

Search results

The electronic search resulted in 6071 studies. After screening title and abstract, 6001 studies were excluded. The remaining 70 studies were analyzed full text, of which another 62 were excluded, mostly because they did not report effects on PA. One additional study[33] was excluded because it was a feasibility study and did not have the objective to measure the effect of the intervention. The remaining seven studies[34-40] were reviewed full text, summarized in a data extraction form and included in this systematic review. Five[35-38, 40] of these seven studies were Randomized Clinical Trials (RCTs) and two studies[34, 39] used a single-group design. One study[38] was a RCT during the first year of the trial and continued with a single-group design during the second year of the trial. Manual search of the reference lists of the included studies identified no additional studies. The selection process is presented in figure 1.

Data extraction

The study characteristics of the studies are reported in table II (page 166 -167).
All seven studies[34-40] included children with Cerebral Palsy (CP) Gross Motor Function Classification System I-II[40] and I-III[34-37] and I-V[38-39], with ages ranging from 6[35] to seventeen[40] years of age. Sample sizes ranged from 15[35] to 102.[40]

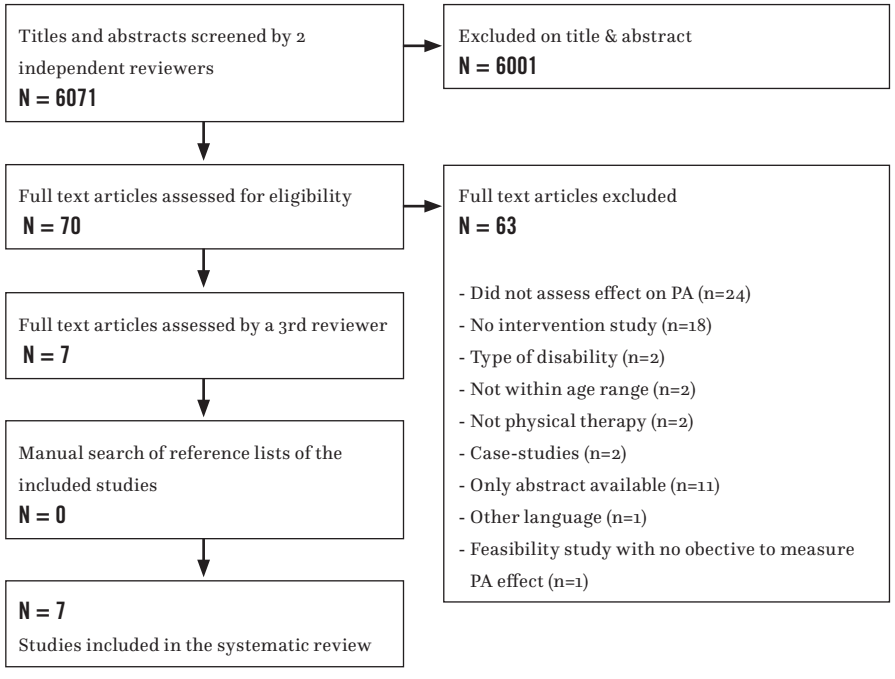

Figure 1. *Overview of the selection process.*

Levels of evidence and methodological quality

The LoE of the studies varied from Level I[40] to Level II[35-38] to Level IV[34, 38, 39] and is reported in table II (page 166-167). The first part of the study from van den Berg-Emons et al. was classified as a RCT and the last part as a pre-postdesign.[38] The methodological quality of the included studies classified as LoE I-III is reported in table III. One Level-I RCT[40] and one Level-II RCT[37] were rated as strong methodological quality. Two level-II RCTs were rated as moderate methodological quality[36, 38] and one as weak methodological quality[35].

Content of the interventions of all included studies

There was a large variation in the content of the interventions. Five studies focused on physical training[34, 35, 38-40] such as strengthening, endurance training and balance

Table 3. *Methodological quality of the studies with level of evidence I-III.*

STUDY	LEVEL / QUALITY	I	II	III	IV	V	VI	VII
1998[38] Van den Berg-Emons et al.	II–M (4/7)	No	Yes	Yes	No	No	Yes	Yes
2007[35] Crompton et al.	II-W (3/7)	Yes	Yes	No	Yes	No	No	No
2010[36] Maher et al.	II-M (5/7)	Yes	Yes	No	Yes	Yes	Yes	No
2014[37] van Wely et al.	II-S (7/7)	Yes	Yes	Yes	Yes	Yes	Yes	Yes
2016[40] Mitchell et al.	I-S (6/7)	Yes	Yes	No	No	Yes	Yes	Yes

W = Weak | M = Moderate | S = Strong

1. Were inclusion and exclusion criteria of the study population well described and followed?
2. Was the intervention well described and was there adherence to the intervention assignment? For 2-group designs, was the control exposure also well described?
3. Were the measures used clearly described, valid and reliable of measuring the outcomes of interest?
4. Was the outcome assessor unaware of the intervention status of the participants (i.e. were there blind assessments)?
5. Did the authors conduct and report appropriate statistical evaluation including power calculations?
6. Were dropouts / loss to follow-up reported and less than 20%? For 2-group designs, were dropouts balanced?
7. Considering the potential within the study design, were appropriate methods for controlling confounding variables and limiting potential biases used?

Table 2. *Summary of studies – interventions and participants.*

STUDY	LEVEL OF EVIDENCE AND CONDUCT RATING RESEARCH DESIGN	PARTICIPANTS	TOTAL N	AGES	INTERVENTION	CONTROL INTERVENTION
1998[38] Van den Berg-Emons et al.	II-M (4/7) Small RCT	Spastic CP 16 diplegic and 4 tetraplegic 10 ambulant and 10 wheel-chair-bound. Normal intelligence and mild mental retardation. 12 male, 8 female	20 ig=10 cg=10	7-13 y ig: mean 9.5 y (SD 1.6) cg: mean 8.8 y (SD 1.1)	Training intervention → Aerobic exercises. - 9 months, 4xpw 45 min. - usual care continued.	Normal therapy program (varied from no therapy to > 2.5 hrs/ week).
1998[38] Van den Berg-Emons et al.	IV Pre-test / Post-test with no control group	CP ambulant and wheelchair-bound. Normal intelligence and mild mental retardation.	18	7-13 y	Training intervention → Aerobic exercises. - 9 months, 2xpw 45 min. - usual care continued.	Na
2007[35] Crompton et al.	II-W (3/7) Small RCT	Spastic bilateral CP, GMFCS I-III 6 male, 9 female	15 ig=7 cg=8	6-14 y ig: mean 9.9 y (SD 2.5) cg: mean 11.2 y (SD 1.9)	Training intervention → Task-related lower limb group training. Activities incorporated elements of strength, coordination and balance. Circuit with 7 exercise stations. Submaximal effort of 70% from maximum, 4 minutes at every station. - 6 weeks, 2xpw 1 hour - usual care continued.	Task-related upper limb dexterity group training. Activities requiring grasp and manipulation of objects. -6 weeks 2xpw 1 hour -three activities of 10-20 minutes each. To develop speed, "mad minutes" were incorporated; working as fast as possible. -usual care continued.
2010[36] Maher et al.	II-M (5/7) Small RCT	CP, GMFCS I-III 16 unilateral, 25 bilateral 25 male, 16female	41 Ig=20 Cg=21	11-16 y, mean 13;7 y (SD 1.8)	Intervention with behavioral component → Highly interactive intenet-based program Get Set, based on social cognitive theory, incorporating education, quizzes, goal-setting, self-reflection, positive role-modeling: - 8 weekly released modules onto site. - One-on-one introduction to site - weekly e-mail or mobile phone text messages - usual care continued.	Usual care
2012[34] Christy et al.	IV Pre-test / Post-test with no control group	CP, GMFCS I-III 12 spastic, 3 hypotonic, 1 athetoid, 1 ataxic. 8 diplegia, 7 quadriplegia, 2 triplegia. 8 male, 9 female	17	4-12 y Mean 7.6 y (SD 2.9)	Training intervention → Intensive physical therapy, focusing on stretching, strengthening, balance, gait training, endurance training. Modalities (TheraSuitTM, cage, weights) of the TheraSuitTM protocol were used. - 3 weeks, 5xpw 4hrs	Na
2013[39] Maniu et al.	IV Pre-test / Post-test with no control group	CP, GMFCS II-V 19 spastic (5 paraparesis, 10 tetraparesis, 4 hemiparesis), 4 dyskinetic, 1 ataxic. 18 male, 6 female	24	8-16 y Mean 12.5 y (SD 2.7)	Training intervention → Aquatic therapy program - 6 months, 2xpw 45 min. - usual care continued.	Na
2014[37] van Wely et al.	II-S (7/7) Small RCT	Spastic CP, GMFCS I-III 33 unilateral, 26 bilateral 28 male, 21 female	49 ig=25 cg=24	7-13 y Ig: mean 9.5 y (SD 1.5) cg: mean 10.0 y (SD 1.8)	Intervention with behavioral component → PA stimulation program, consisted of - 6 months lifestyle intervention (counseling sessions with child and parents at home and home-based physical therapy) - 4 months fitness program in small groups with 2 lower extremity strength exercises and 3 anaerobic exercises (first 2 months 2xpw 1 hr, last 2 months 1xpw 1hr)	Usual pediatric physical therapy at physical therapy practice.
2016[40] Mitchell et al.	I-S (5/7) Large RCT	Spastic unilateral CP, GMFCS I-II 53 male, 49 female	102 ig=51 cg=51	8-17 y Ig: mean 11.3 y (SD 2.4) cg: mean 11.4 y (SD 2.6)	Training intervention → Web-based training - physical activity games interspersed with upper-limb and visual-perceptu al games. - 40% Gross-motor exercises, including sequences of repetitive multi-joint bodyweight functional exercises, such as sit-to-stand and lunging). Lower-limb strength exercises started at approximately 75% from maximum. Weekly increased by physiotherapists, based on individual need. - 20 weeks, 6xpw 30 min. - usual care continued.	Usual care

LEGEND
Ig = intervention group | cg = control group | PA = physical activity
RCT = Randomized Controlled Trial | CP = Cerebral Palsy | y = years
SD = standard deviation | pw = per week | min = minutes | hrs = hours
na = not applicable | GMFCS = Gross Motor Function Classification System
M = moderate | W = weak | S = strong

Table 4. *Summaries of studies with level of evidence I-III: outcomes, measures, and results.*

			COMPONENTS OF HEALTH		
STUDY	**OUTCOME OF INTEREST**	**MEASURE**	**BODY STRUCTURE(S) / FUNCTION(S)**	**ACTIVITIES AND PARTICIPATION**	**CONTEXTUAL FACTORS**
1998[38] van den Berg-Emons et al.	**Objective PA:** TEE, determined from the heart rate according to the FLEX principle (1 weekday)	- ratio of TEE to sleeping metabolic rate or resting metabolic rate. T0 = baseline T1 = 2 months, on a training day (= during intervention) T2 = 9 months, on a training day (= end of intervention)		T1 and T2: ns	
2007[35] Crompton et al.	**Objective PA:** positional activity logger (2 weekdays & 2 weekend days)	- uptime: time in upright position (standing and walking) - mean uptime (hrs/day) T0 = baseline T1 = 6 weeks (=post-intervention) T2 = 12 weeks (= follow-up)		T1 and T2: ns T1 and T2: ns	
2010[36] Maher et al.	**Objective PA:** activity monitor NL-1000 AM (1 week) ***Subjective PA:*** *MARCA (4-days)*	- weekly step counts - weekly MVPA min. - weekly walking distance - *self-reported PA* - *average daily PA level* - *average daily MVPA* T0 = baseline T1 = 10 weeks (=post-intervention) T2 = 20 weeks (= follow-up)		T1 and T2: ns T1 and T2: ns T1: p=0.05 ig / T2:ns *T1 and T2: ns* *T1 and T2: ns* *T1 and T2: ns*	
2014[37] van Wely et al.	**Objective PA:** StepWatch AM (1 week) ***Subjective PA:*** *child-adapted AQuAA (parent-reported PA 7 days)*	- number of strides per day - time spent at medium-to-high stride rate (>15 strides /min) - time spent at high stride rate (>30 strides /min) - *weekly times spent at MVPA* - *PA norm* T0 = baseline T1 = 6 months (=post-intervention) T2 = 12 months (= follow-up)		T1 and T2: ns T1 and T2: ns T1 and T2: ns *T1: p<0.05 ig / T2: ns* *T1 and T2: ns*	
2016[40] Mitchell et al.	Objective PA: ActiGraph GT3X+ tri-axial AM (2 weekdays & 2 weekend days)	- time spent at light activity (101-2295 vertical counts/min) - time spent at MVPA (2296–4011 vertical counts/min) - activity counts - number of steps T0 = baseline T1 = 20 weeks (=post-intervention)		T1: ns T1: ns T1: ns T1: ns	

PA = physical activity | TEE = Total Energy Expenditure | AM = activity monitor | ns = not significant | MVPA = moderate to vigorous physical activity | Min = Minutes

training. The training volumes varied tremendously, from 20 hours a week for 3 weeks[34] to 3 hours per week for 9 months[38]. All these studies[35, 38-40] continued the usual therapy program during the intervention, except for Christy et al.[34] The two other studies[36, 37] focused on a more behavioral[36] or combined behavioral and physical training[37] approach. Maher et al.[36] used an highly interactive internet-based program 'Get Set', based on the cognitive theory, whereas Van Wely et al.[37] used a face-to-face lifestyle intervention, in which they combined group fitness with individual counseling and individual home-based physical therapy. Total length of the program 'Get Set' was 8 weeks and usual physical therapy was continued[36] whereas the lifestyle intervention from Van Wely et al.[37] lasted six months.

Outcome measures of PA of the studies classified as LoE I-III

The outcome measures of the five studies classified as LoE I and II are presented in table IV. Within the focus of this review, the results on PA are described under "Activity and Participation" according to the ICF.[23] All studies[35-38, 40] reported measurements of PA before the intervention and directly after the intervention, with three studies[35-37] also measuring PA during follow up, ranging from 6 weeks[35] to 6 months[37]. Four studies[35-37, 40] measured objective PA by activity monitors, however, a variety of instruments was used and the wear time of the activity monitors varied from four days[35, 40] to one week[36, 37]. Moreover, a wide variety of outcome measures from the activity monitors was used. Crompton[35] defined PA as uptime using a positional activity logger. Other activity monitors were used to report step counts[36, 37, 40], time in light activity[40], time in moderate to vigorous PA (MVPA)[36, 40], weekly walking distance[36], activity counts[40], time spent at medium-to-high stride rate[37], time spent at high stride rate[37]. Van den Berg-Emons et al.[38] measured PA objectively using total energy expenditure.

Two studies[36, 37] measured subjective PA. Maher et al.[36] used the MARCA, a self-reported PA questionnaire about 4 previous days and van Wely et al.[37] used the parent-reported child-adapted AQuAA, with questions about the frequency and duration of PA during the previous 7 days. In addition, both studies used self-reported weekly[37] or daily[36] moderate to vigorous physical activity, self-reported daily level of PA[36] or self-reported compliance with the PA norm[37].

Effects of interventions on PA of the studies classified as LoE I-III

Of the five RCTs[35-38, 40] who measured objective PA, only one study[36] found an effect for objectively measured PA directly after the intervention. They[36] reported an increase in weekly walking distance (effect size 0.96, p=0.05) in favour of the intervention group receiving the internet-based program 'Get Set' compared to usual care. No study reported significant differences during follow-up.[35-37]

Of the two RCTs[36, 37] who measured subjective PA, only one study[37] reported a significant difference at the end of the intervention for parent-reported time at MVPA in favour of the lifestyle intervention compared to usual pediatric physical therapy (between-group change ratio 2.2; 95% CI 1.1-4.4, p=0.04). No significant differences were found at follow-up.[36, 37]

Strength of the evidence

Looking at the content of the interventions of the studies classified as Level I-III, two groups of interventions can be identified: 1) interventions focusing on physical training alone[35, 38, 40] and 2) interventions using a behavioral component[36, 37]. The strength of the evidence is separately analysed for these two groups of interventions and also for objectively and subjectively measured PA. Results are limited to children with CP as no other diagnoses were included.

Physical training

There is one strong Level I study40, one moderate Level II study[38] and one weak Level II study[35] showing no effect, resulting in Level I evidence for no effect of physical training on objectively measured PA in children with CP.

Behavioral component

There is one strong Level II study[37] showing no effect and one moderate Level II study[36] showing a significant effect, resulting in conflicting evidence for interventions with a behavioral component on the increase of objectively measured PA directly after the intervention. Both studies[36, 37] found no effects at follow-up, resulting in Level II evidence for no effect.

With regard to subjectively measured PA one strong Level II study[37] showed a significant effect and one moderate Level II study[36] showed no effect, resulting in conflicting evidence for the increase of subjectively measured PA. As both studies[36, 37] found no effects at follow-up there is Level II evidence for no effects of interventions with a behavioral component on subjectively measured PA during follow-up.

DISCUSSION

The aim of this systematic review was to summarize best evidence of interventions to increase PA in children with physical disabilities in order to provide more insight in effective ways to improve PA in pediatric practice. In this search only seven[34-40] studies were identified; four were RCT's[35-37, 40], two were single-group design studies[34, 39] and one study[38] used a RCT design in the first part of their study and a single-group design in the last part of their study. Five studies[35-38, 40] were classified as Level I-III studies according to the LoE and only results of these studies were included in weighing the strength of the evidence.

Interestingly, all studies included only children with CP. Compared to a systematic review from Bania et al.[41] who analysed the effects of interventions on PA in people with CP and included both quantitative and qualitative studies, four additional studies[34, 37, 39, 40] were included in our review. Bania et al. concluded that structured exercise programmes and online behavioral programs can be effective in increasing PA, but that the results are not maintained.[41] Our evidence showed Level I evidence for no effect of physical training on objectively measured PA in children with CP, conflicting evidence for the effect of interventions with a behavioral component on objectively and subjectively measured PA directly after the intervention and Level II evidence for no effect at follow-up. Unfortunately, the evidence in our review seems less positive than the conclusion from Bania et al.[41] several years ago. It was striking to learn that, even though increasing PA in children with physical disabilities is an important topic in pediatric practice, the knowledge base is still very small and only evidence for children with CP was found.

We found a tremendous variation in the interventions that were applied. It was intriguing to notice that five studies used solely physical training[34, 35, 38-40] and thus focussed on the level of bodily functions of the ICF. This focusing on bodily functions still seems to predominate goal setting and intervention in physical therapy practice. However, both literature[20, 21] and studies included in this review[34, 35, 38-40] show this is insufficient in attaining goals at the level of participation, such as an increase in PA in daily life. This underlines the importance to shift our focus from the level of bodily functions to the level of participation as defined by the ICF and thus taking personal and external factors into account.[23]

Two of the included studies[36, 37] integrated behavioral models in their interventions, with only Maher et al.[36] showing a significant difference in only one objective outcome measure of PA directly after the intervention. Unfortunately, this increase of PA did not sustain until follow-up. Change in participation involves a change in behavior and when aiming to change certain behaviors, models of behavioral change should be integrated in physical therapy practice. One of these models, specifically aimed at PA, is the model of PA for persons with a disability (PAD) by van der Ploeg et al. and combines the ICF with the model of Attitude, Social Influence and Self-Efficacy (ASE).[31] In this model, the intention for being physically active is the central determinant for participation in PA with aspects as attitude, self-efficacy, health condition and facilitators and barriers as personal components and social influence and facilitators and barriers as external components. Using a combination of behavioral models and the ICF is also recommended by Johnston & Dixon.[42] These authors have tested the prediction of behavior using both the ICF, behavioral models and a combination of the ICF and behavioral models and found that the latter performed best in predicting behavior. Interestingly, these authors also hypothesized that non-volitional determinants such as impairments are stronger related to measures of limitations and capacity, whereas volitional determinants are stronger related to measures of performance, in this case participation in PA.[42] This is very much in line with recent studies[24, 25] looking at factors associated with participation in PA. Focus on problem solving, self-efficacy and the presence of positive personal and environmental factors seem crucial for participation in PA.[24, 25] With this emphasis for combining behavioral models with the ICF in mind, what can we learn from the studies[36, 37] using behavioral models in their interventions and why are the effects so marginal?

The "Get Set program" form Maher et al.[36] was described as an eight week "highly interactive internet-based program based on social cognitive theory". The retention rate during the trial was high, so why was there only an effect on one objective PA measure and why did the increase of PA not maintain? An important aspect may be the duration of the program, as it is known that behavioral changes take time to occur and it also takes time and effort to truly maintain these behavioral changes.[43] Another essential aspect may be the fact that a "one size fits all" program may not be the answer, as each child and his or her parents experience specific facilitators and barriers for participation in PA that need to be individually addressed.[24, 25] So, taking this individual approach into account, it is interesting to see that there were no significant findings for objectively measured PA in the RCT from van Wely et al.[37], despite their individually tailored approach. Their intervention consisted of motivational interviewing, together with fitness training and regular home-based physical therapy. Motivational interviewing is a client-centered interview style aiming at behavioral change and all participants received a minimum of three counselling sessions.

However, it is interesting to see that the attitudes towards for example sports of both the participating children and parents were positive from baseline, so the question arises if motivational interviewing was the best treatment option for every participant or if another approach would have been more successful. Moreover, the home-based physical therapy was aimed at increasing the capacity for daily activities. But performance in PA is much more complicated than just capacity for daily activities, as shown by the wide variety of factors associated with PA.[24, 25, 44] So it seems that the content of the home-based physical therapy may not have been focused enough on specific facilitators or barriers that may have been present and are part of the personal and environmental factors as described by both the ICF and the PAD-model. Furthermore, even though the whole intervention lasted for 6 months, this may still not have been long enough to indeed see behavioral change.[43]

Given the very limited evidence for effective interventions to improve PA in children with physical disabilities, it is very interesting to consider future directions. We believe that increasing PA levels in children with physical disabilities is very complex and needs an individual approach. The specific barriers and facilitators, which differ for every child because of differences in context and differences in personal factors, should be analyzed thoroughly.[24, 25] Clinical reasoning should than lead to an individual hypothesis about the causes of the reduced levels of PA. By doing so, the intervention that fits the hypothesis can be identified. Possible future interventions may be developed in co-design[45, 46] with the children with physical disabilities and their parents. This is a relatively new approach that shows positive findings in other healthcare areas.[47-49] These new interventions should first be piloted in for example case studies or case series, after which the interventions can be further developed and improved. If these stages are completed, larger effect studies may be undertaken to analyze the effect on a group level.

An important limitation is that outcome measures for the assessment of PA are limited in their clinimetric properties and feasibility. Both objective and subjective measures were used with results and are not interchangeably, due to low agreement between the two methods.[50] Even more, measuring PA in daily pediatric practice is still a challenge and not implemented within regular care yet.[51]

This systematic review used an extensive and sensitive search in order to identify all possible studies analyzing the effects of interventions on PA in children with physical disabilities. A strength of this review was that both the selection of studies, data extraction and methodological quality assessment were performed by two independent reviewers. Certain limitations should be taken into account when interpreting the results. For example only English and Dutch articles were included, so results from studies using another language may have been missed. Moreover, we chose to exclude qualitative studies, case reports and case-series because we wanted to report statistical differences on a group level. These qualitative studies, case reports and case-series may give insight however, in new possibilities to increase PA in children with physical disabilities. Another important aspect is that only children with CP were included in the studies even though all non-progressive physical disabilities were allowed in this review, of course limiting the generalizability of our results.

CONCLUSION

In conclusion, there is Level I evidence for *no* effect of physical training alone on objectively measured PA in children with CP, conflicting evidence for the effect of interventions with a behavioral component on objectively and subjectively measured PA direct after the intervention and Level II evidence for no effect at follow-up.

ACKNOWLEDGEMENTS

Hanneke Borst and Joyce Benner are acknowledged for their input into the search, data selection, and methodological quality process. This research was funded by Foundation Innovation Alliance - Regional Attention and Action for Knowledge circulation (SIA RAAK PRO), project number SIA RAAK PRO-4-03, and by a personal PhD grant from HU University of Applied Sciences Utrecht. SIA-RAAK had no role in the design, data collection, analysis and interpretation, reporting of this work, or the decision to submit the work for publication. The authors have stated that they had no interest that could be perceived as posing a conflict or bias.

APPENDIX 1

Pubmed total:
(((((((("Child"[Mesh] OR child*[tiab] OR "Minors"[Mesh] OR “minors”[tiab] OR "Puberty"[Mesh] OR "puberty"[tiab] OR "Pediatrics"[Mesh] OR paediatric*[tiab] OR pediatric*[tiab] OR "Adolescent"[Mesh] OR adolescen*[tiab] OR preschool*[tiab] OR "teenager"[tiab] OR "teenagers"[tiab] OR "teen"[tiab] OR "teens"[tiab] OR youth*[tiab] OR "girlhood"[tiab] OR "girl"[tiab] OR "girls"[tiab] OR "boyhood"[tiab] OR "boy"[tiab] OR "boys"[tiab] OR "school age"[tiab] OR "school-aged"[tiab] OR schoolchild*[tiab] OR "kid"[tiab] OR "kids"[tiab] OR underage*[tiab] OR juvenile*[tiab])) AND ("Spinal Dysraphism"[Mesh] OR "spina bifida"[tiab] OR "Cerebral Palsy"[Mesh] OR "cerebral palsy"[tiab] OR "spastic diplegia"[tiab] OR "little disease"[tiab] OR "Motor Skills Disorders"[Mesh] OR "motor skills disorder"[tiab] OR "motor skills disorders"[tiab] OR "motor impairment"[tiab] OR "motor impairments"[tiab] OR "developmental coordination disorder"[tiab] OR "developmental coordination disorders"[tiab] OR "developmental disorder"[tiab] OR "developmental disorders"[tiab] OR "Developmental Disabilities"[Mesh] OR "developmental disability"[tiab] OR "developmental disabilities"[tiab] OR "physically challenged"[tiab] OR handicap*[tiab] OR "disabled"[tiab] OR "disability"[tiab] OR "disabilities"[tiab] OR "mobility impairment"[tiab] OR "mobility impairments"[tiab] OR "physically impaired"[tiab] OR "physical impairment"[tiab] OR "physical impairments"[tiab]))) OR "Disabled Children"[mesh])) AND ("Accelerometry"[Mesh] OR "accelerometry"[tiab] OR "accelerometer"[tiab] OR "stepwatch"[tiab] OR "pedometer"[tiab] OR "activity monitor"[tiab] OR "Actigraphy"[Mesh] OR actigraph*[tiab] OR "physical recreation"[tiab] OR exercis*[tiab] OR "motor activity"[tiab] OR "motor activities"[tiab] OR "energy expenditure"[tiab] OR "Sports"[Mesh] OR "sport"[tiab] OR "sports"[tiab] OR "Motor Activity"[Mesh] OR "physical activity"[tiab] OR "physical activities"[tiab]))) NOT (addresses[pt] OR autobiography[pt] OR bibliography[pt] OR biography[pt] OR "case reports"[pt] OR "classical article"[pt] OR "clinical conference"[pt] OR “collected works”[pt] OR comment[pt] OR “comparative study”[pt] OR congresses[pt] OR “consensus development conference”[pt] OR dataset[pt] OR dictionary[pt] OR directory[pt] OR “duplicate publication”[pt] OR editorial[pt] OR “english abstract”[pt] OR festschrift[pt] OR “government publications”[pt] OR guideline[pt] OR “historical article”[pt] OR “interactive tutorial”[pt] OR interview[pt] OR lectures[pt] OR “legal cases”[pt] OR legislation[pt] OR letter[pt] OR news[pt] OR “newspaper article”[pt] OR overall[pt] OR “patient education handout”[pt] OR “periodical index”[pt] OR “personal narratives”[pt] OR portraits[pt] OR “practice guideline”[pt] OR “published erratum”[pt] OR "research support, american recovery and reinvestment act"[pt] OR "research support, N.I.H., extramural"[pt] OR "research support, N.I.H., intramural"[pt] OR “research Support, non-U.S. Gov't”[pt] OR “Research Support, U.S. Gov't, Non-P.H.S.”[pt] OR “research support, U.S. Gov't, P.H.S.”[pt] OR “retracted publication”[pt] OR “retraction of publication”[pt] OR “support of research”[pt] OR “technical report”[pt] OR “twin study”[pt] OR “video-audio media”[pt] OR webcasts[pt])

REFERENCES

1. Demuth SK, Knutson LM, Fowler EG.
The PEDALS stationary cycling intervention and health-related quality of life in children with cerebral palsy: a randomized controlled trial.
Dev Med Child Neurol 2012;54: 654-661.

2. Peterson MD, Saltarelli WA, Visich PS, Gordon PM.
Strength capacity and cardiometabolic risk clustering in adolescents.
Pediatrics 2014;133: e896-903.

3. Grontved A, Ried-Larsen M, Moller NC, Kristensen PL, Wedderkopp N, Froberg K, et al.
Youth screen-time behaviour is associated with cardiovascular risk in young adulthood: the European Youth Heart Study.
Eur J Prev Cardiol 2014;21: 49-56.

4. Berentzen NE, Smit HA, van Rossem L, Gehring U, Kerkhof M, Postma DS, et al.
Screen time, adiposity and cardiometabolic markers: mediation by physical activity, not snacking, among 11-year-old children.
Int J Obes (Lond) 2014;38: 1317-1323.

5. O'Donovan G, Blazevich AJ, Boreham C, Cooper AR, Crank H, Ekelund U, et al.
The ABC of Physical Activity for Health: a consensus statement from the British Association of Sport and Exercise Sciences.
J Sports Sci 2010;28: 573-591.

6. King G, Law M, King S, Rosenbaum P, Kertoy MK, Young NL.
A conceptual model of the factors affecting the recreation and leisure participation of children with disabilities.
Phys Occup Ther Pediatr 2003;23: 63-90.

7. de Vries AG, Huiting HG, van den Heuvel ER, L'Abee C, Corpeleijn E, Stolk RP.
An activity stimulation programme during a child's first year reduces some indicators of adiposity at the age of two-and-a-half.
Acta Paediatr 2015;104: 414-421.

8. Rimmer JA, Rowland JL.
Physical activity for youth with disabilities: a critical need in an underserved population.
Dev Neurorehabil 2008;11: 141-148.

9. Rimmer JH, Rowland JL, Yamaki K.
Obesity and secondary conditions in adolescents with disabilities: addressing the needs of an underserved population.
J Adolesc Health 2007;41: 224-229.

10. Peterson MD, Gordon PM, Hurvitz EA.
Chronic disease risk among adults with cerebral palsy: the role of premature sarcopoenia, obesity and sedentary behaviour.
Obes Rev 2013;14: 171-182.

11. Keawutan P, Bell K, Davies PS, Boyd RN.
Systematic review of the relationship between habitual physical activity and motor capacity in children with cerebral palsy.
Res Dev Disabil 2014;35: 1301-1309.

12. Maher CA, Williams MT, Olds T, Lane AE.
Physical and sedentary activity in adolescents with cerebral palsy.
Dev Med Child Neurol 2007;49: 450-457.

13. 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.

14. Buffart LM, Roebroeck ME, Rol M, Stam HJ, van den Berg-Emons RJ, Transition Research Group South-West Netherlands.
Triad of physical activity, aerobic fitness and obesity in adolescents and young adults with myelomeningocele.
J Rehabil Med 2008;40: 70-75.

15. van den Berg-Emons HJ, Bussmann JB, Meyerink HJ, Roebroeck ME, Stam HJ.
Body fat, fitness and level of everyday physical activity in adolescents and young adults with meningomyelocele.
J Rehabil Med 2003;35: 271-275.

16. 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: 647-655.

17. Bjornson KF, Belza B, Kartin D, Logsdon R, McLaughlin JF.
Ambulatory physical activity performance in youth with cerebral palsy and youth who are developing typically.
Phys Ther 2007;87: 248-257.

18. Balemans AC, Van Wely L, De Heer SJ, Van den Brink J, De Koning JJ, Becher JG, et al.
Maximal aerobic and anaerobic exercise responses in children with cerebral palsy.
Med Sci Sports Exerc 2013;45: 561-568.

19. 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: 2-14.

20. 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;161: 1075-1081.

21. 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;25: 597-606.

22. van der Ploeg HP, van der Beek AJ, van der Woude LH, van Mechelen W.
Physical activity for people with a disability: a conceptual model.
Sports Med 2004;34: 639-649.

23. Leonardi M, Martinuzzi A.
ICF and ICF-CY for an innovative holistic approach to persons with chronic conditions.
Disabil Rehabil 2009;31 Suppl 1: S83-7.

24. 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;57: 137-148.

25. 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;15: 11-015-0265-9.

26. Shields N, Synnot AJ, Barr M.
Perceived barriers and facilitators to physical activity for children with disability: a systematic review.
Br J Sports Med 2012;46: 989-997.

27. Rowland JL, Fragala-Pinkham M, Miles C, O'Neil ME.
The scope of pediatric physical therapy practice in health promotion and fitness for youth with disabilities.
Pediatr Phys Ther 2015;27: 2-15.

28. Dobbins M, Husson H, DeCorby K, LaRocca RL.
School-based physical activity programs for promoting physical activity and fitness in children and adolescents aged 6 to 18.
Cochrane Database Syst Rev 2013;2: CD007651.

29. Stone EJ, McKenzie TL, Welk GJ, Booth ML.
Effects of physical activity interventions in youth. Review and synthesis.
Am J Prev Med 1998;15: 298-315.

30. Higgins JPT, Green S.
(2011) Cochrane Handbook for Systematic Reviews of Interventions.
Version 5.1.0 [updated March 2011].
www.cochrane-handbook.org: The Cochrane Collaboration. (accessed February 2014.

31. Darrah J, Hickman R, O'Donnell M, Vogtle L, Wiart L. AACPDM
Methodology to Develop Systematic Reviews of Treatment Interventions
(Revision 1.2), 2008 Version.
https://www.aacpdm.org/UserFiles/file/systematic-review-methodology.pdf
Accessed October 18, 2016.

32. Sacket D. Centre for Evidence-based Medicine, 1998.
Oxford Centre for Evidence-based Medicine – Levels of Evidence.
http://www.cebm.net/oxford-centre-evidence-based-medicine-levels-evidence-march-2009/
Accessed October 18, 2016.

33. Sandlund M, Waterworth EL, Hager C.
Using motion interactive games to promote physical activity and enhance motor performance in children with cerebral palsy.
Dev Neurorehabil 2011;14: 15-21.

34. Christy JB, Chapman CG, Murphy P.
The effect of intense physical therapy for children with cerebral palsy.
J Pediatr Rehabil Med 2012;5: 159-170.

35. Crompton J, Imms C, McCoy AT, Randall M, Eldridge B, Scoullar B, et al.
Group-based task-related training for children with cerebral palsy: a pilot study.
Phys Occup Ther Pediatr 2007;27: 43-65.

36. Maher CA, Williams MT, Olds T, Lane AE.
An internet-based physical activity intervention for adolescents with cerebral palsy: a randomized controlled trial.
Dev Med Child Neurol 2010;52: 448-455.

37. Van Wely L, Balemans AC, Becher JG, Dallmeijer AJ.
Physical activity stimulation program for children with cerebral palsy did not improve physical activity: a randomised trial.
J Physiother 2014;60: 40-49.

38. Van den Berg-Emons RJ, Van Baak MA, Speth L, Saris WH.
Physical training of school children with spastic cerebral palsy: effects on daily activity, fat mass and fitness.
Int J Rehabil Res 1998;21: 179-194.

39. Maniu DA, Maniu EM, Benga I.
Effects of an aquatic therapy program on vital capacity, quality of lige and physical activity index in children with cerebral palsy.
Human & Veterinary Medicine 2013;5: 117-124.

40. Mitchell LE, Ziviani J, Boyd RN. A randomized controlled trial of web-based training to increase activity in children with cerebral palsy.
Dev Med Child Neurol 2016;58: 767-773.

41. Bania T, Dodd KJ, Taylor N.
Habitual physical activity can be increased in people with cerebral palsy: a systematic review.
Clin Rehabil 2011;25: 303-315.

42. Johnston M, Dixon D.
Developing an integrated biomedical and behavioural theory of functioning and disability: adding models of behaviour to the ICF framework.
Health Psychol Rev 2014;8: 381-403.

43. 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: 923-930.

44. Verschuren O, Wiart L, Ketelaar M. Stages of change in physical activity behavior in children and adolescents with cerebral palsy.
Disabil Rehabil 2013;35: 1630-1635.

45. McAnuff J, Brooks R, Duff C, Quinn M, Marshall J, Kolehmainen N.
Improving participation outcomes and interventions in neurodisability: co-designing future research.
Child Care Health Dev 2016. [epub ahead of print]

46. Sanders EBN, Stappers PJ.
o-creation and the new landscapes of design.
CoDesign: International Journal of CoCreation in Design and the Arts, 2008;1: 5-18.

47. Hermsen S, Mulder S, Renes RJ, van der Lugt R.
Using the persuasive by design model to inform the design of complex behavior change concepts: two case studies. Proceedings, 11th Conference of the European Academy of Design.
Paris, Université Paris René Descartes. 2015.

48. Hermsen S, Renes RJ, Forst JH. Persuasive by design: A model and toolkit for designing evidence based interventions. Making the Difference. Proceedings, CHI Sparks.
The Hague, Netherlands: The Hague University of Applied Sciences 2014.

49. van Essen A, Hermsen S, Renes RJ.
Developing a theory-driven method to design for behaviour change:
two case studies. P. Lloyd & E. Bohemia, eds., Proceedings of DRS2016: Design + Research + Society – Future-Focused Thinking, Volume 4, pp 1323-1338. 2016.

50. Ekelund U, Tomkinson G, Armstrong N.
What proportion of youth are physically active? Measurement issues, levels and recent time trends.
Br J Sports Med 2011;45: 859-865.

51. Pedisic Z, Bauman A.
Accelerometer-based measures in physical activity surveillance:
current practices and issues.
Br J Sports Med 2015;49: 219-223.

10

General discussion

GENERAL DISCUSSION

In this thesis we analyzed clinimetric measurement properties of physical fitness tests in wheelchair-using youth with SB. Furthermore, the amount of physical behavior in wheelchair-using youth with SB was quantified and associations with age, gender, VO_{2peak} and Hoffer classification were evaluated. Finally, we described the factors associated with physical behavior in youth with SB and youth with physical disabilities, after which the evidence of interventions to improve physical behavior in youth with physical disabilities was analyzed. This last chapter presents the theoretical and clinical implications. At the end, methodological considerations and directions for further research will be discussed after which the overall conclusion is presented.

THEORETICAL CONSIDERATIONS

Physical fitness testing

Assessing and improving physical fitness in youth with physical disabilities like SB are important goals in pediatric physical therapy. The knowledge base regarding physical fitness testing in ambulatory youth with physical disabilities has expanded in the last decades, resulting in several valid and reliable field-based tests for measuring physical fitness that are available for clinicians.[1-4] However, the knowledge base regarding physical fitness testing in wheelchair-using youth is still very small. Even though it is an evolving research area, the limited evidence makes it difficult to interpret both research results and individual clinical results when assessing physical fitness in wheelchair-using youth with SB. [5-7]

Both the Graded Wheelchair Propulsion Test (GWPT) and the Shuttle Ride Test (SRiT) are tests for assessing maximal cardiorespiratory endurance and thus require maximal exercise testing, so results can be interpreted adequately. Subjective criteria and objective criteria to determine maximal cardiorespiratory effort are available for ambulatory youth. The subjective criteria include sweating, blushing and unwillingness to continue despite encouragement. The objective criteria consist of a heart rate peak higher than 180 beats per minute, a respiratory exchange ratio (RER) peak higher than 0.99 and the presence of a VO_2 plateau.[8] However, a VO_2 plateau is seldom observed in ambulatory youth and this last criterion is often not met. Unfortunately, the objective criteria for peak heart rate and RER_{peak} seem not applicable in wheelchair-using youth. We know from evidence in adults that maximal exercise testing using arm muscles by arm cranking shows lower peak heart rate compared to using leg muscles by for example leg cycling because of the lower muscle mass of the arms compared to the legs.[9, 10] This may suggest that the heart rate criterion for maximal exercise testing using wheelchair-propulsion should be adjusted to a lower value. Also an adjustment in the criterion for the RER_{peak} is expected, as the arm muscles consist of more type II fibers and less type I fibers compared to the leg muscles.[11, 12] This may affect the oxygen uptake and carbon dioxide output and thus affect the RER_{peak}. Our results show relatively high RER_{peak}

values compared to values found during leg exercise testing in children, indicating that for RER_{peak} a higher value may be a more appropriate criterion during wheelchair propulsion. In summary, the objective criteria for maximal cardiorespiratory effort do not seem to be applicable during maximal exercise testing using wheelchair propulsion. For now, we advise to use the subjective criteria to distinguish between maximal and submaximal cardiorespiratory effort.

The expertise regarding the physiology of cardiorespiratory exercise testing using wheelchair propulsion is slowly evolving. Currently, there are no reference values available for peak values of the different parameters attained during maximal exercise testing using wheelchair propulsion in children and adolescents and also information about kinematics during maximal exercise testing is lacking. We expect lower VO_{2peak} values due to the smaller muscles of the arms compared to the legs.[13] Furthermore, wheelchair-using youth with SB have less overall muscle mass compared to typically developing peers, because of their spinal cord lesion that results in de-innervation of the leg muscles. This influences oxygen uptake negatively, resulting in lower VO_{2peak} values. There may also be differences in ventilatory parameters when using the arms during testing instead of the legs. For example, the accessory muscles of respiration are also used for the arm task which may affect the contribution to breathing. Verschuren et al. reported a lower minute ventilation at peak exercise during wheelchair propulsion compared to running in youth with Cerebral Palsy.[6] They reported that this could be explained by the lower VO_{2peak} and VCO_{2peak} values during arm exercise, because VCO_2 is the major determinant for the ventilatory drive and thus influences minute ventilation.[6] Moreover, also the seated position of wheelchair-using children and the deformities of the spine that are sometimes present may affect ventilation. Furthermore, various exercise tests using arm muscles (for example wheelchair propulsion or arm cranking) may show also differences in ventilatory parameters because of differences in performance of the tests. In **Chapter 3** we indeed showed significant differences in tidal volume and breathing ventilation comparing the GWPT with the SRiT, probably due to contrasts in test performance (more resistance during continuous propelling versus faster propelling).[14] Consequently, the current lack of reference information regarding outcome parameters during maximal exercise testing and their kinematics when using wheelchair propulsion in wheelchair-using youth limits our interpretation.

The SRiT can be used by clinicians when measuring cardiorespiratory endurance in wheelchair-using youth with SB, because it is a highly valid and highly reliable field-based test as presented in **chapter 3**.[14] However, VO_{2peak} can only be measured directly through the use of a mobile gas analysis system as it is not possible to predict VO_{2peak} accurately using the clinical outcome measure "shuttle". Unfortunately, only a very small amount of clinicians have the availability of a mobile gas analysis system and thus most of the clinicians have to use the clinical outcome measure "shuttle" during their clinical reasoning process. During field-based testing, factors such as the wheelchair-user interface integration and rolling resistance play an important role and influence the results of the tests.[15, 16] For specificity of exercise testing, this is an important advantage above laboratory tests as these field-based tests converge with patient goals. However, this also results in a challenge for clinicians during the clinical reasoning process as all these different factors are reflected in the clinical outcome measure "shuttle" of the SRiT and thus should be taken into account during interpretation. Clinicians should be aware that the clinical outcome measure "shuttle" reflects aerobic fitness and is not identical to VO_{2peak}.

Physical behavior

As defined in the general introduction, physical behavior consists of sedentary activity and physical activity.[17] An important recent development is the attention for the time spent in sedentary activities. A child may for example comply to the guidelines for physical activity (for example as defined by the ACSM to spend a minimum of 60 minutes in moderate physical activity intensity of which 30 minutes in vigorous physical activity intensity per day), but may still spend a large amount of time sitting or lying and thus performing sedentary activities.[18, 19] Evidence has shown benefits of physical activity but also, independently, negative health effects of sedentary activities.[20, 21] This implies that spending time in sedentary activities and physical activities are two independent risk factors for health outcomes in youth. That is why we chose to measure and report both sedentary activity and physical activity in wheelchair-using youth with SB in **chapter 5**.[22] There is a huge challenge however regarding the measurement of sedentary activities and physical activities. Device-based measures are more accurate than self-reported measures, but to our knowledge there is only one valid device available (the VitaMove) that can be used to detect the time spent (duration and frequency) in different activities in wheelchair-using children.[23, 24] Furthermore, measuring the heart rate by for example the Actiheart and analyzing it using the heart rate reserve, seemed the only possibility to gain insight in the intensity of physical behavior in wheelchair-using children.[25, 26] This challenge is not only apparent in wheelchair-using children, measuring physical behavior in ambulating children with physical disabilities is also challenging. Certain devices that are valid for typically developing children are not valid for specific populations because of differences in for example gait pattern.[27] Furthermore, because different constructs of physical behavior are used, there is no consensus about which aspects to measure (such as duration, frequency, type and intensity of activity) and also no consensus about how to measure these different aspects.[28-30] This results in difficulties when interpreting results and comparing results between studies. It is of great importance that international consensus is reached and guidelines are developed about how to measure physical behavior, in order to measure physical behavior validly in youth with disabilities.

Recent years have shown a huge increase in the amount of studies reporting about physical behavior. Various terminology is used in literature, such as physical active behavior, physical activity, physical activity engagement, physical inactivity, sedentary time, sedentary activity, sedentary behavior. We stated in the introduction that physical behavior is performed in a specific context with a certain motivation. We explicitly chose to use the broader term "physical behavior" because of the word "behavior". We believe that there is a wide variety of factors that influence if a person is physically active or not and that context and motivation play an important role. So it is not just an activity as defined by the International Classification of Functioning, Disability and Health- Child and Youth version (ICF-CY), it is much more complex and multifaceted.[31]

We therefor used the Physical Activity for persons with a Disability (PAD) model to present our results regarding factors influencing physical behavior in **chapter 7 and 8**.[32-34]

The PAD model combines the ICF with the psychological model of Attitude, Social Influence and Self-Efficacy (ASE model).[32] This results in a model, enlarging the personal and environmental factors as part of the ICF that either facilitate or hinder the intention to participate in physical activity but also the participation itself and thus converge with the term "behavior". Even though intention is a very important aspect in becoming physically active or not, evidence shows that on average intensions only explain 28% of the variance of future behavior.[35] Johnston and Dixon also proposed an integration of psychological models with the bio-social model that is offered by the ICF because psychology is the study of behavior.[36] They showed that the integrated models predicted activity limitation (as defined by the ICF) better than the ICF-model alone in adults with different diagnoses like chronic pain and hip and knee replacements. They also found however, that it is was much more difficult to predict activity (as defined by the ICF) than activity limitation.[36] Heitzler et al. tried to build a model examining determinants related to physical activity in typically developing youngsters 10-17 years of age.[37] The proposed model only accounted for 14.7% of the variance in objectively measured physical activity and the authors concluded that further research should explore additional factors that may influence participation in physical activity.[37] All these studies show the challenges that still remain in trying to understand physical behavior and thus in trying to develop models regarding physical behavior.

We believe that integration of psychological models with the ICF seems the proper direction to follow. Our experiences in using the PAD-model were positive, because it provided us insight in several barriers that a child experienced on different levels, and also in the opportunities that were present and that may be used to overcome the existing barriers. Nonetheless, is was difficult to incorporate the aspect "growth and development" into the PAD-model. And exactly this aspect, "growth and development", is what children and adolescents distinguish from adults. It may be incorporated in an adapted version of the PAD-model, specifically suitable for children and adolescents.

CLINICAL IMPLICATIONS

Physical fitness testing

When assessing physical fitness in wheelchair-using youth with SB, several tests can be used by clinicians. Wheelchair propulsion, by assessing the GWPT or the SRiT, has the preference above arm cranking when measuring VO_{2peak}, with the remark that a mobile gas analyses system should be used.[14, 38] If one does not have the availability of a mobile gas analyses system, the clinical outcome measure "shuttle" of the SRiT should be interpreted as aerobic fitness and one should take other aspects into account during interpretation and clinical reasoning, like the wheelchair-user interface integration, rolling resistance, etcetera. This is also important when interpreting the field-based skill-related fitness tests. The Muscle Power Sprint Test (MPST) provides an indication of the anaerobic performance and both the 10x5 Meter Sprint Test (10x5MST) and slalom test provide an indication of a child's agility.[39] For evaluation purposes, the wheelchair should be identical because changes in wheelchair configuration may affect the results of the tests. Adequate tire pressure should be checked regularly, as we noticed that in most participants the tire pressure of the wheelchair was much too low, affecting the rolling resistance negatively. In addition, the floor should be identical, because different floors cause different rolling resistance and thus influence field-based measures. The physical fitness tests that are currently available for wheelchair-using youth with SB offer clinicians feasible tools to explicit their clinical reasoning process. Underlying problems can be identified and can thus be evaluated in patient goals like "improving wheelchair basketball".

Physical behavior

Overall, wheelchair-using youth with SB show unfavorable physical behavior. Furthermore, they are more sedentary and less physically active during a weekend day compared to a school day.[22] While there also may be possibilities to improve physical behavior during school days, there seems to be a great opportunity during weekend days because of the larger amount of time spent sedentary. How to achieve improvement in physical behavior is a very complex issue, considering the immense variety of barriers and facilitators. Generally, clinicians should be aware of the influence of age and ambulation level on physical behavior, with older age and the inability to walk influencing physical behavior negatively.[40] Furthermore, we believe that it is essential to start as early as possible with stimulating healthy active lifestyles so it becomes part of normal routine behavior and thus becomes habitual, especially in young children with a Hoffer classification 4 or 5.[41] There seems to be an important opportunity during regular hospital visits that are part of usual care for these children. During our qualitative study presented in **chapter 7**, parents explicitly mentioned the scarce attention for and information about healthy active lifestyles for children with SB during their regular hospital visits.[33] Implementing healthy active lifestyles as a standard component in usual care will lead to early identification of problems with the possibility of early intervention and will most probably support and encourage parents and their children. A clinical report from Murphy et al. offers practical suggestions to pediatricians for the improvement of physical behavior for children with disabilities.[42] A recent paper reported about the importance of physical activity assessment and promotion in pediatric healthcare and proposed to implement the "pediatric physical activity vital sign", the Peds PAVS.[43] The Peds PAVS reports aspects of physical behavior of children, so opportunities are created for clinicians to provide domain specific interventions and to enhance opportunities for children to be active every day.[43] This Peds PAVS may be a good opportunity to raise attention towards physical behavior during regular hospital visits.

As a result of the individual variation of the barriers and facilitators together with the variation in physical strain observed during different activities, an individually tailored intervention program seems to be most appropriate in wheelchair-using children with SB.[22, 33, 34] An important challenge is to properly identify the individual possibilities

that can overcome barriers, especially because they may be situated on different levels of the PAD-model and can be both personal and environmental. This requires an individual assessment using proper clinical reasoning including the concept of behavioral change, taking all the different levels of the PAD-model into account.
A second challenge is to create or design an individually tailored intervention that fits the individual possibilities and barriers and is multi-faceted. Unfortunately, evidence about effective interventions for improving physical behavior in children with physical disabilities is lacking at this moment as presented in **chapter 9**.[44]
It seemed though that interventions (partly) using a psychological and thus behavioral change approach may be promising.[44] However, behavioral change is complex and it is difficult to clearly describe and identify behavioral change techniques. Mitchie et al. used a Delphi-type exercise and created a behavioral change techniques taxonomy of 16 clusters (of 93 hierarchically-clustered individual) behavioral change techniques.[45] Even though this is a first version and the authors anticipate on further development and interdisciplinary consensus, this taxonomy provides insight in possible techniques and provides clear descriptions of the techniques.[45] It may help when developing and describing a (partly) behavioral change intervention. Furthermore, we should take in mind that evidence shows that there is a considerable variation in how long it takes people to develop new habits and even more, that it can take a very long time.[46]
Considering all evidence, we believe that a multidisciplinary approach focusing on facilitators and (partly) using a behavioral change approach may be the best possibility.[33, 34, 44] Facilitators that were mentioned explicitly during our qualitative research were wheelchair skills training, improving self-efficacy, using a solution oriented approach during training, the use of role models and an adequate level of fitness.[33]

METHODOLOGICAL CONSIDERATIONS

Strengths

This thesis regarding physical fitness testing and physical behavior in wheelchair-using youth with SB has several strengths. Overall, we were able to measure physical fitness and physical behavior in a relatively large number of wheelchair-using youth with SB during the "*Let's Ride… Study*".[14, 22, 38-40] We are not aware of any studies that were able to include more wheelchair-using children. Because of this, we were able to analyze different aspects of validity and reliability of the physical fitness tests for wheelchair-using youth with SB. During the reliability analyses, we did not only report the ICCs as a measure of agreement, but also analyzed smallest detectable changes or limits of agreement that can be used in clinical practice. By doing so, clinicians can interpret if changes of an individual child exceed the measurement error and thus represent a true change.[47, 48] Furthermore, we were able to measure VO_{2peak} directly during the incremental maximal exercise tests with respiratory gas analysis and thus report the gold standard measure for cardiorespiratory endurance. Even though we were not able to predict VO_{2peak} using the clinical outcome measure "shuttle" of the SRiT, it did provide us insight in the construct of the "shuttle", often used in clinical practice and clinical reasoning.[14]

A strength of the physical behavior data of the "*Let's Ride… Study*" was that it was gathered with valid devices, measuring both the type of activity and the intensity.[22, 40] Because we gathered these data simultaneously, we were able to merge the data and present results about the physical strain of different activities in wheelchair-using youth with SB.[22] Moreover, because we measured VO_{2peak} straight before or after collecting physical behavior data, we were able to analyze associations between physical behavior and VO_{2peak}.[40] To our knowledge, this is unique knowledge about physical behavior in wheelchair-using youth as we did not find other studies reporting these different aspects of physical behavior and associations with VO_{2peak} in wheelchair-using youth.

A second strength concerning physical behavior was that we were able to additionally use qualitative research identifying children's and parental perspectives about facilitators and barriers for physical activity.[33] Information gathered with this qualitative research contributes to awareness and understanding of problems that may arise if a child with SB wants to become physically active. Moreover, it helped us to understand how we can use facilitators and possibilities to overcome the experienced barriers, as the children and their parents are the genuine experts concerning participation in physical activity.

Finally, in trying to understand the construct physical behavior, we decided to evaluate current evidence by two systematic reviews. One review reported the factors to consider when aiming to improve physical behavior and the other review reported the effect of available interventions aiming to improve physical behavior in children with physical disabilities. [34, 44] The information presented in both quantitative studies concerning physical behavior, combined with information gathered in the qualitative study and information collected in the systematic reviews, provide extensive knowledge that we can use when developing tailored interventions.

Limitations

There are also several limitations of our studies which should be kept in mind when interpreting the results presented in this thesis. We were not able to apply objective criteria to determine if a child showed a true maximal effort in the studies that analyzed the validity and reliability of physical fitness testing.[8] For the maximal cardiorespiratory exercise tests (the GACT, the GWPT and the SRiT) we therefore chose to use the subjective criteria that are available, like flushing, sweating and stopping despite encouragement.[8] Furthermore, the test leader, an experienced pediatric physical therapist, determined during all physical fitness tests if the participants were showing maximal effort or not. We also used the OMNI scale of perceived exertion during all tests.[49] Unfortunately we were not able to use the results, as most youngsters found it too difficult to use this scale. The answers we obtained did often not correspond with our observations, for example children showing heartrates of 200 beats per minute at the end of the SRiT, but explaining that they were not tired at all at the OMNI scale.

Children with SB frequently have cognitive impairments and nonverbal learning disabilities, which may result in difficulties understanding questionnaires like the OMNI scale.[50] Another issue may be the problem of socially desirable answers.

An important limitation concerning the physical behavior data was the fact that we had approximately 35% missing data.[22] This was mostly due to technical problems, but in certain cases participants did not want to wear the devices. Especially the Vitamove is relatively large for using in wheelchair-using children, which makes it difficult for them to wear. It was also very challenging to measure physical behavior in both walking and wheelchair-using participants, as they had to wear five devices for the Vitamove.[24] The devices placed on their legs were prone to be displaced during transfers in and out of the wheelchair, which caused some difficulties in analyzing the results.

Even though we obtained a relatively large sample size of wheelchair-using children, we were still limited in statistical methods due to the sample size, For example, in the manuscript where we explored the relations between physical behavior and age, gender, VO_{2peak} and Hoffer classification (n = 34 and 36), we were limited in the amount of independent variables that we could use.[40] This of course affects the interpretability of our results and still leaves a large proportion of the variance in physical behavior unexplained. In addition, the *Let's Ride... Study* had a cross-sectional design which limited the interpretation of our results. For example, we do not know how physical behavior changes during childhood and adolescence.

Finally, with this thesis we obtained insight into the levels of physical behavior in wheelchair-using youth with SB and possible factors that may influence this physical behavior. And while this information will help us to develop interventions aiming to improve physical behavior, we were still not able to improve physical behavior yet. This was just a small step in our ultimate goal, to facilitate and stimulate wheelchair-using children with SB and their parents to obtain healthy active lifestyles. In other words, there is still much more to do.

DIRECTIONS FOR FUTURE RESEARCH

What can we learn from the research presented in this thesis, as directions for further research? For adequate interpretation of the smallest detectable changes for the physical fitness tests in wheelchair-using youth with SB, we need more information about possible changes after training programs in this population. As these information is lacking, it is difficult to estimate if the smallest detectable changes are acceptable or too large. So future research should try to identify possible improvements of the physical fitness tests after systematical training. Furthermore, future research should estimate the minimal important changes of the tests so clinical important changes can be identified during evaluations.[47]

Even though we developed valid and reliable tests to measure physical fitness in wheelchair-using youth with SB, the knowledge base about fitness testing in wheelchair-using youth is still small. It would be very interesting to combine research results to get more power and develop objective criteria for maximal exercise testing in wheelchair-using youth. Also knowledge about physiologic responses and kinematics during maximal cardiorespiratory exercise testing should be developed so maximal exercise testing can be adequately interpreted in this population.

Despite the fact that knowledge about physical behavior has expanded in recent years, there is still a challenge in developing international guidelines about terminology, how and what to measure and how to analyze and interpret results. Moreover, valid and reliable feasible devices that can easily be used in clinical practice and do not require difficult analyses systems should be developed for wheelchair-using children.

Due to the relative small sample size we were not able to analyze associations between physical behavior and other determinants (than age, gender, VO_{2peak} and Hoffer classification) or health outcomes. Future longitudinal research may provide evidence about these associations, so we can understand this complex and multifaceted construct better in order to develop effective interventions. Furthermore, future research should analyze the effect of wheelchair-skills training, mentioned explicitly by the children and parents during our qualitative study. Moreover, the concept of improving self-efficacy in wheelchair-using children and the impact it may have on physical behavior seems promising. Traditionally, most interventions are developed by healthcare professionals. However, the development of individually tailored interventions to improve physical behavior may require a novel approach. A collaboration between healthcare professionals, social professionals, behavioral professionals and design professionals may be able to bridge the gap between healthcare and participation in physical activities such as playing outside or playing sports. Even more, co-creation of interventions together with the children with disabilities, their parents and (health-care) professionals seems a promising concept.[51, 52]

CONCLUSIONS

Several valid and reliable field-based physical fitness tests have been developed for wheelchair-using youth with SB, which can be easily used by clinicians. These tests offer clinicians feasible tools to explicit their clinical reasoning process. Moreover, these tests can be used for evaluation purposes after training interventions, to identify true changes in wheelchair-using youth with SB.

Physical behavior of wheelchair-using youth with SB is unfavorable compared to typically developing youth. Opportunities for improving this behavior seem especially present during weekend days. The challenge is how to achieve healthy active lifestyles in wheelchair-using youth with SB. In general, older age and the inability to walk

influence physical behavior negatively. Furthermore, there is a large variety of personal and environmental barriers and facilitators influencing physical behavior. Up till now, no interventions succeeded in improving physical behavior in children with physical disabilities. Individually tailored interventions, using the facilitators to overcome barriers, seem a starting point when aiming to improve physical behavior.

REFERENCES

1. 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;25: 25-28.

2. 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: 108-115.

3. 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: 1107-1117.

4. de Groot JF, Takken T, Gooskens RH, Schoenmakers MA, Wubbels M, Vanhees L, et al.
Reproducibility of maximal and submaximal exercise testing in "normal ambulatory" and "community ambulatory" children and adolescents with spina bifida: which is best for the evaluation and application of exercise training?
Phys Ther 2011;91: 267-276.

5. Verschuren O, Ketelaar M, De Groot J, Vila Nova F, Takken T.
Reproducibility of two functional field exercise tests for children with cerebral palsy who self-propel a manual wheelchair.
Dev Med Child Neurol 2013;55: 185-190.

6. Verschuren O, Zwinkels M, Ketelaar M, Reijnders-van Son F, Takken T.
Reproducibility and validity of the 10-meter shuttle ride test in wheelchair-using children and adolescents with cerebral palsy.
Phys Ther 2013;93: 967-974.

7. Bongers BC, Rijks EB, Harsevoort AG, Takken T, van Brussel M. 10-m
Shuttle Ride Test in Youth With Osteogenesis Imperfecta Who Use Wheelchairs: Feasibility, Reproducibility, and Physiological Responses.
Phys Ther 2016;96: 679-686.

8. Rowland T. (1996)
Developmental exercise physiology. Champaign,
IL: Human Kinetics.

9. Orr JL, Williamson P, Anderson W, Ross R, McCafferty S, Fettes P.
Cardiopulmonary exercise testing: arm crank vs cycle ergometry.
Anaesthesia 2013;68: 497-501.

10. Schrieks IC, Barnes MJ, Hodges LD.
Comparison study of treadmill versus arm ergometry.
Clin Physiol Funct Imaging 2011;31: 326-331.

11. Koppo K, Bouckaert J, Jones AM.
Oxygen uptake kinetics during high-intensity arm and leg exercise.
Respir Physiol Neurobiol 2002;133: 241-250.

12. Schneider DA, Wing AN, Morris NR.
Oxygen uptake and heart rate kinetics during heavy exercise: a comparison between arm cranking and leg cycling.
Eur J Appl Physiol 2002;88: 100-106.

13. Larsen RT, Christensen J, Tang LH, Keller C, Doherty P, Zwisler AD, et al.
A Systematic Review and Meta-Analysis Comparing Cardiopulmonary Exercise Test Values obtained from the Arm Cycle and the Leg Cycle Respectively in Healthy Adults.
Int J Sports Phys Ther 2016;11: 1006-1039.

14. Bloemen MAT, de Groot JF, Backx FJG, Benner J, Kruitwagen CLJJ, Takken T.
The Shuttle Ride Test for assessing aerobic performance in youth with spina bifida who are wheelchair-dependent; a validity and reliability study.
Physical Therapy Accepted;.

15. Goosey-Tolfrey VL, Leicht CA. Field-based physiological testing of wheelchair athletes.
Sports Med 2013;43: 77-91.

16. van der Woude LH, de Groot S, Janssen TW.
Manual wheelchairs: Research and innovation in rehabilitation, sports, daily life and health.
Med Eng Phys 2006;28: 905-915.

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.

18. American College of Sports Medicine.
Exercise prescription for healthy populations & special considerations. (2010)
Philadelphia: Lippincott Williams & Wilkins.

19. Van Hecke L, Loyen A, Verloigne M, van der Ploeg HP, Lakerveld J, Brug J, et al.
Variation in population levels of physical activity in European children and adolescents according to cross-European studies: a systematic literature review within DEDIPAC.
Int J Behav Nutr Phys Act 2016;13: 70-016-0396-4.

20. Carson V, Hunter S, Kuzik N, Gray CE, Poitras VJ, Chaput JP, et al.
Systematic review of sedentary behaviour and health indicators in school-aged children and youth: an update.
Appl Physiol Nutr Metab 2016;41: S240-65.

21. 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.

22. Bloemen MAT, van den Berg-Emons RHJG, Tuijt M, Nooijen CF, Takken T, Backx FJG, et al.
Physical behavior in wheelchair-using children and adolescents with spina bifida .
Submitted.

23. Ekelund U, Tomkinson G, Armstrong N.
What proportion of youth are physically active? Measurement issues, levels and recent time trends.
Br J Sports Med 2011;45: 859-865.

24. 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: 11-015-0004-x.

25. Brage S, Brage N, Franks PW, Ekelund U, Wareham NJ.
Reliability and validity of the combined heart rate and movement sensor Actiheart.
Eur J Clin Nutr 2005;59: 561-570.

26. Karvonen MJ, Kentala E, Mustala O.
The effects of training on heart rate; a longitudinal study.
Ann Med Exp Biol Fenn 1957;35: 307-315.

27. de Groot JF, de Jong AS, Visser T, Takken T.
Validation of the Actical and Actiheart monitor in ambulatory children with spina bifida.
J Pediatr Rehabil Med 2013;6: 103-111.

28. 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.

29. Wijndaele K, Westgate K, Stephens SK, Blair SN, Bull FC, Chastin SF, et al.
Utilization and Harmonization of Adult Accelerometry Data:
Review and Expert Consensus.
Med Sci Sports Exerc 2015;47: 2129-2139.

30. Fulton JE, Carlson SA, Ainsworth BE, Berrigan D, Carlson C, Dorn JM, et al.
Strategic Priorities for Physical Activity Surveillance in the United States.
Med Sci Sports Exerc 2016;48: 2057-2069.

31. International Classification of Functioning Disability and Health:
Children & Youth version:
ICF-CY. (2007) Switzerland: World Health Organization.

32. van der Ploeg HP, van der Beek AJ, van der Woude LH, van Mechelen W.
Physical activity for people with a disability: a conceptual model.
Sports Med 2004;34: 639-649.

33. 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;15: 11-015-0265-9.

34. 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;57: 137-148.

35. Sheeran P.
Intention—Behavior Relations: A Conceptual and Empirical Review.
European Review of Social Psychology 2002;12: 1-36.

36. Johnston M, Dixon D.
Developing an integrated biomedical and behavioural theory of functioning and
disability: adding models of behaviour to the ICF framework.
Health Psychol Rev 2014;8: 381-403.

37. Heitzler CD, Lytle LA, Erickson DJ, Barr-Anderson D, Sirard JR, Story M.
Evaluating a model of youth physical activity.
Am J Health Behav 2010;34: 593-606.

38. 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: 432-437.

39. Bloemen MA, Takken T, Backx FJ, Vos M, Kruitwagen CL, de Groot JF.
Validity and Reliability of Skill-Related Fitness Tests for Wheelchair-Using
Youth With Spina Bifida.
Arch Phys Med Rehabil 2016.

40. Bloemen MAT, Takken T, de Groot JF, Kruitwagen CLJJ, Rook RA,
van den Berg-Emons RHJG, et al.
Determinants of physical behavior in wheelchair-using youth with spina bifida.
Submitted.

41. Verplanken B.
Beyond frequency: habit as mental construct.
Br J Soc Psychol 2006;45: 639-656.

42. 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: 1057-1061.

43. Joy EA, Lobelo F.
Promoting the athlete in every child: physical activity assessment and promotion
in healthcare.
Br J Sports Med 2016;.

44. Bloemen MAT, Van Wely L, Mollema J, Dallmeijer A, de Groot JF.
Inreasing physical activity in children with physical disability:
what is the evidence? A systematic review.
Dev Med Child Neurol Accepted.

45. Michie S, Richardson M, Johnston M, Abraham C, Francis J, Hardeman W, et al.
The behavior change technique taxonomy (v1) of 93 hierarchically clustered
techniques: building an international consensus for the reporting of behavior
change interventions.
Ann Behav Med 2013;46: 81-95.

46. Lally P, van Jaarsveld,C.H.M.,Potts,H.W.W., Wardle J.
How are habits formed: Modelling habit formation in the real world.
Eur J Soc Psychol 2010;40: 998-1009.

47. de Vet H, Terwee C, Mokkink L, Knol D. (2013)
Measurement in Medicine. Practical Guide to Biostatistics and Epidemiology.
New York, United States of America: Cambridge University Press.

48. de Vet HC, Terwee CB, Knol DL, Bouter LM.
When to use agreement versus reliability measures.
J Clin Epidemiol 2006;59: 1033-1039.

49. Robertson RJ, Goss FL, Boer NF, Peoples JA, Foreman AJ, Dabayebeh IM, et al.
Children's OMNI scale of perceived exertion: mixed gender and race validation.
Med Sci Sports Exerc 2000;32: 452-458.

50. Kelly NC, Ammerman RT, Rausch JR, Ris MD, Yeates KO, Oppenheimer SG, et al.
Executive functioning and psychological adjustment in children and youth with spina bifida.
Child Neuropsychol 2012;18: 417-431.

51. McAnuff J, Brooks R, Duff C, Quinn M, Marshall J, Kolehmainen N.
Improving participation outcomes and interventions in neurodisability: co-designing future research.
Child Care Health Dev 2016;.

52. Revenas A, Opava CH, Martin C, Demmelmaier I, Keller C, Asenlof P.
Development of a web-based and mobile app to support physical activity in individuals with rheumatoid arthritis: results from the second step of a co-design process.
JMIR Res Protoc 2015;4: e22.

5

Summary

SUMMARY

Chapter 1 described the introduction of this thesis. Spina bifida (SB) is the most frequently seen congenital deformity of the neural tube. The malformation of the spinal cord and often the brain can result in both motor and sensor impairment, incontinence for bowel and bladder and cognitive impairment. Depending on the type of SB and the height of the lesion level of the spinal cord, children and adolescents with SB experience difficulties with ambulation. The ambulation level is classified according to the Hoffer classification adjusted by Schoenmakers et al. and ranges from normal ambulatory (level 1) to non-ambulatory (Level 5). A large part of children and adolescents with SB will use a manual wheelchair for different purposes. In this thesis, "wheelchair-using" is defined as using a wheelchair for either daily activities but also as using a wheelchair for solely long distances or sports participation.

Due to advances in medical approach, most children with SB can now be expected to live to be adults. So we should not only focus on pathological aspects, but also at preventable medical and social consequences of the congenital disorder. In general, adults with SB have lower physical fitness and unfavorable physical behavior, higher prevalence of obesity and lower health-related quality of life compared to peers. Moreover, adults with SB who are not able to walk show lower physical fitness and more unfavorable physical behavior compared to ambulating adults with SB.

Physical fitness testing

Even though assessment and optimizing physical fitness in youth with chronic conditions like SB are important goals in pediatric rehabilitation, there are no valid and reliable tests available for clinicians to measure physical fitness in wheelchair-using children and adolescents with spina bifida. Physical fitness consists of health-related fitness and skill-related fitness. An important component of health-related fitness is cardiorespiratory endurance, with peak oxygen uptake (VO2peak) as the single best indicator of the cardiorespiratory system. Skill-related fitness consists of power, speed, agility, coordination, balance and reaction time and is reflected in activities such as playing outside or playing wheelchair sports.

In wheelchair-using adults, arm cranking protocols are often used to assess VO_{2peak}. However, arm cranking protocols lack specificity compared to wheelchair propulsion and therefore the validity of these types of protocols are questioned. Consequently, wheelchair propulsion might be a more appropriate way of testing VO_{2peak} in wheelchair-using youth with SB. **Chapter 2** reported which laboratory test should be used to measure VO_{2peak} in wheelchair-using youth with SB. The Graded Wheelchair Propulsion Test (GWPT) showed significantly higher heart rate peak and higher VO_{2peak} values compared to the Graded Armcranking Exercise Test (GAET). Furthermore, the reliability of the GWPT was good. Based on these findings, we advised to use wheelchair propulsion and not arm cranking for measuring VO_{2peak} in wheelchair-using youth with SB.

After determining the best laboratory test to measure VO_{2peak}, **chapter 3** analyzed the validity and reliability of the Shuttle Ride Test (SRiT) in wheelchair-using youth with

SB, a field-based test using wheelchair propulsion to measure cardiorespiratory endurance. Results showed that the SRiT is highly valid and highly reliable. The clinical outcome measure "number of completed shuttles" represents aerobic fitness, while also being highly correlated with both anaerobic performance and agility. A mobile gas analysis system should be used to truly measure VO_{2peak} as is was not possible to accurately predict VO_{2peak} using the "number of completed shuttles". The individual prediction intervals were too wide and thus indicating too much prediction error.

Chapter 4 described the clinimetric properties of four skill-related fitness tests, the Muscle Power Sprint Test (MPST), the 10x5 Meter Sprint Test (10x5MST), the slalom test and the One Stroke Push Test (1SPT). The MPST, adjusted to four sprints, is highly valid and moderately reliable to measure anaerobic performance. The 10x5MST and slalom test were both highly valid and highly reliable for measuring agility. The results for the 1SPT showed that the validity and the reliability are not yet established.

Physical behavior

Physical behavior consists of sedentary activity and physical activity and is performed in a specific context with a certain motivation. Sedentary activity is defined as "sitting or lying during waking hours with an energy expenditure lower than 1.5 metabolic equivalent task (MET)" whereas physical activity has been defined as "any bodily movement that results in energy expenditure". There is no evidence in the literature that presents an overview of physical behavior in wheelchair-using youth with SB. Also relations with VO2peak or other determinants such as age, gender and ambulatory status are lacking. Knowing the level of physical behavior in wheelchair-using youth with SB and understanding its relations with certain determinants will help us to tailor and optimize interventions specific for this population.

In **Chapter 5** we showed that physical behavior (expressed as type of activities and intensity) of wheelchair-using youth with SB was unfavorable compared to typically developing peers, with weekend days being even more unfavorable compared to school days. The participants spent less time performing sedentary activities, more time performing physical activities and showed higher intensities during a school day compared to a weekend day. Of all participants, only 19% met the Guidelines of Physical Activity (> 60 minutes moderate to vigorous intensity of which 30 minutes > vigorous intensity) during school days and 8% during weekend days. We also evaluated the intensities of different activities, which varied extensively between participants. The different intensities during activities indicate the importance of individually tailored assessments and interventions.

The associations between physical behavior and age, gender, VO_{2peak} and Hoffer classification were analyzed in **Chapter 6**. Results demonstrated that physical behavior was associated with age and Hoffer classification in wheelchair-using youth with SB, with older age and the inability to walk influencing physical behavior negatively. Gender and VO_{2peak} were not associated with physical behavior in wheelchair-using youth with SB. Interestingly, still a large percentage of the variance in physical behavior remained unexplained (61%-86%), implicating that there are other important personal or environmental factors that should be explored regarding the improvement of physical behavior.

In **Chapter 7 and 8** we presented a wide variety of personal and environmental factors that were either positively or negatively associated with physical behavior in both children with SB and in children with physical disabilities on all levels of the PAD (Physical Activity for persons with a Disability) model. Bowl and bladder care, medical events and the decreased intention to be physically active seemed to be negative personal factors specific for youth with SB. Overall, competence in skills, sufficient fitness and self-efficacy were important personal factors for youth with SB and for youth with physical disabilities. Environmental factors that were associated with physical behavior included the contact with and support from other people, the use of assistive devices for mobility and care, adequate information regarding possibilities for adapted sports and availability and accessibility of playgrounds and sports facilities.

Finally, the evidence of interventions for increasing physical activity was evaluated in **chapter 9**. This will help us to understand which aspects of interventions that are already used show effectiveness and which aspects not. Results showed that there is level-I evidence for no effect of training on improving physical behavior in children with physical disability. Furthermore, there is conflicting evidence for the effect of interventions with a behavioral component on short term physical behavior and level-II evidence for no effect of interventions with a behavioral component on long term physical behavior in children with physical disability. More research using innovative approaches are needed to develop and investigate interventions for improving physical behavior.

Conclusions

Chapter 10 presented the theoretical and clinical implications, methodological considerations, directions for future research and the conclusions. In summary, several valid and reliable field-based physical fitness tests have been developed for wheelchair-using youth with SB, which can be easily used by clinicians. Physical behavior of wheelchair-using youth with SB is unfavorable compared to typically developing youth. Furthermore, older age and the inability to walk influence physical behavior negatively in these children and adolescents. Moreover, there is a large variety of personal and environmental barriers and facilitators related to physical behavior in children and adolescents with SB or other physical disabilities. Up till now, no interventions succeeded in improving physical behavior in children and adolescents with physical disabilities. Individually tailored interventions, using the facilitators to overcome barriers, seem a starting point when aiming to improve physical behavior.

5

Samenvatting

SAMENVATTING

In **Hoofdstuk 1** is de inleiding beschreven van dit proefschrift. Spina bifida (SB) is de meest frequente aangeboren aandoening van het ruggenmerg. De malformatie van het ruggenmerg en vaak ook de hersenen kunnen resulteren in zowel motorische als sensorische stoornissen, incontinentie en cognitieve beperkingen. Afhankelijk van het type SB en de hoogte van de laesie zullen kinderen en jongeren problemen ervaren met lopen. De manier van voortbewegen wordt geclassificeerd door middel van de Hoffer classificatie, aangepast door Schoenmakers et al en varieert van “zelfstandig buitenhuis lopen zonder hulpmiddelen” (Hoffer 1) tot “volledig rolstoel gebonden” (Hoffer 5).
Een groot gedeelte van kinderen en adolescenten met SB zal een handbewogen rolstoel gebruiken voor bijvoorbeeld dagelijkse activiteiten maar ook voor het overbruggen van lange afstanden of tijdens sportparticipatie. In dit proefschrift is “rolstoel-rijdend” gedefinieerd als het gebruik van een rolstoel voor dagelijkse activiteiten maar ook voor alleen lange afstanden of tijdens sport.

Vanwege vooruitgang in de medische zorg groeien de meeste kinderen met SB op tot volwassenen. Dit betekent dat we ons niet alleen moeten focussen op pathologische aspecten, maar ook op secundaire consequenties van de aangeboren aandoening op latere leeftijd die wellicht voorkomen kunnen worden. In het algemeen hebben volwassenen met SB een verlaagde fysieke fitheid, ongunstig fysiek gedrag, een hogere prevalentie van obesitas en een lager gezondheids-gerelateerde kwaliteit van leven vergeleken met gezonde volwassenen. Daarnaast hebben volwassenen met SB die rolstoel gebonden zijn een lagere fysieke fitheid en vertonen zij ongunstiger fysiek gedrag dan volwassenen met SB die lopen.

Testen van fysieke fitheid
Alhoewel het testen en verbeteren van fysieke fitheid van kinderen en jongeren met lichamelijke beperkingen zoals SB belangrijke doelen zijn binnen de kinderrevalidatie, zijn er geen valide en betrouwbare testen beschikbaar voor clinici om fysieke fitheid te meten bij rolstoel-rijdende kinderen en jongeren met SB. Fysieke fitheid bestaat uit gezondheids-gerelateerde fitheid en vaardigheids-gerelateerde fitheid. Een belangrijke component van gezondheids-gerelateerde fitheid is het cardiorespiratoire uithoudings-vermogen, met piek zuurstofopname (VO_{2piek}) als de gouden standaard. Vaardigheids-gerelateerde fitheid bestaat uit power, snelheid, behendigheid, coördinatie, balans en reactie tijd en is gereflecteerd in activiteiten zoals buitenspelen en rolstoelsporten.

In rolstoel-rijdende volwassenen worden vaak arm-fiets protocollen gebruikt om VO_{2piek} te meten. Deze protocollen missen echter specificiteit vergeleken met rolstoel propulsie en daarom wordt de validiteit van deze arm-fiets protocollen bediscussieerd. Wellicht is rolstoel propulsie een geschiktere manier om VO_{2piek} te meten bij rolstoel-rijdende kinderen en jongeren met SB. In **Hoofdstuk 2** is gerapporteerd welke test (arm-fietsen of rolstoel propulsie) het beste gebruikt kan worden om VO_{2piek} te meten bij rolstoel-rijdende kinderen en jongeren met SB. De resultaten lieten significant hogere hartslagen en hogere VO_{2piek} zien bij de oplopende rolstoel propulsie test (Graded Wheelchair Propulsion Test – GWPT) vergeleken met de oplopende arm-fiets test (Graded Arm-

cranking Exercise Test – GAET). Daarbij was de betrouwbaarheid van de GWPT goed. Gebaseerd op deze bevindingen, adviseren wij om bij rolstoel-rijdende kinderen en jongeren met SB VO_{2piek} te meten door middel van rolstoel propulsie in plaats van arm-fietsen.

Nadat we bepaald hebben wat de beste laboratorium test is om VO_{2piek} te meten, is in **hoofdstuk 3** de validiteit en betrouwbaarheid bepaald van de Shuttle Rij Test (Shuttle Ride Test – SRiT) bij rolstoel-rijdende kinderen en jongeren met SB. Dit is een veldtest waarbij gebruik wordt gemaakt van rolstoel propulsie om het cardiorespiratoire uithoudingsvermogen te meten. De resultaten lieten zien dat de SRiT valide en betrouwbaar is. De klinische uitkomstmaat "aantal volbrachte trappen" representeert aerobe fitheid en hangt samen met zowel anaerobe fitheid als behendigheid. Wanneer clinici een indruk willen krijgen van de VO_{2piek}, dan moet er een mobiel gas analyse systeem gebruik worden tijdens de test, aangezien het niet mogelijk was om VO_{2piek} goed te voorspellen aan de hand van de uitkomstmaat "trap". De individuele predictie intervallen waren te groot en hadden dus een te grote foutmarge bij het voorspellen van VO_{2piek}.

In **Hoofdstuk 4** zijn de klinimetrische eigenschappen beschreven van vier vaardigheids-gerelateerde fitheidstesten: de Muscle Power Sprint Test (MPST), de 10x5 Meter Sprint Test (10x5MST), de slalom test en de Een Afzet Test (One Stroke Push Test – 1SPT). De MPST, aangepast naar 4 sprints, is valide en matig betrouwbaar voor het meten van anaerobe fitheid. De 10x5MST en slalom test zijn beiden valide en betrouwbaar om behendigheid te meten. De resultaten voor de 1SPT lieten zien dat deze test nog niet valide en betrouwbaar bevonden is bij rolstoel-rijdende kinderen en jongeren met SB.

Fysiek gedrag

Fysiek gedrag bestaat uit sedentaire activiteiten en fysieke activiteiten en vindt plaats in een bepaalde context met een bepaalde motivatie. Sedentaire activiteit wordt gedefinieerd als "zitten of liggen gedurende de tijd dat men wakker is met een energie verbruik van minder dan 1.5 metabole equivalent (Metabolic Equivalent Task)". Fysieke activiteit wordt gedefinieerd als "elke beweging van het lichaam die resulteert in energie verbruik". Er is geen overzicht in de literatuur die het fysieke gedrag laat zien van rolstoel-rijdende kinderen en jongeren met SB. Tevens zijn er geen gegevens te vinden over relaties tussen fysiek gedrag en VO_{2piek}, leeftijd, geslacht en manier van voortbewegen bij rolstoel-rijdende kinderen met SB. Informatie over het fysieke gedrag van rolstoel-rijdende kinderen en jongeren met SB en over eventuele relaties tussen het fysieke gedrag en bepaalde variabelen zal ons helpen om interventies te ontwikkelen specifiek voor deze doelgroep.

In **hoofdstuk 5** hebben we laten zien dat fysiek gedrag (uitgedrukt in type activiteit en intensiteit) van rolstoel-rijdende kinderen en jongeren met SB ongunstig is vergeleken met normaal ontwikkelende kinderen, waarbij het fysiek gedrag op weekenddagen nog slechter was dan op schooldagen. De kinderen en jongeren waren minder sedentair en meer fysiek actief gedurende een schooldag ten opzichte van een weekenddag. Daarnaast lieten ze hogere intensiteiten van fysieke activiteit zien gedurende de schooldag ten opzichte van de weekenddag. Slechts 19% van alle deelnemers voldeden aan de Fysieke Activiteiten Richtlijn (>60 minuten matig tot zware intensiteit waarvan 30 minuten zware intensiteit, volgens de American College of Sports Medicine) gedurende een schooldag en slechts 8% gedurende een weekenddag. De intensiteiten van de verschillende activiteiten varieerden enorm tussen de kinderen en jongeren, wat het belang weergeeft van individueel onderzoek en behandeling bij hulpvragen gericht op fysiek gedrag.

De relaties tussen fysiek gedrag en leeftijd, geslacht, VO_{2piek} en Hoffer classificatie zijn geanalyseerd in **hoofdstuk 6**. De resultaten demonstreerden dat fysiek gedrag gerelateerd was aan leeftijd en Hoffer classificatie bij rolstoel-rijdende kinderen met SB, waarbij een oudere leeftijd en niet kunnen lopen het fysieke gedrag negatief beïnvloedden. Geslacht en VO_{2piek} waren niet gerelateerd aan fysiek gedrag bij rolstoel-rijdende kinderen met SB. Een groot gedeelte van de variantie bleef onverklaard (61%-86%) hetgeen impliceert dat er andere belangrijke persoonlijke en omgevingsfactoren zijn die geëxploreerd moeten worden wanneer we kijken naar fysiek gedrag.

In **hoofdstuk 7 en 8** zijn een grote variatie aan persoonlijke en omgevingsfactoren gepresenteerd op alle niveaus van het PAD (Fysieke Activiteit voor mensen met een Beperking / Physical Activity for persons with a Disability – PAD) model die ofwel positief ofwel negatief samen hangen met fysiek gedrag bij zowel kinderen en jongeren met SB als bij kinderen en jongeren met lichamelijke beperkingen. Verzorging in verband met incontinentie, medische ingrepen en de verminderde intentie om fysiek actief te zijn leken negatieve persoonlijke factoren specifiek voor kinderen en jongeren met SB. In het algemeen waren het competent zijn in vaardigheden, een voldoende fitheidsniveau en zelfvertrouwen belangrijke persoonlijke factoren voor kinderen en jongeren met SB of andere lichamelijke beperkingen. Omgevingsfactoren die geassocieerd werden met fysieke activiteit waren het contact met en de ondersteuning van andere mensen, het gebruik van hulpmiddelen voor verzorging en mobiliteit, adequate informatievoorziening met betrekking tot mogelijkheden voor aangepast sporten en de aanwezigheid en toegankelijkheid van speeltuinen en sportfaciliteiten.

In **hoofdstuk 9** is tenslotte de bestaande evidentie uit wetenschappelijke literatuur geëvalueerd en geanalyseerd met betrekking tot interventies en het verbeteren van de fysieke activiteit bij kinderen en jongeren met een lichamelijke beperking. Dit overzicht zal ons inzicht geven welke aspecten van interventies die al gebruikt worden effectief zijn en welke niet. Resultaten lieten zien dat er niveau-I bewijs is voor geen effect van training op het verbeteren van fysieke activiteit in kinderen en jongeren met een cerebrale parese. Daarnaast is er conflicterend bewijs voor het effect van interventies met een gedragsmatige component op de fysieke activiteit op de korte termijn en niveau-II bewijs voor geen effect van interventies met een gedragsmatige component op de fysieke activiteit op de lange termijn bij kinderen met een cerebrale parese. Er is meer innovatief onderzoek nodig om interventies met betrekking tot het verbeteren van fysiek gedrag te ontwikkelen voor kinderen met een lichamelijke beperking.

Conclusies

In **Hoofdstuk 10** zijn de theoretische en klinische implicaties, de methodologische overwegingen, de richtingen voor toekomstig onderzoek en de conclusies gepresenteerd. Geconcludeerd kan worden dat er verschillende valide en betrouwbare veldtesten ontwikkeld zijn om fysieke fitheid te meten bij rolstoel-rijdende kinderen en jongeren met SB, welke gemakkelijke gebruikt kunnen worden door clinici. Rolstoel-rijdende kinderen en jongeren met SB laten ongunstig fysiek gedrag zien ten opzichte van normaal ontwikkelende leeftijdgenoten. Daarnaast beïnvloeden een oudere leeftijd en rolstoel gebondenheid fysiek gedrag negatief in deze kinderen en jongeren.
Er is een grote variëteit aan persoonlijke en omgevingsfactoren gerelateerd aan fysieke activiteit in kinderen en jongeren met SB en andere lichamelijke beperkingen. Er is tot op heden helaas nog geen wetenschappelijk bewezen effectieve interventie beschikbaar om fysiek gedrag positief te beïnvloeden. Individueel aangepaste interventies, waarbij de facilitators ingezet worden om de barrieres te beslechten, lijken een startpunt wanneer het doel is om fysieke gedrag te verbeteren.

Dankwoord

DANKWOORD

Wat was het een mooie reis! Ontzettend veel geleerd, veel mooie momenten gehad, veel gelachen, veel gediscussieerd en veel meegemaakt... Graag wil ik hier iedereen bedanken die mij bij deze reis heeft geholpen!

Als eerste natuurlijk mijn promotieteam. Janke; bij jou kreeg ik de kans om te starten met het onderzoek, nadat je de subsidie binnen haalde voor de Healthy Active Living for Youth with a Neuromotor Disability studie (HALYNeD studie). Een geweldige kans die ik met beide handen aan heb gepakt en waar ik je nog steeds dankbaar voor ben! Je was mijn directe begeleider en hebt me tijdens deze jaren in alle verschillende aspecten van onderzoek doen ondersteund. Elke keer kon ik me zo weer een stapje verder ontwikkelen, elke keer daagde je me weer wat meer uit en nu mijn promotietraject is afgerond mag ik nog steeds van je begeleiding gebruik maken als mijn mentor. Ik wil je bedanken voor alle inspirerende gesprekken, discussies en al het brainstormen. Je was altijd bereikbaar, altijd positief en hebt me altijd ondersteund. Je kent me door en door en hebt me ook geholpen met de balans tussen werk en privé. Ik kan me geen fijnere dagelijkse begeleider wensen. Ik hoop dat we samen blijven werken in de toekomst en kijk uit naar al onze toekomstige inspirerende gesprekken!

Beste Tim, jij was mijn 2de dagelijkse begeleider. Je hebt me meegenomen in de wereld van de inspanningsfysiologie en interpretatie van gasanalysedata bij kinderen. Ik heb me altijd gerealiseerd dat ik hierin geen betere begeleiding kon krijgen. Je was altijd bereid om samen naar de data te kijken en om mee te denken bij problemen. Je expertise ondersteunde mijn ontwikkeling en kennis over de inspanningsfysiologie. Ik werd uitgedaagd tijdens onze discussies over meten van fitheid bij rolstoel-rijdende kinderen en de interpretatie van de data bij deze doelgroep. Je snelle en concrete feedback heeft me erg geholpen bij het schrijven van de manuscripten. Ik hoop nog heel lang met je samen te mogen werken en zo de zorg voor kinderen met beperkingen te verbeteren!

Beste Frank, wat fijn dat je mijn promotor wilde zijn en je expertise vanuit de sport-geneeskunde wilde toepassen bij de kinderen en jongeren met SB. Als ik denk aan je begeleiding, dan denk ik aan je helikopterview, je stimulans om telkens een stap verder te gaan, je drive om mezelf te ontwikkelen. Je manier van begeleiden paste perfect bij me en ik keek altijd uit naar onze gesprekken. Met veel plezier denk ik terug aan het ACSM-congres waar we tijdens een etentje inspirerende gesprekken hadden over onderzoek en onderwijs. Ik hoop dat we in de toekomst elkaar blijven ontmoeten, zodat ik van je mag blijven leren.

Beste kinderen, jongeren en ouders, wat hebben jullie me veel geleerd! Alle gesprekken die we hebben gehad, alle ervaringen die jullie met ons hebben gedeeld, al die dagen van testen waar dan ook maar in Nederland... Zonder jullie hadden we alle kennis niet kunnen verzamelen, zonder jullie zouden we niet zoveel stappen hebben gezet. Hiervoor wil ik jullie hartelijk bedanken! Ik hoop jullie in de toekomst nog vaak tegen te komen en te mogen blijven leren van jullie.

Beste Harriët. Ik heb in jouw lectoraat (Lectoraat Leefstijl en Gezondheid, LLG) het onderzoek mogen uitvoeren en de promotievoucher mogen aanvragen. Je hebt me hierdoor een unieke kans gegeven om deze reis te maken. Ik moet nog vaak denken aan onze gesprekken, vaak op vrijdagmiddagen op de BL101, waarbij we verschillende onderzoeksthema's en -mogelijkheden bespraken, vaak samen met Janke O. Waardevol om aan terug te denken en het heeft zeker bijgedragen aan mijn ontwikkeling als onderzoeker. Nu de volgende stap, die ik ook mag zetten binnen jouw lectoraat; op naar nog vele mooie onderzoeken!

Lieve (oud-)collega's van het LLG: Janke O, Marleen, Marike, Imke, Jacqueline N, Jacqueline O, Jan, Stefan, Michiel, Karin, Jolanda, Tim, Huib, Kristel, Saskia, Francois, Edwin, Marlies, Roelof, Geert, Henri, Hans, Leendert! Dank voor alle cappuccino-momenten, waarbij we onze onderzoeken bespraken, zowel de kansen als de uitdagingen. Of het nu op de HU was of met een lekkere :-) koffie bij The Village. Johanna; ik heb ervan genoten, het heeft me geïnspireerd en gevormd! Daarnaast al onze PSGs (Peer Support Groepen), of het nu ging om kwalitatief onderzoek, kwantitatieve analyses of schrijven; dank voor al jullie feedback, adviezen en hulp! Ik hoop dat er nog veel cappuccino's, borrels en etentjes volgen :-) ! Speciaal wil ik Jacqueline O nog bedanken, voor alle momenten die plaatsvonden in je tuin en onder het genot van je kookkunsten, samen met Janke O en Marlies. Ze waren een aangename rode draad tijdens de gehele promotie. Marike, dank voor al je advies en hulp betreffende opvoedperikelen :-) en de balans vinden tussen werk en privé. Daarnaast natuurlijk al onze gesprekken over onderzoek binnen de kinderfysiotherapie en de blik naar de toekomst; ik heb er zin in! Imke, altijd fijn om te sparren en altijd tijd om mee te denken. Stefan dank voor je 'psychologische blik' en het meedenken over gedragsverandering, ik heb veel van je geleerd! Karin en Jolanda, helaas geen collega's meer, maar gelukkig hebben we nog steeds contact. Jullie input betreffende inspanningsfysiologie heeft me ontzettend geholpen, wat was het fijn om op zo een vertrouwde manier van jullie te kunnen leren. Huib dank voor al je input en inzet tijdens mijn promotie, ik kon altijd van je op aan: fijn als we samen klaar zijn :-). Jan, altijd bereid en tijd om mee te denken en te helpen, wat fijn dat je er bent! Marleen, samen sparren, brainstormen, discussiëren over ons onderzoek. Of het nu op de HU was, tijdens koffiemomenten bij jou of bij mij of in San Diego; het was altijd inspirerend en ik kijk dan ook uit naar alle toekomstige gesprekken! Fijn om een collega als jij te hebben en dank dat je geholpen hebt als ceremoniemeester om 7 juni tot een mooie dag te maken! Last but not least; Janke O. We zijn samen gestart en je bent gedurende het gehele traject mijn maatje geweest. Samen heel het land door om te interviewen, kwalitatieve analyses, statistiek cursussen, reflectieve leermomenten, inspirerende gesprekken zowel over werk als privé. Ik kon altijd bij je terecht, je had altijd tijd en was altijd bereid om mee te denken. Mijn dank is groot en ik vind het dan ook superfijn dat je mijn paranimf wil zijn!

Beste Jacqueline N., als hoofd van de Master Kinderfysiotherapie heb jij me altijd gestimuleerd om te gaan promoveren. Dankzij jou zijn de eerste stappen gezet en samen met Rob heb je me gefaciliteerd tijdens deze eerste fase. Hiervoor wil ik jullie beiden hartelijk danken! En dan mijn huidige en oud collega's van de Master Kinderfysiotherapie, lieve Jacqueline N, Bert, Mirjam, Eline, Barbara, Anjo, Kitty, Ida, Ron en Marianne.

Het is een prachtige combinatie: onderzoek en onderwijs. Jullie hebben me altijd geïnspireerd binnen het onderwerp kinderfysiotherapie. De maandagen zijn altijd een prettige start van de week, met tijd om te discussiëren over interessante onderwerpen binnen ons mooie vakgebied en uitdagingen binnen ons onderwijs. Lieve Mirjam jou wil ik nog extra bedanken; je bent mijn maatje als derdejaars tutor en hebt me tijdens de laatste fase van mijn promotie bijgestaan met al je adviezen en tips. Ik kon altijd bij je terecht waardoor de laatste fase duidelijk en overzichtelijk werd. Daarnaast heb je me ook nog ondersteund als ceremoniemeester; wat fijn dat je dit hebt gedaan en zeker ook hoe je dit hebt gedaan! Dank! En dan natuurlijk Kitty, je bent mijn vaste collega en maatje bij de Master vanaf het moment dat ik daar werkzaam ben. Ook heb je mij vervangen tijdens mijn zwangerschapsverlof; ik kon geen betere vervanger wensen. Zonder probleem kon ik alles aan je overlaten, wat een heerlijk gevoel was dat! En dan al onze gesprekken over onderzoek, onderwijs, werk, privé, toekomst, keuzes... Dank hiervoor en ook dat je mijn paranimf wilde zijn!

Ook wil ik graag al mijn co-auteurs bedanken, zonder jullie was het niet mogelijk om alle manuscripten te schrijven. Beste Rita, dank voor al je support en expertise met betrekking tot het gebruik en het analyseren van de VitaMove en de discussies en verdieping betreffende het concept fysiek gedrag. Carla, jij was mijn rots in de branding betreffende alle analyses van de VitaMove en was altijd bereid om mee te denken; mijn dank is groot! Beste Annet en Leontien, dank voor al jullie ondersteuning, feedback en het meedenken betreffende de literatuurstudie over de effecten van interventies gericht op fysieke activiteit; jullie expertise heeft voor verdieping gezorgd. Matthijs, wat ontzettend fijn dat je me geholpen hebt met MATLAB waardoor we de data van de twee objectieve activiteiten monitors aan elkaar konden koppelen. Zonder jou was dit nooit gelukt! En dan natuurlijk Jurgen; je was altijd bereid om mee te denken en mee te werken aan de zoekstrategieën van de literatuurstudies. Zo ontzettend fijn dat ik van je expertise gebruik heb mogen maken. Tussendoor was er altijd even tijd om bij te kletsen: ik ben onder de indruk van hoe je in het leven staat. Dan alle statistische hulp, Cas was daarin mijn vaste aanspreekpunt en altijd bereid om mee te denken. Je hield me scherp met je kritische vragen en stimuleerde me om hierin stappen te maken. Harriët, dank voor je input tijdens de eerste systematic review; het heeft me heel erg geholpen! Beste Olaf, jouw input in het kwalitatieve stuk heeft voor de nodige diepgang gezorgd. Dank daarvoor! Beste Joyce, nadat je was gestart als student, heb je mijn zwangerschapswaarneming op je genomen nadat je je diploma behaald had. Erg prettig voor mij, ik kon zonder probleem alles loslaten; en dat is heel wat voor mij :-) . Dank daarvoor! Rosalyne, Marleen V, Rosanne, Claudia, Hanneke, Arina en Marsha, jullie extra inzet tijdens jullie studie heeft me geholpen om de studies uit te werken. Wat fijn dat ik met jullie mocht samenwerken.

Alle studenten die deelgenomen hebben aan onze studies. Onderzoek doen binnen de HU is onderzoek doen samen met studenten: iets dat ik nooit zou willen missen. Of het nu de Bachelor-studenten zijn, de Minor-studenten, de Ster-studenten of de Master-studenten; al jullie input is essentieel geweest! Bij het verzamelen van data, bij het invoeren van data, bij het analyseren en bij het schrijven; jullie hielden me scherp en jullie waren er altijd! Dank, dank, dank!!

Alle kinderfysiotherapeuten van de deelnemende revalidatiecentra, kinderfysiotherapie-praktijken, ziekenhuizen en Stichting Fitkids; wat was het prettig om met jullie samen te werken. Dankzij jullie inzet was het mogelijk om alle kinderen te werven, alle kinderen te testen en alle kinderen en ouders te spreken. Daarnaast waren jullie altijd bereid om mee te denken en input te leveren. Op deze manier ontstond er een prachtige wisselwerking welke van groot belang is bij praktijkgericht onderzoek. Hopelijk gaan we in de toekomst samen nog veel mooie projecten uitwerken.

De fysiologie-overleggen op het WKZ op dinsdag waren een vast leermoment tijdens mijn promotie. Ik wil iedereen die daarbij aanwezig was danken voor de input, de kritische blik en het meedenken. Speciale dank naar de vaste mensen van de afgelopen jaren; Tim, Erik, Janke, Elles, Bart, Maremka en Patrick. Het was altijd mogelijk om feedback te vragen waardoor keuzes gemaakt konden worden en de manuscripten verbeterden. Natuurlijk alle collega's van het Instituut Bewegingsstudies van de Hogeschool Utrecht. Dank voor alle gesprekken, al het meedenken, alle ideeën! Nogmaals excuses voor alle geluidsoverlast tijdens het testen :-)

Nico en collega's van de ICT; zonder jullie hadden we deze onderzoeken nooit uit kunnen voeren en was menig planning in de soep gelopen... Jullie waren altijd bereid om mee te denken als er weer eens ICT-uitdagingen waren, zoals onderzoeksprogramma's die geïnstalleerd moesten worden hetgeen natuurlijk voor problemen zorgde... Jullie waren altijd bereid om net iets meer te doen waardoor alles altijd weer op zijn pootjes terecht kwam. Wat zou ik zonder jullie hebben moeten doen.... In dat kader ook Salvador en collega's van facilitair; soms kneep je een oogje dicht bij inleverdata van apparatuur als er weer eens interviews afgenomen moesten worden, laptops die we toch nog echt even nodig hadden, en zo zijn er nog vele voorbeelden te noemen. Zonder jullie meedenken was het absoluut niet gelukt.

Ik heb mijn best gedaan om mijn sociale leven ook te onderhouden, hoewel het soms een uitdaging was om alles te combineren. Dank aan alle lieve vrienden, die er altijd weer waren om even niet met werk bezig te zijn, altijd geïnteresseerd waren, altijd tijd hadden om te eten, te drinken en te lachen, te genieten van elkaar. Wat ben ik blij dat jullie er zijn!

Bart, dank voor al je werk betreffende de vormgeving! Meer tijd dan verwacht, maar ook nog mooier dan ik me ooit voor kon stellen. Zo fijn dat je dit op je genomen hebt, dat was me nooit zelf gelukt...

Pa en ma, zonder jullie had ik dit nooit bereikt! Helaas kan pa dit niet meer meemaken, maar in gedachten is hij er altijd bij. Jullie hebben me altijd gestimuleerd en ondersteund om te gaan voor mijn toekomst, om verder te leren, om een volgende stap te maken. Daarbij hebben jullie me ook geleerd om bij mezelf te blijven en te waken voor de balans tussen werk en privé. Ma en Henny, dank voor al jullie hulp en oppas momenten voor Faas en Lilly, waardoor het voor mij mogelijk is om gewoon mijn werk te blijven doen. Zo ontzettend fijn dat er altijd twee lieve oma's voor ze klaarstaan. Verder Hayco en Femke, jullie waren altijd geïnteresseerd in hoe alles ging en vroegen altijd even hoe mijn promotie verliep, fijn om dat zo te ervaren.

Als laatste Martijn, Faas en Lilly. Lieve Faas en lieve Lilly, zo klein als jullie nog zijn, wat heb ik al veel van jullie geleerd! Loslaten, relativeren, balans, keuzes, maar vooral genieten, genieten en genieten! Wat ben ik dankbaar dat jullie in mijn leven zijn! En lieve Martijn, we hebben al zoveel meegemaakt en zoveel gedaan. Dank voor al je steun tijdens mijn promotie, al die dagen dat ik je auto mocht gebruiken :-), al die momenten waarop we de leuke dingen gevierd hebben, al die verhalen die je hebt moeten aanhoren; ik kon echt altijd op je terugvallen. Dank voor ons leven samen, ik geniet er elke keer weer van!

A

About the Author

CURRICULUM VITAE

Manon Bloemen was born on February 24, 1976 in Oldenzaal, the Netherlands. After graduating from the University of Applied Sciences Enschede as a Physical Therapist in 1999, she started working as a physical therapist in pediatric rehabilitation and finished her specialization as a Pediatric Physical Therapist (PPT) in 2004 at 'Transfergroep Rotterdam'. After finishing this education, she lived in Reykjavik (Iceland) for 1.5 years; combining her work as a PPT with exploring this beautiful country. She moved back to the Netherlands at the end of 2005 and started working as a PPT at Rehabilitation center De Hoogstraat (school for special education Mytylschool Ariane de Ranitz). As she wanted to be part of improving the scientific base of pediatric physical therapy and the quality of care, she started her education as a Clinical Health Scientist (Clinical Health Science, Physical Therapy Science at the Faculty of Medicine) at University Utrecht, while continuing working as a PPT. The central theme during this study was testing and training physical fitness of children with physical disabilities. Her Master thesis resulted in two English publications (second author) and one Dutch publication (first author) and she received her Master of Science degree in 2009. Overall, Manon has more than 10 years of working experience as a PPT in the field of pediatric rehabilitation and has also worked as a PPT in several private practices. During the last year of her Master of Science education, she combined her work as a PPT with working as a lecturer at the Master Program Physical Therapy, specialization Pediatric Physical Therapy, of the HU University of Applied Sciences Utrecht. Furthermore, to develop herself as a researcher, she started as a research assistant at the Erasmus Medical Center Rotterdam in 2010. In 2011 she got the opportunity to start as a researcher in the Research group Lifestyle and Health of the HU University of Applied Sciences Utrecht, within the HALYNeD study (Healthy Active Living Youth with Neuromotor Disability: 'Active now, Healthy Later'). Since then, she combined research with lecturing activities; integrating research into the educational program. She is part of the TULIPS PhD program (Training Upcoming Leaders In Pediatric Science) 2016 - 2017. Currently, she is living with her two children, Faas and Lilly Bazen, and her boyfriend, Martijn Bazen, in Utrecht, the Netherlands.

LIST OF PUBLICATIONS

Bloemen MAT, Takken T, de Groot JF, Kruitwagen CLJJ, Rook RA, vd Berg-Emons HJG, Backx FJG.
Determinants of physical behavior in wheelchair-using youth with spina bifida.
Submitted

Bloemen MAT, vd Berg-Emons HJG, Tuijt M, Nooijen CFJ, Takken T, Backx FJG, Vos M, de Groot JF.
Physical behavior in wheelchair-using youth with Spina Bifida, an observational study.
Submitted

Bloemen MAT, de Groot JF, Backx FJG, Benner J, Kruitwagen CLJJ, Takken T.
The wheelchair shuttle test for assessing aerobic fitness in youth with Spina Bifida; a validity and reliability study.
Accepted for publication in Physical Therapy.

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 Apr 4. [Epub ahead of print]

Bloemen MA, Takken T, Backx FJ, Vos M, Kruitwagen CL, de Groot JF.
Validity and reliability of skill-related fitness tests for wheelchair-using youth with Spina Bifida.
Arch Phys Med Rehabil. 2016 Sep 12. [Epub ahead of print]

Bloemen MA, Verschuren O, van Mechelen C, Borst HE, de Leeuw AJ, van der Hoef M, de Groot JF.
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.

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 May;47(5):432-7.

Bloemen MA, Backx FJ, Takken T, Wittink H, Benner J, Mollema J, de Groot JF.
Factors associated with physical activity in children and adolescents with a physical disability: a systematic review.
Dev Med Child Neurol. 2015 Feb;57(2):137-48.

Verschuren O, Bloemen M, Kruitwagen C, Takken T.
Reference values for anaerobic performance and agility in ambulatory children and adolescents with cerebral palsy.
Dev Med Child Neurol. 2010 Oct;52(10):e222-8.

Verschuren O, Bloemen M, Kruitwagen C, Takken T.
Reference values for aerobic fitness in children, adolescents, and young adults who have cerebral palsy and are ambulatory.
Phys Ther. 2010 Aug;90(8):1148-56.

Bloemen MAT, Verschuren O, Kruitwagen CLJJ, Takken T.
Referentiewaarden voor de aerobe prestatie, anaerobe prestatie en de behendigheid bij lopende kinderen en jongeren met spastische cerebrale parese.
Nederlands Tijdschrift voor Fysiotherapie 2011;12(4).

Bloemen MAT, van Dolder R, Helders PJM.
Functionele spierkracht- en conditietraining bij een vijfjarige jongen.
FysioPraxis Juli: 2-5.

LIST OF ABSTRACTS AND PRESENTATIONS AT SCIENTIFIC CONGRESSES

Bloemen MAT, vd Berg-Emons HJG, Tuijt M, Nooijen CFJ, Takken T, Backx FJG, Vos ML, de Groot JF.
Physical behavior in wheelchair-using children with spina bifida.
European Academy on Childhood Disabilities.
Amsterdam, The Netherlands. May 2017.
Oral presentation.

Bloemen MAT, Sol ME, van der Klooster KJ, van der Krocht M.
***Let's Ride... Study*. Wheelchair skills training to stimulate participation.**
European Academy on Childhood Disabilities.
Amsterdam, The Netherlands. May 2017.
Workshop.

Bloemen MAT, Takken T, Backx FJ, Vos M, Kruitwagen CL, de Groot JF.
Validity and reliability of skill-related fitness tests for wheelchair-using youth with Spina Bifida.
World Congress on Spina Bifida.
San Diego, United States of America. March 2017.
Oral presentation.

Bloemen MAT, Takken T, de Groot JF, Kruitwagen CLJJ, Rook RA, vd Berg-Emons RHJG, Backx FJG.
Determinants for physical behavior in wheelchair-using children with Spina Bifida.
World Congress on Spina Bifida.
San Diego, United States of America. March 2017.
Oral presentation.

Bloemen MAT, Verschuren O, van Mechelen C, Borst HE, de Leeuw AJ, van der Hoef M, de Groot JF.
Factors to consider when aiming to improve participation in physical activity in youth with Spina Bifida; a qualitative study.
World Congress on Spina Bifida.
San Diego, United States of America. March 2017.
Oral presentation.

Bloemen MAT, vd Berg-Emons HJG, Tuijt M, Nooijen CFJ, Takken T, Backx FJG, Vos ML, de Groot JF.
Physical behavior in wheelchair-using youth with Spina Bifida
World Congress on Spina Bifida.
San Diego, United States of America. March 2017.
Poster presentation.

Bloemen MAT, de Groot JF, Backx FJG, Benner J, Westerveld RA, Takken T.
Testing VO_{2peak} in wheelchair-using youth with Spina Bifida: arm cranking or wheelchair propulsion? World Congress on Spina Bifida.
San Diego, United States of America. March 2017.
Poster presentation.

Bloemen MAT, de Groot JF, Backx FJG, Benner J, Westerveld RA, Takken T.
Testing VO_{2peak} in wheelchair-using youth with spina bifida: arm cranking or wheelchair propulsion?
European Workshop on Clinical Pediatric Exercise Testing.
Zeist, The Netherlands, October 2016.
Poster presentation.

Bloemen MAT, Takken T, Backx FJ, Vos M, Kruitwagen CL, de Groot JF.
Validity and reliability of skill-related fitness tests for wheelchair-using youth with Spina Bifida.
European Workshop on Clinical Pediatric Exercise Testing.
Zeist, The Netherlands, October 2016.
Poster presentation.

Bloemen MAT, de Groot JF.
Het Kennis Transfer Systeem voor Kinderfysiotherapeuten.
Najaarscongres Nederlandse Vereniging voor Kinderfysiotherapie (NVFK).
Maarssen, The Netherlands. November 2016.
Oral Presentation.

Bloemen MAT, Takken T, Backx FJ, Vos M, Kruitwagen CL, Benner J, de Groot JF.
Veldtesten voor het meten van fitheid bij rolstoel-rijdende kinderen met Spina Bifida.
Najaarscongres Nederlandse Vereniging voor Kinderfysiotherapie (NVFK).
Maarssen, The Netherlands. November 2015.
Oral Presentation.

De Groot JF, Bloemen MAT, Sol ME.
Inspanningsgerelateerde diagnostiek bij kinderen.
Najaarscongres Nederlandse Vereniging voor Kinderfysiotherapie (NVFK).
Maarssen, The Netherlands. November 2015.
Workshop.

Bloemen MAT, de Groot JF, Backx FJG, Benner J, Westerveld RA, Takken T.
Aerobe fitheidstesten bij rolstoel-rijdende jeugd met Spina Bifida: armfietsen of rolstoelpropulsie?
Jaarcongres Vereniging Sport Geneeskunde. November 2015.
Oral presentation.

Bloemen MAT, de Groot JF, Backx FJG, Benner J, Westerveld RA, Takken T.
Arm cranking versus wheelchair propulsion for testing aerobic fitness in children with Spina Bifida who are wheelchair dependent.
Pediatric Work Physiology.
Utrecht, The Netherlands. September 2015.
Oral presentation.

Bloemen MAT, Verschuren O, van Mechelen C, Borst HE, de Leeuw AJ, van der Hoef M, de Groot JF.
Personal and environmental factors to consider when aiming to improve participation in physical activity in children with Spina Bifida: a qualitative study.
Pediatric Work Physiology,
Utrecht, The Netherlands. September 2015.
Poster presentation.

Bloemen MAT, Verschuren O, van Mechelen C, Borst HE, de Leeuw AJ, van der Hoef M, de Groot JF.
Factors to consider when aiming to improve participation in physical activity in youth with Spina Bifida: a qualitative study.
World Congress Physical Therapy,
Singapore. May 2015.
Oral presentation.

Bloemen MAT, de Groot JF, Backx FJG, Benner J, Kruitwagen CLJJ, Takken T.
Field-based testing for aerobic performance in youth with Spina Bifida who use a manual wheelchair.
World Congress Physical Therapy, Singapore. May 2015.
Oral presentation.

Bloemen MAT, Takken T, Verschuren O, Wittink H, Backx FJG, de Groot JF.
Actief of niet? Dat is de vraag!
Dag van het sportonderzoek. October 2014.
Zeist, The Netherlands.
Oral presentation.

Bloemen MAT, de Groot JF, Backs FJG, Westerveld RA, Takken T.
Arm ergometry versus wheelchair propulsion for testing aerobic fitness in wheelchair-using children.
Annual congress of American College of Sports Medicine.
Orlando, United States of America. May 2014.
Poster presentation.

Bloemen MAT, de Groot JF, Backx FJG, Westerveld RA, Takken T.
Arm ergometry versus wheelchair propulsion for testing aerobic fitness in wheelchair-using children.
5th International state-of-the-art congress Rehabilitation: mobility, exercise and sports.
Groningen, The Netherlands. April 2014.
Oral presentation.

Bloemen MAT, Verschuren O, van Mechelen C, Borst HE, de Leeuw AJ, van der Hoef M, de Groot JF.
Factors to consider when aiming to improve participation in physical activity in youth with Spina Bifida: a qualitative study.
5th International state-of-the-art congress Rehabilitation: mobility, exercise and sports.
Groningen, The Netherlands. April 2014.
Poster presentation.

Bloemen MAT, de Groot JF, Backx FJG, Benner J, Takken T.
Field-based testing for aerobic performance in wheelchair dependent youth with spina bifida.
5th International state-of-the-art congress Rehabilitation: mobility, exercise and sports.
Groningen, The Netherlands. April 2014.
Poster presentation.

Bloemen MAT, Zwinkels ME, Takken T, Verschuren O, de Groot JF.
Fitheidstesten bij rolstoel-rijdende kinderen.
Congres Nederlandse Vereniging voor Kinderfysiotherapie (NVFK). June 2013.
Workshop.

Bloemen MAT, Verschuren O, van Mechelen C, Borst HE, de Leeuw AJ, van der Hoef M, de Groot JF.
Belemmerende en bevorderende factoren voor fysieke activiteit bij kinderen met Spina Bifida.
Congres Nederlandse Vereniging voor Kinderfysiotherapie (NVFK). June 2013.
Poster presentation.

Bloemen MAT, Verschuren O, van Mechelen C, Borst HE, de Leeuw AJ, van der Hoef M, de Groot JF.
Determinants for physical activity in youth with Spina Bifida.
World congress on Exercise is Medicine and annual congress of American College of Sports Medicine.
Indianapolis, United States of America. May 2013.
Poster presentation.

OTHER PUBLICATIONS, SYMPOSIA AND ACTIVITIES

Bloemen MAT, Smits F, Hermsen S, Wigmans J, Kotte E, de Groot JF.
Wat beweegt jou?!
BOSK magazine. June 2016.
Publication.

Bloemen MAT, Takken T, van den Berg-Emons HJG, Backx FJG, Nooijen CF, Tuijt M, Westerveld RA, Benner J, Rook RA, Vos ML, Kruitwagen CLJJ, de Groot JF.
Fysieke fitheid en fysiek gedrag bij rolstoel-rijdende kinderen met Spina Bifida.
Vereniging Revalidatie Artsen Nederland.
Huizen, The Netherlands. June 2016.
Oral presentation.

Bloemen MAT, Smits F, Hermsen S, Wigmans J, Kotte E, de Groot JF.
Wat beweegt jou?!
Terugkomdag ouders en kinderen op de Supportbeurs.
Utrecht, The Netherlands. May 2016.
Oral presentation.

Bloemen MAT, Blatter T.
Inspanningstesten bij rolstoel-rijdende kinderen.
Dag VVBN (vereniging van Bewegingswetenschappers Nederland).
Utrecht, The Netherlands. May 2016.
Workshop.

De Groot JF, Bloemen MAT.
Praktijkgericht onderzoek op de Hogeschool Utrecht.
Dag VVBN (vereniging van Bewegingswetenschappers Nederland).
Utrecht, The Netherlands. May 2016.
Oral presentation.

Bloemen MAT, Takken T, Backx FJG, de Groot JF.
Voorlopige resultaten HALYNeD studie en *Let's Ride... Study*. Fit For the Future!
Symposium HU University of Applied Sciences Utrecht,
Utrecht, The Netherlands. June 2015.
Oral presentation.

Bloemen MAT, Takken T, Backx FJG, Westerveld RA, Benner J, Vos ML, Kruitwagen CLJJ, de Groot JF.
Fitheidstesten bij rolstoel-rijdende kinderen met Spina Bifida.
Afdeling kinderfysiotherapie van Revalidatiecentrum Leijpark.
Tilburg, The Netherlands. May 2015.
Workshop.

Bloemen MAT, Takken T, Backx FJG, Westerveld RA, Benner J, Vos ML, Kruitwagen CLJJ, de Groot JF.
Fitheidstesten bij rolstoel-rijdende kinderen met Spina Bifida.
Afdeling kinderfysiotherapie van Revalidatiecentrum de Hoogstraat locatie Mytylschool Ariane de Ranitz.
Utrecht, the Netherlands. May 2015.
Oral presentation.

Bloemen MAT, Takken T, Backx FJG, de Groot JF.
Voorlopige resultaten HALYNeD studie.
Terugkomdag ouders en kinderen op de Supportbeurs,
Utrecht, The Netherlands. May 2014.
Oral presentation.

Bloemen MAT, de Groot JF, Backx FJG, Westerveld RA, Benner J, Kruitwagen CLJJ, Takken T.
HALYNeD *Let's Ride... Study*. Een maximale aerobe inspanningstest bij rolstoel rijdende jeugd met Spina Bifida.
Alumnidag Master Opleiding Fysiotherapie, HU University of Applied Sciences Utrecht.
Utrecht, The Netherlands. March 2014.
Oral presentation.

Bloemen MAT, de Groot JF.
Healthy Active Living for Youth with Neuromotor Disability; active now, healthy later!
BOSK magazine. November 2013.
Publication.

Bloemen MAT, de Groot JF, Backx FJG, Westerveld RA, Kruitwagen CLJJ, Takken T.
Inspanningstesten bij rolstoelgebonden kinderen.
HALYNeD symposium HU University of Applied Sciences Utrecht,
Utrecht, The Netherlands. June 2013.
Oral presentation.

Bloemen MAT, de Groot JF, Verschuren O, Backx FJG, Westerveld RA, Benner J, Takken T.
Het uitvoeren van fitheidstesten bij rolstoelgebonden kinderen.
HALYNeD symposium HU University of Applied Sciences Utrecht,
Utrecht, The Netherlands. June 2013.
Workshop.

Bloemen MAT, de Groot JF, Backx FJG, Takken T.
Ontwikkelen van meetinstrumenten voor rolstoel-rijdende kinderen met Spina Bifida.
HALYNeD symposium HU University of Applied Sciences Utrecht,
Utrecht, the Netherlands. March 2012.
Oral presentation.

FUNDING

Foundation Innovation Alliance – Regional Attention and Action for Knowledge circulation (SIA RAAK), project number 2011-13-35P. Healthy Active Living for Youth with Neuromotor Disability: active now, healthy later (HALYNeD).
Foundation Innovation Alliance - Regional Attention and Action for Knowledge circulation (SIA RAAK PRO), project number SIA RAAK PRO-4-03. Fit For the Future!
HU University of Applied Sciences Utrecht: personal PhD grant (promotie voucher).