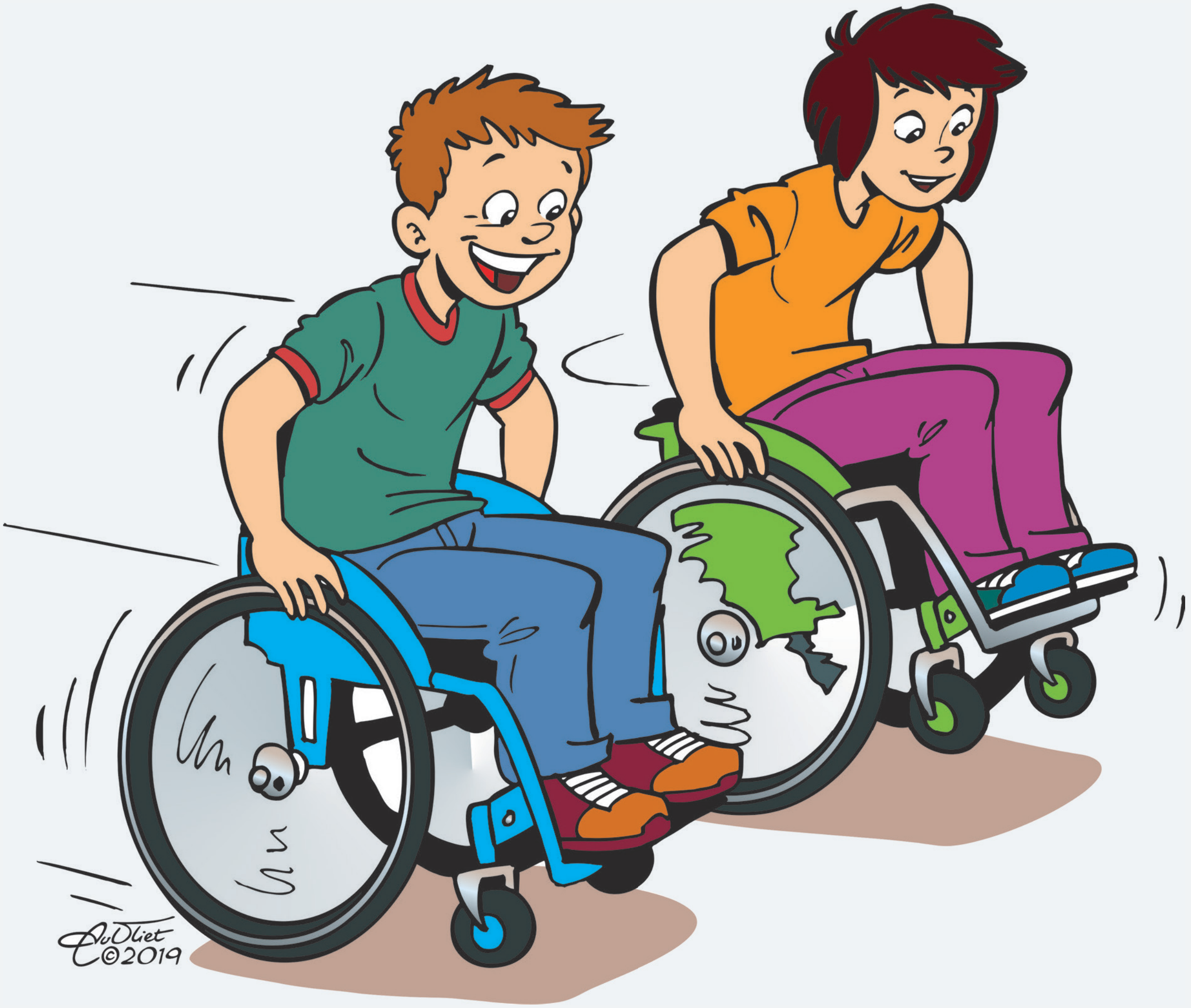
Wheelchair Mobility Skills in youth using a manual wheelchair

From Test to Training



UMC Utrecht Brain Center

Marleen Sol

Wheelchair Mobility Skills in youth using a manual wheelchair

From Test to Training

Marleen Sol

Drawing: Arie van Vliet; Sam en Sarah; samensarah.nl
Printed by: ProefschriftMaken || www.proefschriftmaken.nl
Layout: ProefschriftMaken || www.proefschriftmaken.nl
ISBN: 978-94-6423-294-3

Wheelchair Mobility Skills in youth using a manual wheelchair

From Test to Training

Rolstoelvaardigheid van test tot training
(met een samenvatting in het Nederlands)

Proefschrift

ter verkrijging van de graad van doctor aan de
Universiteit Utrecht
op gezag van de
rector magnificus, prof.dr. H.R.B.M. Kummeling,
ingevolge het besluit van het college voor promoties
in het openbaar te verdedigen op

woensdag 7 juli 2021 des middags te 12.15 uur

door

Marleen Elisabeth Sol

geboren op 8 augustus 1985
te Breda

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

Copromotoren:
Dr. J.F. de Groot
Dr. O.W. Verschuren

The studies in this thesis were financially supported by the Dutch Foundation Innovation Alliance (SIA RAAK) project number SIA RAAK PRO-4-03 and the Dutch Foundation for disabled children.
Financial support for the printing of this thesis has been generously provided by *steunfonds faculteit gezondheidszorg HU university of Applied Science, Brain center Rudolph Magnus, the Scientific College Physical Therapy (WCF) of the Royal Dutch Society for Physical Therapy (KNGF), ProCare B.V. and the Wheelchair Skill Team NL.*

Contents

Voorwoord

'Mis jij het niet, het kunnen lopen?' vraagt een vriendinnetje.
Ze kijkt haar verbaasd aan.
'Mis jij het niet, dat je niet in een rolstoel rijdt?' stelt zij de vraag terug, terwijl ze in een wheelie voor de tv balanceert.

Onze dochter Tess (16 jaar, Spina Bifida) is nog heel klein als ze haar eerste rolstoel krijgt, 18 maanden. Zij en haar rolstoel trekken al vroeg de wereld in en groeien uit tot een eenheid.

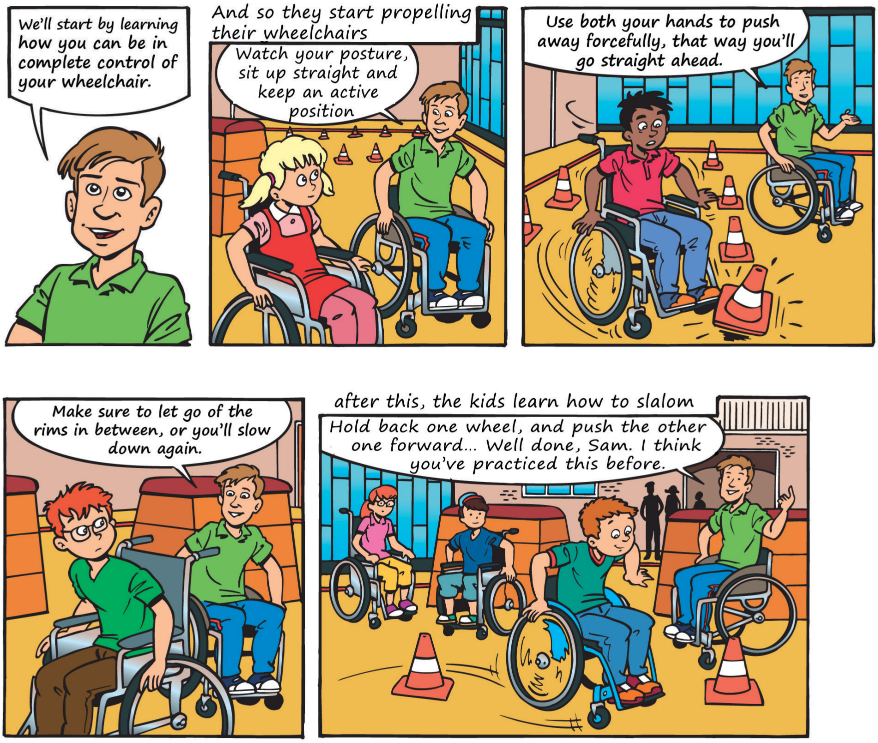
'Gelukkig heb ik heel goed geleerd hoe ik met mijn rolstoel moet omgaan. Andere mensen weten vaak niet dat je heel veel zelf kunt en vragen vaak of ze even moeten helpen. En eigenlijk moeten mensen wachten tot je zelf om hulp vraagt. Je wilt het namelijk zelf doen; alles wat je zelf kunt, wil je graag zelf doen. Ik weet precies wanneer ik hulp nodig heb.'

'Rolstoelvaardigheidstraining is heel belangrijk voor mij, ik heb daar veel geleerd. Hoe ik moet omgaan met mijn rolstoel, maar vooral dat ik kan denken in mogelijkheden. Ik kan echt super veel met mijn rolstoel en dat geeft ook een gevoel van vrijheid. Dat je niet meer afhankelijk bent van iemand anders. En je gewoon kan gaan en staan waar je wilt.'

Tess brengt het heel mooi onder woorden. Kunnen gaan en staan waar je wilt, minder afhankelijk zijn van anderen en met het volste vertrouwen de hobbels en uitdagingen op eigen kracht aan gaan. Dat is waar iedere ouder naar streeft. Opvoeden is gericht op eindigheid en heeft als doel om je kind zo zelfstandig mogelijk in de wereld te laten komen. Dat vraagt van ouders om los te laten op het juiste moment en vertrouwen te hebben in je eigen kind. De training is niet alleen voor de kinderen van belang. Ook ouders worden uitgedaagd om hun grenzen op te zoeken en te vertrouwen op de vaardigheden en bredere ontwikkeling van hun kind. Zij groeien mee in de zelfstandigheid en zelfredzaamheid van hun kind. Vanuit mijn rol als ervaringsdeskundige ouder en pedagoog, gun ik alle kinderen en ouders dit mooie en waardevolle proces van vaardigheden, (zelf)vertrouwen en op het juiste moment los laten.

Marleen draagt met haar onderzoek bij aan het versterken en verbeteren van het leven van kinderen en hun ouders. Het is een eerste, succesvolle stap in de emancipatie van kinderen in een rolstoel, waarvan er nog vele mogen volgen. Rolstoelvaardigheid als basis voor een actief, gezond en waardevol leven als volwaardige burgers.

Marleen van de Krogt en Tess Boekhoud


We'll start by learning how you can be in complete control of your wheelchair.
And so they start propelling their wheelchairs
Watch your posture, sit up straight and keep an active position
Use both your hands to push away forcefully, that way you'll go straight ahead.
Make sure to let go of the rims in between, or you'll slow down again.
after this, the kids learn how to slalom
Hold back one wheel, and push the other one forward... Well done, Sam. I think you've practiced this before.

Chapter 1

General introduction

Mother of a child with Spina Bifida*: "Wheelchair mobility skills 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 wheelchairs ....and you can just go....your life becomes a lot more fun"* [1].

YOUTH USING A MANUAL WHEELCHAIR

Spina bifida (SB) and cerebral palsy (CP) are two of the most common motor disorders in childhood leading to physical disability. The prevalence of SB is 3 to 4 per 10000 live births and stillbirths[2]. The malformation of the spinal cord and/or brain in SB can lead to sensory, cognitive and motor impairments[3]. Children and adolescents (youth) with SB can have different levels of ambulation (table 1), which can be classified with the adapted Hoffer classification[4] into non-ambulatory (completely wheelchair dependent and therapeutic ambulatory) and ambulatory (normal, community and household ambulation). Schoenmakers et al.[5] reported on the ambulation level in 103 Dutch children with SB and found 52% to be non-ambulatory and 48% to be functional ambulatory.

Table 1. Hoffer classification as adapted by Schoenmakers[5].

Level of ambulation		Description
Functional ambulatory	Normal ambulation	Independent and unrestricted ambulation without use of assistive devices
	Community ambulation	Independent outdoor ambulation without use of braces or assistive devices; using wheelchair for longer distances
	Household ambulation	Independent indoor ambulation with or without use of braces or assistive devices; using wheelchair outdoors
Non-ambulatory	Non-functional ambulation	Walking only in therapeutic situations
	Non-ambulation	Wheelchair dependent

Cerebral palsy has a prevalence ranging from 1.7 to 3 per 1000 live births[6]. The severity of CP can be classified with the Gross Motor Function Classification System (GMFCS)[7], with a higher level indicating more functional limitations. The distribution of youth with CP among the GMFCS classifications is approximately: 34% level I, 25% level II, 11% level III, 14% level IV, 16% level V[8]. Youth with CP who are classified as level IV and V use a wheelchair for all mobility, level III use a combination of wheelchair or ambulatory mobility device, level II use a wheelchair over long distances and level I are completely ambulatory[9].

PEDIATRIC REHABILITATION

In the Netherlands, youth with physical disabilities such as CP and SB are usually referred to a pediatric rehabilitation team to support their health and their development from the time of the diagnosis. Pediatric rehabilitation care is aimed at supporting optimal functioning and participation in activities of daily life for youth with physical disabilities[8]. Until recently, rehabilitation for youth with SB and CP has focused primarily on increasing ambulatory abilities with limited attention for increasing mobility using a wheelchair. This lack of attention for wheelchair mobility in pediatric rehabilitation has been confirmed in several systematic reviews[10–14] and is also reflected in a guideline for SB from the Netherlands Society of rehabilitation medicine which is entitled: "ambulatory abilities in youth with Spina Bifida"[15]. At the same time, the International Classification of Functioning and Health for Children and Youth (ICF-CY)[16] has over 20 classifications of describing 'walking', i.e. 'walking short distance', 'walking on different surfaces', 'walking and moving around', but only one description of wheelchair mobility, as part of 'moving around using equipment': *"Moving the whole body from place to place, on any surface or space, by using specific devices designed to facilitate moving or create other ways of moving around, such as with skates, skis, scuba equipment, swim fins, or moving down the street in a **wheelchair** or a walker"* (ICF-CY d465)[16].

Where it is common in pediatric rehabilitation practice and research to give functional gait training in ambulatory youth with orthoses/crutches/walkers[17–19] or powered wheelchair training for youth using a powered wheelchair[20,21], no or little attention has been given to manual wheelchair mobility skills (WMS) training in youth using a manual wheelchair[22]. Youth using a manual wheelchair often experience problems with participation in physical activity (PA)[1,23]. In a recent qualitative study towards describing factors that influence PA in youth with Spina Bifida (SB), youth using a manual wheelchair and their parents reported on the importance of WMS as a facilitating factor towards being physically active[1]. An example of one of the responses of a parent was quoted at the start of this introduction. Parents also mentioned that self-confidence seems to be crucial in the process of learning new WMS skills and using these skills in daily life[1].

ASSESSING WHEELCHAIR MOBILITY AND CONFIDENCE

There is no evidence from literature towards a validated outcome tool for assessing or improving WMS in youth using a manual wheelchair. There is only limited research towards wheelchair mobility in youth using a manual wheelchair, with one small pilot study (n=6) reporting on possible positive effects of WMS training on WMS in youth[22]. There has been far more attention for WMS training in adult wheelchair users, where

WMS training has proven to have a positive effect on WMS[24], confidence in wheelchair mobility[25] and participation[26].

In adult wheelchair users several outcome measures have been developed to assess WMS[27] or confidence in wheelchair mobility[28,29]. It was found important to assess both the capacity and confidence, as they determine whether a skill is actually performed[30], for example going up a curb or going up/down a slope[28].

It remains unclear whether these outcome tools from the adult population for assessing WMS and confidence in wheelchair mobility are also valid for the use in youth using a manual wheelchair. There is a clear difference between adult wheelchair users and youth, as youth is still developing physically and psychologically and participate in different activities than adults, e.g. playing outdoors or going to school, for which they might need different WMS. Moreover, parents may play an important role in the development of (confidence in) wheelchair mobility in youth, which is different to the role of a caregiver in adult wheelchair users. In order to ascertain content validity of an assessment instrument, it is important to assess the relevance of items for a specific population[31], i.e. youth using a manual wheelchair.

ASSESSING PHYSICAL ACTIVITY

There has been ample research towards the objective assessment of PA in youth using a manual wheelchair[32]. Only the VitaMove[33] was validated for youth using a manual wheelchair. Unfortunately, the three large body fixed sensors and complex data analysis makes the VitaMove less suitable for use in clinical practice[34]. There is an urgent need for the development of an activity monitor that can be used in clinical practice and is valid for assessing wheelchair activities in youth.

INCREASING PHYSICAL ACTIVITY

While research towards interventions aimed at increasing PA in youth using a wheelchair is lacking, there is research available about ambulatory youth with a disability[10,12–14]. Important lessons for the development of successful interventions in youth using manual wheelchair can be learned from (1) (un)successful interventions aimed at increasing PA in ambulatory youth with a physical disability, (2) qualitative descriptive research on (un)modifiable facilitators and barriers towards PA in youth with a disability (including wheelchair users) and (3) successful interventions on three of these modifiable factors (WMS, confidence in wheelchair mobility, physical fitness) in adult wheelchair users.

1. Two systematic reviews[10,13] on the effectiveness of PA intervention in youth with a disability, concluded that focusing on solely physical training in ambulatory youth with a physical disability is not effective at increasing PA. Both reviews suggested to include a behavioral component when aiming to increase PA through physical training.
2. Two qualitative studies[1,23] in youth with a disability included wheelchair users in their sample and aimed to describe facilitators and barriers towards PA. Multiple factors on all the levels of the ICF-CY were described, including the need for early attainment of (wheelchair) mobility skills, importance of physical fitness and the confidence in wheelchair mobility.
3. Recently Kirby et al.[35] concluded there are significant associations between physical fitness, WMS and confidence in wheelchair mobility in adult manual wheelchair users with a spinal cord injury and suggested that both WMS training and exercise training may be useful during rehabilitation of people with a spinal cord injury. Exercise training[36] and WMS training[24] have demonstrated effectiveness in improving physical fitness, wheelchair mobility skills and/or confidence in wheelchair mobility in adult wheelchair users.

Based on the evidence from literature in the three before mentioned fields and the clinical expertise of the partners in this research project[37], a new intervention towards increasing PA in youth using a manual wheelchair was developed in this thesis.

AIM AND OUTLINE OF THIS THESIS

Based on the lack of validated outcome measures to assess WMS and confidence in wheelchair mobility in youth using a manual wheelchair, the first aim of this thesis was to:

1. Develop or validate assessment tools

Chapter 2 describes the development and validation of the Utrecht Pediatric Wheelchair Mobility Skills Test (UP-WMST) using the current knowledge from literature in combination with the clinical expertise of health care professionals, children and their parents. In **Chapter 3,** the psychometric properties of this WMS test for youth using a manual wheelchair were assessed. This Chapter explains the adaptation of the scoring method, reliability, validity and responsiveness of the UP-WMST 2.0.

In **Chapter 4** the development of a new tool to assess confidence in wheelchair mobility in Dutch youth is described. In this study, a forward-backward translation of the original Wheelchair Confidence Scale for adults (WheelCon-M) was performed and adaptations for youth were made based on focus groups with youth using a manual wheelchair, parents and health care professionals. Furthermore, the internal consistency and construct validity of the newly developed WheelCon-Mobility Dutch Youth were assessed.

The second aim of this thesis was to assess the validity of an activity monitor in youth using a manual wheelchair to:

2. Objectively quantify physical activity

In **Chapter 5** the criterion validity of the adapted algorithm of an activity monitor (Activ8) for youth using a manual wheelchair was assessed. In this study the outcomes of the Activ8 to detect 'active wheelchair use' were compared to observations through video recording as a gold standard.

The last aim of this thesis was to develop and evaluate an intervention in youth using a manual wheelchair aimed at:

3. Increasing (determinants of) physical activity

Chapter 6 shows the results of a practice based intervention study in youth using a manual wheelchair using the previously described outcome measures. This study evaluates the short term and long term effect of a combined exercise and WMS training on PA, physical fitness, confidence in wheelchair mobility and WMS. A secondary aim was to evaluate if the order of training (exercise before or after WMS training) had a significant different effect on the outcome measures.

Finally, Chapter 7 discusses the main findings of this thesis in the theoretical and methodological considerations. It also describes the important clinical implications and directions for future research that resulted from the research process performed in this thesis.

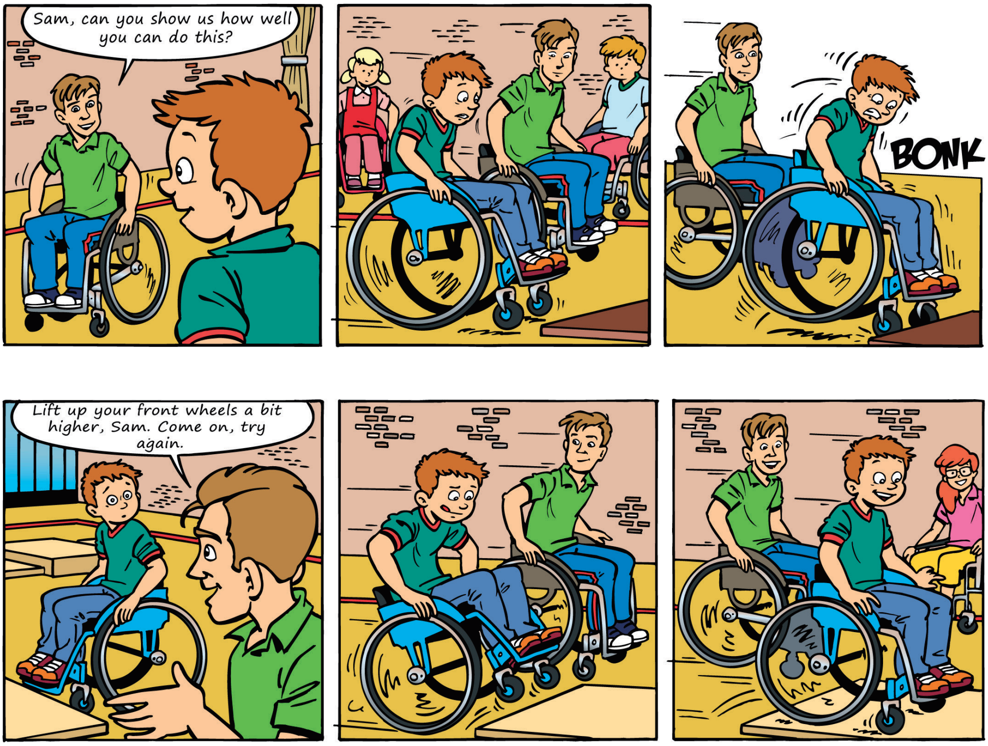
REFERENCES

1. Bloemen MAT, Verschuren O, van Mechelen C, 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(1):11. doi:10.1186/s12883-015-0265-9
2. Kondo A, Kamihira O, Ozawa H. Neural tube defects: Prevalence, etiology and prevention. *Int J Urol.* 2009;16(1):49-57. doi:10.1111/j.1442-2042.2008.02163.x
3. Lomax-Bream LE, Barnes M, Copeland K, Taylor HB, Landry SH. The impact of Spina Bifida on development across the first 3 years. *Dev Neuropsychol.* 2007;31(1):1-20. doi:10.1207/s15326942dn3101_1
4. Schoenmakers MAGC, Gulmans VAM, Gooskens RHJM, Helders PJM. Spina bifida at the sacral level: More than minor gait disturbances. *Clin Rehabil.* 2004;18(2):178-185. doi:10.1191/0269215504cr718oa
5. Schoenmakers MAGC, Uiterwaal CSPM, Gulmans VAM, Gooskens RHJM, Helders PJM. Determinants of functional independence and quality of life in children with Spina Bifida. *Clin Rehabil.* 2005;19(6):677-685. doi:10.1191/0269215505cr865oa
6. Reid SM, Meehan E, Mcintyre S, Goldsmith S, Badawi N, Reddihough DS. Temporal trends in cerebral palsy by impairment severity and birth gestation. *Dev Med Child Neurol.* 2016;58:25-35. doi:10.1111/dmcn.13001
7. Boyce WF, Gowland C, Rosenbaum PL, et al. The gross motor performance measure: Validity and responsiveness of a measure of quality of movement. *Phys Ther.* 1995;75(7):603-613. doi:10.1093/ptj/75.7.603
8. Hadders-Algra M, Maathuis K, eds. Kinderrevalidatie. In: *Edition: 5.* Koninklijke van Gorcum; 2015:3-27, 351-393, 417-435.
9. Palisano R, Rosenbaum P, Walter S, Russell D, Wood E, Galuppi B. Development and reliability of a system to classify gross motor function in children with cerebral palsy. *Dev Med Child Neurol.* 1997;39(4):214-223. doi:10.1111/j.1469-8749.1997.tb07414.x
10. 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;59(10):1004-1010. doi:10.1111/dmcn.13422
11. Zwinkels M, Verschuren O, Janssen TW, et al. Exercise training programs to improve hand rim wheelchair propulsion capacity: a systematic review. *Clin Rehabil.* 2014;28(9):847-861. doi:10.1177/0269215514525181
12. Ganz F, Hammam N, Pritchard L. Sedentary behavior and children with physical disabilities: a scoping review. *Disabil Rehabil.* March 2020:1-13. doi:10.1080/09638288.2020.1723720
13. Reedman S, Boyd RN, Sakzewski L. The efficacy of interventions to increase physical activity participation of children with cerebral palsy: a systematic review and meta-analysis. *Dev Med Child Neurol.* 2017;59(10):1011-1018. doi:10.1111/dmcn.13413
14. O'Brien TD, Noyes J, Spencer LH, Kubis H-P, Hastings RP, Whitaker R. Systematic review of physical activity and exercise interventions to improve health, fitness and well-being of children and

young people who use wheelchairs. *BMJ Open Sport Exerc Med*. 2016;2(1):e000109. doi:10.1136/bmjsem-2016-000109

15. Loopvaardigheid Spina Bifida. https://richtlijnendatabase.nl/richtlijn/loopvaardigheid_spina_bifida/loopvaardigheid_spina_bifida_-_startpagina.html. Published 2013.
16. World Health Organization. *International Classification of Functioning, Disability, and Health–Children and Youth Version*. Geneva: World Health Organization; 2011.
17. Booth ATC, Buizer AI, Meyns P, Oude Lansink ILB, Steenbrink F, van der Krogt MM. The efficacy of functional gait training in children and young adults with cerebral palsy: a systematic review and meta-analysis. *Dev Med Child Neurol*. 2018;60(9):866-883. doi:10.1111/dmcn.13708
18. Baym CL, Hedgecock JB, Rapport MJK. Functional Mobility Improved after Intensive Progressive Resistance Exercise in an Adolescent with Spina Bifida. *Pediatr Phys Ther*. 2018;30(2):E1-E7. doi:10.1097/PEP.0000000000000497
19. Moreau NG, Bodkin AW, Bjornson K, Hobbs A, Soileau M, Lahasky K. Effectiveness of rehabilitation interventions to improve gait speed in children with cerebral palsy: Systematic review and Meta-Analysis. *Phys Ther*. 2016;96(12):1938-1954. doi:10.2522/ptj.20150401
20. Field DA, Livingstone RW. Power mobility skill progression for children and adolescents: a systematic review of measures and their clinical application. *Dev Med Child Neurol*. 2018;60(10):997-1011. doi:10.1111/dmcn.13709
21. Livingstone R. A critical review of powered mobility assessment and training for children. *Disabil Rehabil Assist Technol*. 2010;5(6):392-400. doi:10.3109/17483107.2010.496097
22. Sawatzky B, Rushton PW, Denison I, McDonald R. Wheelchair skills training programme for children: A pilot study. *Aust Occup Ther J*. 2012;59(1):2-9. doi:10.1111/j.1440-1630.2011.00964.x
23. Shields N, Synnot A. Perceived barriers and facilitators to participation in physical activity for children with disability: a qualitative study. *BMC Pediatr*. 2016;16(1):9. doi:10.1186/s12887-016-0544-7
24. Keeler L, Kirby RL, Parker K, McLean KD, Hayden JA. Effectiveness of the Wheelchair Skills Training Program: a systematic review and meta-analysis *. *Disabil Rehabil Assist Technol*. 2019;14(4):391-409. doi:10.1080/17483107.2018.1456566
25. Best KL, Miller WC, Huston G, Routhier F, Eng JJ. Pilot study of a peer-led wheelchair training program to improve self-efficacy using a manual wheelchair: A randomized controlled trial. *Arch Phys Med Rehabil*. 2016;97(1):37-44. doi:10.1016/j.apmr.2015.08.425
26. Smith EM, Sakakibara BM, Miller WC. A review of factors influencing participation in social and community activities for wheelchair users. *Disabil Rehabil Assist Technol*. 2016;11(5):361-374. doi:10.3109/17483107.2014.989420
27. Fliess-Douer O, Vanlandewijck YC, Lubel Manor G, Van Der Woude LHV. A systematic review of wheelchair skills tests for manual wheelchair users with a spinal cord injury: Towards a standardized outcome measure. *Clin Rehabil*. 2010;24(10):867-886. doi:10.1177/0269215510367981
28. Rushton PW, Miller WC, Kirby RL, Janice J. Measure for the ass essment of confidence wit h manua l wheelchair use (wheelcon-m) version 2.1: Relia bility and validity. *J Rehabil Med*. 2013;45(1):61-67. doi:10.2340/16501977-1069

29. Fliess-Douer O, Vanlandewijck YC, Van Der Woude LHV. Reliability and validity of perceived self-efficacy in wheeled mobility scale among elite wheelchair-dependent athletes with a spinal cord injury. *Disabil Rehabil*. 2013;35(10):851-859. doi:10.3109/09638288.2012.712198
30. Bandura A, Freeman W, Lightsey R. *Self-Efficacy: The Exercise of Control*. New York; 1999. https://connect.springerpub.com/content/sgrjcp/13/2/158.full.pdf. Accessed June 9, 2020.
31. Mokkink LB, Terwee CB, Knol DL, et al. The COSMIN checklist for evaluating the methodological quality of studies on measurement properties: A clarification of its content. *BMC Med Res Methodol*. 2010;10. doi:10.1186/1471-2288-10-22
32. Lankhorst K, Oerbekke M, van den Berg-Emons R, Takken T, de Groot J. Instruments Measuring Physical Activity in Individuals Who Use a Wheelchair: A Systematic Review of Measurement Properties. *Arch Phys Med Rehabil*. 2020;101(3):535-552. doi:10.1016/j.apmr.2019.09.006
33. Nooijen CFJ, De Groot JF, Stam HJ, Van Den Berg-Emons RJG, Bussmann HBJ. Validation of an activity monitor for children who are partly or completely wheelchair-dependent. *J Neuroeng Rehabil*. 2015;12(1). doi:10.1186/s12984-015-0004-x
34. Bloemen MAT, van den Berg-Emons RJG, Tuijt M, et al. Physical activity in wheelchair-using youth with Spina Bifida: an observational study. *J Neuroeng Rehabil*. 2019;16(1):9. doi:10.1186/s12984-018-0464-x
35. Kirby RL, de Groot S, Cowan RE. Relationship between wheelchair skills scores and peak aerobic exercise capacity of manual wheelchair users with spinal cord injury: a cross-sectional study. *Disabil Rehabil*. 2020;42(1):114-121. doi:10.1080/09638288.2018.1493545
36. Valent L, Dallmeijer A, Houdijk H, Talsma E, van der Woude L. The effects of upper body exercise on the physical capacity of people with a spinal cord injury: A systematic review. *Clin Rehabil*. 2007;21(4):315-330. doi:10.1177/0269215507073385
37. Fit for the Future. https://husite.nl/fitforthefuture/samenwerking/. Accessed December 16, 2020.


Sam, can you show us how well you can do this?
BONK
Lift up your front wheels a bit higher, Sam. Come on, try again.

Chapter 2

Development of a wheelchair mobility skills test for children and adolescents: combining evidence with clinical expertise

Marleen Elisabeth Sol MSc[1,3,4], Olaf Verschuren PhD[3,4], Laura de Groot PT[4], Janke Frederike de Groot PhD[1,2], Fit-For-the-Future!-consortium.

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, Utrecht, The Netherlands
3. Brain Center Rudolf Magnus and Center of Excellence for Rehabilitation Medicine, University Medical Center Utrecht, Utrecht, The Netherlands
4. De Hoogstraat Rehabilitation, Utrecht, The Netherlands

BMC Pediatr. 2017;17:51

ABSTRACT

Background: Wheelchair mobility skills (WMS) training is regarded by children using a manual wheelchair and their parents as an important factor to improve participation and daily physical activity. Currently, there is no outcome measure available for the evaluation of WMS in children. Several wheelchair mobility outcome measures have been developed for adults, but none of these have been validated in children. Therefore the objective of this study is to develop a WMS outcome measure for children using the current knowledge from literature in combination with the clinical expertise of health care professionals, children and their parents.

Methods: Mixed methods approach. Phase 1: Item identification of WMS items through a systematic review using the 'COnsensus-based Standards for the selection of health Measurement Instruments' (COSMIN) recommendations. Phase 2: Item selection and validation of relevant WMS items for children, using a focus group and interviews with children using a manual wheelchair, their parents and health care professionals. Phase 3: Feasibility of the newly developed Utrecht Pediatric Wheelchair Mobility Skills Test (UP-WMST) through pilot testing.

Results: Phase 1: Data analysis and synthesis of nine WMS related outcome measures showed there is no widely used outcome measure with levels of evidence across all measurement properties. However, four outcome measures showed some levels of evidence on reliability and validity for adults. Twenty-two WMS items with the best clinimetric properties were selected for further analysis in phase 2. Phase 2: Fifteen items were deemed as relevant for children, one item needed adaptation and six items were considered not relevant for assessing WMS in children. Phase 3: Two health care professionals administered the UP-WMST in eight children. The instructions of the UP-WMST were clear, but the scoring method of the height difference items needed adaptation. The outdoor items for rolling over soft surface and the side slope item were excluded in the final version of the UP-WMST due to logistic reasons.

Conclusions: The newly developed 15 item UP-WMST is a validated outcome measure which is easy to administer in children using a manual wheelchair. More research regarding reliability, construct validity and responsiveness is warranted before the UP-WMST can be used in practice.

Keywords: Children, Wheelchair mobility skills, Wheelchair mobility, Outcome measure

BACKGROUND

Two of the most common motor disorders in childhood in the Netherlands are Cerebral Palsy with a prevalence of 2.5 per 1000 births[1], and neural tube defects with a prevalence of 6.52 per 10.000 births[2]. A large proportion of these children use a manual wheelchair for their daily mobility[3]. In adults, several studies have reported on the importance of wheelchair mobility skills (WMS) to overcome mobility problems and improve participation [4, 5]. Moreover, it has been shown that WMS training in adults can decrease their mobility problems by improving their WMS[6-9]. In children, evidence is limited, with only one pilot study by Sawatzki et al. looking at the effects of WMS training in six children using a manual wheelchair[10]. At the same time though, the importance of WMS training in children was recently confirmed in a qualitative study exploring factors associated with levels of physical activity[11]. One of the facilitating factors identified by children and their parents was WMS training. This can be illustrated by a quote from one of the parents: *"Wheelchair training, that is very important I think, .she can do much more now….a lot of places are not adjusted for wheelchairs ….and you can just go….your life becomes a lot more fun"*[11].

In the last decade a large variety of WMS related outcome measures has been developed for adults using a manual wheelchair[12]. In order to evaluate a WMS training for children, there is a need for such an outcome measure in this population as well. The pilot study by Sawatzki et al. was the only intervention study reporting on the use of a WMS outcome measure in children and used an adapted version of the WST 3.2[10]. However, this WMS outcome measure was developed for adult manual wheelchair users and has not been validated for use in children. It is recommended to validate an outcomes measure again if it is applied in a new population[13]. This is important because certain items could be irrelevant, other items might need adaptation or new items need to be included for different populations. In this case wheelchair outcome measures have been developed for adults with spinal cord injury, stroke or amputation, whereas children more often use a manual wheelchair due to congenital defects such as cerebral palsy or neural tube defects.

To the best of our knowledge, no WMS outcome measure has been specifically developed for or validated in children using a manual wheelchair.

The best available WMS outcome measures for adults could potentially be used for validation in children. Unfortunately, there is currently no consensus among clinicians and researchers on the best outcome measure in adults to evaluate WMS[12, 14, 15]. One of the reasons for this lack in consensus could be the difference in definitions used for the selection of items, including wheelchair user function, manual wheelchair use, wheelchair driving or wheelchair mobility[12, 16]. In this paper we use the term WMS, as skills that address aspects of wheelchair mobility. In the International Classification of Functioning (ICF) [17] wheelchair mobility is classified in Chapter 4 (Mobility) as moving around using equipment (d465) and defined as "moving the whole body from place to place, on any surface or space, by using specific devices designed to facilitate moving or create other ways of moving around, such as a wheelchair". This definition excludes other activities in a wheelchair such as transferring oneself or handling objects.

There is currently no outcome measure available for the evaluation of WMS training in children. Therefore, the objective of this study was to develop (based on available literature and expert opinion) a WMS outcome measure for children using a manual wheelchair.

METHODS

In this study, the recommendations for the development of outcomes measures by the 'COnsensus-based Standards for the selection of health Measurement INstruments' (COSMIN) checklist[18] was followed. The COSMIN checklist was developed in a Delphi study by an international team of leading experts in epidemiology, psychometrics, and health care[18]. One of these recommendation involves combining evidence from literature with clinical expertise, i.e. opinion of the target population and health care professionals[13].This process is illustrated in Figure 1 and included the following phases: (1) Identification of potentially relevant WMS items with good measurement properties through a systematic review and best evidence synthesis regarding validity, reliability and responsiveness of existing WMS outcome measures (2) Selection of WMS items relevant for children using the opinion of children, their parents and health care professionals, (3) Pilot testing the feasibility of WMS items in children using a manual wheelchair.

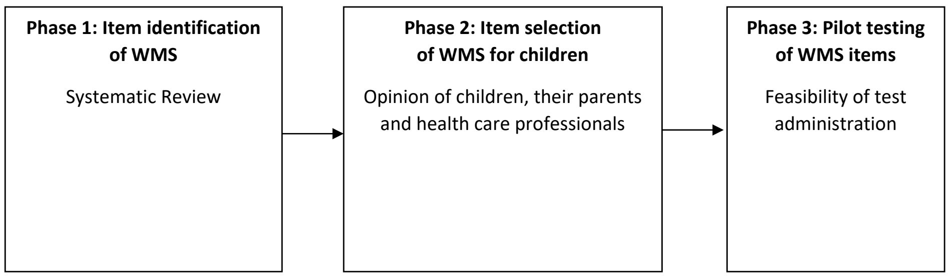


Figure 1. Methodological process of the development of a WMS outcome measure for children.

Phase 1 Item identification of WMS

Data Sources and Searches We updated the most recent systematic review on WMS from 2010 by Fliess-Douer et al.[12] The same search string as Fliess- Douer et al.[12] was applied to the following databases: Pubmed, Cochrane and Web of Science up to July 2015. The full search strategy for Pubmed is described in additional file 1.

Study Selection The selection of articles was independently performed by 2 reviewers (MS and JdG). While the search string was similar to Fliess-Douer et al.[12] the criteria used for selection were adapted to include WMS outcome measures for people with all types of disability, instead of only those for people with a spinal cord injury (SCI). This resulted in the following inclusion criteria : (1) aim of the study was to assess wheelchair skill performances in a wheelchair, (2) outcome measure is constructed for people using a manual wheelchair, (3) available statistical data regarding reproducibility or validity (4) full report written in English and publication date January 2010–July 2015. Studies were excluded when: (1) constructed for people using power wheelchairs, (2) developed for assessing in virtual environment, (3) focused on 'body function and structures' (measuring specific physiological and/or biomechanical variables which do not comply with the terms of 'activity' or 'participation' domains as defined in the ICF[17].

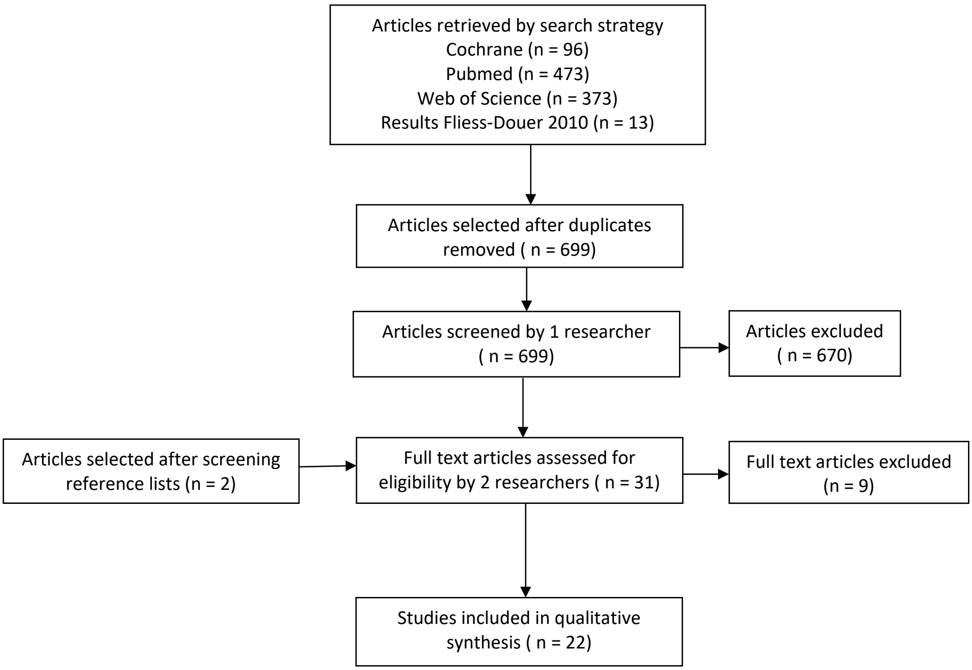


Figure 2. Flow chart of the search strategy till July 2015 and selection of articles.

Assessment of methodological quality Studies reporting a total and item score were divided into sub studies to be able to differentiate between statistical methods being used. Two reviewers (MS and JdG) independently evaluated the methodological quality of the included studies using the COSMIN checklist[19]. The COSMIN checklist contains twelve boxes, which assess the methodological quality of the studies regarding reliability, measurement error, content validity, hypothesis testing, cross-cultural validity, structural validity, criterion validity, and responsiveness. The items in each box are rated with a 4-point scoring system; excellent, good, fair, and poor. A quality score per measurement property was obtained by taking the lowest rating of any item in a box ("worst score counts"). One item in each box concerns the sample size requirements, with a minimal requirement of n>30 for an adequate sample size. As the COSMIN checklist was originally developed for health related questionnaires, sample size requirements might differ for performance based measures and can alternatively be based on power calculations as earlier discussed by Bartels et al.[20]. Therefore, the sample size requirement for assessment of methodological quality of reliability was adjusted to N ≥ 20, based on a sample size determination for a WMS outcome measure with power calculation from Kirby et al.[21].

Data extraction and best evidence synthesis Two reviewers (MS, OV) independently performed the data extraction and assessed the results of the studies based on the quality criteria described by Terwee et al.[22]. The possible ratings per measurement property were "positive," "indeterminate" and "negative". Studies looking at different measurement properties of the same outcome measure were pooled for best evidence synthesis. This synthesis combines the methodological quality of the studies with the consistency of their results[23]. The level of evidence for each outcome measure was subsequently rated as "strong", "moderate", "limited", "conflicting", or "unknown" per measurement property. This method is similar to the method used for the systematic review of clinical trials as suggested by the Cochrane Collaboration Back Review Group[23].

Selection of WMS outcome measure The WMS outcome measures with some level of evidence across reliability and validity were grouped together for item selection in phase 2.

Phase 2: Item selection of WMS for children

The resulting list of WMS items identified in phase 1 was assessed on their relevance for children using a manual wheelchair. Relevance checking was performed through a focus group or individual interviews with children using a manual wheelchair, their parents and health care professionals. The children and their parents were recruited from a voluntary WMS training program, which was set to start a few weeks later. Physiotherapy students were trained by an experienced qualitative researcher to conduct interviews with parents and children following a topic list. Individual interviews were conducted with the children and their parents separately or, in case this was preferred by the child, together. The parents and children were asked open ended questions about their current limitations regarding wheelchair mobility, their expectations of the WMS training and training goals. Open ended interview questions were preferred over relevance checking per item as this method assured an open mind regarding WMS which are relevant for children, without being influenced by WMS for adults. All interviews were recorded by video and transcribed verbatim. After transcription, a qualitative Framework Method Analyses[24] was performed for all interviews by 2 independent researchers to determine relevant items. The coding framework was based on the compiled list of items from the results of phase 1. Concurrently with the individual interviews, a focus group interview was conducted with health care professionals with clinical expertise in pediatric rehabilitation. All health care professionals were currently working at a special needs school and employed by De Hoogstraat rehabilitation centre, the Netherlands. Every potential WMS item from phase 1 was assessed in the focus group with health care professionals on the appropriateness for children and rated as 'relevant', 'relevant with adaptations' or 'not relevant'. Professionals were asked to keep in mind a total test duration of an hour, to make sure all items were critically assessed on relevance. One researcher (LdG) documented the answers given by the professionals. The results of the qualitative framework analyses of the target population was combined with the opinion of the health care professional to develop a new assessment tool with the work name: Utrecht Pediatric Wheelchair Mobility Skills Test (UP-WMST).

Phase 3: Pilot testing of WMS items

One occupational therapist and one physiotherapist were asked to provide written comments and answer question regarding: 1) the feasibility to assess WMS within one hour; 2) the ease of handling material; and 3) clarity of instructions when administering the UP-WMST to children using a manual wheelchair. This was followed by individual interviews with the therapists. Both health care professionals received a manual of the UP-WMST with instructions about test set-up and instructions per item. Children who use a manual wheelchair were recruited from a special needs school in Utrecht, the Netherlands.

Table 1. Characteristics of WMS outcome measures

Instrument	Version	Study	Construct	Target population		# WMS items/ # total items	Duration	Outcome Parameters (Scale)
				Disease characteristics	Age			
WPT		**Askari (2013)**	Manual wheelchair propulsion	Manual wheelchair user	≥ 17y	1/1	≤ 50 seconds	Propulsion method
TOWM & Wheelie test		**Fliess-Douer (2012, 2013a, 2013b)**	Manual wheelchair mobility	SCI	18-65y	36/38	40 minutes	Ability (0-1), Quality (0-5), Time (s), Anxiety (0-10)
WC		**Kilkens (2002, 2004)**	Manual wheelchair mobility	SCI	18-65y	5/8	na	Ability (0-1), Performance time (s), Physical strain (max HR)
	AM WC	**Cowan (2011)**	Manual wheelchair mobility	SCI	> 18y	12/14	na	Ability (0-1), Performance time (s)
	WAIMS	**Vereecken (2012)**	Manual wheelchair driving	MS	na	7/8	20-30 minutes	Ability (0-2), Performance time (s), Covered distance (m)
Slalom test		**Gagnon (2011)**	Manual wheelchair propulsion	SCI	≥ 18y	1/1	17 minutes	Time (s)
WST	1.0	**Kirby (2002)**	Manual wheelchair user function in daily life	Manual wheelchair user	≥ 18y	14/33	29 minutes	Safety completion (0-2),
	2.4	**Kirby (2004)**	Manual wheelchair user function	Manual wheelchair user	≥ 17y	30/50	27 minutes	Ability (pass/fail/NA), GAS
	4.1	**Lindquist (2010)**	Manual wheelchair user function	Manual wheelchair user	> 16y	24/30	31 minutes	Ability (pass/fail). Safety (safe/unsafe)
HMAT		**Harvey (1998)**	Manual wheelchair mobility	SCI	na	3/6	< 15 minutes	Level of assistance (1-6)
FIM + 5AML		**Middleton (2006)**	Manual wheelchair user functional independence	SCI	18-65y	3/5	na	Level of assistance (1-7)
OCAWUP		**Routhier (2004, 2005)**	Wheelchair mobility	Manual and power wheelchair user	18-65y	10/10	60-90 minutes	Time (s), Degree of ease (0-3)

Continue

Continued

Instrument Version	Study	Construct	Target population		# WMS items/ # total items	Duration	Outcome Parameters (Scale)
			Disease characteristics	Age			
TMT	**Chafetz (2004)**	Manual wheelchair user function	SCI	8-14y	2/6	60-90 minutes	Time (s)
WUFA	**Stanley (2003)**	Manual wheelchair user function	Manual wheelchair user	na	6/13	60-90 minutes	Level of independence (1-7)
WC-PFP	**Cress (2002)**	Manual wheelchair user function	Manual wheelchair user	18-67y	1/11	40 minutes	Speed (1/time)
VFM	**Taricco (2000)**	Manual wheelchair user function	SCI	All age groups	9/65	30-50 minutes	Level of independence (1-5)
TOMP	**Gans (1988)**	Physical function and motor performance	All disabilities	> 6y	2/32	< 60 minutes	Assistance (0-4), Approach (0-1), Pattern (0-1), Proficiency (0-2)
TM	**Jebsen (1970)**	Mobility	Locomotor disorders	> 20y	2/31	60 minutes	Time (s)

WMS = wheelchair mobility skills, WPT = wheelchair propulsions test, TMT = Timed Motor Test for wheelchair users, AM-WC = adapted manual wheelchair circuit, WC-PFP = wheelchair physical functional performance test, TOWM = test of wheeled mobility, TOMP = tufts assessment of motor performance, HMAT = Harvey mobility assessment tool, TM = measurement of time, WC = wheelchair circuit, WST = wheelchair skills test, FIM = functional independence measure, 5-AML= five additional mobility and locomotor items, OCAWUP = obstacle course assessment of wheelchair user performance, WUFA = wheelchair user functional assessment, VFM = Valutazione Funzionale Mielolesi, WAIMS = wheelchair assessment instrument for people with multiple sclerosis, NA = not applicable, SCI = spinal cord injury, MS = multiple sclerosis, HR = heart rate, GAS = Goal attainment scale.

Table 2. General information per study

Study	Instrument	Participants (N)	Disease characteristics	Mean Age (mean years ± SD)	Sex (M/F)
Askari et al. 2013	WPT	N = 58 *	Amputation, Spinal Cord Injury, Stroke, Traumatic Brain Injury, others	58 ± 17.9	35/13
Chafetz et al. 2004	TMT	N = 11	Spinal Cord Injury	10.5 ± 2.1	6/5
Cowan et al. 2011	AM WC	N = 50	Spinal Cord Injury	41.9 ± 13.4	42/8
Cress et al. 2002	WC-PFP	N = 18 *	Multiple Sclerosis, Stroke, Spinal Cord Injury, Polio, Arthritis, Brain injury	49.4 ± 10.6	13/5
Fliess-Douer 2012	TOWM & Wheelie	N = 126	Spinal Cord Injury	na	91/35
Fliess-Douer 2013(a)	TOWM & Wheelie	N = 30 *	Spinal Cord Injury	38.8 ± 8.0	na
Fliess-Douer 2013(b)	TOWM & Wheelie	N = 30 *	Spinal Cord Injury	38.8 ± 8.0	na
Gagnon et al. 2011	Slalom test	N = 15	Spinal Cord Injury	40.7 ± 12.6	na
Gans et al. 1988	TOMP	N = 40	Neurological disorder, musculoskeletal disorder	25.6 ± 19.5	14/26
Harvey et al. 1998	HMAT	N = 20	Spinal Cord Injury	45.6 ± 16.8	na
Jebsen et al. 1970	TM	N = 18	Stroke, Amputation, Peripheral neuropathy, Polio, Spinal Cord Injury, others	49.7 ± na	na
Kilkens et al. 2002	WC	N = 27	Spinal Cord Injury	34.7 ± 12.5	18/9
Kilkens et al. 2004	WC	N = 74	Spinal Cord Injury	40.5 ± 14.5	51/23
Kirby et al. 2002	WST 1.0	N = 24	Amputation, Stoke, Musculoskeletal disorder, Spinal Cord Injury, Neuromuscular disorder	59 ± 19	16/8
Kirby et al. 2004	WST 2.4	N = 298 *	Amputation, Musculoskeletal, Spinal Cord Injury, Stroke, Able bodied	43.8 ± 22.5	158/140
Lindquist et al. 2010	WST 4.1	N = 11	Spinal Cord Injury, Stroke, other	42.1 ± 16.2	9/2
Middleton et al. 2006	FIM +5-AML	N = 39	Spinal Cord Injury	28 ± 6.5	32/7

Continue

Continued

Study	Instrument	Participants (N)	Disease characteristics	Mean Age (mean years ± SD)	Sex (M/F)
Routhier et al. 2004	OCAWUP	N = 17	Spinal Cord Injury, Neuromuscular disorder, Stroke, Amputation	50.9 ± 12.1	10/7
Routhier et al. 2005	OCAWUP	na	na	na	na
Stanley 2003	WUFA	N = 5	Spinal Cord injury, Cerebral Palsy, Transverse myelitis	34.2 ± 10.3	4/1
Taricco 2000	VFM	N = 100	Spinal Cord Injury	37 ± na	77/23
Vereecken 2012	WAIMS	N = 50	Multiple Sclerosis	50 ± 12	30/20

M = male, F = female, * Subset of participants differs per measurement property used for reliability sample, na = no information available, WPT = wheelchair propulsions test, TMT = Timed Motor Test for wheelchair users, AM-WC = adapted manual wheelchair circuit, WC-PFP = wheelchair physical functional performance test, TOWM = test of wheeled mobility, TOMP = tufts assessment of motor performance, HMAT = Harvey mobility assessment tool, TM = measurement of time, WC = wheelchair circuit, WST = wheelchair skills test, FIM = functional independence measure, 5-AML= five additional mobility and locomotor items, OCAWUP = obstacle course assessment of wheelchair user performance, WUFA = wheelchair user functional assessment, VFM = Valutazione Funzionale Mielolesi, WAIMS = wheelchair assessment instrument for people with multiple sclerosis.

RESULTS

Phase 1: Item identification of WMS

Search results The search strategy combined with the previous results from Fliess-Douer et al.[12] resulted in a total of 699 unique articles, of which 31 were selected for full text assessment (Figure 2). Nine studies were excluded after full text assessment. After exclusion, 22 studies were considered eligible for this review. The main reasons for exclusions were; the absence of psychometric properties of the outcome measure being used[10, 25-28] ; outcome measures focused on the level of 'body function and structures'[26, 29] and one outcome measure was a questionnaire[30].

Study characteristics The 22 studies reported on 15 different outcome measures. The general characteristics of these outcome measures are presented in Table 1. The Wheelchair Circuit[31-34] (WC) and the Wheelchair Skills test [21, 35-37] (WST) have been further developed into additional versions. Most outcome measures were constructed to assess either wheelchair mobility (TOWM, Wheelie test, Wheelchair Circuit, HMAT and OCAWUP)[31-33, 38-43] or wheelchair user function (WST, AML, TMT, WUFA, WC-PFP, VFM)[21, 35, 36, 44-48]. Three outcome measures focused on a specific aspect of wheelchair mobility; wheelchair propulsion (WPT, slalom test)[49, 50] or wheelchair driving (WC-WAIMS)[34]. Two outcome measures were constructed to reflect a broad overview of physical function[51] and mobility[52]. All fifteen outcome measures contained items specifically related to wheelchair mobility, ranging from 1/11 WMS items[47] to 10/10 WMS[42, 43] items per total number of items.

Table 2. shows the general characteristics of the studies; number of participants, disease characteristics, mean age and sex. Due to sample sizes requirements of N ≥ 20, eight studies[36, 42, 43, 45-47, 50, 52] were excluded for further data synthesis and analysis. This exclusion includes the TMT[45] which was the only outcome measure specifically developed for children.

Measurement properties The methodological quality and level of evidence of the studies are presented in Table 3 and 4 for each measurement property, arranged per outcome measure. No studies assessed all measurement properties. Reliability and hypothesis testing were the most frequently reported properties. Different methods were used to assess inter-rater reliability; some studies used 2 raters to separately assess the same video recording, whereas other studies used 2 raters to separately administer the test. Only 3 studies[46, 47, 49] demonstrated levels of evidence on content validity. Criterion validity was not assessed as there is no gold standard available. Some studies reported on the Smallest Detectable Change or Limits of Agreement, but no studies calculated the Minimal Important Change needed to determine the level of evidence for the measurement error of an instrument. Therefore no levels of evidence were found for any of the outcome measures on criterion validity and interpretability.

Wheelchair Skills Test (WST)

The WST 1.0 was originally developed by Kirby et al.[35] consisting of 33 items measuring wheelchair user functional skills in daily life for adults using a manual wheelchair. Fourteen of these items assess WMS, the other items assess other activities in a wheelchair, such as transfers or handling objects. The level of evidence for content validity of this outcome measure is unknown. A number of items and the outcome parameter were adapted in the WST 2.4 by Kirby et al.[21] The WST 2.4 demonstrated good methodological quality for the reliability of the total score. The scoring of individual items reached a poor methodological quality, due to statistical flaws. Overall the WST shows moderate levels of positive evidence on reliability of the total score, moderate positive levels of evidence for hypothesis testing and unknown or no information on the other measurement properties.

Wheelchair Propulsion Test (WPT)

Askari et al.[49] reported on the WPT, which is a quick test consisting of one WMS item measuring several parameters of wheelchair propulsion. This studies demonstrates limited to moderate levels of positive evidence on reliability. Moderate levels of positive evidence on content validity and hypothesis testing. Even though the structural validity showed good methodological quality, the level of evidence is unknown as the explained variance was not mentioned in the results.

Test of Wheeled Mobility (TOWM) and Wheelie Test

Fliess-Douer et al.[38-40] demonstrated poor content validity. All 38 items, except the wheelchair transfer, assess WMS. Although a large sample size was used to create a list of essential WMS, there was no assessment if all items together comprehensively reflect the construct to be measured. The statistical method regarding the reliability of item quality scores was inadequate, however the method used for all other scores was appropriate. Therefore the level of positive evidence is moderate for test-retest reliability of all scores, except for the item quality scores. There is unknown or no level of evidence for all other measurement properties.

Tufts Assessment of Motor Performance (TOMP)

This assessment tool for functional motor skills in all disabilities was developed by Gans et al.[51] The tool consists of 32 items in total with 2 items assessing WMS. This study demonstrated a limited level of positive evidence for inter rater reliability. No other measurement properties were assessed.

Table 3. Methodological quality of measurement properties on reliability and best evidence synthesis

Instrument version	Author	Reliability				
		Internal consistency	Test-retest	Intra rater	Inter rater	Measurement error
WST						
1.0	Kirby 2002					
	Substudy 1: total score	.	poor/indeterminate	fair/positive	fair/positive [a]	.
	Substudy 2: Item score	.	poor/indeterminate	poor/indeterminate	poor/indeterminate [a]	.
2.4	Kirby 2004					
	Substudy 1: total score	.	good/positive	good/positive	good/positive [a]	.
	Substudy 2: Item score	.	poor/indeterminate	poor/indeterminate	poor/indeterminate [a]	.
Level of evidence		na	moderate positive	moderate positive	moderate positive	na
WPT						
	Askari 2013	.	fair/positive	.	good/positive [a]	.
Level of evidence		na	limited positive	na	moderate positive	na
WC						
	Kilkens 2002	.	good/positive	.	good/positive	.
	Kilkens 2004	.	.	.	.	.
AM-WC	Cowan 2011	.	good/positive	.	.	good/indeterminate
WAIMS	Vereecken 2012	.	fair/positive	.	fair/positive	.
Level of evidence		na	strong positive	na	moderate positive	unknown
TOWM Wheelie			.			
	Fliess-Douer 2012	.	.	.	.	.
	Fliess-Douer 2013(a)					
	Substudy 1: quality item score	.	poor/indeterminate	poor/indeterminate	poor/indeterminate [a]	good/indeterminate
	Substudy 2: other scores	.	good/positive	.	.	good/indeterminate

Continue

Continued

Instrument version	Author	Reliability				
		Internal consistency	Test-retest	Intra rater	Inter rater	Measurement error
	Fliess-Douer 2013(b)	.	.	.	.	.
Level of evidence		na	moderate positive	unknown	unknown	unknown
TOMP						
	Gans 1988	.	.	.	fair/positive [a]	.
Level of evidence		na	na	na	limited positive	na
HMAT						
	Harvey 1998	.	.	.	good/positive	.
Level of evidence		na	na	na	moderate positive	na
FIM + 5-AML						
	Middleton 2006	fair/positive	.	.	.	.
Level of evidence		limited positive	na	na	na	na
VFM						
	Taricco 2000	good/positive	.	.	.	.
Level of evidence		moderate positive	na	na	na	na

[a] Based on video rating

Table 4. Methodological quality of measurement properties on validity, responsiveness and best evidence synthesis

Instrument Version	Author	validity			
		Content validity	**Structural validity**	**Hypothesis testing**	**Responsiveness**
WST					
1.0	Kirby 2002				
	Substudy 1: total score	fair/indeterminate	.	fair/positive	.
	Substudy 2: Item score	.	.	.	.
2.4	Kirby 2004				
	Substudy 1: total score	.	.	fair/positive	.
	Substudy 2: Item score	.	.	.	.
Level of evidence		unknown	na	moderate positive	na
WPT					
	Askari 2013	good/positive	good/indeterminate	good/positive	.
Level of evidence		moderate positive	unknown	moderate positive	na
WC					
	Kilkens 2002	.	.	.	.
	Kilkens 2004	.	.	fair/positive	fair/positive
AM-WC	Cowan 2011	.	.	good/negative	.
WAIMS	Vereecken 2012	.	.	fair/positive	.
Level of evidence		na	na	conflicting	limited positive
TOWM Wheelie					
	Fliess-Douer 2012	poor/indeterminate	.	.	.
	Fliess-Douer 2013(a)				
	Substudy 1: quality item score	.	.	.	.
	Substudy 2: other scores	.	.	.	.

Continue

Continued

Instrument Version	Author	validity			
		Content validity	Structural validity	Hypothesis testing	Responsiveness
	Fliess-Douer 2013(b)	poor/indeterminate	.	poor/indeterminate	.
Level of evidence		unknown	na	unknown	na
TOMP					
	Gans 1988	.	.	.	.
Level of evidence		na	na	na	na
HMAT					
	Harvey 1998	.	.	.	.
Level of evidence		na	na	na	na
FIM + 5-AML					
	Middleton 2006	.	fair/positive	fair/positive	fair/positive
Level of evidence		na	limited positive	limited positive	limited positive
VFM					
	Taricco 2000	.	fair/indeterminate	poor/indeterminate	fair/indeterminate
Level of evidence		na	unknown	unknown	unknown

WST = wheelchair skills test, WPT = wheelchair propulsions test, WC = wheelchair circuit, AM-WC = adapted manual wheelchair circuit, WAIMS = wheelchair assessment instrument for people with multiple sclerosis, TOWM = test of wheeled mobility, TOMP = tufts assessment of motor performance, HMAT = Harvey mobility assessment tool, FIM = functional independence measure, 5-AML= five additional mobility and locomotor items, VFM = Valutazione Funzionale Mielolesi, na = no information available.

Table 5. Methodological quality of measurement properties on validity, responsiveness and best evidence synthesis

Instrument Version	Author	validity			
		Content validity	Structural validity	Hypothesis testing	Responsiveness
WST					
1.0	Kirby 2002				
	Substudy 1: total score	fair/indeterminate	.	fair/positive	.
	Substudy 2: Item score	.	.	.	.
2.4	Kirby 2004				
	Substudy 1: total score	.	.	fair/positive	.
	Substudy 2: Item score	.	.	.	.
Level of evidence		unknown	na	moderate positive	na
WPT					
	Askari 2013	good/positive	good/indeterminate	good/positive	.
Level of evidence		moderate positive	unknown	moderate positive	na
WC					
	Kilkens 2002	.	.	.	.
	Kilkens 2004	.	.	fair/positive	fair/positive
AM-WC	Cowan 2011	.	.	good/negative	.
WAIMS	Vereecken 2012	.	.	fair/positive	.
Level of evidence		na	na	conflicting	limited positive
TOWM Wheelie					
	Fliess-Douer 2012	poor/indeterminate	.	.	.
	Fliess-Douer 2013(a)				
	Substudy 1: quality item score	.	.	.	.
	Substudy 2: other scores	.	.	.	.

Continue

Continued

Instrument Version	Author	validity			
		Content validity	Structural validity	Hypothesis testing	Responsiveness
	Fliess-Douer 2013(b)	poor/indeterminate	.	poor/indeterminate	.
Level of evidence		unknown	na	unknown	na
TOMP					
	Gans 1988	.	.	.	.
Level of evidence		na	na	na	na
HMAT					
	Harvey 1998	.	.	.	.
Level of evidence		na	na	na	na
FIM + 5-AML					
	Middleton 2006	.	fair/positive	fair/positive	fair/positive
Level of evidence		na	limited positive	limited positive	limited positive
VFM					
	Taricco 2000	.	fair/indeterminate	poor/indeterminate	fair/indeterminate
Level of evidence		na	unknown	unknown	unknown

WST = wheelchair skills test, WPT = wheelchair propulsions test, WC = wheelchair circuit, AM-WC = adapted manual wheelchair circuit, WAIMS = wheelchair assessment instrument for people with multiple sclerosis, TOWM = test of wheeled mobility, TOMP = tufts assessment of motor performance, HMAT = Harvey mobility assessment tool, FIM = functional independence measure, 5-AML= five additional mobility and locomotor items, VFM = Valutazione Funzionale Mielolesi, na = no information available.

Harvey Mobility Assessment Tool (HMAT)

Harvey et al.[41] developed an outcome measure that could quantify the mobility of wheelchair dependent paraplegics. Three out of the six items assessed WMS. Information on measurement properties were only reported on inter rater reliability. The methodological quality of reliability was rated as good with a moderate level of positive evidence.

Functional Independence Measure (FIM) + 5 Additional Mobility and Locomotor items (5-AML)

Middleton et al.[44] developed an additional three WMS items and two other wheelchair items to improve the sensitivity of the FIM for people with a SCI. Although they do not show any evidence on content validity or test-retest reliability they are the only study who demonstrated limited levels of positive evidence for internal consistency, structural validity, hypothesis testing and responsiveness.

Valutazione Funzionale Mielolesi (VFM)

The VFM was developed in Italy by Tarrico et al.[48] and consists of 65 items of which nine items assess WMS. This study used one of the largest sample sizes of all studies in this review and demonstrated good methodological quality for internal consistency. No other aspects of reliability were mentioned. This study did not report on content validity. The other properties of validity were of unknown level of evidence due to poor methodological for hypothesis testing and fair methodological quality for structural validity without explaining the variance.

Conclusion phase 1: Item identification of WMS

There is no widely used WMS outcome measure with levels of evidence across all measurement properties e.g. validity, reliability and responsiveness. However, the WST[21, 35], WPT[49], WC[31-34] and 5AML[44] already showed some level of evidence on aspects of reliability and validity. The individual WMS items of these four outcome measures seem to be the best WMS items available from literature for validation in children. The WST, WPT, WC and 5AML were combined into an overall list of 22 unique WMS items, excluding items not related to mobility as defined by the ICF d465 [17].The first column in Table 5 shows the compiled list of WMS items and the original outcome measures they were selected from.

Results Phase 2: Item selection of WMS for children

Individual interviews took 30-60 minutes and were conducted with three girls, eight boys and their parents. The children's age ranged from six to thirteen years old. The group consisted of two children with cerebral palsy, seven children with Spina Bifida, one child with congenital sodium diarrhea and one child with congenital myeasthenic syndrome.

Parents and children gave descriptions of different community activities in daily life in which the WMS of the child were inadequate or where they would like to improve on. Framework data analysis resulted in WMS which were literally part of the compiled list of potentially relevant *items as can be seen in the fourth column of Table 5. F*or example, children would like to improve in their ability to go over a steep ramp or to go up and down a high or low curb. In addition, there were new codes developed for the coding framework to categorize recurring themes which could not be attributed to a single WMS item. The subsequent four columns in Table 5 show these categories: 'crossing the road', 'maneuver in crowded places' or 'small rooms' and 'propel over uneven surfaces' and their match to existing WMS items or if not available a new WMS item.

The focus group conducted with health care professionals consisted of five occupational therapists and five physiotherapists, with an average age of 34.4 (SD = 7.8) years and 8.0 (SD = 4.8) years of experience in working with children in a wheelchair. All 22 potentially relevant items were assessed in the focus group. Most items were considered relevant for children, however six items were deemed not appropriate for a WMS outcome measure for children: 'ascending or descending stairs', 'propelling in a wheelie', 'turn 180° in wheelie position left and right' and 'get over a pothole'. Total time of administration was considered important due to the extra instruction time and shorter attention span of children when administering an outcome measure. When considering these time restraints health care professionals suggested that while 'holding a wheelie' is a useful skill, it is already part of 'ascending a platform' and therefore not needed to be tested separately. The item 'avoids moving obstacles' was suggested to be adapted into an item measuring the ability to perform a 'sudden stop' as this was seen to be more relevant for children.

Conclusion phase 2: Item selection of WMS for children

The WMS items which were deemed relevant by both the children or their parents and by the health care professionals were selected for further pilot testing in phase 3. The item 'avoiding moving obstacles' was adapted into 'sudden stop'. Even though holding a wheelie was seen as not relevant by health care professionals, it was retained as a separate item as this WMS was regarded as highly relevant by children and their parents. This resulted in a 16 item WMS outcome measure, from here on called the UP-WMST.

Results Phase 3: Pilot testing of WMS items

One physiotherapist (30 years old, 4.5 years of experience) and one occupational therapist (28 years old, 5 years of experience) jointly administered the UP-WMST in eight children. All items were scored with an ability score (pass/fail) and a performance time score. The children's age ranged from 5 to 11 years old, with five children diagnosed with Cerebral Palsy and three with other disabilities. The two health care professionals commented on the ease of administering the UP-WMST in an hour. For most items both health care professionals confirmed that the items had clear instructions and were easy to administer. However, the following items were less easy to administer. The dependability of weather conditions and the extra time burden of testing in- and outdoors made the outdoor items for rolling over soft surface 'propel over grass' and 'propel over gravel' too difficult to administer. The indoor item for rolling over soft surface 'propel over a mat' was retained. The material for the item 'side slope' was seen as too big and difficult to handle when setting up the test. Table 6 shows the remaining UP-WMST items after excluding the outdoor and side slope items. Therapists also suggested future changes in the scoring method of the items with a height difference. When a child passes the ability score, the quality of execution could be a more important indicator of the performance than the time it takes to complete the item.

Table 6. Selected items of the Utrecht Pediatric Wheelchair Mobility Skills Test after phase 3.

Utrecht Pediatric Wheelchair Mobility Skills Test		**Outcome parameter**	
		Ability	**Time**
1. Propulsion forward[a,b,c,d]	10 meter	Yes/No	Seconds
2. Propulsion backwards[a]	5 meter	Yes/No	Seconds
3. Rolls on soft surface (mat) [a,c]	2 meter	Yes/No	Seconds
4. Turns 90° while moving forward[a]	Left	Yes/No	Seconds
	Right	Yes/No	Seconds
5. Turns 90° while moving backward[a]	Left	Yes/No	Seconds
	Right	Yes/No	Seconds
6. Turns 180º in place[a]	Left	Yes/No	Seconds
	Right	Yes/No	Seconds
7. Sudden stop			Seconds
8. Opening/Closing a door[a,c]	Open toward	Yes/No	Seconds
	Open away	Yes/No	Seconds
9. Figure-of-8-shape[c]		Yes/No	Seconds
10. Holding a Wheelie[a,c]	30 seconds	Yes/No	Seconds
11. Slope ascent[a,c,d]	20%	Yes/No	Seconds
12. Slope descent[a,d]	20%	Yes/No	Seconds
13. Platform ascending[a,c,d]	5,10 centimeter	Yes/No	Seconds
14. Platform descending[a]	5,10 centimeter	Yes/No	Seconds
15. Doorstep[a,c]	2 centimeter	Yes/No	Seconds

[a.] Wheelchair Skills Test, [b.] Wheelchair Propulsion Test, [c.] Wheelchair Circuit, [d.] 5 Additional Mobility and Locomotor items

DISCUSSION

The objective of this article was to develop a WMS outcome measure for children. The results of the literature review in phase 1 are in accordance with previous systematic reviews[12, 14, 15] and show the wide range of available outcome measures used for assessing WMS. Only the TMT[45] was developed for children, but due to the small sample size (n= 11) this instrument was excluded for data synthesis and analysis. There are two WMS items in the TMT 'propelling down the hall' and 'propelling up a ramp'. These two items are part of WMS outcome measures for adults and were therefore assessed on relevance in phase 2. No other WMS outcome measure has been developed or validated for children using a manual wheelchair. Furthermore none of the identified outcome measures showed good levels of evidence across all measurement properties. For example, most outcome measures showed a low level of evidence on content validity. Content validity is defined by COSMIN as 'the degree to which the content of a measurement instrument is an adequate reflection of the construct to be measured' [22]. Without good content validity, it is impossible to select the best outcome measure for a specific goal[53]. The construct the UP-WMST aims to assess skills of 'wheelchair mobility' as defined by the ICF d465 [17]. Phase 1 of this study shows that most existing WMS outcome measures do not assess 'wheelchair mobility' as defined by the ICF d465, but rather related concepts such as wheelchair user function or manual wheelchair use. While also important, these are different constructs and therefore only sections of these outcome measure were included that were relevant for assessing WMS.

In addition to assessing content validity and reliability, it is important to assess whether an outcome measure is responsive to detect change over time. The results of phase 1 showed limited levels of evidence on responsiveness available for only one outcome measure[32]. However, while there is no evidence regarding the responsiveness of the WST, the WST has been used in randomized controlled trials and seems responsive to measure change[6-9, 54]. Based on all the available psychometric data assessed, there was not a single WMS outcome measure suitable for validation in children. Therefore the second best option was to select outcome measures with some level of evidence on reliability and validity. The WST[21, 35], WPT[49], WC[31-34] and 5AML[44] already showed some level of evidence on an aspect of reliability and validity and both the WC and WST proved to be responsive to measure change in randomized controlled trials. The WMS items of these four outcome measures are the best available WMS items in current literature and were used or adapted for validation in children.

While there was little evidence available on the content validity of the identified WMS items, the results of phase 2 in this study show that most of the items on WMS were deemed as relevant by parents, children and health care professionals. These results can also be corroborated with a recent Delphi Survey[16], which reported on similar relevant items for a new WMS test for adults with an acute SCI. The only WMS items considered not relevant for children, were the more advanced WMS skills, such as descending or ascending stairs. Parents, children and health care professionals advised to include basic maneuvering tasks and include several height difference items for the outcome measure to be applicable for children. In contrast to adult manual wheelchair users, children are still developing as they grow older and different WMS will become more or less relevant. Furthermore, the size and weight of wheelchairs for young children are relatively large for their own size and strength, which makes it more difficult to execute WMS with height differences. When compared to WMS outcome measures for adults, it is important to include a higher proportion of more basic WMS in an outcome measure for children. The TMT[45] which was the only outcome measure developed for children contained two basic WMS items. These were 'propelling down the hall' and 'propelling up a ramp', and four other skills such as donning clothes or transfers. Similar adaptations were made in the WST 3.2 by Sawatsky et al.[10] for a pilot of WMS training in six children. They decreased the level of difficulty of the WST 3.2 by lowering the level change and incline for application in children. Even though outcome measures for wheelchair mobility had not been validated before in children, this study shows that most items related to basic wheelchair mobility with acceptable level of evidence in adults were considered relevant for assessment in children. Therefore the UP-WMST can be seen as an adapted version of adult outcome measures, specifically aimed at assessing basic wheelchair mobility, excluding more advanced items and items assessing different domains of the ICF [17]. However, as was mentioned by Sawatsky et al.[10] and confirmed by parents, children and health care professionals in this study, there could be a group of children with a high level of WMS and a basic WMS outcome measure might show a ceiling effect for these children. More advanced WMS are already assessed in WMS outcome measures for adults, but these outcome measures have not yet been validated for children. For example, TOWM and Wheelie test[40] consist of more advanced items with high platforms (20 cm) and several WMS skills in wheelie position. Future research should be aimed at developing and validating a similar advanced WMS outcome measure for children.

In addition to selecting relevant WMS items for children, it is also important to evaluate the applicability of the outcome measure in clinical practice. As suggested by Kirby [55] there are more assessment criteria which are useful to assess when selecting an outcome measure, such as time burden, availability of materials and ease of administering the test. Therefore, we examined the feasibility of administering the UP-WMST in phase 3 of this study. While the outdoor items were previously seen as relevant in phase 2, they were excluded from the UP-WMST after the results of phase 3 due to time burden of testing both in- and outdoors. When developing a more advanced test for wheelchair mobility in children, these outdoor items should be reconsidered for inclusion. Results of phase 3 also showed the need for an additional outcome parameter for the height difference items. All items are currently assessed on performance time and ability. The combination of these two outcome parameters seem to be in line with recent findings by Sawatzky et al.[56] that propulsion speed and ability are related. However, according to the results of phase 3, a more extensive scoring method should be included for the height difference items. Such a method could include a five point scoring method as used in the TOWM and Wheelie test[40], a performance score as used in the WC[32], or a safety score as described in the WST[36]. We are currently continuing with the validation of the UP-WMST and development of a qualitative scoring method which is able to distinguish between beginner or more advanced execution methods on an item.

This study was limited to WMS items with good measurement properties available in current literature. Surprisingly there was not one WMS outcome measure available with good levels of evidence across all measurement properties. The second best option was to select the best available WMS outcome measures for adults with some level of evidence across reliability and validity. The levels of evidence of these selected WMS items for responsiveness, minimal detectable change and minimal important change remain unknown. The feasibility of the UP-WMST was assessed by 2 health care professionals from the same rehabilitation center. It would be interesting to assess if the administration of the UP-WMST in a different setting or with different health care professionals would lead to the same results. Before the UP-WMST can be used in clinical practice, additional research towards responsiveness, interpretability, reliability and construct validity of the newly developed UP-WMST is warranted. Furthermore, the necessity of including basic WMS items could have been enhanced by the sampling of children and their parents used in this study for relevance checking. Children were recruited from a voluntary wheelchair mobility skills training program and interviewed a few weeks before the start of the program. Therefore, their level of wheelchair mobility could have been lower and this could have resulted in some bias towards more basic WMS. At the same time this is the group of children who attend a wheelchair skill training program and therefore the group of children the UP-WMST is developed for. Nevertheless, interviews only took place before the start of the wheelchair skill training program and children and parents might have underestimated the possible WMS a child is able to learn. Therefore, future research should evaluate possible ceiling effects of the UP-WMST.

CONCLUSION

No single WMS outcome measure with good levels of evidence across all measurement properties was available for validation in children. However, four outcome measures did show levels of evidence on reliability and validity. The individual WMS items of these four outcome measure is the best knowledge available from literature and were used for relevance checking and validation in children. Parents, children using a manual wheelchair and health care professionals agreed on the necessity of including more basic WMS in an outcome measure for children compared to adults. The resulting 15 item UP-WMST outcome measure is easy to administer and demonstrates content validity for assessing WMS in children using a manual wheelchair. While this is the first step towards developing a WMS outcome measure for children, further assessment of reliability, construct validity and responsiveness is needed.

ACKNOWLEDGEMENTS

This study is part of the Fit For the Future collaboration; H. Wittink, A. Dallmeijer, R van den Berg. We would like to thank all participating children, their parents and health care professionals for sharing their experiences with us. We also thank the physiotherapists, occupational therapists and physical education teachers from Arianne de Ranitz and mytylschool De Trappenberg for their help in the pilot phase and organization of the focus group interviews. We would like to acknowledge the BOSK for their help in distributing the information flyers and recruitment of children for participation in this study. We especially like to thank KJ-Projects for sharing their knowledge on WMS training in children. Furthermore we would like to thank all the participating students who have enthusiastically contributed to the data collection and analysis in this study.

APPENDIX

Search filter PUBMED

Search (((wheelchair AND rehabilitation AND mobility)) OR (wheelchair AND measurement AND assessment)) OR ((Wheelchair AND Mobility AND Skill) OR (Wheelchair AND Mobility AND Task) OR (Wheelchair AND Mobility AND Measurement) OR (Wheelchair AND Mobility AND Test) OR (Wheelchair AND Mobility AND ADL) OR (Wheelchair AND Mobility AND Functional) OR (Wheelchair AND Mobility AND Instrument) OR (Wheelchair AND Mobility AND Performance) OR (Wheelchair AND Mobility AND SCI) OR (Wheelchair AND Mobility AND Validity) OR (Wheelchair AND Mobility AND Reliability) OR (Wheelchair AND Mobility AND Pathology) OR (Wheelchair AND Mobility AND Behavior) OR (Wheelchair AND Mobility AND Activity) OR (Wheelchair AND Mobility AND Disability) OR (Wheelchair AND Mobility AND Assessment) OR (Wheelchair AND Mobility AND Quality of life)) Filters: Publication date from 2010/01/01

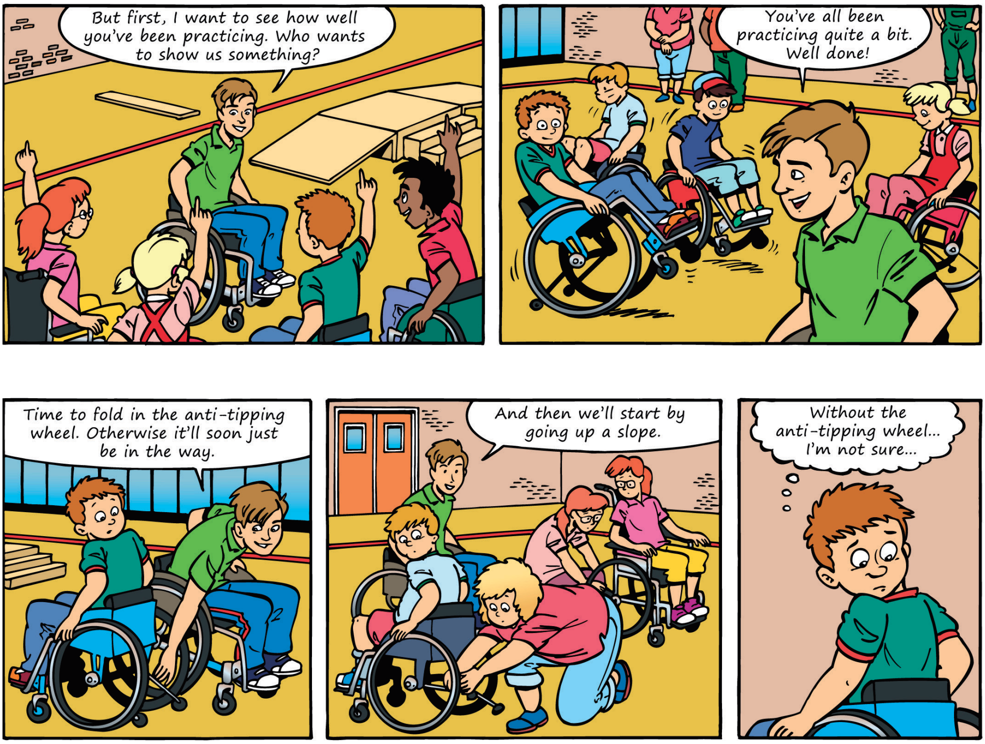
REFERENCES

1. Odding E, Roebroeck ME, Stam HJ: The epidemiology of cerebral palsy: incidence, impairments and risk factors. Disabil Rehabil 2006, 28(4):183-191.
2. Busby A, Abramsky L, Dolk H, Armstrong B, Addor MC, Anneren G, Armstrong N, Baguette A, Barisic I, Berghold A, Bianca S, Braz P, Calzolari E, Christiansen M, Cocchi G, Daltveit AK, De Walle H, Edwards G, Gatt M, Gener B, Gillerot Y, Gjergja R, Goujard J, Haeusler M, Latos-Bielenska A, McDonnell R, Neville A, Olars B, Portillo I, Ritvanen A, Robert-Gnansia E, Rosch C, Scarano G, Steinbicker V: Preventing neural tube defects in Europe: a missed opportunity. Reprod Toxicol 2005, 20(3):393-402.
3. Rodby-Bousquet E., Hagglund G.: Use of manual and powered wheelchair in children with cerebral palsy: A cross-sectional study. BMC Pediatr 2010, 10.
4. Kilkens OJ, Dallmeijer AJ, Nene AV, Post MW, van der Woude LH: The Longitudinal Relation Between Physical Capacity and Wheelchair Skill Performance During Inpatient Rehabilitation of People With Spinal Cord Injury. Arch Phys Med Rehabil 2005, 86(8):1575-1581.
5. Mortenson W, B., Miller W, C., Backman C, L., Oliffe J, L.: Association Between Mobility, Participation, and Wheelchair-Related Factors in Long-Term Care Residents Who Use Wheelchairs as Their Primary Means of Mobility. J Am Geriatr Soc 2012, 60(7):1310-1315
6. Routhier F, Kirby RL, Demers L, Depa M, Thompson K: Efficacy and Retention of the French-Canadian Version of the Wheelchair Skills Training Program for Manual Wheelchair Users: A Randomized Controlled Trial. Arch Phys Med Rehabil 2012, 93(6):940-948.
7. Ozturk A., Ucsular F.D.: Effectiveness of a wheelchair skills training programme for community-living users of manual wheelchairs in Turkey: a randomized controlled trial. Clin Rehabil 2011, 25(5):416-424.
8. Best KL, Kirby RL, Smith C, MacLeod DA: Wheelchair Skills Training for Community-Based Manual Wheelchair Users: A Randomized Controlled Trial. Arch Phys Med Rehabil 2005, 86(12):2316-2323.
9. MacPhee AH, Kirby RL, Coolen AL, Smith C, MacLeod DA, Dupuis DJ: Wheelchair skills training program: a randomized clinical trial of wheelchair users undergoing initial rehabilitation1. Arch Phys Med Rehabil 2004, 85(1):41-50.
10. Sawatzky B, Rushton PW, Denison I, McDonald R: Wheelchair skills training programme for children: a pilot study. Aust Occup Ther J 2012, 59(1):2-9.
11. 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, 15:11.
12. Fliess-Douer O, Vanlandewijck YC, Lubel Manor G, Van Der Woude LHV: A systematic review of wheelchair skills tests for manual wheelchair users with a spinal cord injury: towards a standardized outcome measure. Clin Rehabil 2010, 24(10):867-886.
13. de Vet HC, Terwee CB, Mokkink LB, Knol DL: *Measurement in Medicine:* New York: Cambridge University Press; 2011.
14. Mortenson WB, Miller WC, Auger C: Issues for the Selection of Wheelchair-Specific Activity and Participation Outcome Measures: A Review. Arch Phys Med Rehabil 2008, 89(6):1177-1186.

15. Kilkens OJ, Post MW, Dallmeijer AJ, Seelen HA, van der Woude LH: Wheelchair skills tests: a systematic review. Clin Rehabil 2003, 17(4):418-430.
16. Gollan EJ, Harvey LA, Simmons J, Adams R, McPhail SM: Development, reliability and validity of the queensland evaluation of wheelchair skills (QEWS). Spinal Cord 2015, 53(10):743-749.
17. [http://apps.who.int/classifications/icfbrowser/]
18. Mokkink LB, Terwee CB, Knol DL, Stratford PW, Alonso J, Patrick DL, Bouter LM, de Vet HC: The COSMIN checklist for evaluating the methodological quality of studies on measurement properties: a clarification of its content. BMC Med Res Methodol 2010, 10:22.
19. Terwee CB, Mokkink LB, Knol DL, Ostelo RW, Bouter LM, de Vet HC: Rating the methodological quality in systematic reviews of studies on measurement properties: a scoring system for the COSMIN checklist. Qual Life Res 2012, 21(4):651-657.
20. Bartels B, de Groot JF, Terwee CB: The six-minute walk test in chronic pediatric conditions: a systematic review of measurement properties. Phys Ther 2013, 93(4):529-541.
21. Kirby RL, Dupuis DJ, Macphee AH, Coolen AL, Smith C, Best KL, Newton AM, Mountain AD, Macleod DA, Bonaparte JP: The wheelchair skills test (version 2.4): measurement properties. Arch Phys Med Rehabil 2004, 85(5):794-804.
22. Terwee CB, Bot SD, de Boer MR, van der Windt DA, Knol DL, Dekker J, Bouter LM, de Vet HC: Quality criteria were proposed for measurement properties of health status questionnaires. J Clin Epidemiol 2007, 60(1):34-42.
23. van Tulder M, Furlan A, Bombardier C, Bouter L, Editorial Board of the Cochrane Collaboration Back Review Group: Updated method guidelines for systematic reviews in the cochrane collaboration back review group. Spine (Phila Pa 1976) 2003, 28(12):1290-1299.
24. Gale NK, Heath G, Cameron E, Rashid S, Redwood S: Using the framework method for the analysis of qualitative data in multi-disciplinary health research. BMC Med Res Methodol 2013, 13:117.
25. Walker K.A., Morgan K.A., Morris C.L., DeGroot K.K., Hollingsworth H.H., Gray D.B.: Development of a community mobility skills course for people who use mobility devices. Am J Occup Ther 2010, 64(4):547-554.
26. Koontz AM, Lin Y, Kankipati P, Boninger ML, Cooper RA: Development of custom measurement system for biomechanical evaluation of independent wheelchair transfers. J Rehabil Res Dev 2011, 48(8):1015-1028.
27. Smith C, Kirby RL: Manual Wheelchair Skills Capacity and Safety of Residents of a Long-Term-Care Facility. Arch Phys Med Rehabil 2011, 92(4):663-669.
28. Lamberg A.S., Friden J.: Changes in skills required for using a manual wheelchair after reconstructive hand surgery in tetraplegia. J Rehabil Med 2011, 43(8):714-719.
29. Tsai CY, Rice LA, Hoelmer C, Boninger ML, Koontz AM: Basic psychometric properties of the transfer assessment instrument (version 3.0). Arch Phys Med Rehabil 2013, 94(12):2456-2464.
30. Mills TL, Holm MB, Schmeler M: Test-retest reliability and cross validation of the Functioning Everyday With a Wheelchair instrument. Assist Technol 2007, 19(2):61-77.

31. Kilkens OJ, Post MW, van der Woude LH, Dallmeijer AJ, van den Heuvel WJ: The wheelchair circuit: Reliability of a test to assess mobility in persons with spinal cord injuries. Arch Phys Med Rehabil 2002, 83(12):1783-1788.
32. Kilkens OJ, Dallmeijer AJ, de Witte LP, van der Woude LH, Post MW: The wheelchair circuit: construct validity and responsiveness of a test to assess manual wheelchair mobility in persons with spinal cord injury. Arch Phys Med Rehabil 2004, 85(3):424-431.
33. Cowan RE, Nash MS, de Groot S, van der Woude LH: Adapted Manual Wheelchair Circuit: Test-Retest Reliability and Discriminative Validity in Persons With Spinal Cord Injury. Archives of Physical Medicine and Rehabilitation 2011, 92(8):1270-1280.
34. Vereecken M, Vanderstraeten G, Ilsbroukx S: From "wheelchair circuit" to "wheelchair assessment instrument for people with multiple sclerosis": reliability and validity analysis of a test to assess driving skills in manual wheelchair users with multiple sclerosis. Arch Phys Med Rehabil 2012, 93(6):1052-1058.
35. Kirby RL, Swuste J, Dupuis DJ, MacLeod DA, Monroe R: The Wheelchair Skills Test: a pilot study of a new outcome measure. Arch Phys Med Rehabil 2002, 83(1):10-18.
36. Lindquist NJ, Loudon PE, Magis TF, Rispin JE, Kirby RL, Manns PJ: Reliability of the Performance and Safety Scores of the Wheelchair Skills Test Version 4.1 for Manual Wheelchair Users. Arch Phys Med Rehabil 2010, 91(11):1752-1757.
37. [http://www.wheelchairskillsprogram.ca/eng/publications.php]
38. Fliess-Douer O, Van DW, Vanlandewijck YC: Test of Wheeled Mobility (TOWM) and a short wheelie test: a feasibility and validity study. Clin Rehabil 2013, 27(6):527-537.
39. Fliess-Douer O., Vanlandewijck YC, Van Der WL: Most essential wheeled mobility skills for daily life: An international survey among paralympic wheelchair athletes with spinal cord injury. Arch Phys Med Rehabil 2012, 93(4):629-635.
40. Fliess-Douer O, Van Der Woude LH, Vanlandewijck YC: Reliability of the test of wheeled mobility (TOWM) and the short Wheelie test. Arch Phys Med Rehabil 2013, 94(4):761-770.
41. Harvey LA, Batty J, Fahey A: Reliability of a tool for assessing mobility in wheelchair-dependent paraplegics. Spinal Cord 1998, 36(6):427.
42. Routhier F, Vincent C, Desrosiers J, Nadeau S, Guerette C: Development of an obstacle course assessment of wheelchair user performance (OCAWUP): a content validity study. TECHNOL DISABIL 2004, 16(1):19-31.
43. Routhier F, Desrosiers J, Vincent C, Nadeau S: Reliability and construct validity studies of an obstacle course assessment of wheelchair user performance. Int J Rehabil Res 2005, 28(1):49-56.
44. Middleton JW, Harvey LA, Batty J, Cameron I, Quirk R, Winstanley J: Five additional mobility and locomotor items to improve responsiveness of the FIM in wheelchair-dependent individuals with spinal cord injury. Spinal Cord 2006, 44(8):495-504.
45. Chafetz R, McDonald C, Mulcahey MJ, Betz R, Anderson C, Vogel L, Gaughan JP, Martin S, O'Dell MA, Flanagan A: Timed motor test for wheelchair users: initial development and application in children with spinal cord injury. J Spinal Cord Med 2004, 27 Suppl 1:S38-43.

46. Stanley RK, Stafford DJ, Rasch E, Rodgers MM: Development of a functional assessment measure for manual wheelchair users. J Rehabil Res Dev 2003, 40(4):301-307.
47. Cress ME, Kinne S, Patrick DL, Maher E: Physical functional performance in persons using a manual wheelchair. J Orthop Sports Phys Ther 2002, 32(3):104-113.
48. Taricco M, Apolone G, Colombo C, Filardo G, Telaro E, Liberati A: Functional status in patients with spinal cord injury: A new standardized measurement scale. Arch Phys Med Rehabil 2000, 81(9):1173-1180.
49. Askari S., Kirby R.L., Parker K., Thompson K., O'Neill J.: Wheelchair propulsion test: Development and measurement properties of a new test for manual wheelchair users. Arch Phys Med Rehabil 2013, 94(9):1690-1698.
50. Gagnon D, Decary S, Charbonneau MF: The timed manual wheelchair slalom test: a reliable and accurate performance-based outcome measure for individuals with spinal cord injury. Arch Phys Med Rehabil 2011, 92(8):1339-1343.
51. Gans B, M.: Description and interobserver reliability of the Tufts. American Journal of Physical Medicine & Rehabilitation 1988, 67(5):202-210.
52. Jebsen RH, Trieschmann RB, Mikulic MA, Hartley RB, McMillan JA, Snook ME: Measurement of time in a standardized test of patient mobility. Arch Phys Med Rehabil 1970, 51(3):170-175.
53. Mokkink LB, Prinsen CA, Bouter LM, Vet HC, Terwee CB: The COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) and how to select an outcome measurement instrument. Braz J Phys Ther 2016, .
54. Best KL, Miller WC, Huston G, Routhier F, Eng JJ: Pilot Study of a Peer-Led Wheelchair Training Program to Improve Self-Efficacy Using a Manual Wheelchair: A Randomized Controlled Trial. Arch Phys Med Rehabil 2016, 97(1):37-44.
55. Kirby RL: Survey of wheelchair skills tests: making the case for broadening the search and assessment criteria. Clin Rehabil 2011, 25(3):287.
56. Sawatzky B, Hers N, MacGillivray MK: Relationships between wheeling parameters and wheelchair skills in adults and children with SCI. Spinal Cord 2015, 53(7):561-564.


But first, I want to see how well you've been practicing. Who wants to show us something?
You've all been practicing quite a bit. Well done!
Time to fold in the anti-tipping wheel. Otherwise it'll soon just be in the way.
And then we'll start by going up a slope.
Without the anti-tipping wheel... I'm not sure...

Chapter 3

Utrecht pediatric wheelchair mobility skills test: reliability, validity, and responsiveness in youth using a manual wheelchair

Sol M[1,2], de Groot JF[1,3], Zwinkels M[2], Visser-Meily JMA[2], Kruitwagen C[4], Verschuren O[2]

1. Research Group Lifestyle and Health, HU University of Applied Sciences Utrecht, Utrecht, the Netherlands
2. Brain Center Rudolf Magnus and Center of Excellence for Rehabilitation Medicine, University Medical Center Utrecht and De Hoogstraat Rehabilitation, Utrecht, University Utrecht, The Netherlands
3. Netherlands Institute for Healthcare Services Research (NIVEL), Utrecht, the Netherlands
4. Department of Biostatistics and Research Support, Julius Center, University Medical Center Utrecht, Utrecht, The Netherlands

Phys Ther 2019; 99(8):1098–106

ABSTRACT

Background: The assessment of wheelchair mobility skills (WMS) is important in youth using a manual wheelchair. More information is needed regarding the psychometric properties of the newly developed Utrecht Pediatric Wheelchair Mobility Skills Test (UP-WMST).

Objective: The purpose of this study was to evaluate the reliability, content validity, construct validity and responsiveness of the UP-WMST 2.0 in youth using a manual wheelchair.

Design: Repeated measurements and cross-sectional study design.

Methods: A total of 117 children and adolescents who use a manual wheelchair participated in this study. The UP-WMST 2.0 contains the same 15 WMS items as the original UP-WMST with an adaptation of the scoring method. Test-retest reliability was estimated in 30 participants. Content validity was assessed through floor and ceiling effect analyses. Construct validity was assessed through hypothesis testing. Initial responsiveness was assessed in 23 participants who participated in a WMS training program.

Results: Test-retest reliability analysis showed weighted Cohen's kappa values ranging from 0.63-0.98, for all but one (sub) item. The total UP-WMST 2.0 score had an intraclass correlation coefficient of 0.97. No floor or ceiling effects were detected. Independent sample T-Test analysis confirmed our hypotheses regarding direction and difference in scores between age and diagnostic groups. Within group analysis in the responsiveness study showed a positive significant change in UP-WMST 2.0 score (8.3 points).

Limitations: The small sample size used in the responsiveness study.

Conclusion: This study shows evidence towards test-retest reliability, content and construct validity of the UP-WMST 2.0. It also shows initial evidence towards responsiveness of the UP-WMST 2.0 to measure change in WMS in youth using a manual wheelchair.

Keywords: Psychometric properties; wheelchair mobility; skills; youth; wheelchair user; reliability; validity

INTRODUCTION

People who use a manual wheelchair in daily life rely on their wheelchair and mobility skills to move around and overcome physical barriers, such as uneven surfaces, curbs and slopes. More advanced wheelchair mobility skills (WMS) are associated with a higher quality of life, higher life satisfaction and more community participation[1,2].

Although WMS training can improve the skill level of adult wheelchair users and is an important part of clinical rehabilitation programs[3-7], only scarce evidence exists for the impact of WMS training in youth (children and adolescents). Two recent studies have reported on the importance of WMS training for youth using a manual wheelchair[8,9]. In order to evaluate the effect of WMS training in youth, there is a need for a WMS outcome measure specifically developed for this population. Existing WMS outcome measures have been developed for adult wheelchair users, but none of these outcome measures have been validated in youth who use a manual wheelchair. It is important to validate an outcomes measure again if it is applied in a new population, because certain items could be irrelevant or need adaptation10. Therefore, we recently developed a new WMS outcomes measure, the Utrecht Pediatric Wheelchair Mobility Skills Test (UP-WMST)[11]. Earlier research on the development of the UP-WMST focused on content validity, through selection of WMS items from adult outcome measures and subsequently identifying relevant WMS items for youth using the clinical expertise of youth health care professionals, youth and their parents. Afterwards the feasibility of administering the UP-WMST in youth using a manual wheelchair was assessed in a pilot study[11]. Outcomes of this previous study resulted in a list of 15 items to assess skills related to wheelchair mobility as defined by the International Classification of human Functioning d465 (ICF)[12] in youth. To further develop and support clinical use of the UP-WMST, more information about its psychometric properties is needed.

Reliability refers to the extent to which scores of participants who have not changed are the same for repeated measurements over time (test-retest reliability) and to the degree to which the UP-WMST is free from measurement error[13]. Content validity refers to the degree to which the content of the UP-WMST is an adequate reflection of WMS in youth[13]. The previous study[11] focused on content validity through the development of a comprehensive list of relevant items to assess WMS in youth. A final step in assessing content validity is to assess if the UP-WMST includes a range of items which are comprehensive enough to demonstrate change, i.e. without floor or ceiling effects[14]. Construct validity refers to the degree to which the scores of the UP-WMST represent WMS in youth and is assessed through hypothesis testing[13]. We hypothesize that age is positively associated with UP-WMST score due to more experience in wheelchair mobility and/or increased strength or neuromotor development in older wheelchair users[15,16]. Furthermore we expect a difference between diagnostic groups. In adult manual wheelchair users the success rate of WMS differs per patient group, with people with a spinal cord injury having a higher success rate than people with a stroke or acquired brain disorder[17]. In children who use a manual wheelchair, agility and anaerobic performance tests show better results in children with Spina Bifida (SB) compared to children with cerebral palsy (CP)[18,19]. We therefore hypothesize that youth with SB will have a higher score on the UP-WMST compared to youth with CP. Responsiveness refers to the ability of the UP-WMST to detect change over time[13]. A WMS outcome measure in adults has shown to be responsive after WMS training[6]. In youth there is only data available from one small pilot study (n=6), that reported on improvements in WMS after a two day WMS training program[9]. To determine if the UP-WMST is able to detect change over time, we hypothesize to find a significant improvement in the outcome after a six month WMS training program.

The purpose of this study is to evaluate the reliability, content validity, construct validity and responsiveness of the UP-WMST in youth using a manual wheelchair.

METHODS

Sample

This study is part of a currently ongoing larger Let's Ride study (n=117)[20]. The database consists of youth who participated in a reliability study (n = 30) and a WMS training program study (n = 97) (trialregister.nl, registration number NTR5791). The WMS training program study consisted of a pilot study (n=23), from which the data was used to obtain initial estimates of responsiveness and a larger intervention study (n=74), from which the pre-intervention measurements are used in this study. When youth participated in more than one study (n=10), only the first measurement of the first enrolled study was used for analysis of content and construct validity.

All participants were household or community bimanual wheelchair users in daily life. Children (5-11 years) and adolescents (12-18 years) were included if they were able to follow simple instructions. Informed consent was obtained from all parents and all adolescents. To ascertain the sample was a good reflection of the total manual wheelchair youth population, we included participants with different diagnoses and youth with varying cognitive levels who received regular education or special education.

UP-WMST and UP-WMST 2.0

Table 1 shows the different WMS assessed in this measurement tool, as well as the cut-off times that we used to modify the scoring method to create a modified version, the

Table 1 Distribution of time scores per item and determination of cut-off times.

UP-WMST item list		N Completed item	Mean	SD	25%	75%	Cut-off time 0	1	2	3
1. Propulsion forwards (sec)		117	6.92	3.04	5.14	7.45	NC	>7.5	5 -7.5	<5
2. Propulsion backwards (sec)		113	8.23	5.23	5.37	9.15	NC	>9	5.5-9	<5.5
3. Rolls on soft surface (sec)		115	2.98	1.57	2.11	3.31	NC	>3.5	2-3.5	<2
4. Turns 90° while moving forward (sec)		117	5,07	2.22	3.85	5.49	NC	>5.5	4-5.5	<4
5. Turns 90° while moving backward (sec)		115	9,35	7.61	5.34	10.02	NC	>10	5.5-10	<5.5
6. Turns 180° in place (sec)		114	4.00	3.20	2.34	4.56	NC	>4.5	2.5-4.5	<2.5
7. Sudden stop (sec)		115	1.57	5.14	0.78	1.34	NC	>1.5	1-1.5	<1
8. Opening/Closing a door (sec)	Away	114	12.56	7.43	6.83	17.43	NC	>17.5	7-17.5	<7
	Towards	98	18.37	12.69	10.13	22.54	NC	>22.5	10-22.5	<10
9. Figure-of-8-shape (sec)		115	11.48	6.18	8.41	12.18	NC	>12	8.5-12	<8.5
10. Holding a Wheelie (sec)		69	11.67	13.51	0.79	30.00	NC	<2	2-30	>30
11. Slope ascent (sec)		83	4.97	2.39	3.48	6.03	NC	>6	3.5-6	<3.5
12. Slope descent (sec)		117	3.12	2.92	1.88	3.12	NC	>3	2-3	<2
13. Platform ascending (sec)	5 cm	92	5.58	4.42	3.03	6.55	NC	>6.5	3-6.5	<3
	10 cm	25	4.79	3.65	2.85	4.46	NC	>4.5	3-4.5	<3
14. Platform descending (sec)	5 cm	116	2.34	1.64	1.50	2.33	NC	>2.5	1.5-2.5	<1.5
	10 cm	93	3.68	2.52	2.08	4.32	NC	>4.5	2-4.5	<2
15. Doorstep (sec)		113	3.85	5.09	2.18	3.87	NC	>4	2-4	<2

UP-WMST = Utrecht Pediatric Wheelchair Mobility Skills Test, sec = time in seconds, SD = standard deviation, NC = not completed

UP-WMST 2.0. All items, apart from item 10 "holding a wheelie", are performed at maximum speed. Item 8 "door open and away", item 13 "platform ascending" and item 14 "platform descending" contain two sub items with different degrees of difficulty and are scored per sub item.

In close collaboration with pediatric physical therapists and a biostatistician a few changes have been made to the scoring method in comparison with the original UP-WMST. To distinguish between the original version and the new version with the adapted scoring method, the new version will be called UP-WMST 2.0. Pediatric physical therapists collaborating in our research reported difficulties with timing the item "holding a wheelie" in participants who were unable to control a wheelie position. The duration of holding a wheelie was too short to time with a stopwatch. Therefore we developed a different scoring method: a score of 1 is awarded when the participant can pop up their front casters off the ground, a score of 2 is awarded if the participant is able to maintain a wheelie for more than 2 seconds and a score of 3 is awarded if the participant is able to maintain a wheelie for more than 30 seconds.

At the same time, the original time scoring method of the UP-WMST in Sol et al.[11] has been further developed into an ordinal scale for the UP-WMST 2.0. The use of an ordinal scale has several advantages over a time score per (sub)item. The ordinal ranks of the (sub) items can be summed up to a total score. A total score facilitates interpretation of test results and can be used to evaluate changes within or between individuals or groups. With support of a biostatistician, the time scores per (sub) item were converted into a 4-point ordinal scale per item. A rank of 0 is given when participants are unable to complete an item or the time to task completion takes over 60 seconds.

The UP-WMST times of the 117 participants in this study were used to determine the cut-off time per (sub) item. The times of the participants who were able to complete an item within 60 seconds were used to calculate the mean times and determine the cut-off times at the 25th and 75th percentile. Table 1 shows per (sub) item the cut-off times for the ordinal ranks. A total UP-WMST 2.0 score, with a maximum of 54 points, was calculated from the sum of the ordinal ranks per (sub) item. A higher score on the UP-WMST 2.0 indicates more advanced WMS.

Table 2. Ordinal scale of the UP-WMST 2.0

0	Not able to complete item or time to task completion > 60 seconds.
1	Above the 75th percentile
2	Between the 25th and 75th percentile
3	Below the 25th percentile

Procedures

To assess test-retest reliability (n=30), the UP-WMST 2.0 was administered twice within a two week period (median = 7 days) . Ethical approval was granted by the Medical Ethical Committee of University Medical Centre Utrecht (UMCU) in the Netherlands for the reliability part of this study (#12-586). As the UP-WMST 2.0 is a new outcome measure, no information was available for sample size calculations. We have therefore followed calculations from a similar study in adult wheelchair users of Kilkens et al.[21] A group of 27 participants was needed to detect an ICC >0.80. With a 10 % drop-out or missing data, alpha of 0.05 and power of 0.80, a sample size of 30 participants will be required. All re-tests of the UP-WMST 2.0 were conducted using the same material, gym floor, tire pressure, test administrator and instructions as the first administration of the UP-WMST 2.0. The administer explained every item verbally and when necessary showed the execution of the item by walking (e.g. figure of eight shape). Participants could use one trial attempt and subsequently a maximum of three attempts were allowed. The best time was used and converted to a rank score.

To assess content and construct validity (n=117), the descriptive statistics of the baseline UP-WMST 2.0 scores of the total sample were used. The UP-WMST 2.0 was administered according to the protocol by a physical therapist, occupational therapist or research team member. All administrators received a 4 hour training (theory and practice) on the administration of the UP-WMST 2.0. The Institutional Review Board of the UMCU approved the study protocol for Let's Ride intervention study (#15-136).

A subsample of participants (n=23) participated in the Let's Ride pilot study[22]. The results of this pilot study were used to estimate initial responsiveness. Participants followed a WMS training program aimed at propelling efficiently, going up and down curbs, holding a wheelie and negotiating the physical environment outdoors. This program was organized by a Dutch patient organization and consisted of 4 training sessions of 2.5 hours each over a six month period. An experienced manual wheelchair user with over 10 years of experience in teaching WMS conducted the training sessions. After each training session, participants were instructed and motivated to practice at home and received an individual video instruction about how to practice their skills. The research team administered the UP-WMST 2.0 before and after the training period using the same material, gym floor, tire pressure and instructions.

Statistical analysis

Reliability

The test-retest reliability of the individual (sub) items was assessed using a quadratic weighted Cohen's Kappa. In addition, the test-retest reliability of the total score was calculated using a two-way mixed intraclass correlation coefficient ($ICC_{agreement}$). A priori, a weighted Cohen's Kappa of 0.61 or higher and an $ICC_{agreement}$ of 0.80 or higher was defined as an indication of good reliability[23]. Standard error of measurement ($SEM_{agreement}$) and smallest detectable change (SDC), were calculated for the total UP-WMST 2.0 score[10].

$$SDC = 1.96 \times \sqrt{2} \times SEM_{agreement}$$

Content validity
Floor and ceiling effects of the total UP-WMST 2.0 score were determined as part of content validity. Floor and ceiling effects were considered present if 15% or more of the participants scored a lowest or highest possible score[10].

Construct validity
The construct validity of the UP-WMST 2.0 was evaluated using an independent samples t-test analysis to assess between group differences in UP-WMST 2.0 score within age groups (children and adolescents) and diagnoses (SB and CP).

Responsiveness
A paired samples t-test was used to assess the change of the UP-WMST 2.0 after a WMS training program.

RESULTS

Reliability
Table 3 shows the descriptive statistics of the participants included per study. Thirty participants were included in the test-retest reliability study. One participant became ill and could not perform the retest of the UP-WMST 2.0 within the two week period. The scores of this participant were not used in the reliability analysis. The results for the test-retest reliability per (sub) item and total score are shown in Table 4. The weighted kappa coefficients ranged from 0.63-0.98 for all but one item, an indication of good reliability based on definitions the research team developed a priori. Item 7 'sudden stop' with a coefficient of 0.50, was moderately reliable based on the definitions the research team developed a priori. Due to this moderate value of reliability, the 'sudden stop' item has been removed from the UP-WMST 2.0 and will not be part of the total score. The new maximal score on the UP-WMST 2.0 is 51 points. The $ICC_{agreement}$ for the total UP-WMST 2.0 score was 0.97 with a $SEM_{agreement}$ of 1.80 points and SDC of 4.98 points.

Table 3. Participant characteristics and UP-WMST 2.0 scores

Parameter	Reliability study	Validity study	Responsiveness (pilot) study
Number of participants, n	30	117	23
Children	12	56	21
Adolescents	18	61	2
Age in years, mean (SD)	12.5 (3.4)	12.4 (3.4)	9.3 (1.9)
Gender, n			
Male	18	71	16
Female	12	46	7
Diagnoses, n			
Spina bifida	15	31	11
Cerebral palsy	13	47	5
Muscle disorder	1	11	1
Other	1	28	6
UP-WMST 2.0, mean (SD)			
Baseline	29.4 (11.0)	29.4 (10.2)	29.2 (9.0)
Re-test (n=29)	31.3(10.4)	-	-
Post-training (n=20)	-	-	38.9 (8.8)

UP-WMST = Utrecht Pediatric Wheelchair Mobility Skills Test, SD = standard deviation

Content validity

Figure 1 shows the variation of UP-WMST 2.0 scores in 117 participant with a normal distribution and a range of 9 – 51 points. No floor or ceiling effect were present, as none of the participants had a minimum score and only 1.7% of the participants reached the maximum score. Fifteen percent of the participants reached a score of 40 points or higher on the UP-WMST 2.0.

Construct validity

Table 5 shows the mean score, standard deviation and range of the UP-WMST 2.0 score per age group and per diagnosis. The UP-WMST 2.0 scores were significantly lower, 5.4 points with $p<0.01$, in children (n=56) compared to adolescents (n=61). The UP-WMST 2.0 scores were significantly higher, 9.1 points with $p<0.001$, in youth with SB (n=31) compared to youth with CP (n=47). No confounding influence of age was detected when comparing youth with SB to youth with CP.

Table 4 UP-WMST 2.0 Descriptive statistics of items and total score

		Test					Re-test					Reliability	
		n	Median	Mean	Q1-Q3	SD	n	Median	Mean	Q1-Q3	SD	Weighted Cohen's Kappa	$ICC_{agreement}$
1. Propulsion forwards		29	2		2.00-3.00		29	2		2.00-2.00		0.80	
2. Propulsion backwards		29	2		1.00-3.00		29	2		1.50-3.00		0.63	
3. Rolls on soft surface		29	2		1.00-2.00		29	2		1.50-2.00		0.71	
4. Turns 90° while moving forward		29	2		1.50-3.00		29	2		1.00-3.00		0.78	
5. Turns 90° while moving backward		29	2		1.00-2.50		29	2		1.00-3.00		0.77	
6. Turns 180º in place		29	2		1.00-2.50		29	2		2.00-2.50		0.63	
7. Sudden stop		29	2		2.00-3.00		29	2		2.00-3.00		0.50	
8. Opening/Closing a door	Away	28	2		2.00-3.00		29	2		2.00-3.00		0.67	
	Towards	29	2		1.25-3.00		27	2		2.00-3.00		0.88	
9. Figure-of-8-shape		29	2		2.00-3.00		29	2		1.00-3.00		0.74	
10. Holding a Wheelie		28	1		0.00-2.00		27	1		1.00-3.00		0.80	
11. Slope ascent		29	1		0.00-3.00		29	2		0.00-3.00		0.89	
12. Slope descent		29	2		1.00-3.00		29	2		1.00-3.00		0.67	
13. Platform ascending	5cm	29	2		0.50-2.00		29	2		1.00-3.00		0.88	
	10cm	28	0		0.00-0.75		29	0		0.00-0.50		0.98	
14. Platform descending	5cm	29	2		1.00-2.00		29	2		1.50-2.50		0.79	
	10cm	27	2		1.00-2.00		29	2		1.00-2.50		0.74	
15. Doorstep		29	2		2.00-2.00		29	2		1.00-2.00		0.69	
Total UP-WMST 2.0 score		29		30.0		10.7	29		31.3		10.4		0.97

UP-WMST = Utrecht Pediatric Wheelchair Mobility Skills Test, SD = Standard deviation, Q1 and Q3 = first and third quartile

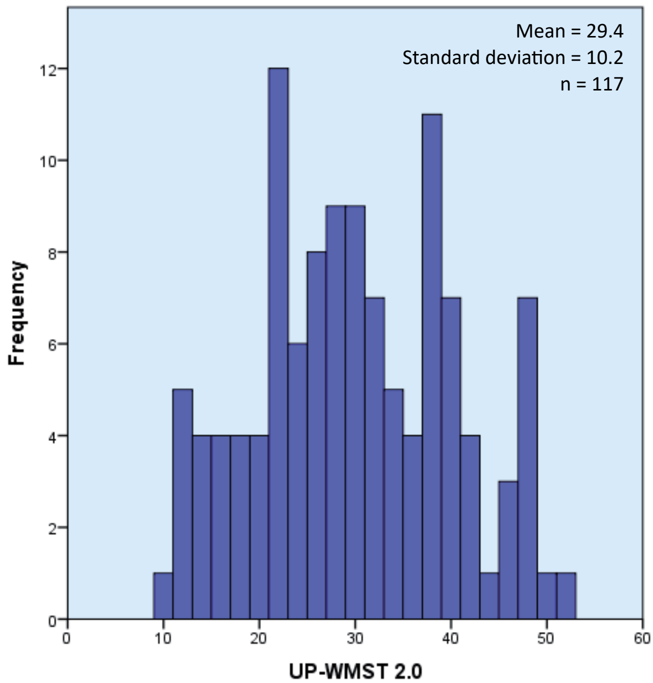


Figure 1. Distribution of baseline UP-WMST 2.0 scores in total sample

Table 5 Comparison of total UP-WMST 2.0 scores between and within groups

	n	Mean UP-WMST 2.0	Standard deviation	Range	P values
Age					
Children	56	26.6	9.3	10-47	
Adolescents	61	32.0	10.5	11-51	< 0.01*
Diagnosis					
Cerebral Palsy	47	26.1	9.4	11-48	
Spina Bifida	31	35.2	9	13-48	< 0.001*
Responsiveness					
Pre-training	20	30.6	8.8	14-47	
Post-training	20	38.9	8.8	26-50	< 0.001**

UP-WMST = Utrecht Pediatric Wheelchair Mobility Skills Test, * from independent samples T-Test, ** from paired samples T-Test

Responsiveness

Table 3 showed the descriptive statistics of the 23 participants who participated in a WMS training with their pre-training and post-training UP-WMST 2.0 score. Two participants could not complete the program due to unrelated medical treatments and one participant was not available for assessment post-training. The results of these 3 participants were not included in the statistical analysis. The paired sample t-test of 20 participants shows a significant ($p<0.001$) improvement of 8.3 points (95% confidence interval, 5.29–11.31).

DISCUSSION

This is the first study to evaluate measurement properties of a WMS outcome measure in youth using a manual wheelchair. Our results show positive estimates of reliability, content validity and construct validity and preliminary evidence of the responsiveness of the UP-WMST 2.0 for youth who use a manual wheelchair.

Reliability

A low kappa value for the item 'sudden stop' let to the removal of this item from the UP-WMST 2.0. In earlier research, the item 'sudden stop' was found to be relevant to assess in youth using a manual wheelchair[11]. The results of this study show that the current administration is not a reliable method and was therefore removed from the current version. Given the relevance though, future research should look into a different method of assessing this item. The total score of the UP-WMST 2.0 showed an excellent $ICC_{agreement}$ (0.97), comparable to the ICC's reported for WMS outcome measures in adults (0.90-0.99)[17,25,26]. In addition to the ICC, we calculated the $SEM_{agreement}$ and SDC. This information is useful in clinical practice. The SDC of 4.98 points indicates that an individual improvement or decline of 5 or more points reflects a true change in UP-WMST 2.0 score. This change in score is 9.4% of the total range of the UP-WMST 2.0 and is in line with previously reported detectable change score (5-15 %) in two adult WMS outcome measures[26]. Thus, repeated periodically, the UP-WMST 2.0 can be used as a criterion for the effectiveness of rehabilitation treatment focusing on WMS in youth who use a manual wheelchair.

Content validity

In addition to the evidence reported on content validity in the previous study on the development of the UP-WMST[11], this study assessed the comprehensiveness of the UP-WMST 2.0 through the analysis of floor and ceiling effects. The results show no floor or ceiling effect detectable in our sample and further supports the content validity of the measure. Even though no ceiling effect occurred in this study, 15% of our study sample scored in the upper ranges of this test (40 points or more). Youth with more advanced WMS may require a different outcome measure[9,11].

Construct validity

Independent t-test analysis confirmed our hypothesis regarding age and diagnoses. Youth with SB and adolescents scored significantly higher on the UP-WMST 2.0 compared to youth with CP and children respectively. Even though the group of adolescents scored on average higher than the group of children, their lower and upper UP-WMST 2.0 limits did not differ much. Similarly, the range in scores between youth with SB and CP did not differ much, despite the fact that youth with SB on average scored 9.1 points higher. Clinicians should be aware of these large variations in WMS within age groups or different diagnoses. Bloemen et al.[19] already mentioned possible explanations for the variation in wheelchair skill related fitness for children with SB, such as wheelchair features, physical factors or propulsion techniques. Sawatzky et al.[27] found that speed and mechanical effectiveness explained 36% of the variance in wheelchair skills scores in adult and children wheelchair users. In adults who use a wheelchair, wheelchair weight[17] and upper-extremity strength[28] already showed to have an influence on wheelchair skill outcome measures. Future research may provide insight into how these factors contribute to WMS in youth.

Responsiveness

On group level we found a 8.3 points improvement, which is a 27% mean relative improvement in WMS on the UP-WMST 2.0 after a WMS training program. These results show a larger relative improvement compared to the 14% improvement reported by Sawatsky et al.[9] in six children following a WMS training program. These findings show the UP-WMST 2.0 to be sensitive to measure change in this group of participants over a 6 month period. On an individual level, participants showed variation in the magnitude of improvement. This variation could be due to possible wheelchair or physical related factors as mentioned above, different effects of WMS training per participant, or lower sensitivity of the UP-WMST 2.0 in some children. The sample size for this portion of the study was too small to explore possible explanations for the observed variations. The small sample size also made it impossible to determine the Minimal Important Change (MIC). The MIC is the smallest change in score of the UP-WMST 2.0 which patients perceive as important[10]. The MIC together with the SDC are very useful parameters for clinicians to interpret change in individual UP-WMST 2.0 scores. For use in clinical practice, future research with larger samples should examine the MIC and evaluate if the UP-WMST 2.0 is sensitive to measure change in a broad sample of youth who use a manual wheelchair.

Strength and limitations

The sample size used in this study is large compared to other studies in youth who use a manual wheelchair. The inclusion of a heterogeneous group of participants who use a wheelchair leads to an increase in the clinical relevance and generalizability of the results of this study[10]. Moreover, to ascertain clinical relevance, a group of physical therapists, occupational therapists and physical education teachers of the Fit-For-The-Future Consortium[20] have been advising the research team through all the phases of the development of the UP-WMST 2.0. One of the limitations of this study was the use of several test administrators to administer the UP-WMST 2.0. Even though the administrators were trained to administer the test according to protocol, it was impossible to determine if test administrators had a different influence on the participants' performance. A participant might for example perform differently when the test is administered by a familiar therapist compared to an unknown research team member. At the same time, this too reflects daily clinical practice and increases the generalizability of the results. The interrater reliability of the UP-WMST 2.0 was not assessed in this study. Another limitation was the sample size in the responsiveness study. The sample size was small due to the set-up of the Let's Ride pilot study. The results do give a good indication whether the test is responsive to measure change on group level, but no general statements can be made towards the responsiveness of the UP-WMST 2.0. The results of this study show the UP-WMST 2.0 can be used in further research towards wheelchair mobility in youth. Furthermore, the average age of the responsiveness group was younger than the average age of the total sample. Future research should evaluate the interrater reliability and responsiveness of the UP-WMST 2.0 in a larger and slightly older sample. Future outcomes of our currently ongoing Let's Ride study into the responsiveness of the UP-WMST 2.0 after a WMS training in a larger sample will give additional valuable information about the SDC compared to the MIC. These future outcomes combined with the results of this study will make the UP-WMST 2.0 more suitable for use on individual level in clinical practice.

CONCLUSION

This study supports the reliability, content validity and construct validity of the UP-WMST 2.0 to measure WMS in youth who use a manual wheelchair and show preliminary evidence that suggests responsiveness to change over a 6 month period. Further research is needed to improve the interpretation of UP-WMST 2.0 scores for use in daily practice.

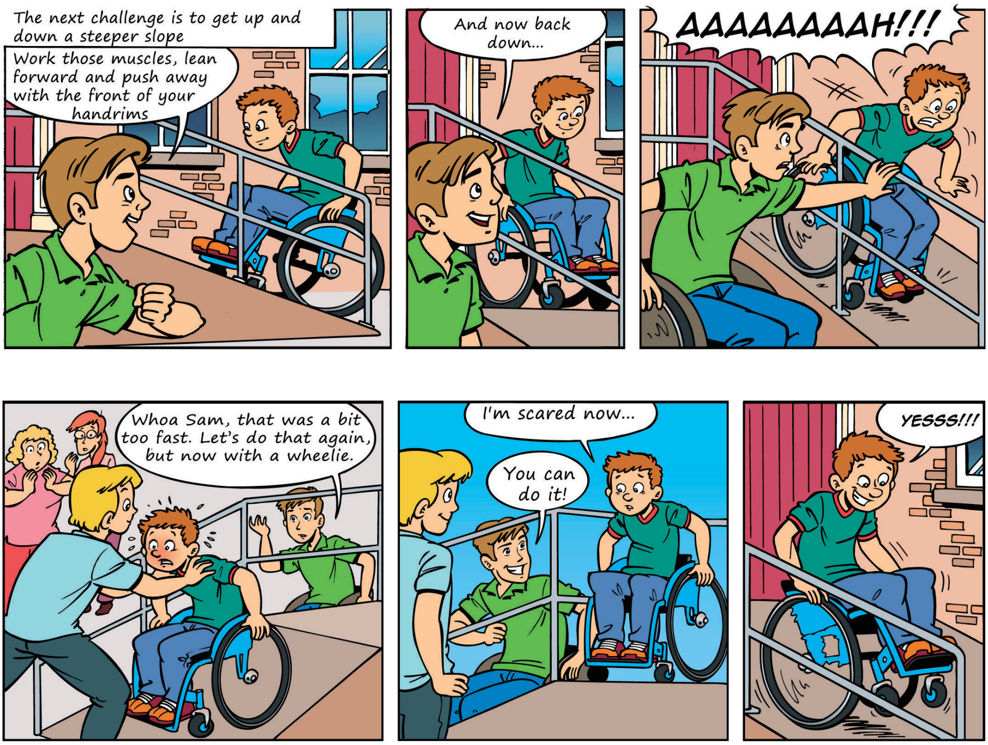
ACKNOWLEDGEMENTS

This study is part of the Fit For the Future collaboration; H. Wittink, A. Dallmeijer, R van den Berg-Emons. We would like to thank all participating children, their parents and health care professionals for volunteering their time and sharing their experiences with us. We would like to thank all the participating students who have enthusiastically contributed to the data collection and analysis in the Let's ride study. We are especially grateful to the BOSK patient organization and KJ-projects for sharing their expertise and involvement in the Wheelchair Mobility Skills Training program.

REFERENCES

1. Smith EM, Sakakibara BM, Miller WC. A review of factors influencing participation in social and community activities for wheelchair users. . 2016;11(5):361-374. doi: 10.3109/17483107.2014.989420 [doi].
2. Hosseini SM, Oyster ML, Kirby RL, Harrington AL, Boninger ML. Manual wheelchair skills capacity predicts quality of life and community integration in persons with spinal cord injury. Arch Phys Med Rehabil. 2012;93(12):2237-2243.
3. MacPhee AH, Kirby RL, Coolen AL, Smith C, MacLeod DA, Dupuis DJ. Wheelchair skills training program: A randomized clinical trial of wheelchair users undergoing initial rehabilitation1. Arch Phys Med Rehabil. 2004;85(1):41-50. doi: http://dx.doi.org/10.1016/S0003-9993(03)00364-2.
4. Kirby RL, Mitchell D, Sabharwal S, McCranie M, Nelson AL. Manual wheelchair skills training for community-dwelling veterans with spinal cord injury: A randomized controlled trial. PLoS One. 2016;11(12):e0168330. doi: 10.1371/journal.pone.0168330 [doi].
5. Ozturk A., Ucsular F.D. Effectiveness of a wheelchair skills training programme for community-living users of manual wheelchairs in turkey: A randomized controlled trial. Clin Rehabil. 2011;25(5):416-424.
6. Worobey LA, Kirby RL, Heinemann AW, et al. Effectiveness of group wheelchair skills training for people with spinal cord injury: A randomized controlled trial. Arch Phys Med Rehabil. 2016;97(10):1777-1784.e3. doi: 10.1016/j.apmr.2016.04.006 [doi].
7. Best KL, Routhier F, Miller WC. A description of manual wheelchair skills training: Current practices in canadian rehabilitation centers. Disabil Rehabil Assist Technol. 2014. doi: 10.3109/17483107.2014.907367 [doi].
8. Bloemen MA, Verschuren O, van Mechelen C, 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. doi: 10.1186/s12883-015-0265-9 [doi].
9. Sawatzky B, Rushton PW, Denison I, McDonald R. Wheelchair skills training programme for children: A pilot study. Aust Occup Ther J. 2012;59(1):2-9. doi: 10.1111/j.1440-1630.2011.00964.x [doi].
10. de Vet HCW, Terwee CB, Mokkink LB, Knol DL. Measurement in medicine. New York: Cambridge University Press; 2011.
11. Sol ME, Verschuren O, de Groot L, de Groot JF, Fit-For-the-Future!-consortium. Development of a wheelchair mobility skills test for children and adolescents: Combining evidence with clinical expertise. BMC Pediatr. 2017;17(1):51-017-0809-9. doi: 10.1186/s12887-017-0809-9 [doi].
12. World Health Organization. International classification of functioning, disability and health (ICF). http://apps.who.int/classifications/icfbrowser/. Accessed 01/04, 2016.
13. Mokkink LB, Terwee CB, Patrick 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(7):737-745. doi: 10.1016/j.jclinepi.2010.02.006 [doi]
14. Mokkink LB, Terwee CB, Knol DL, et al. The COSMIN checklist for evaluating the methodological quality of studies on measurement properties: A clarification of its content. BMC Med Res Methodol. 2010;10:22-2288-10-22. doi: 10.1186/1471-2288-10-22 [doi]

15. Larson JC, Mostofsky SH, Goldberg MC, Cutting LE, Denckla MB, Mahone EM. Effects of gender and age on motor exam in typically developing children. Dev Neuropsychol. 2007;32(1):543-562. doi: 10.1080/87565640701361013 [doi].
16. Ploegmakers JJ, Hepping AM, Geertzen JH, Bulstra SK, Stevens M. Grip strength is strongly associated with height, weight and gender in childhood: A cross sectional study of 2241 children and adolescents providing reference values. J Physiother. 2013;59(4):255-261. doi: 10.1016/S1836-9553(13)70202-9 [doi].
17. Kirby RL, Dupuis DJ, Macphee AH, et al. The wheelchair skills test (version 2.4): Measurement properties. Arch Phys Med Rehabil. 2004;85(5):794-804. doi: S0003999303009754 [pii].
18. Verschuren O, Zwinkels M, Obeid J, Kerkhof N, Ketelaar M, Takken T. Reliability and validity of short-term performance tests for wheelchair-using children and adolescents with cerebral palsy. Dev Med Child Neurol. 2013;55(12):1129-1135. doi: 10.1111/dmcn.12214 [doi].
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. 2017;98(6):1097-1103. doi: S0003-9993(16)30961-3 [pii].
20. Https://Husite.nl/fitforthefuture/project-informatie/.
21. Kilkens OJ, Post MW, van der Woude LH, Dallmeijer AJ, van den Heuvel WJ. The wheelchair circuit: Reliability of a test to assess mobility in persons with spinal cord injuries. Arch Phys Med Rehabil. 2002;83(12):1783-1788. doi: http://dx.doi.org/10.1053/apmr.2002.36066.
22. Sol ME, De Groot JF, Verschuren O, Visser-Meily JMA. Let's ride…. effectiveness of wheelchair skills training in manual wheelchair-using children. World Confederation of Physical Therapy congress. 2017;Poster presentation.
23. Portney LG, Watkins MP. Foundations of clinical research: Application to practice (3rd edition). Prentice Hall; 2009.
24. de Vet HC, Bouter LM, Bezemer PD, Beurskens AJ. Reproducibility and responsiveness of evaluative outcome measures. theoretical considerations illustrated by an empirical example. Int J Technol Assess Health Care. 2001;17(4):479-487.
25. Cowan RE, Nash MS, de Groot S, van der Woude LH. Adapted manual wheelchair circuit: Test-retest reliability and discriminative validity in persons with spinal cord injury. Arch Phys Med Rehabil. 2011;92(8):1270-1280. doi: http://dx.doi.org/10.1016/j.apmr.2011.03.010.
26. Fliess-Douer O, Van der Woude LHV, Vanlandewijck YC. Reliability of the test of wheeled mobility (TOWM) and the short wheelie test. ARCHIVES OF PHYSICAL MEDICINE AND REHABILITATION}. 2013;94(4):761-770. doi: 10.1016/j.apmr.2012.09.023.
27. Sawatzky B, Hers N, MacGillivray MK. Relationships between wheeling parameters and wheelchair skills in adults and children with SCI. Spinal Cord. 2015;53(7):561-564. doi: 10.1038/sc.2015.29 [doi].
28. Kilkens OJ, Dallmeijer AJ, Nene AV, Post MW, van der Woude LH. The longitudinal relation between physical capacity and wheelchair skill performance during inpatient rehabilitation of people with spinal cord injury. Arch Phys Med Rehabil. 2005;86(8):1575-1581. doi: http://dx.doi.org/10.1016/j.apmr.2005.03.020.


The next challenge is to get up and down a steeper slope
Work those muscles, lean forward and push away with the front of your handrims
And now back down...
AAAAAAAAH!!!
Whoa Sam, that was a bit too fast. Let's do that again, but now with a wheelie.
I'm scared now...
You can do it!
YESSS!!!

Chapter 4

Wheelchair mobility confidence scale for dutch youth using a manual wheelchair

Sol ME[1-3], Bloemen MAT[1], Verschuren O[2], Rushton PW[4], Visser-Meily JMA[2], de Groot JF[1,5]

1. Research Group Lifestyle and Health, HU University of Applied Sciences Utrecht, Utrecht, the Netherlands
2. Brain Center Rudolf Magnus and Center of Excellence for Rehabilitation Medicine, University Medical Center Utrecht and De Hoogstraat Rehabilitation, Utrecht, The Netherlands
3. Special Needs School, De Hoogstraat Rehabilitation, Utrecht, The Netherlands
4. École de réadaptation, Université de Montréal , Montréal, QC , Canada, Canada
5. Netherlands Institute for Healthcare Services Research (NIVEL), Utrecht, the Netherlands

Disabil Rehabil Assist Technol 2020 Aug;15(6):684-691.

ABSTRACT

Purpose: The objective of this study was to develop a questionnaire to assess confidence in wheelchair mobility in Dutch Youth (WheelCon-Mobility Dutch Youth).

Methods: (1) A forward-backward translation process was used to translate the original WheelCon-M from English into Dutch. (2) Items related to wheelchair mobility in Dutch youth were selected and adapted based on focus groups with youth, parents and health care professionals to create the WheelCon-Mobility Dutch Youth. (3) The WheelCon-Mobility Dutch Youth and the Utrecht Pediatric Wheelchair Mobility Skills Test 2.0 (UP-WMST 2.0) were administered to 62 participants to evaluate internal consistency and construct validity.

Results: Translation and cultural adaptation led to general adaptations in instructions, sentence structure and response scale. At the item level, 24 items were included with (n=17) and without (n=7) adaptation, 10 items were deleted and 7 new items were included. The WheelCon-Mobility Dutch Youth had an excellent Cronbach's alpha of 0.924 and a significant correlation ($r = 0.44$, $p < 0.001$) with the UP-WMST 2.0.

Conclusions: This study resulted in the adaptation of the WheelCon-M into the WheelCon-Mobility for Dutch youth using a manual wheelchair. Our study suggests there is evidence supporting the internal consistency and construct validity of the WheelCon-Mobility Dutch Youth.

Keywords: WheelCon; wheelchair mobility; youth; translation; validity; confidence

INTRODUCTION

When looking at physical activity behavior, studies have shown that youth (children and adolescents) using a manual wheelchair is more sedentary and less physically active than able-bodied peers [1-3] and peers with a disability who are ambulatory [4]. In a study of Dutch youth using a manual wheelchair, Bloemen et al.[5] reported that youth with Spina Bifida was approximately 72 minutes physically active on a school day compared to 175 minutes of physical activity time for typically developing peers.

To improve physical activity behavior in youth with a disability, recent reviews concluded that interventions should not only focus on body functions and impairments, but also take personal factors, such as confidence, and environmental factors, such as the importance of a good assistive device, into account [5, 6]. Similarly, two qualitative studies [7, 8] have described that confidence and wheelchair mobility skills are important factors in facilitating participation in physical activity. These results are in line with Bandura's Social Cognitive Theory, where confidence is strongly related to motivation and an important factor for changes in behavior [9]. Therefore, to improve physical activity in youth using a manual wheelchair, confidence-specific targeted interventions and measurement instruments need to be developed and implemented.

To the best of our knowledge, there is currently no questionnaire available to assess confidence in wheelchair mobility in youth. However, to assess confidence in wheelchair use in adults, there are two questionnaires available: the Wheelchair Use Confidence Scale for people who use a manual wheelchair (WheelCon-M) [10] and the Self-Efficacy in Wheeled Mobility (SEWM) [11]. In the literature, the WheelCon-M [10, 12-19] has been reported more extensively than the SEWM [11]. Measurement properties of the WheelCon-M have been studied thoroughly and evidence has shown that the WheelCon-M is a valid and reliable questionnaire to measure confidence in wheelchair use in adults [13].

The original WheelCon-M is a self-report 65-item questionnaire on a 101-point response scale for English-speaking adults using a manual wheelchair. It measures several areas of confidence in wheelchair use including 'negotiating the environment', 'activities performed in the wheelchair', 'knowledge and problem solving', 'advocacy', 'managing social situations' and 'managing emotions' [12]. Studies using the WheelCon-M have demonstrated an association between lower confidence in wheelchair use and lower levels of participation frequency and life space mobility [10, 19]. It stands to reason that there would be similar associations in the pediatric population.

The aim of this study was to develop a measure to assess confidence in wheelchair mobility among Dutch youth, where confidence in wheelchair mobility is defined as the belief individuals have in their ability to use their wheelchair in a variety of physically challenging situations. Specific study objectives were (1) to translate the original WheelCon-M into the WheelCon-M Dutch, (2) to select items specific to confidence in wheelchair mobility and to culturally adapt these items for Dutch Youth using a manual wheelchair (WheelCon-Mobility Dutch Youth) and (3) to evaluate the internal consistency and construct validity of the WheelCon-Mobility Dutch Youth.

METHODS

Design

The WheelCon-Mobility Dutch Youth was developed using a three-phase mixed-methods design. *Phase 1: Translation* consisted of a forward-backward translation process. *Phase 2: Item Selection and Cultural Adaptation* was accomplished using focus groups. *Phase 3:* Baseline results of a larger intervention study were used for the *Evaluation of Internal Consistency and Construct Validity*. The Medical Ethics Committee of the University Medical Center Utrecht, the Netherlands, approved the study procedures (15-136). All participants provided informed consent.

Phase 1: Translation

For the translation of the original WheelCon-M from English into Dutch we used the Translation and Cultural Adaptation of Patient Reported Outcomes Measures–Principles of Good Practice guidelines [20], the same method used by Rushton et al. [15] who translated the original WheelCon-M into French. This method consists of nine steps.

In step one, the translation team was assembled which consisted of: three members (MS, OV and JdG) whose first language is Dutch and second language is English, who live in the Netherlands and who have a background in the subject area of pediatric manual wheelchair users; one member (TS) who is a native English speaker with Dutch as his second language who has lived in the Netherlands for 17 years; and one Canadian member (PWR) whose first language is English, with extensive expertise in wheelchair confidence being the developer of the original WheelCon-M. The second step involved independent forward translations from English into Dutch by two team members (MS and OV). For the third step, both forward translations were compared and discrepancies were resolved through consensus by the Dutch members of the team. The fourth step was a parallel back translation from Dutch into English by two team members (JdG and TS). In step five, the back translation was compared to the original version by the Canadian researcher and discrepancies were resolved through consensus by the research team. Steps six to eight, the harmonization, cognitive debriefing and review of cognitive debriefing were not included as part of this study. Step nine, proofreading of the Dutch translation of the WheelCon-M was conducted by the Dutch team members in preparation for Phase 2.

Phase 2: Item Selection and Cultural Adaptation

Participants

Purposive sampling was used to recruit health care professionals / educators, children who use a manual wheelchair (aged 6 to 12) and their parents, adolescents who use a manual wheelchair (aged 13 to 18) and their parents. Health care professionals / educators were eligible to participate if they were an OT, a PT, a physical education teacher, a pediatric psychologist or an independent wheelchair mobility skills trainer (educator) with over 3 years of experience working with youth who use a wheelchair. They were recruited from two rehabilitation centers (De Hoogstraat and Merem) in the Netherlands and from a company (K-J projects) that teaches wheelchair mobility skills trainings in the Netherlands. Youth were eligible to participate if either their parents or their health care professional thought they were capable of speaking about confidence in wheelchair mobility, used their wheelchair for more than one year and for more than 4 hours on a daily basis. They were recruited through local physiotherapists at the same two rehabilitation centers from where the health care professionals were recruited or through the Let's Ride database, a database of youth using a manual wheelchair who previously participated in research of the Research Group of Lifestyle and Health and consented to being contacted again. Parents were recruited through their corresponding participating child. The ability to speak in Dutch and to participate in a focus group were eligibility criterion for all participants.

Procedure

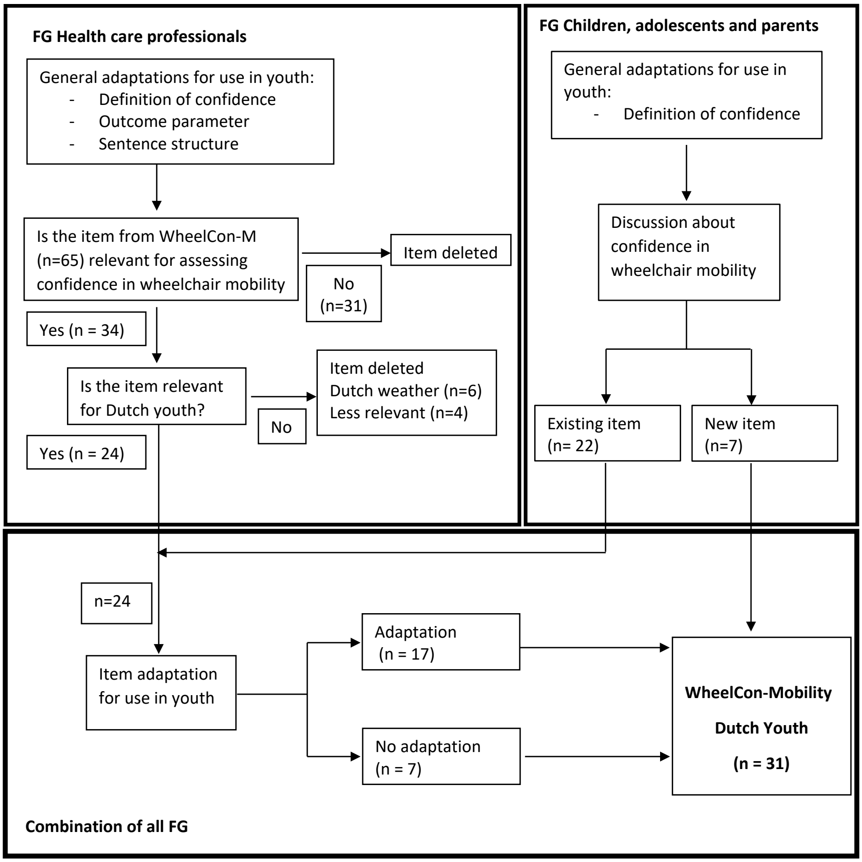
Five focus groups were held with the health care professionals / educators (n=1), children (n=1), adolescents (n=1) and parents (n=2). Each focus group was conducted by an experienced moderator and assistant moderator using a stakeholder-specific focus group guide. The aim of the session with health care professionals / educators was to discuss (1) children's ability to understand the term 'confidence', (2) identify general adaptations of the WheelCon-M Dutch translation for Dutch youth, (3) discuss on item level selection of items related to wheelchair mobility and (4) determine relevancy of selected items for Dutch youth. Prior to this focus group, participants received a demographic questionnaire, a brief overview of the study rationale and the translated WheelCon-M. Participants were asked to review in advance the translated WheelCon-M and to prepare to discuss aims 3 and 4 (described above). The focus groups with children, adolescents and parents were conducted with the aims of (1) discussing children's ability to understand the term 'confidence' and (2) to identify additional relevant items for measuring confidence with wheelchair mobility, which are not yet included in the WheelCon-M Dutch translation. Prior to these focus groups, the parents received a demographic questionnaire and a brief overview of the study rationale. Parents and youth did not receive the translated WheelCon-M to assure an unbiased opinion about relevant topics on confidence in wheelchair mobility. All focus groups were audio recorded and transcribed verbatim.

Data analysis
Two independent researchers analyzed the transcripts of the focus groups using a content framework approach [21]. Data from the focus group with health care professionals / educators were coded according to the four aims of the session. Segments of the focus groups with youth and their parents were coded as 'definition of confidence', 'existing items from the WheelCon-M' or as 'new items'. Differences in coding were resolved through discussion between three Dutch team members (MS, OV, JdG) until consensus was reached. After analysis of the focus group with health care professionals/educators and the analysis of the combined four focus groups with youth and their parents, both results were combined to decide if an item was relevant for assessing wheelchair mobility in Dutch youth. When an item was deemed less relevant by health care professionals / educators and not mentioned by youth or their parents it was considered to be irrelevant for Dutch youth. The results were discussed in the research group, which led to the adaptation of the WheelCon-M into the WheelCon-Mobility Dutch Youth. The WheelCon-Mobility Dutch Youth was digitalized in an online survey program for ease of administration using a tablet in phase three of this study.

Phase 3 Internal consistency and construct validity

Participants
This phase of the study was part of an ongoing larger intervention study evaluating the effectiveness of a wheelchair mobility skills training program for youth attending rehabilitation centers and schools for special education in the Netherlands (trialregister.nl, registration number NTR5791). A convenience sample of participants was recruited by local physiotherapists and occupational therapists in the participating rehabilitation centers / schools for special education (n=6). To be included in this study, participants needed to be: between 7-18 years of age, (bimanual) wheelchair user on a daily basis, able to understand the spoken Dutch language and be able to understand simple instructions. Participants were excluded if they had undergone a medical intervention during the previous six months that could have affected the intervention study outcomes (e.g., botox-injections).

Outcome Measures
The resulting WheelCon-Mobility Dutch Youth developed in phase 2 (figure 1 and appendix 1) was used in phase 3 for assessment of internal consistency and construct validity.



FG = Focus group, WheelCon-Mobility Dutch Youth = Wheelchair Mobility Confidence scale for Dutch youth

Figure 1. Results of adaptation of WheelCon-M into WheelCon-Mobility Dutch Youth in Phase 1

The Utrecht Pediatric Wheelchair Mobility Skills Test 2.0 (UP-WMST 2.0) [22, 23] is a recently developed and validated objective, performance-based measure of wheelchair mobility skills for children and adolescents using a manual wheelchair. It consists of 15 items, such as propulsion forward/backward, turning, holding a wheelie and ascending and descending slopes and platforms. The total score may range from 0 to 51, with a higher score representing more advanced wheelchair mobility skills [23]. We hypothesized that the WheelCon-Mobility Dutch Youth would have a positive low-moderate correlation (r = 0.3-0.7) with the UP-WMST 2.0, given that wheelchair mobility skills and wheelchair confidence in the adult population have demonstrated a positive, moderate correlation [19].

Procedure
All measures were administered at baseline. The WheelCon-Mobility Dutch Youth was administered with support from a researcher or research assistant when necessary. The UP-WMST 2.0 was administered by a local PT or OT, who had received a four-hour training (theory and practice) on how to administer this outcome measure.

Data analyses
Cronbach's alpha was calculated as a parameter of internal consistency. A Cronbach's alpha of 0.5-0.6 was considered as poor, 0.6-0.7 as questionable, 0.7-0.8 as acceptable, 0.8-0.9 as good and > 0.9 as excellent [24].

For construct validity we visually checked if the data were normally distributed using histograms, QQ-plots and assessed normality using the Kolmogorov-Smirnov Test. Consecutively, we analyzed the linearity of the data graphically by scatterplots. Depending on the normality of the data, Pearson correlation coefficients or Spearman correlation coefficients were used to calculate the correlation. Low correlation was defined as r = 0.3-0.5, moderate correlation as r = 0.5 – 0.7, high correlation as r = 0.7 – 0.9 and excellent correlation as r = 0.9 – 1.0 [24]. Finally, we assessed if there was a change in correlation between the WheelCon-Mobility Dutch Youth and the UP-WMST 2.0 when corrected for age.

RESULTS

Phase 1: Translation
The translation of the WheelCon-M from English to Dutch revealed some important topics. As the research team felt the concept of confidence is difficult to understand, especially for children, they decided it was an important topic that had to be discussed during the focus groups. In addition, adjustment to Dutch standard building codes of items regarding ramps, slopes or curbs were considered. Because the degrees and heights of the ramps, slopes and curbs are similar in Canada and the Netherlands no adjustments were necessary.

Phase 2: Item Selection and Cultural Adaptation
Seven participants took part in the health care professional / educator focus group. Participant characteristics are presented in table 1. In terms of aim (1), minimal age to understand concept of confidence, after discussion consensus was reached that at eight years of age a child with normal cognitive function would be able to understand the concept of confidence and independently complete the questionnaire. This decision was based on the expert-opinion of the pediatric psychologist. Furthermore, the health

Table 1. Characteristics of participants of focus group in phase 2

	Youth (n=8)	Parents (n=9)	Health care professionals/ educator (n=7)
Age (mean (range))	12 (8-17)	45 (41 – 52)	36 (25-59)
Sex (M/F)	5/3	2/7	3/4
Diagnosis/profession	2 Cerebral Palsy 6 Spina Bifida		1 pediatric psychologist 2 occupational therapists 2 wheelchair skills trainers 2 pediatric physical therapists
Wheelchair/working experience (years(range))	8 (6 – 15)		11 (3-19)
Sports participation (yes / no)	6/2		
Type of education	8 regular education 1 special education		

care professionals suggested that "confidence" should be clearly defined at the beginning of the questionnaire in simple and concrete language. In terms of aim (2), general adaptations, participants suggested to modify the original 101-point response scale, as this was considered too complicated for younger participants. Consensus was reached about using a 5-point scale ranging from 1 (no confidence) to 5 (very high confidence). It was proposed that the scale be visually supported by 5 circles in ascending size, from small (no confidence) to big (very high confidence). Furthermore, the appropriateness of the sentence structure for youth was discussed. This discussion led to adjustment of the stem to make it grammatically correct in Dutch and to include the words 'independently' and 'safely' to remind the participants the items are about an independent performance. To visually support the questions and to make it easier for youth to understand and answer the questions, participants advised to include photos. As a result, the research team added photos to 15 items (appendix 1).

In terms of aim (3), relevance to wheelchair mobility, only the items in the domain "negotiating the environment (n=34)" were deemed relevant for assessing confidence in wheelchair mobility among Dutch youth. The other items (n = 31) from the WheelCon-M are related to other aspects of wheelchair use. In terms of aim (4), relevance for Dutch youth, five items were deemed not relevant in Dutch weather conditions. Twenty-nine of the items in the domain "negotiating the environment" were considered relevant for youth. However, when discussing the importance of making the questionnaire as short as possible for youth, some items were deemed less relevant (n = 5) and could possibly be removed from the questionnaire.

Demographic characteristics of the participants in the four focus groups with children (n = 5), adolescents (n = 3) and parents (n = 3 and n = 6) are presented in Table 1. Data from these focus group transcripts were analyzed according to children's ability to understand the term confidence (aim 1) and items to measure confidence with wheelchair mobility (aim 2). In terms of aim (1), participants gave different descriptions and suggestions on how to explain confidence to a child. For aim (2), participants in these focus groups identified 22 items that were already present in the WheelCon-Mobility Dutch Youth and 7 new items, including cross a busy street without a crosswalk or traffic light, cross over a road with a 5° incline, move wheelchair down 3 steps, move wheelchair up and down an escalator, propelling the wheelchair without anti-tipping wheels and performing a wheelie.

These results were combined with the outcome of the focus group with health care professionals / educators. All items that were deemed less relevant (n = 10) by health care professionals / educators were not mentioned in the focus groups with youth and their parents. Therefore, these items were deleted from the questionnaire. This process led to a total of 31 items in the WheelCon-Mobility Dutch Youth, with a minimal score of 31 and a maximal score of 155.

Phase 3 Internal consistency and construct validity

The characteristics of the 62 participants included in this phase of the study are presented in table 2. Four participants were unable to complete the questionnaire due to cognitive disabilities and thus their data was removed from the analyses. The remaining 58 participants, with a mean age of 13 years, completed the WheelCon-Mobility Dutch Youth with a mean score of 109 (SD 22, range 55-154) in 15-25 minutes. The WheelCon-Mobility Dutch Youth and UP-WMST 2.0 (mean 31.4, standard deviation 10.1) data were normally distributed.

Table 2. Characteristics of participants in phase 3

	Participants (n=58)
Age	13.0 (SD 3.3 range 7.1-18.8)
Sex	34 boys, 24 girls
Diagnosis	23 Cerebral Palsy 10 Spina Bifida 8 Neuromuscular Disease 15 Other
Progressive disorder	47 no 11 yes
Wheelchair experience (years)	8.4 (mean 4.0, range 1-15)
Level of education	37 No learning problems 21 Mild or severe learning problems

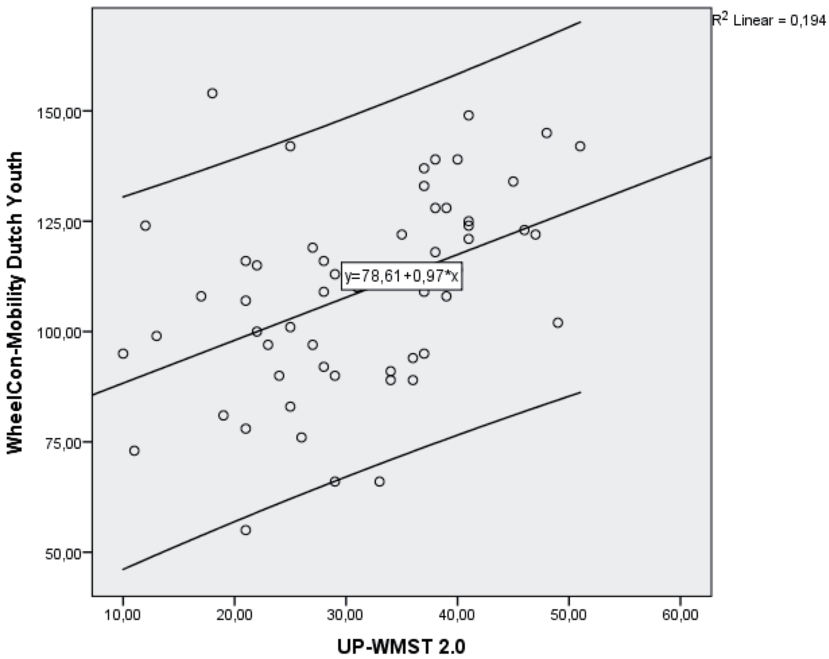


Figure 2. Scatterplot of the Wheelchair Mobility Confidence Scale for Dutch Youth (WheelCon--Mobility Dutch Youth) and the Utrecht Pediatric Wheelchair Mobility Skills Test (UP-WMST 2.0) with the confidence interval.

Cronbach's alpha of the 31-item WheelCon-Mobility Dutch Youth was excellent (0.924). Analyzing "Cronbach's alpha if item deleted" showed that no items needed to be deleted.

Figure 2 shows the scatterplot of the correlation between the WheelCon-Mobility Dutch Youth and the UP-WMST 2.0 with the confidence interval. Table 3 shows the correlations of the WheelCon-Mobility Dutch Youth with the UP-WMST 2.0. We found a positive correlation between the WheelCon-Mobility Dutch Youth and the UP-WMST 2.0 ($r = 0.44$, $p < 0.001$). No changes in significance of correlations were found when corrected for age ($r = 0.43$, $p < 0.001$).

DISCUSSION

To the best of our knowledge, this is the first study reporting on an outcome measure to assess confidence in wheelchair mobility in youth using a manual wheelchair.

An important consideration in phase 2 of this study was the ability of youth to understand the concept of confidence and answer questions about their own confidence. Our finding of a minimum age of 8 years is in line with evidence from the literature, where a child with normal cognitive abilities should be able to understand the concept of reflection and comparing themselves to peers at the age of 7-8 years old [25, 26]. Several adaptations were made in the WheelCon-Mobility Dutch Youth to increase the understanding of the question (photos) and answer options (five point response scale) for youth. These adaptations are similar to the use of examples and 5-point Likert Scale in a Quality of Life Questionnaire for people with an intellectual disability [27]. Given these adaptations for youth, one should keep in mind that decreased cognitive abilities may affect the ability to complete the WheelCon-Mobility Dutch Youth. Our advice is to accompany a child when administering the WheelCon-Mobility Dutch Youth, to allow a child to ask clarification if needed. A similar method is used in the administration of the Self Perception Profile for Children in young children at kindergarten level [28]. However, even with adult support four children (aged 10-12 years old) with severe learning problems out of the 17 participants with mild to severe learning problems in phase 3 of this study were unable to complete the WheelCon-Mobility Dutch Youth. Future research, by using a think-aloud process, may give more insight into how children, and specifically children with decreased cognitive abilities understand and answer items regarding confidence. This may lead to further refinement of the questionnaire or development of proxy reports by parents.

Another aim of this study was to select items related to wheelchair mobility. The selection of these items based on the qualitative results of the focus groups are in line with the short-form list of items selected through a Rasch analyses of the WheelCon-M [10], contributing to the content validity of this questionnaire. Interestingly, most of the wheelchair mobility items from the existing questionnaire for adults were deemed relevant for youth. These results are similar to the development of the UP-WMST, were most of the wheelchair mobility skills for adults were also relevant to assess in youth and show that youth encounter similar barriers when negotiating their wheelchair [22]. There are however some important basic additions specifically for youth in this questionnaire, such as propelling the wheelchair without anti-tippers, and performing a wheelie, which is also in line with the development of the UP-WMST 2.0, where more basic skills were included in a wheelchair mobility skills test for youth [22]. Surprisingly, there was also the addition of more advanced wheelchair mobility related items, such as going up and down an escalator. It was not possible to distinguish from this data, if the use of an escalator with a wheelchair might be more relevant for the Dutch situations or for youth specifically. Further research towards validating the WheelCon-M in the Dutch adult population, could give more insight.

Furthermore, this study shows similar evidence towards internal consistency (Cronbach's alpha 0.92) of the WheelCon-Mobility Dutch Youth as the original WheelCon-M and translated versions of the Wheelcon-M (Cronbach's alpha 0.92-0.98) [12, 14, 15]. Deletion of items did not lead to a higher Cronbach's alpha and this finding supports the inclusion of all current items. Nevertheless, the current time to complete the questionnaire is 15-25 minutes and future research using Rasch analyses in a larger sample size could lead to further adaptation of the WheelCon-Mobility Dutch Youth into a short form as has been done in the adult English [10] and Italian version [14].

For evidence regarding construct validity, we confirmed our hypotheses regarding a significant low to moderate positive correlation (r_s=.44) between the WheelCon-Mobility Dutch Youth and the UP-WMST 2.0 [23]. The strength of this correlation was lower than the correlation reported in adult manual wheelchair users (r=0.52) [19]. A lower correlation in youth could possibly be explained by a larger overestimation of their own capabilities compared to adults [26]. When looking at the distribution of scores in this study, there were four participants who had a high confidence in performing wheelchair mobility skills, but a low capacity of performing skills. Interestingly, these conflicting scores were also found in adult wheelchair users [19]. It is therefore important to asses both the confidence in wheelchair mobility and the wheelchair mobility skill level in youth in clinical practice.

Strengths and limitations

Collaborating with the original developer of the WheelCon-M and the use of the Translation and Cultural Adaptation of Patient Reported Outcomes Measures [20] during translation from English to Dutch were strengths in our research. Furthermore, we used qualitative research with a variety of stakeholders, including health care professionals, parents and youth in order to decide which adjustments should be made for the youth version. In addition, a sample size of 58 youth for quantitative analyses was larger than the sample size used in the Italian [14] and French translation [15]. The evidence for internal consistency and construct validity was proven in a heterogeneous sample, with different diagnoses, ages and learning difficulties. The heterogeneity of this sample is a good representation of the Dutch youth who use a manual wheelchair in daily life, supporting the use of this outcome measure in clinical practice. The design of the focus groups with children, adolescents and parent ascertained an open mind, but this also made it impossible to check if all items together reflect the whole topic of confidence in wheelchair mobility in youth. Further research into the structure of the WheelCon-Mobility Dutch Youth using a factor analyses is necessary to confirm if these original, adapted and new items belong to the same domain of confidence in wheelchair mobility in youth. Due to our research design of the larger ongoing study, we were not able to analyze the test-retest reliability and therefore we were not able to look at agreement and measurement error. Although the original and translated versions do show good test-retest reliability, it would still be recommended to study test-retest reliability of this adapted version of the WheelCon-Mobility Dutch Youth[14, 15].

CONCLUSION

This study resulted in the adaptation of the WheelCon-M into the WheelCon-Mobility for Dutch Youth using a manual wheelchair. It shows positive evidence towards internal consistency and construct validity of the WheelCon-Mobility Dutch Youth. Future studies should lead to further refinement of the WheelCon-Mobility Dutch Youth, assess test-retest agreement and measurement error. In clinical practice, it is important to assess confidence in wheelchair mobility and the capacity of performing a wheelchair mobility skill to choose the most appropriate treatment method.

ACKNOWLEDGEMENTS

This study is part of the Fit For the Future collaboration; H. Wittink, A. Dallmeijer, R van den Berg-Emons. We would like to thank all participating children, their parents and health care professionals for volunteering their time and sharing their experiences with us. We would like to thank Tim Schilling for his help with the translations. We would like to thank all the participating students who have enthusiastically contributed to the data collection and analysis in the Let's ride study. We are especially grateful to the BOSK patient organization and KJ-projects for sharing their expertise and involvement in the Wheelchair Mobility Skills Training program.

APPENDIX 1

Items related to wheelchair mobility from original WheelCon and new items	FG Health Care Professionals Relevant (Yes/No)	FG Children (C), Adolescents (A) and Parents (P)	Item number (#) and adaptation
1. can move your wheelchair over carpet?	Yes	A	#1
2. can move your wheelchair around furniture in your own home?	Yes	P	#2
3. can move your wheelchair over thresholds, such as between rooms?	Yes	P	#3 Added photo
4. can manoeuvre your wheelchair in small spaces, such as a bathroom?	Yes	A,P	#4
5. can move your wheelchair through a door that opens automatically?	No	-	Deleted, less relevant
6. can open, go through, and then close a standard 81cm (32”) lightweight door?	Yes	-	#5 Added photo
7. can open and go through a spring loaded door, such as a door at your local mall?	No	-	Deleted, less relevant
8. can move your wheelchair up a standard ramp, built to code (5º incline)?	Yes	C,A,P	#6 Added photo
9. can move your wheelchair down a standard ramp, built to code (5º incline)?	Yes	C,A,P	#7 Added photo
10. can move your wheelchair up a dry steep slope (> 5º incline)?	Yes	C,A,P	#8 Added photo
11. can move your wheelchair down a dry steep slope (> 5º incline)?	Yes	C,A,P	#9 Added photo
12. can move your wheelchair down a dry steep slope (> 5º incline) and stop as soon as you are off the slope?	Yes	A	#10 Added photo
13. can move your wheelchair up a curb cut?	Yes	C,P	#11 Added photo
14. can move your wheelchair down a curb cut?	Yes	C,P	#12 Added photo
15. can move your wheelchair over a drainage grate and then up a curb cut?	Yes	P	#13 Added photo
16. can move your wheelchair down a curb cut then over a drainage grate?	Yes	P	#14 Added photo
17. can move your wheelchair through a puddle then up a curb cut?	No	-	Deleted, less relevant
18. can move your wheelchair down a curb cut then through a puddle?	No	-	Deleted, less relevant
19. can move your wheelchair through slush then up a curb cut?	No	-	Deleted, less relevant Dutch weather conditions
20. can move your wheelchair down a curb cut then through slush?	No	-	Deleted, less relevant Dutch weather conditions
21. can move your wheelchair down a curb cut then though 5cm (2”) snow?	No	-	Deleted, less relevant Dutch weather conditions
22. can move your wheelchair through 5cm (2”) snow then up a curb cut?	No	-	Deleted, less relevant Dutch weather conditions
23. can move your wheelchair up a standard height curb 15cm (6”) without a curb cut?	Yes	C,A,P	#15 Added photo + remove words ‘without a curb cut’
24. can move your wheelchair down a standard height curb 15cm (6”) without a curb cut?	Yes	C,A,P	#16 Added photo + remove words ‘without a curb cut’

Continue

Continued

Items related to wheelchair mobility from original WheelCon and new items	FG Health Care Professionals Relevant (Yes/No)	FG Children (C), Adolescents (A) and Parents (P)	Item number (#) and adaptation
25. can manoeuvre your wheelchair to press the crosswalk button and cross the street before the traffic light changes?	Yes	P	#17 Added photo + include words 'changes to red'.
26. can cross a street with light traffic at a crosswalk with no traffic lights?	Yes	P	#18
27. can move your wheelchair across 3m (10ft) of flat, freshly mowed, dry grass?	Yes	C,P	#19 Remove words 'flat, freshly mowed, dry'
28. can move your wheelchair through a pothole that is wider than your wheelchair and 5cm (2") deep?	Yes	C	#20
29. can move your wheelchair along a paved sidewalk that is cracked and uneven?	Yes	-	#21 Added photo
30. can move your wheelchair along a flat dirt path or trail with some tree roots and rocks?	Yes	P	#22
31. can move your wheelchair across 3m (10ft) of flat, unpacked gravel?	Yes	A	#23 Added photo + remove words 'flat, unpacked'
32. can move your wheelchair along a sidewalk with 5cm (2") of snow?	No	-	Deleted, less relevant Dutch weather conditions
33. can move your wheelchair through a crowd of people without hitting anyone?	Yes	P	#24
42. can move your wheelchair down a store aisle that has just enough room for your wheel-chair without knocking items over?	No	-	Deleted, less relevant
New			
can cross a busy street, without a crosswalk or traffic light, with your wheelchair?		P	#25
can cross over a road with a 5° incline with your wheelchair?		P	#26
can move your wheelchair up an escalator?		C,P	#27
can move your wheelchair down an escalator?		C,P	#28
can move your wheelchair down 3 steps without a handrail?		C,A,P	#29
can move you wheelchair without anti-tipping wheels?		C,P	#30
can balance your wheelchair on the rear wheels (wheelie).		C,A,P	#31

REFERENCES

1. Bloemen M, vd Berg-Emons R, Tuijt M, Nooijen C, Takken T, Backx F, Vos M, de Groot J: Physical activity in wheelchair-using youth with Spina Bifida: an observational study. Accepted in J Neuroeng Rehabil .
2. Buffart LM, Westendorp T, van den Berg-Emons RJ, Stam HJ, Roebroeck ME: Perceived barriers to and facilitators of physical activity in young adults with childhood-onset physical disabilities. J Rehabil Med 2009, 41(11):881-885.
3. Nooijen CFJ, Post MWM, Spooren AL, Valent LJ, Broeksteeg R, Sluis TA, Stam HJ, Act-Active Research Group, van den Berg-Emons RJG: Exercise self-efficacy and the relation with physical behavior and physical capacity in wheelchair-dependent persons with subacute spinal cord injury. J Neuroeng Rehabil 2015, 12:10.1186/s12984-015-0099-0.
4. 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):1856-1861.
5. 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, 59(10):1004-1010.
6. 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):989-997.
7. 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, 15:11-015-0265-9.
8. Shields N, Synnot A: Perceived barriers and facilitators to participation in physical activity for children with disability: a qualitative study. BMC Pediatr 2016, 16:9-016-0544-7.
9. Bandura A: Self-Efficacy: The exercise of control: New York: W. H. Freeman and Company; 1997.
10. Sakakibara BM, Miller WC, Rushton PW: Rasch analyses of the wheelchair use confidence scale. Arch Phys Med Rehabil 2015, 96(6):1036-1044.
11. Fliess-Douer O., van der WL, Vanlandewijck Y.C.: Development of a new scale for perceived self-efficacy in manual wheeled mobility: a pilot study. J Rehabil Med 2011, 43(7):602-608.
12. Rushton PW, Miller WC, Lee Kirby R, Eng JJ, Yip J: Development and content validation of the Wheelchair Use Confidence Scale: a mixed-methods study. Disabil Rehabil Assist Technol 2011, 6(1):57-66.
13. Rushton PW, Miller WC, Kirby RL, Eng JJ: Measure for the assessment of confidence with manual wheelchair use (WheelCon-M) version 2.1: reliability and validity. J Rehabil Med 2013, 45(1):61-67.
14. Berardi A, De Santis R, Tofani M, Marquez MA, Santilli V, Rushton PW, Mollica R, Galeoto G: The Wheelchair Use Confidence Scale: Italian translation, adaptation, and validation of the short form. Disabil Rehabil Assist Technol 2017, :1-6.
15. Rushton PW, Routhier F, Miller WC, Auger C, Lavoie MP: French-Canadian translation of the WheelCon-M (WheelCon-M-F) and evaluation of its validity evidence using telephone administration. Disabil Rehabil 2015, 37(9):812-819.

16. Rushton PW, Smith E, Miller WC, Vaughan K: Measuring wheelchair confidence among power wheelchair users: an adaptation of the WheelCon-M using focus groups and a think aloud process. 2017, 12(1):39-46.
17. Sakakibara BM, Miller WC, Routhier F, Backman CL, Eng JJ: Association between self-efficacy and participation in community-dwelling manual wheelchair users aged 50 years or older. Phys Ther 2014, 94(5):664-674.
18. Sakakibara BM, Miller WC, Eng JJ, Backman CL, Routhier F: Influences of wheelchair-related efficacy on life-space mobility in adults who use a wheelchair and live in the community. Phys Ther 2014, 94(11):1604-1613.
19. Sakakibara BM, Miller WC: Prevalence of low mobility and self-management self-efficacy in manual wheelchair users and the association with wheelchair skills. Arch Phys Med Rehabil 2015, 96(7):1360-1363.
20. Wild D, Grove A, Martin M, Eremenco S, McElroy S, Verjee-Lorenz A, Erikson P, ISPOR Task Force for Translation and Cultural Adaptation: Principles of Good Practice for the Translation and Cultural Adaptation Process for Patient-Reported Outcomes (PRO) Measures: report of the ISPOR Task Force for Translation and Cultural Adaptation. Value Health 2005, 8(2):94-104.
21. Gale NK, Heath G, Cameron E, Rashid S, Redwood S: Using the framework method for the analysis of qualitative data in multi-disciplinary health research. BMC Med Res Methodol 2013, 13:117-2288-13-117.
22. Sol ME, Verschuren O, de Groot L, de Groot JF, Fit-For-the-Future!-consortium: Development of a wheelchair mobility skills test for children and adolescents: combining evidence with clinical expertise. BMC Pediatr 2017, 17(1):51-017-0809-9.
23. Sol M, de Groot J, Zwinkels M, Visser-Meily A, Kruitwagen C, Verschuren O: The Utrecht Pediatric Wheelchair Mobility Skills Test: validity, reproducibility and responsiveness in children and adolescents using a manual wheelchair. Submitted, .
24. Portney LG, Watkins MP: Foundations of Clinical Research: Application to Practice (3rd edition): Prentice Hall; 2009.
25. Veerman JW, Straathof MAE, Treffers PDA, Van den Bergh B, Ten Brink LT: Handleiding Competentiebelevingsschaal voor Kinderen (CBSK) (Manual for the Dutch version of the SPPC). Lisse, The Netherlands: Swets & Zeitlinger; 1997.
26. Fredricks JA, Eccles JS: Children's competence and value beliefs from childhood through adolescence: growth trajectories in two male-sex-typed domains. Dev Psychol 2002, 38(4):519-533.
27. Smith EM, Sakakibara BM, Miller WC: A review of factors influencing participation in social and community activities for wheelchair users. 2016, 11(5):361-374.
28. Noordstar JJ, van der Net J, Jak S, Helders PJ, Jongmans MJ: The change in perceived motor competence and motor task values during elementary school: A longitudinal cohort study. Br J Dev Psychol 2016, 34(3):427-446.


Then, it's time for recess
You seem to have become heavier since last year, Sam!
Whoops, sorry!
BONK
Shall I leave you here? In the sunshine?
I'll pick you up in a little bit.
Hey Sam; do you want to play with us?
No, that's alright.

Chapter 5

The criterion validity of the Activ8 to measure physical activity in youth using a wheelchair

Kristel Lankhorst[1,2], Marleen Sol[1,3], Rita van den Berg-Emons[4], Herwin Horemans[4], Janke de Groot[1,5]

1. Research Group Lifestyle and Health, HU University of Applied Sciences Utrecht, Utrecht, The Netherlands
2. Institute for Human Movement Studies, HU University of Applied Sciences Utrecht, Utrecht, The Netherlands
3. Center of Excellence for Rehabilitation Medicine, UMC Utrecht Brain Center, University Medical Center Utrecht, and De Hoogstraat Rehabilitation, Utrecht, The Netherlands.
4. Department of Rehabilitation Medicine, Erasmus University Medical Center, Rotterdam, the Netherlands
5. Knowledge Institute of the Federation of Medical Specialists, Utrecht, The Netherlands.

Submitted

ABSTRACT

Purpose: The ability to objectively assess physical activity in people using a manual wheelchair is important for both clinical practice and research. It remains unclear whether the Activ8 is valid for youth using a manual wheelchair.

Methods: The aim of this study was to assess the criterion validity of the Activ8 (1) to detect 'active wheelchair use' and (2) to distinguish six types of wheelchair activities using video recordings as a gold standard. Ten participants (10-18 years old) who use a manual wheelchair were included.

Results: Criterion validity for detecting 'active wheelchair use' showed a relative time difference of 7.4%, agreement of 96%, sensitivity of 98.3% and positive predictive value of 90%. Results for distinguishing six types of wheelchair activities showed an agreement of 73%, sensitivity of 67.1% and positive predictive value of 65.5%.

Conclusions: The Activ8 is able to detect 'active wheelchair use' in youth using a manual wheelchair. Further development of the algorithm is necessary to distinguish between different types of wheelchair activities.

INTRODUCTION

The ability to objectively assess physical activity (PA) is important to determine whether a person meets the PA guidelines and whether programs aimed at improving PA in clinical practice are indicated and effective in people using a manual wheelchair. Especially in children, objective assessment is preferred over subjective assessment as their PA mainly consists of repeated brief bouts which is difficult to accurately report in questionnaires [1–4]. Moreover, a recent systematic review [5] showed there was no evidence for validated questionnaires and advised to use objective assessments in youth using a manual wheelchair.

The same systematic review[6] reported on the criterion validity of commercially available activity monitors in people using a manual wheelchair and found several devices to be valid for detecting intensity of PA, type of PA or both in people using a wheelchair. However, there are still some challenges to overcome before activity monitors can be broadly applied in research or clinical practice [5,7]. For example, data was mainly collected in a laboratory setting, limiting the external validity of activity monitors. Furthermore, the installation, analysis and interpretation of results was complex, limiting the feasibility in clinical practice [7]. Moreover, selection of an appropriate device to measure PA depends on the population for which it was developed and validated. The predictive models used in the development of activity monitors are based on the data of the sample used and therefore only valid in a similar population, which was primarily adult wheelchair users with a spinal cord injury [6–9]. For youth, congenital disorders such as cerebral palsy and Spina Bifida are more common diagnosis that lead to using a manual wheelchair[10,11]. More research towards the validity of activity monitors in youth using a manual wheelchair is required [5,12].

There are likely to be differences between youngsters and adults in types of activities and in movement patterns for wheelchair propulsion, for example due to a different ratio of arm length/wheelchair size. Of all the commercially available activity monitors, only the Vitamove [13] was validated in youth using a manual wheelchair. However, the costs, the complex data analysis, size (4 x 8 x 1,5 cm) and measurement burden of wearing three body fixed devices (two on wrists, one on sternum) makes the Vitamove less feasible for use in clinical practice [14]. For the appliance of accelerometers in youth, Jang et al. [4] suggested to use no more than two small wearable devices. For detection of 'active wheelchair user', the combination of two monitors is necessary to distinguish between active wheelchair activities, such as propelling the wheelchair at normal speed or maneuvering, and non-propulsive wheelchair activities, such as being pushed around i.e. movement of wheel and minimal movement of wrist, or using a laptop i.e. movement of wrist and no movement of wheel [15].

The Activ8 meets these criteria with the combination of one monitor attached at the wrist and one monitor on the wheel. The Activ8 Activity monitor (2M Engineering Ltd., Valkenswaard, The Netherlands) is a small, lightweight triaxial accelerometer (3.0 x 3.2 x 1.0 cm), which is relatively inexpensive and has a short Epoch data collection window (5 seconds). Data analysis is simple which makes it suitable for use in research and clinical practice[16,17]. Moreover, the Activ8 has already successfully been used in ambulatory youth with a physical disability, where it was found valid for detecting type of PA [17]. In adult able-bodied wheelchair users, the Activ8 has been found valid for detecting 'active wheelchair use'[18]. Whether the Activ8 is also able to detect 'active wheelchair use' in youth using a manual wheelchair remains unclear. At the same time, Leving et al. [18] found the Activ8 not valid for distinguishing different types of wheelchair activities such as 'maneuvering' and 'driving fast' and suggested future research should be aimed at improving the accuracy of the algorithm of the Activ8. Therefore, the aim of this study was to assess the criterion validity of the adapted algorithm of the Activ8 to (1) detect 'active wheelchair use' and (2) to distinguish between six different types of wheelchair activities in youth using a manual wheelchair.

METHODS

Participants

Participants were recruited through a research database of a larger research group (Fit-for-the-Future consortium) in the Netherlands, which included research in youth with Spina Bifida and research in youth with a chronic disability. Participants were included if they used a wheelchair for daily mobility, were able to perform the wheelchair activities in the activity protocol, able to understand the instructions and if they were 10 to 18 years old. Participants were excluded if they used a power wheelchair. In total, ten participants who used a wheelchair for daily mobility were included in this study. Sample size was based on the number of participants in comparable studies[13,16,17,19]. In line with Dutch laws, participants over twelve years old and all parents gave written informed consent.

Ethics approval

The study was approved by the Medical Ethical Committee of the Erasmus MC University Medical Center, Rotterdam, the Netherlands (MEC number: MEC-2013-404). The study was conducted in accordance with the principles of the Declaration of Helsinki (www.wma.net) and in accordance with the Medical Research Involving Human Subjects Act (WMO).

Testing procedure

The semi-structured activity protocol was executed by participants in a natural environment (home or school environment). The protocol was based on earlier research and consisted of eight basic activities and seven complex activities that each lasted 90 seconds (table 2) [8,18,20]. A basic activity was defined as an activity consisting of a single body posture or movement, for example wheelchair propulsion. A complex activity was defined as a combination of postures and movements of short duration. For instance, a ball game could consist of both driving (normal speed) and non-driving (sitting). The basic and complex activities in this protocol are representative of the everyday life of youth using a manual wheelchair [13]. Participants were asked to perform each activity in their own manner and interim rest periods were allowed. The total duration of the measurement time was approximately 45 minutes.

Activ8 activity monitor

One Activ8 monitor was placed on the dorsal side of the right wrist with Tegaderm (3M) skin tape to ensure a fixed position on the wrist (Figure 1)[18]. The second Activ8 monitor was attached with tape as close as possible to the axle of the right wheel[18]. The raw acceleration signals (12.5Hz) of each monitor were filtered with an exponential moving average filter and converted to counts. A build-in algorithm produced predefined postures and movements for an upright population based on counts and orientation of the monitor, which were summated and stored on the monitor in epochs of 5 seconds[16,18].

Reference method

The gold standard was observation through video recording made with a handheld digital video camera of every participant by a trained research assistant. Camera positions were adapted to the movement of the participant to ensure visibility of the motion of the right wrist and right wheel of the wheelchair. The activities from the activity protocol on the video were independently scored by two trained researchers with an epoch of five seconds (same interval as Activ8) for each participant[13,21]. During complex activities,

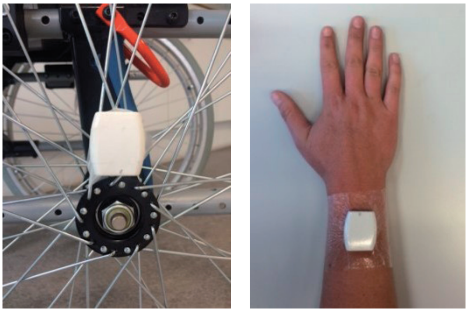


Figure 1. Placement of Activ8 on wheel and wrist

multiple activities can be performed during one epoch. As a ball game, the participant propelled and manoeuvred the wheelchair within one epoch of five seconds. The type of wheelchair activity with the longest duration during those five seconds determined the score of that five second epoch. A similar method was used for determining type of wheelchair activity per five second video observation. Thereafter, the five second Activ8 epochs were synchronized with the five second video data.

Algorithm for activity classification

Similarly to Leving et al.[18] the classification of activities was based on combining the synchronized vector counts of the wrist and wheel sensor using a custom written Matlab algorithm to automate the process of assigning a class to a given 5s epoch interval. This algorithm has been previously developed with data of wheelchair users and able bodied individuals[18]. In this study 'Active wheelchair use' is a grouped category that consists of: 'wheelchair propulsion normal speed', 'wheelchair propulsion fast speed' and 'maneuvering' (momentary movement such as a twist). The three other types of wheelchair activities: 'assistive driving' (being pushed around), 'sitting sedentary' and 'sitting active' are grouped in a 'non-propulsive wheelchair use' category.

In this study, the algorithm was adapted for youth based on the data of two at random selected participants, i.e. the activity count thresholds between classes for the wrist and wheel sensor were adapted. Average counts for both wrist and wheel for each class were considered as range midpoints and thresholds as midpoints between ranges.

Data analysis

The criterion validity was defined as 'the degree to which the scores of a measurement instrument, in this case the Activ8, are an adequate reflection of a gold standard, in this case observation through video recordings [22]. The primary aim of this study was to assess the absolute and relative time difference, agreement, sensitivity and positive predictive value of the adapted algorithm of the Activ8 to detect 'active wheelchair use' compared to observations through video recording in youth using a manual wheelchair. Comparisons were made for every 5 second Epoch window.

- Absolute (seconds) and relative (percentage) time difference between the Activ8 and video recording in the total duration of 'active wheelchair use' was determined per participant.
- Agreement was calculated per participant as the proportion of time the Activ8 identified 'active wheelchair use' correctly as identified by observation through video recording.
- Sensitivity was calculated per participant as: (correct detection of 'Active wheelchair use' by Activ8)/ (correct detection of 'Active wheelchair use' by video)* 100[23].

- Positive predicted value was calculated per participant as: (correct detection of 'Active wheelchair use' by Activ8)/(total detection of 'active wheelchair use' (correct and incorrect) by Activ8)*100[23].

The secondary aim of this study was to assess the absolute and relative time difference, agreement, sensitivity and positive predicted value between the adapted algorithm of the Activ8 and video recordings for six types of wheelchair activities (sitting sedentary, sitting active, assisted driving, normal driving, fast driving, manoeuvring).

The mean and range of absolute and relative time differences and agreement and the mean sensitivity and positive predicted value were calculated for detection of 'active wheelchair use' and six types of wheelchair activities in the total sample. Relative time differences of less than 10% were considered acceptable, which is similar to other similar studies [13,15,18].

Table 1: Characteristics of the participants.

Participant		Gender	Age (y)	Diagnosis	Weight (kg)	Length (m)
Criterion validity	1	Female	14	SB, FMS 1	52	1.62
	2	Female	11	SB, FMS 1	50	1.50
	3	Female	18	SB, FMS 1	52	1.85
	4	Male	12	SB, FMS 2-2-1	28	1.43
	5	Female	14	SB, FMS 1	69	1.64
	6	Female	10	SB, FMS 1	47	1.41
	7	Female	15	SB, FMS 1	68	1.53
	8*	Male	10	CP, FMS 2-1-1	60	1.58
Adaption of algorithm	1	Female	18	SB, FMS 1	63	1.77
	2	Male	16	SB, FMS 1	67	1.53

SB = Spina Bifida; FMS = Functional Mobility Score, CP = Cerebral Palsy, * Missing data criterion validity due to wheelchair malfunction.

Table 2: The semi-structured activity protocol

Basic activities	Complex Activities
Sitting	Slalom
Slouched sitting	Ball game
Driving slow	Dribbling
Driving normal	Table covering
Driving fast	Handcrafting
Assistive driving	Reading
20 meter driving at preferred pace	Transfer
Hand-biking	

Values for agreement, sensitivity and positive predicted value were considered excellent when more than 90%, good when 80% to 90%, moderate when 70% to 80% and weak when less than 70% [13,15,18].

RESULTS

A total of ten participants aged 10 to 18 years old were included in this study (Table 1). One participant was unable to perform the activities in the protocol due to wheelchair malfunction and therefore not included in data analysis. Data of two participants were used for adaptation of the algorithm and therefore excluded from analyses of criterion validity. All activities in the activity protocol were performed by all participants, except for handbiking where two participants did not have a handbike and were therefore unable to perform this activity.

1. Active wheelchair use

For the detection of 'active wheelchair use' the relative time difference between the video and Activ8 was 7.4% with an agreement of 96% (standard deviation 2.1%; range 93-100%), sensitivity of 98.3% and positive predicted value of 90.0% (Table 3).

2. Six types of wheelchair activities

The relative time difference between the video and Activ8 in distinguishing six types of wheelchair activities was >10% for all activities. The overall agreement was 73% (standard deviation 8.5%; range 59-86%) (Table 4). Absolute time difference, sensitivity and positive predictive value are described in Table 4.

DISCUSSION

The primary aim of this study was to evaluate the criterion validity of the Activ8 to detect 'active wheelchair use' in youth using a manual wheelchair. The results of our study indicate that the Activ8, with the newly adapted algorithm for youth, appears to be valid for detecting 'active wheelchair use' with an excellent agreement, an acceptable small relative time difference, excellent sensitivity and positive predictive value.

The results for criterion validity of the adapted algorithm Activ8 are more accurate than the results reported by Leving et al.[18], who reported moderate to good levels of agreement, sensitivity and positive predicted value in able bodied adults using a wheelchair. A possible explanation for the higher agreement found in our study could be the adapted algorithm used. For the wheel sensor, the threshold for activity counts between 'sitting sedentary/

Table 3: Criterion validity of the Activ8 to detect 'active wheelchair use'.

	Video (mean sec)	Activ8 (mean sec)	Agreement (SD) [Range]	Absolute time difference (sec)*		Relative time difference (%)**		Sensitivity	Positive Predictive value
				Mean	Range	Mean	Range		
Active Wheelchair Use	560	602	96 % (2.1 %) [93-100%]	42.1	[-70–0]	7.4	[0–12.1]	98.3	90.0
Non-propulsive wheelchair use	465	423							

*Calculated as time identified by Activ8 minus time identified by video recording. ** Calculated as time identified by Activ8 minus time identified by video recording, divided by the time of the video observation, times 100. SD = standard deviation

Table 4. Criterion validity of the Activ8 to distinguish six types of wheelchair activities.

	Video (mean sec)	Activ8 (mean sec)	Agreement (SD) [Range]	Absolute time difference (sec)*		Relative time difference (%)**		Sensitivity	Positive Predictive value
				Mean	[Range]	Mean	[Range]		
Types of wheelchair activities			73% (8.5 %) [59-86%]						
Sitting sedentary	271	199		-71.4	[-130–-15]	-26.3	[-45.6–-5.6]	72.1	97.9
Sitting active	124	163		38.6	[-35–90]	27.7	[-36.8–69.2]	70.1	60.7
Assisted Driving	70	61		-9.3	[-25–0]	-13.1	[-35.7–0]	86.9	100
Normal Driving	421	231		-190.7	[-320–-90]	-44.2	[-75.3–-20.4]	50.6	85.4
Fast Driving	98	274		176.4	[40–280]	196.4	[34.8–400]	89.4	35.3
Manoeuvring	41	97		56.4	[20–115]	385.8	[25–850]	32.7	13.2
Mean								**67.1**	**65.5**

*Calculated as time identified by Activ8 minus time identified by video recording. ** Calculated as time identified by Activ8 minus time identified by video recording, divided by the time of the video observation, times 100. SD = standard deviation active' and 'maneuvering' was three times higher compared to Leving et al.[18], resulting in less overestimation of 'active wheelchair use'. These results together with the low costs of the device and easy to use software make the Activ8 an appropriate device for measuring the frequency and duration of 'active wheelchair use' in youth using a manual wheelchair. This information can be used in clinical practice to measure the time spent in 'active wheelchair use' in daily life, to evaluate the effectiveness of tailored interventions aimed at increasing 'active wheelchair use' or to give tailored advice on how active (duration) clients use their wheelchair and how this activity is distributed (frequency) over the day or weekend [14]. For example, if playing during school breaks or leisure time after school is mostly classified as non-propulsive wheelchair use (i.e. 'sitting sedentary/active or 'assisted driving'), physiotherapy can be aimed at stimulating active play on the playground at school or near their home to pursue healthy active behavior.

Stimulating active play could possibly be enhanced with a real-time feedback function on PA goals [24]. Regrettably the Activ8 does not have a real-time feedback option for wheelchair users yet, while this is already available for ambulatory persons.

A secondary aim was to assess the criterion validity of the Activ8 to distinguish six types of wheelchair activities. Even though there was a moderate agreement of 73% (range 59-86%), there were large relative time difference between the observations through video recording and Activ8 measurements. Sensitivity and positive predictive value values were also weak, indicating that the adapted algorithm of the Activ8 is not able to distinguish six different types of wheelchair activity accurately. In this study, the largest error of misclassification between the Activ8 and video recording was found for 'maneuvering' which appeared to have a large relative time difference and low values of sensitivity and positive predicted value. This was partly due to the low amount of time spent maneuvering the wheelchair in the used protocol, but also due to challenges in classifying this type of wheelchair activity correctly [13,18]. Another misclassification was found for distinguishing 'driving fast' from 'driving normal'. For several basic and complex activities we found an overestimation of 'driving fast' and an underestimation of 'driving normal'. In ambulatory youth with or without a disability the Activ8 also overestimated running and underestimated walking[17]. This shows that there are large individual differences between angular speed of movement in ambulatory and wheelchair using youth, which are difficult to correctly classify with set threshold values for activity counts between classes. A possible solution for this problem is an individual calibration with the development of algorithms that correct for differences in individual movement speed.

These results are similar to other studies using the Activ8 in different populations [17,18,20], commercial available monitors in adult wheelchair users [5,25] and to a study of Nooijen et al. [13] in youth using a manual wheelchair using three large body-fixed monitors (the Vitamove). While accelerometers are valid for detecting grouped activities, there is misclassification when aiming to distinguish different types of wheelchair activities [5,9,25].

There is a limited amount of research towards assessing objectively measured physical activity in youth using a wheelchair [6,12], therefore results from this study offer valuable information for this population. One of the strengths of this study, was the selection of a small accelerometer that is feasible to use in clinical practice and is already validated in ambulatory youth with a physical disability[17]. Youth with cerebral palsy[11] or Spina Bifida[10] use different methods of mobility depending on the severity of the disorder and can be ambulatory indoors while using a wheelchair outdoors. Although further research is necessary, the Activ8 seems to be a promising tool for simultaneously assessing ambulatory and wheelchair activities in youth with a disability. Another strength was the use of an activity protocol in the natural environment increasing the validity of the Activ8 in real life. Further research in sports, recreational and free-living activities is necessary to improve the generalizability of the results[9].

The results of the criterion validity of our study are somewhat limited by the homogeneity of the sample with all participants diagnosed with Spina Bifida. Moreover, this study only included youth aged 10 – 18 years and therefor excluded young children. It is likely that younger children also perform physical activities outside the wheelchair, such as crawling and playing on the ground. Further validation of the Activ8 in a more heterogeneous population and in youth with other types of wheelchair propulsion, such as single hand with or without feet, electric assistance in wheelchair and sport specific mobility devices is warranted.

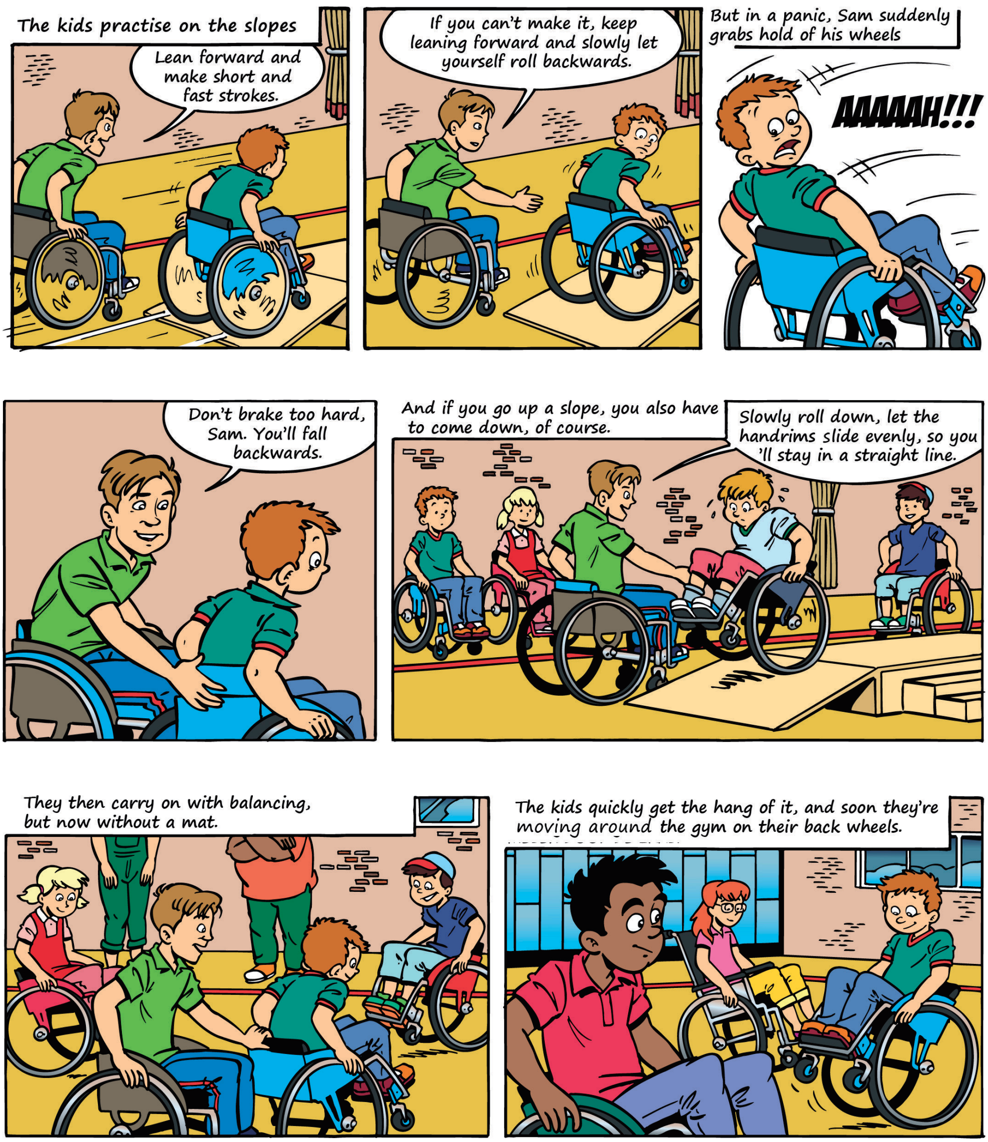
CONCLUSION

This study further adds to the validity of the Activ8 to measure 'active wheelchair use' in youth using a manual wheelchair. The Activ8 is not able to accurately distinguish between different types of wheelchair activity. Further development is necessary and is recommended to study in a broader population including younger children to improve the accuracy of the algorithm of the Activ8 in youth using a manual wheelchair.

REFERENCES

1. Adamo KB, Prince SA, Tricco AC, Connor-Gorber S, Tremblay M. A comparison of indirect versus direct measures for assessing physical activity in the pediatric population: A systematic review [Internet]. Vol. 4, International Journal of Pediatric Obesity. Int J Pediatr Obes; 2009 [cited 2020 Sep 8]. p. 2–27. Available from: https://pubmed.ncbi.nlm.nih.gov/18720173/
2. Bailey RC, Olson J, Pepper SL, Porszasz J, Barstow TJ, Cooper DM. The level and tempo of children's physical activities: An observational study. Med Sci Sports Exerc [Internet]. 1995 [cited 2020 Jul 6];27(7):1033–41. Available from: https://pubmed.ncbi.nlm.nih.gov/7564970/
3. Welk GJ, Corbin CB, Dale D. Measurement issues in the assessment of physical activity in children. Res Q Exerc Sport. 2000;71:59–73.
4. Jang Y, Kim S, Kim K, Lee D. Deep learning-based classification with improved time resolution for physical activities of children. PeerJ [Internet]. 2018 [cited 2020 Jul 6];6. Available from: https://pubmed.ncbi.nlm.nih.gov/30364555/
5. Lankhorst K, Oerbekke M, van den Berg-Emons R, Takken T, de Groot J. Instruments Measuring Physical Activity in Individuals Who Use a Wheelchair: A Systematic Review of Measurement Properties. Vol. 101, Archives of Physical Medicine and Rehabilitation. W.B. Saunders; 2020. p. 535–52.
6. Lankhorst K, Oerbekke M, van den Berg-Emons R, Takken T, de Groot J. Instruments Measuring Physical Activity in Individuals Who Use a Wheelchair: A Systematic Review of Measurement Properties. Arch Phys Med Rehabil [Internet]. 2020 Mar 1 [cited 2020 Apr 9];101(3):535–52. Available from: http://www.ncbi.nlm.nih.gov/pubmed/31606452
7. Routhier F, Lettre J, Miller WC, Borisoff JF, Keetch K, Mitchell IM. Data logger technologies for manual wheelchairs: A scoping review. Assist Technol. 2018 Mar 15;30(2):51–8.
8. Hiremath S V., Intille SS, Kelleher A, Cooper RA, Ding D. Detection of physical activities using a physical activity monitor system for wheelchair users. Med Eng Phys. 2015 Jan 1;37(1):68–76.
9. Tsang K, Hiremath S V., Crytzer TM, Dicianno BE, Ding D. Validity of activity monitors in wheelchair users: A systematic review. Vol. 53, Journal of Rehabilitation Research and Development. Rehabilitation Research and Development Service; 2016. p. 641–58.
10. Johnson KL, Dudgeon B, Kuehn C, Walker W. Assistive technology use among adolescents and young adults with Spina Bifida. Am J Public Health [Internet]. 2007 Feb [cited 2020 Sep 22];97(2):330–6. Available from: https://pubmed.ncbi.nlm.nih.gov/17194874/
11. Palisano RJ, Tieman BL, Walter SD, Bartlett DJ, Rosenbaum PL, Russell D, et al. Effect of environmental setting on mobility methods of children with cerebral palsy. Dev Med Child Neurol. 2003 Feb 1;45(2):113–20.
12. Ganz F, Hammam N, Pritchard L. Sedentary behavior and children with physical disabilities: a scoping review [Internet]. Disability and Rehabilitation. Taylor and Francis Ltd; 2020 [cited 2020 Apr 9]. p. 1–13. Available from: http://www.ncbi.nlm.nih.gov/pubmed/32116041
13. Nooijen CFJ, De Groot JF, Stam HJ, Van Den Berg-Emons RJG, Bussmann HBJ. Validation of an activity monitor for children who are partly or completely wheelchair-dependent. J Neuroeng Rehabil. 2015;12(1).

14. Bloemen MAT, van den Berg-Emons RJG, Tuijt M, Nooijen CFJ, Takken T, Backx FJG, et al. Physical activity in wheelchair-using youth with Spina Bifida: an observational study. J Neuroeng Rehabil [Internet]. 2019 Dec 14 [cited 2020 Apr 14];16(1):9. Available from: https://jneuroengrehab.biomedcentral.com/articles/10.1186/s12984-018-0464-x
15. Kooijmans H, Horemans HLD, Stam HJ, Bussmann JBJ. Valid detection of self-propelled wheelchair driving with two accelerometers. Physiol Meas [Internet]. 2014 Nov [cited 2020 May 12];35(11):2297–306. Available from: http://www.ncbi.nlm.nih.gov/pubmed/25340938
16. Horemans H, Kooijmans H, van den Berg-Emons R, Bussmann H. The Activ8 activity monitor: Validation of posture and movement classification. J Rehabil Assist Technol Eng [Internet]. 2020 Jan [cited 2020 May 6];7:205566831989053. Available from: http://www.ncbi.nlm.nih.gov/pubmed/32206335
17. Lankhorst K, Van Den Berg-Emons RJ, Bussmann JBJ, Horemans HLD, De Groot JF. A Novel Tool for Quantifying and Promoting Physical Activity in Youths with Typical Development and Youths Who Are Ambulatory and Have Motor Disability. Phys Ther [Internet]. 2019 Mar 1 [cited 2020 Sep 8];99(3):354–63. Available from: https://academic.oup.com/ptj/article/99/3/354/5289874
18. Leving MT, Horemans HLD, Vegter RJK, De Groot S, Bussmann JBJ, van der Woude LHV. Validity of consumer-grade activity monitor to identify manual wheelchair propulsion in standardized activities of daily living. PLoS One. 2018 Apr 1;13(4).
19. Postma K, Berg-Emons Van Den HJG, Bussmann JBJ, Sluis TAR, 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 Sep;43(9):550–7.
20. Fanchamps MHJ, Horemans HLD, Ribbers GM, Stam HJ, Bussmann JBJ. The accuracy of the detection of body postures and movements using a physical activity monitor in people after a stroke. Sensors (Switzerland). 2018 Jul 5;18(7).
21. Bussmann JBJ, Tulen JHM, Van Herel ECG, Stam HJ. Quantification of physical activities by means of ambulatory accelerometry: A validation study. Psychophysiology [Internet]. 1998 Sep [cited 2020 Oct 9];35(5):488–96. Available from: https://pubmed.ncbi.nlm.nih.gov/9715093/
22. Mokkink LB, Terwee CB, Knol DL, Stratford PW, Alonso J, Patrick DL, et al. The COSMIN checklist for evaluating the methodological quality of studies on measurement properties: A clarification of its content. BMC Med Res Methodol [Internet]. 2010 [cited 2020 Jul 6];10. Available from: https://pubmed.ncbi.nlm.nih.gov/20298572/
23. Portney L, Watkins M. Foundations of clinical research: Applications to practice. Upper Saddle River NJ. PH, editor. Pearson/Prentice Hall; 2009.
24. Ridgers ND, McNarry MA, Mackintosh KA. Feasibility and Effectiveness of Using Wearable Activity Trackers in Youth: A Systematic Review. JMIR mHealth uHealth [Internet]. 2016 Nov 23 [cited 2020 Jul 7];4(4):e129. Available from: https://pubmed.ncbi.nlm.nih.gov/27881359/
25. Nightingale TE, Rouse PC, Thompson D, Bilzon JLJ. Measurement of Physical Activity and Energy Expenditure in Wheelchair Users: Methods, Considerations and Future Directions. Vol. 3, Sports Medicine–Open. Springer; 2017.


The kids practise on the slopes
Lean forward and make short and fast strokes.
If you can't make it, keep leaning forward and slowly let yourself roll backwards.
But in a panic, Sam suddenly grabs hold of his wheels
AAAAAH!!!
Don't brake too hard, Sam. You'll fall backwards.
And if you go up a slope, you also have to come down, of course.
Slowly roll down, let the handrims slide evenly, so you 'll stay in a straight line.
They then carry on with balancing, but now without a mat.
The kids quickly get the hang of it, and soon they're moving around the gym on their back wheels.

Chapter 6

The effects of wheelchair mobility skills and exercise training on physical activity, fitness, skills and confidence in youth using a manual wheelchair

Sol M.E.[1,2], Verschuren O.[2], Horemans H.[3], Westers P.[4,],Visser-Meily J.M.A.[2,5], Fit-for-the-Future Consortium, De Groot J.F.[1,6]

1. Research Group Lifestyle and Health , HU University of Applied Sciences Utrecht , Utrecht , The Netherlands.
2. Center of Excellence for Rehabilitation Medicine, UMC Utrecht Brain Center, University Medical Center Utrecht and De Hoogstraat Rehabilitation, Utrecht, The Netherlands.
3. Department of Rehabilitation Medicine, Erasmus MC University Medical Centre Rotterdam, PO Box 2040, 3000 CA Rotterdam, The Netherlands
4. Department of Biostatistics and Research Support, Julius Center, University Medical Center Utrecht, Utrecht, The Netherlands
5.Department of Rehabilitation, Physical Therapy Science & Sports, UMC Utrecht Brain Center, University Medical Center Utrecht, The Netherlands.
6. Knowledge Institute of the Federation of Medical Specialists, Utrecht, The Netherlands.

Dis Rehab, accepted

ABSTRACT

Purpose: To evaluate the effects of a combination of wheelchair mobility skills (WMS) training and exercise training on physical activity (PA), WMS, confidence in wheelchair mobility and physical fitness.

Methods: *Y*outh using a manual wheelchair (n=60) participated in this practice-based intervention, with a waiting list period (16wk), exercise training (8wk), WMS training (8wk) and follow-up (16wk). Repeated measures included: PA (Activ8), WMS (Utrecht Pediatric Wheelchair Mobility Skills Test), confidence in wheelchair mobility (Wheelchair Mobility Confidence scale) and physical fitness (cardiorespiratory fitness, (an)aerobic performance,) and were analysed per outcome parameter using a multilevel model analyses. Differences between the waiting list and training period were determined with an unpaired sample t-test.

Results: Multilevel model analysis showed significant positive effects for PA ($p = 0.01$), WMS ($p< 0.001$), confidence in wheelchair mobility ($p<0.001$), aerobic ($p<0.001$) and anaerobic performance ($p<0.001$). Unpaired sample t-tests underscored these effects for PA ($p<0.01$) and WMS ($p <0.001$). There were no effects on cardiorespiratory fitness. The order of training (exercise before WMS) had a significant effect on confidence in wheelchair mobility.

Conclusion: A combination of exercise and WMS training appears to have significant positive long term effects on PA, WMS, confidence in wheelchair mobility and (an)aerobic performance in youth using a manual wheelchair.

Keywords: Mobility, Children, Physical Behavior, Self-efficacy, Wheelchair Skills, Performance

INTRODUCTION

Youth with or without a disability benefit physically and mentally from a physical active lifestyle[1,2]. It has been shown that low levels of physical activity (PA) are more prevalent among youth (children and adolescents) with a physical disability compared to their typically developing peers[3,4]. Youth using a manual wheelchair are markedly less physically active than their ambulatory peers with or without a physical disability[5–8]. Bloemen et al.[5] found that youth using a manual wheelchair with Spina Bifida were 2.5 times less physically active than typically developing peers. These low levels of PA in youth using a wheelchair are worrisome and need attention.

Evidence for the effectiveness of interventions aimed at increasing PA in youth using a manual wheelchair is lacking, as there has been very limited research undertaken in this population[9]. Most intervention studies in youth with a physical disability focused on increasing PA in youth with cerebral palsy and were limited to participants who were able to stand or walk[8–11]. Recent systematic reviews[10,11] in youth with a physical disability concluded there was no or conflicting evidence on effectiveness of interventions aimed at increasing PA.

In this study we were interested in increasing the wheelchair propulsion time in youth using a manual wheelchair, as wheelchair propulsion is the largest component of PA in wheelchair users. We aimed to increase wheelchair propulsion time through improving three modifiable determinants of PA: wheelchair mobility skills (WMS), confidence in wheelchair mobility and physical fitness [12–14]. Two studies in children with a disability[15,16] showed high-intensity interval training (HIIT) to have positive results on physical fitness. High intensity interval training consist of intermittent bouts of activity and rest, which is similar to the active behaviour of youth[17]. Current literature in adult wheelchair users[18,19] shows that WMS and confidence in wheelchair mobility and physical fitness are modifiable factors, either through exercise training (physical fitness)[18] or WMS-training (WMS and confidence in wheelchair mobility)[19]. These studies have focused on the effect of one type of training, i.e. WMS training or exercise training, on respectively WMS and confidence in wheelchair mobility or physical fitness. Recently, Kirby et al.[20] suggested to focus on both WMS-training and exercise training during rehabilitation of people with a spinal cord injury, as there are significant positive relationships between WMS, confidence in wheelchair mobility and physical fitness. Whether the relationships and benefits of these training programs in adults are similar in youth using a manual wheelchair is unclear as there is barely any research in this population for WMS-training[21] and exercise training[9]. Moreover, and to the best of our knowledge, the effect of a combination of WMS-training and exercise training on PA is unknown in adults and youth using a manual wheelchair.

Therefore, the aim of this study was to: (1) study both the short term and long term effects of a combined WMS-training and exercise training on PA and modifiable determinants of PA, including WMS, confidence in wheelchair mobility and physical fitness in youth using a manual wheelchair and (2) explore differences in outcomes based on the order of training, i.e. WMS-training before or after exercise training.

METHODS

PARTICIPANTS

In this practice based study, a convenience sample of participants was recruited at six rehabilitation centres / schools for special education in the Netherlands. Participants were included if they were: between 7-18 years of age, bimanually propel their wheelchair, use wheelchair on a daily basis, able to understand the spoken Dutch language, able to understand simple instructions and had a problem related to WMS, physical fitness and/or PA. Participants were excluded if they had undergone a medical intervention during the previous six months that could have affected the intervention study outcomes (e.g. botox-injections). Local physiotherapists (PT), occupational therapists (OT) or physical education (PE) teachers approached and informed suitable participants and their parents.

PROCEDURE

This intervention study is part of the Let's Ride study from the Fit-for-the-Future consortium and registered at trialregister.nl, registration number NTR5791. The Institutional Review Board of the University Medical Center Utrecht approved the study protocol for the Let's Ride intervention study (protocol number15-136). Participants aged 12 years and over and all parents signed the informed consent form before enrolment in this study. Participants were placed in peer groups of four to seven participants per rehabilitation centre or school. Groups were allocated to one of the four programs in this study (figure 1). Program A and B were placed on the 16-week waiting list after the first assessment. This group was created due to limited facilities of running multiple groups simultaneously in a rehabilitation centre or school. Program C and D started training after the first assessment. All participants followed 8-weeks WMS training followed by 8-weeks exercise training (program A and C) or 8-weeks exercise training followed by 8-weeks WMS training (program B and D).Training programs were carried out during the school year and were not interrupted by school holidays that lasted more than one week. The group training sessions were given twice per week by two or three local PT, OT or PE teachers and each session lasted 30 minutes. The presence of the participant was documented in every session and participants were included in the analysis of the training results if they attended ≥ 10 sessions of WMS-training and ≥ 10 sessions of exercise training.

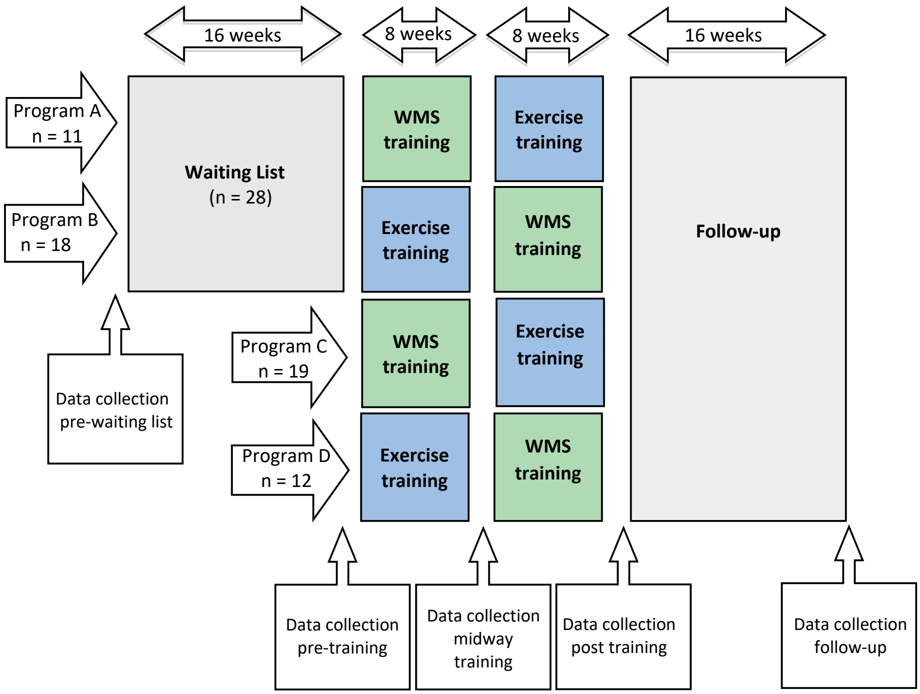


Figure 1. Study design

All participants had four assessments; pre-training, midway-training, post-training and follow-up. Participants in program A and B had one extra assessment before the waiting list period. All tests during an assessment period were administered within a two-week time frame at the participant's rehabilitation centre/school for special education in the gymnasium or laboratory under similar conditions (e.g. optimal tire pressure, floor).

INTERVENTION

WMS Training

The WMS-training program was developed by 'KJ Projects', a WMS school with over 10 years of experience in teaching WMS to both children and adults[22]. WMS sessions were aimed at learning to propel efficiently, going up and down curbs, holding a wheelie and negotiating the physical environment outdoors. Before starting the training sessions, the PT,OT and PE teachers received a three hour training by the research team and the KJ Projects. In combination with the two weekly training sessions by local PT, OT or PE teachers, there were three training sessions at the start, in the middle and at the end of the WMS-training program given by KJ projects. Parents and/or close relatives observed and participated in these three training sessions to stimulate and motivate the participants to practice at home. In addition, all participants received an individual video instruction on how to practice their skills at home.

Exercise training

A HIIT protocol was chosen as a form of exercise training, as this form of training is a time-effective method of improving physical fitness in youth[23]. This training protocol has recently been used in a study with children with a disability, including a small sample of wheelchair users[15]. Results showed a significant improvement in anaerobic performance and aerobic performance after HIIT training[15]. The used HIIT protocol in our study, number of series (8–12 series) and active recovery time (90-120 seconds), is described by Zwinkels et al.[15] for children with a disability. All participants performed a HIIT in their own wheelchair, aimed at improving their physical fitness (aerobic performance, cardiorespiratory fitness and anaerobic performance) through series of 30-s all-out exercises. The 30-s all out exercises were easily executable sprint exercises such as, go back and forward between 2 cones. The group training sessions were given in the school's gym by two or three PT,OT or PE teachers, who had received a training on the intervention by the research team.

DEMOGRAPHIC AND MORPHOLOGIC PARAMETERS

Parents of the participant completed one general questionnaire at the first assessment regarding age, diagnosis, functional mobility level[24], type of wheelchair and level of education of their child. They also completed a short questionnaire at the other assessments about possible factors that could influence the participants performance (e.g. illness or injuries). At each assessment body mass and wheelchair mass was determined using a calibrated (wheelchair) scales (Kern MWS 300K100M) from the local rehabilitation centre/ school for special education. Height was assessed in supine position with an non-stretchable tape from head to heel or with arm span (fingertip to fingertip, with arms abducted 90°and elbow and wrists straight) when participants were unable to fully extent their hips or knees due to contractures[25].

OUTCOME MEASURES

Physical Activity: active wheelchair use

An objective assessment of time spent in 'active wheelchair use' was measured with a small accelerometer, the Activ8 activity monitor (2M Engineering BV, Valkenswaard, the Netherlands)[26]. The Activ8 has shown to be valid for distinguishing the classification 'active wheelchair use' from the classification 'non-propulsive wheelchair use'[27]. The counts thresholds used to separate active from non-propulsive wheelchair use were modified for youth using a manual wheelchair based on earlier pilot data. For the purpose of this study, active wheelchair use is defined as independent wheelchair propulsion at normal speed, high speed or manoeuvring. Participants were asked to wear an Activ8 in a stretchable wristband on the dorsal side of the wrist on their dominant arm for seven consecutive days. A second Activ8 was securely fastened as close as possible to the axle of the wheel on the same side as the dominant arm. The use of this combination of accelerometers makes it possible to distinguish active propulsion from assisted driving. The total amount of PA consists of more than active wheelchair use, including activities such as swimming and transfers in/out of wheelchair. However, the aim of this intervention is to increase the active propulsion of the everyday wheelchair and the Activ8 is a valid device for detecting this component of PA[27]. Only data of participants with a minimal wear time of eight hours/day on at least two school-days and one weekend-day were analysed[28]. The main outcome for PA is the amount of 'active wheelchair use' expressed as a percentage of wear time per day.

Wheelchair mobility skills

The WMS were assessed using the recently developed Utrecht Pediatric Wheelchair Mobility Skills Test 2.0 (UP-WMST 2.0)[29,30]. The UP-WMST 2.0 is a performance-based measure of WMS with good validity and test-retest reliability [29]. It consists of 15 items measuring different WMS such as propelling forwards/backwards, turning, ascending a platform and holding a wheelie. The UP-WMST 2.0 was administered by a PT, OT or PE teacher, who had received a two hour training (theory and practice) on how to administer the UP-WMST 2.0 in youth using a manual wheelchair. The main outcome is a total score ranging from 0 to 51, with a higher score representing more advanced wheelchair mobility skills.

Confidence in wheelchair mobility

For confidence in wheelchair mobility, the recently developed and validated Wheelchair Mobility Confidence Scale (WheelCon-Mobility) for Dutch Youth was used[31]. In this questionnaire participants are asked to rate on a 5 point Likert scale how confident they feel about doing different wheelchair mobility activities independently and safely, such as manoeuvring your wheelchair in small spaces or going up and down a curb. Participants were asked to complete the questionnaire independently or with the help of a research team member when they were unable to read the questions themselves. The main outcome of this 31-item questionnaire is a total score ranging from 31 to 155, with a higher score indicating a higher confidence in wheelchair mobility.

Physical fitness

Aerobic fitness

The Shuttle Ride Test (SRiT) is a maximal graded aerobic exercise field test where participants propel their wheelchair back and forth over a distance of 10 meters with increasing speed. This test has shown to be reliable and valid in youth (cerebral palsy, Spina Bifida and osteogenesis imperfecta) using a manual wheelchair[32–34]. The SRiT was administered by two experienced researchers following the protocol of Verschuren et al.[33]. The VO_2 (ml/min) during the SRiT was recorded with a calibrated mobile gas analysis system (Cortex Metamax B3; Cortex Medical GmbH, Leipzig, Germany). The relative

VO_{2peak} (ml/min/kg) was determined as the highest value of VO_2 (ml/min) during the last 30 seconds of the SRT, divided by the body mass (kg). The researcher used subjective criteria (lack of motivation, pain, distraction) to determine if the participant had shown real maximal effort to achieve the highest number of shuttles. The main outcome of the SRiT is the number of shuttles (ranging from 0.5-23) as a measure of aerobic performance and the relative VO_{2peak} (ml/min/kg,) as a measure of cardiorespiratory fitness. Data were excluded from analysis for 'number of shuttles' and 'relative VO_{2peak}' when maximal effort was not achieved.

Anaerobic performance

The Muscle Power Sprint Test (MPST) is an anaerobic performance test in youth using a manual wheelchair, where participants propel their wheelchair six times at maximal speed over a distance of 15 meters with a break of 10 seconds between each sprint to turn around and get ready for the next sprint. This test has shown to be reliable and valid in youth using a manual wheelchair[35,36]. The MPST was administered by a member of the research team who recorded the time per sprint. Afterwards the time per sprint was converted to power as a measure of anaerobic performance.

Power = (total mass x distance^2)/time^3.

Total mass is calculated as body mass plus wheelchair mass. Participants with power assisted wheels (Ewheels) were excluded from analysis. The main outcome of the MPST is the mean and peak power of the six sprints.

DATA ANALYSES

Statistical analysis was performed using SPSS for Windows (version 25.0, SPSS Inc., Chicago, IL, USA). Characteristics of the participants, such as gender, diagnosis and functional mobility level are described categorically and the participants age, height, weight and years of experience in wheelchair use are presented as a mean with the standard deviation for the total group (Program A-D), waiting list group (Program A,B) and split for order of training (Program A,C or Program B,D). Data was checked for normality and characteristics and baseline scores were compared between the waiting list group (Program A,B) and total group (Program A–D) and between the orders of training (Program A and C or Program B and D) using chi-square test or independent sample t-tests.

Aim (1): The short term and long term effect of combined WMS and exercise training per outcome parameter was calculated using a multilevel model analyses with a random intercept for participant and time (pre-training, post-training and follow-up) as a categorical factor. The pre-waiting list assessment was not added as a time value in the model, as we did not expect different outcomes for participants who were placed on the waiting list before commencing the combined training program. As a secondary analysis, we performed unpaired sample t-tests for all outcome measures to determine if the change in outcomes after the training period (post-training minus pre-training of program A–D) were significantly different from the normal variation in outcomes during the waiting list period (pre-training minus pre-waiting list of program A and B). Due to the heterogeneity of this population, analyses in the total sample using an unpaired sample t-test (program A-D) was the preferred method over a paired t-test in a half of the sample (program A and B). Due to the use of multiple testing we have set the significance level at $p<0.01$ to prevent type-I errors. Cohen's D effect size was calculated for the unpaired sample t-tests with effect sizes classified as small (d=0.2), medium (d=0.5), large (d=0.8) and very large (d=1.3)[37].

Aim (2) The order of training (WMS training before or after exercise training) was added to the multilevel model analyses per outcome parameter to determine if program A and C had a significant different effect from program B and D.

RESULTS

A total of 60 youth using a manual wheelchair in daily life participated in this study. For the effectiveness of the intervention, data of 12 participants was discarded. They could either not continue training due to unrelated medical problems (n=3), left school during the training period (n=1), declined to participate (n=2) or did not attend ≥ 10 trainings sessions per type of training (n=6). From the remaining group of 48 participants, the mean adherence for exercise training was 14.2 (standard deviation (SD) 1.6) out of the 16 training sessions and the mean adherence for WMS training was 14.3 (SD 1.7) out of the 16 training sessions. The characteristics of the total group (program A–D) are described in Table 1 and subsequently split for waiting list group (program A,B), WMS before Exercise training (program A,C) and Exercise before WMS training (program B,D). There were no significant differences in characteristics or baseline scores between the waiting list group and total group (Table 1, Figure 2). There were no significant difference in characteristics or pre-training scores between the orders of training, except for more years of experience in wheelchair use in program A and C compared to program B and D (Table 1).

Effects of combined WMS-training and exercise training on PA

The median amount of days with sufficient wear time of the Activ8 for pre-training, post-training and follow-up are respectively: 6 days, 7 days, and 6 days. Missing data per measurement instrument is reported in Appendix A. No significant differences were

Table 1. Characteristics of participants

			Split per order of training	
	Total group Program A-D (n = 48)	**Waiting list group Program A & B (n= 24)**	**WMS before Exercise Program A & C (n = 23)**	**Exercise before WMS Program B & D (n = 25)**
Gender(M/F)	28/20	16/8	12/11	16/9
Age (years) mean (SD)	12.8 (3.1)	14.0 (3.3)	12.9 (3.4)	12.9 (3.0)
Diagnosis				
Cerebral palsy	21	11	9	12
Spina bifida	8	6	6	2
Neuromuscular	5	1	2	3
Other	14	6	6	8
Height (cm) mean (SD)	149 (16)	153.9 (14.0)	145.9 (18.1)	151.5 (14.1)
Weight (kg) mean (SD)	44.3 (16.3)	50.5 (15.2)	41.1 (17.7)	46.9 (15.0)
Wheelchair mass* (kg)	19.4 (3.7)	20 (3.9)	18.6 (3.7)	20.0 (3.8)
Power assisted wheels	7	3	5	2
Experience in wheelchair (years) mean (SD)	8.4 (3.8)	9.3 (4.0)	9.6 (3.8)**	7.2 (3.6)**
Ambulation level				
Non ambulatory	28	15	15	13
Partly ambulatory	20	9	8	12
Level of education				
Regular	30	15	13	9
Special	18	9	10	16

* Power assisted wheels not included, ** statistical difference between program A&C and B&D, M = male, F = Female, n = number of participants, WMS = Wheelchair mobility Skills, SD = Standard deviation

found for baseline characteristics of participants with missing data of the Activ8 at pre-training, post-training and follow-up, except for gender at pre-training (Appendix A).

For the total group, the combined intervention of WMS and exercise training had a significant positive effect ($p = 0.01$) on physical activity as measured with the Activ8 (pre-training 6.5% (standard Error (SE) 2.4), post-training 8.1% (SE 3.2) and follow-up 7.5% (SE 2.5)) (Table 2). Post-hoc analysis showed that the short term effect was an absolute average increase from 53 minutes/day before training to 66 minutes/day after the combined training. Looking at the follow up data, there was a sustained improvement with no significant ($p=0.73$, 95% $CI^{-0.7,1.1}$) differences at follow-up in 'active wheelchair use' per day.

Table 2. Short and long term effects per outcome parameter of combined exercise and wheelchair mobility skills training using a multilevel model analyses

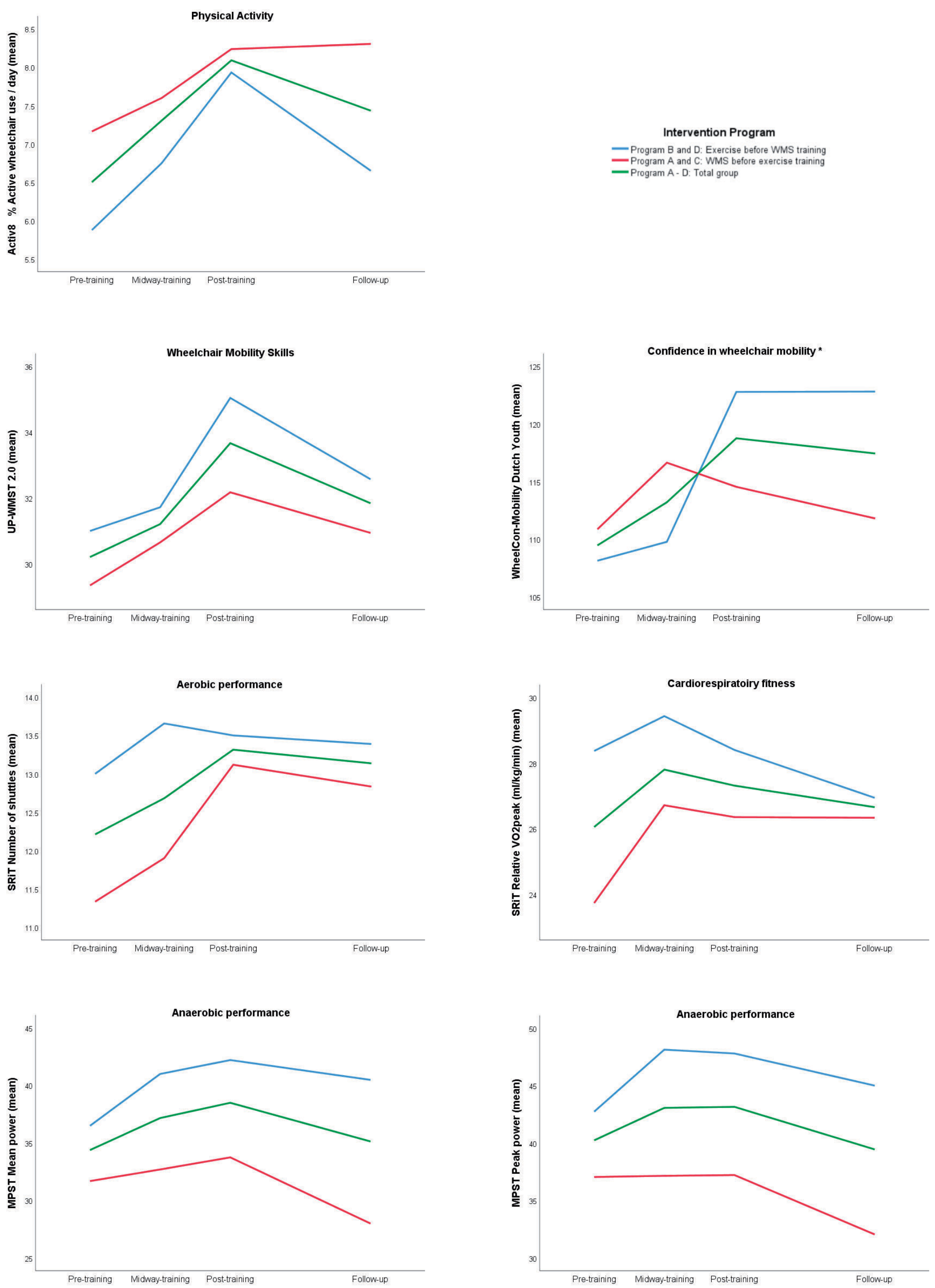
	Pre-training		Post-training		Follow-up		p-value overall effect	Post vs Pre-training		Post-training vs Follow-up	
Outcome measure	**N**	**Mean (SE)**	**N**	**Mean (SE)**	**N**	**Mean (SE)**		**(95 % CI)**	**P-value**	**(95 % CI)**	**P-value**
Physical activity											
Active wheelchair use per day (% wear time)	35	6.5 (2.4)	27	8.1 (3.2)	19	7.5 (2.5)	**0.01**	(0.4;2.0)	**<0.01**	(-0.7;1.1)	0.73
Wheelchair Mobility Skills											
UP-WMST 2.0	48	30.2 (10.7)	48	33.7 (10.1)	37	32.0 (10.0)	**<0.001**	(2.4;4.5)	**<0.001**	(-1.1;1.1)	0.99
Confidence in wheelchair mobility											
WheelCon-Mobility Dutch Youth	45	109.5 (23.4)	45	118.8 (22.0)	41	117.4 (24.0)	**<0.001**	(4.5;14.0)	**<0.001**	(-5.4;4.5)	0.85
Physical fitness											
Shuttle Ride Test											
Number of shuttles	38	12.2 (3.6)	35	13.3 (3.7)	33	13.1 (2.9)	**<0.001**	(0.6;1.6)	**<0.001**	(-0.8;0.2)	0.24
Relative VO_{2peak} (ml/kg/min)	38	26.1 (9.2)	32	27.3 (8.5)	32	26.7 (8.9)	0.71	(-1.4;1.9)	0.77	(-2.1;1.4)	0.67
Muscle Power Sprint Test											
Mean power (W)	41	34.4 (27.8)	41	38.5 (28.8)	35	35.1 (21.2)	**<0.001**	(2.1;6.2)	**<0.001**	(-4.3;0.2)	0.07
Peak Power (W)	41	40.2 (33.0)	41	43.2 (32.0)	35	39.5 (23.4)	**<0.001**	(0.4;5.4)	0.02	(-5.3;0.1)	0.05

UP-WMST = Utrecht Pediatric Wheelchair Mobility Skills test, WheelCon-Mobility = Wheelchair mobility confidence scale for Dutch Youth. W = Watt, CI = Confidence Interval, SE = Standard Error N = number of participants

Table 3. Unpaired sample t-test total group and waiting list group

	Waiting list period Program A and B (n=24)			Training period Program A – D (n=48)			Unpaired sample t-test p-value	Cohen's D
	n	**Pre-waiting list mean (SD)**	**Mean change* (SD)**	**n**	**Pre-training mean (SD)**	**Mean change* (SD)**		
Active wheelchair use (% wear time)	14	6.5 (1.8)	-1.1 (1.2)	21	6.3 (2.4)	1.1 (2.1)	**<0.01**	1.2
UP-WMST 2.0	24	32.6 (10.4)	0.3 (3.7)	48	30.2 (10.7)	3.5 (3.6)	**0.001**	0.88
WheelCon-mobility Dutch Youth	23	106.7 (23.6)	0.0 (14.8)	45	109.5 (23.4)	9.3 (17.4)	0.03	0.56
Shuttle Ride Test								
Number of shuttles	18	12.5 (3.1)	0.4 (1.7)	28	13.0 (2.7)	1.2 (1.2)	0.07	0.57
Relative VO_{2peak} (ml/kg/min)	18	24.6 (6.9)	3.9 (4.0)	28	27.2 (9.0)	0.4 (5.8)	0.03	-0.68
Muscle Power Sprint Test								
Mean power (W)	20	33.9 (18.9)	2.6 (4.8)	41	34.4 (27.8)	4.1 (6.5)	0.37	0.25
Peak Power (W)	20	38.3 (21.2)	3.9 (5.9)	41	40.2 (32.3)	2.9 (8.7)	0.62	-0.12

* Mean change = post- training or waiting list period minus respectively pre- training or waiting list period. There are no significant differences between pre-waiting list period and pre-training period scores. UP-WMST = Utrecht Pediatric Wheelchair mobility Skills test, WheelCon-Mobility Dutch youth = Wheelchair Confidence scale for Dutch Youth, SD = Standard Deviation, W = Watt.



*Significant difference in outcome for order of training. UP-WMST = Utrecht Pediatric Wheelchair Mobility Skills Test, SRiT = Shuttle Ride Test, MPST = Muscle Power Sprint Test

Figure 2. Line graph of the change in mean score over time for the total group (green), program A and C (red) and program B and D (blue) per outcome parameter.

Unpaired sample t-test (see Table 3) showed a large effect (*d*=1.2), with an increase in 'active wheelchair use' of 1.1 % (SD 2.1%) (n=21) after the training period that was significantly different (p<0.01) from the decline of 1.1 (SD 1.2%) that occurred during the waiting list period (n=14).

Effects of combined WMS-training and exercise training on determinants of PA

The WMS, confidence in wheelchair mobility and physical fitness, with the exception of relative VO_{2peak}, improved significantly over time (pre-training, post-training and follow-up) in the total group (Table 2, figure 2). Missing data per assessment and parameter is reported in Appendix A.

When looking at differences in change with the unpaired sample t-test during the training period compared to the waiting list period, there was a significant change of 3.5 (SD 3.6) points in WMS after the training period compared to a change of 0.3 (SD 3.7) after the waiting list period. We found a non-significant difference between the waiting list period and training period for confidence in wheelchair mobility (p = 0.03), number of shuttles (p=0.07), relative VO_{2peak} (p = 0.03), mean power (p=0.37) and peak power (p=0.62).

Effect of the order of WMS and exercise training

We found a significant effect of order of training when added to the multilevel model for confidence in wheelchair mobility (p=0.01) (Figure 2). We found no significant effect of order of the training for all the other outcomes.

DISCUSSION

The aim of this intervention in youth using a manual wheelchair was to evaluate the short term and long term effect of a combined WMS-training and exercise training on PA and three determinants of PA: WMS, confidence in wheelchair mobility and physical fitness. In this study we found increasing levels of PA and positive changes in determinants of PA after the combined training program, which were maintained at follow-up, with the exception of cardiorespiratory fitness.

Despite the heterogeneity of the participants in this practice based intervention study, positive results in increased PA over time are supported by a significant difference, with a large effect size, in change of PA during the training period compared to the waiting list period. The increase of 13 minutes per day is a relative increase of 25% in time spent physically active per day. This is an important and clinically relevant increase, as any improvement of PA can lead to numerous health benefits[1]. Moreover, greater health benefits can be achieved by people who have an inactive lifestyle[1]. The results of this study also show that youth using a manual wheelchair are very inactive with 53 minutes/day of active wheelchair use before commencing the combined training programs.

For the assessment of PA we were able to assess the largest component of PA for wheelchair users, which is the percentage of time spent actively propelling their manual wheelchair per day. While it would be preferable to assess the total amount of PA, at the start of this study in 2015, the Activ8 was the best available activity monitor for wheelchair users, which did appear to be user friendly[27]. In this study, we did experience some technological challenges in the use of the Activ8. Even though the soft stretchable armband did appear to be child friendly, participants reported more and more a dislike to wear the armband due to an itchy feeling or esthetical reasons. This led to decreased willingness to wear to monitor for multiple days and resulted in more missing data of the Activ8 in the final assessments, i.e. post-training and follow-up. Consequently, the results reported for PA are collected in a smaller sample. Even with this small heterogeneous sample, significant sustainable changes were seen after the 16 week combined training program.

When looking at the determinants of PA, we aimed to improve three determinants through a combined WMS-training and exercise training. We found a significant improvement in WMS of 7.1% (3.5 points) on the UP-WMST 2.0, which was maintained at follow-up. This improvement in WMS is smaller than the effects of WMS training in adults (14.0%, 95% CI[7.4, 20.8])[19]. Subgroup analysis in the meta-analysis of Keeler et al.[19] showed that training was more effective in new wheelchair users. Youth using a manual wheelchair in this study had an average of 8 years of experience in using a wheelchair, which could explain the smaller effects on WMS. Nonetheless, small improvements in WMS, such as being able to go up a curb (+1 point), could already lead to more independence outdoors and have a positive effect on PA. And more importantly, there is no significant decline in training effects between post-training and at 16-weeks of follow-up as is similar in adult wheelchair users[19], which implies WMS were maintained during the follow up period.

For confidence in wheelchair mobility we found a significant increase in the WheelCon-Mobility for Dutch youth[31] after the combined training. The growth in confidence levels was mainly gained after WMS training (figure 2), with a significantly larger increase in the group that started with exercise training (Program B and D). This could be explained by Bandura's social cognitive theory[38], were the experience of mastering of a new skill, i.e. WMS, is the most effective way of improving ones confidence. Participants in program B and D mastered on average more WMS skills, possibly leading to a bigger increase in confidence compared to participants in program A and C. These results suggest that exercise before WMS training might be the preferred order of training when aiming to improve confidence in wheelchair mobility.

The short term effect of the combined training on anaerobic performance was a significant increase in mean (+4.1 Watt) and peak power (+3.0 Watt). In the longer term, we found a non-significant decline between post-training and follow-up, which was especially prominent in program A and C. This trend for a decline could be explained by one of the limitations of this study, where we were unable to assess participants who had graduated from school (n=4) and left the program between post-training and follow-up. This led to missing data not ad random, but with missing data of older and heavier participants, leaving younger and therefore lighter participants at follow-up assessment. For the power calculations in the MPST the total weight of the participant is an important factor for the outcome. The four participants who had left school all had a mean power at post-training that was two to four times higher than the average mean power of the total group. Secondary analysed without these four participants showed a smaller decline in anaerobic performance at follow-up.

For aerobic fitness, we found similar positive results as Zwinkels et al.[15], with a significant increase of shuttles on the SRiT (+1 shuttle) as a measure of aerobic performance and no change in relative VO_{2peak} as a measure of cardiorespiratory fitness. Surprisingly, we did find a non-significant positive effect on relative VO_{2peak} after the waiting list period. It is unclear what caused these results during the waiting list period. Possibly, there were seasonal effects (pre-waiting list measurement was assessed at the start of school year) that might have influenced the cardiorespiratory fitness during the waiting list period. For the results of the combined training we found an increase in shuttles with similar O_2 uptake, which implies more efficient propulsion after the combined training and at follow-up. These results are in line with a functional exercise study in ambulatory children with Spina Bifida[39]. The increase in efficiency may be explained by the fact that the training intensity was limited by the propelling capacities of the child rather than the cardiorespiratory limitation. Recently, Zwinkels et al.[15] also concluded that the HIIT training protocol used is not an effective form of exercise training to increase cardiorespiratory fitness in youth who use a wheelchair. It is possible that reaching high intensities in 30 second exercise bouts is less attainable through wheelchair propulsion due smaller active muscle mass than running based exercises[40].

One of the limitations of this study was the 'one size fits all' training approach, where all participants received a WMS-training and exercise training, regardless of the large variation at baseline in PA and the determinants of PA. In figure two, the changes in scores over time are visually presented per order of training, and show that the greatest improvement for PA, confidence in wheelchair mobility and aerobic performance are in the groups that had lower scores at pre-training. It is possible that participants with a relatively high physical fitness or more advanced WMS did not benefit from the exercise or WMS-training as much as the participants with a relatively low physical fitness or more basic WMS. At the same time, HIIT training did challenge each child to exercise at his/her own maximal level during the high intensity intervals. The small training groups with peers seemed motivating to go as fast as possible during exercise training and encouraging to learn new WMS from seeing their peers mastering a new skill.

While the obvious strength of this study was the practice based approach, with outcomes measures that can be applied in clinical practise and a heterogeneous sample that is representative for this population, this type of study does come with methodological imitations. The heterogeneity of the sample makes it is more difficult to show significant results due to the large confidence intervals. Even so, this study showed positive results which are immediately relevant for daily practice in schools for special education or rehabilitation centres. The intervention in a school setting has disadvantages, such as limited time for assessments and drop-outs due to graduation, which leads to missing data that potentially could have confounded the results of this study. Future research towards PA in school settings should also take into account possible seasonal variations due to the school program, e.g. decreased or increased PA over the summer holidays. A benefit of the practice based design was the involvement of PT, OT and PE teachers in assessments and interventions. This will help future implementation of study results, which has already happened in the rehabilitation centres in the Netherlands that were involved in this study.

CONCLUSION

A combination of exercise and WMS training appears to have a clinically relevant and significant increase in PA in youth using a manual wheelchair. The combined training also had a positive effect on WMS, confidence in wheelchair mobility, aerobic performance and anaerobic performance. More insight is needed towards finding an effective form of exercise training for improving cardiorespiratory fitness in youth using a manual wheelchair. Exercise before WMS training is the preferred order of training, when aiming to improve confidence in wheelchair mobility.

REFERENCES

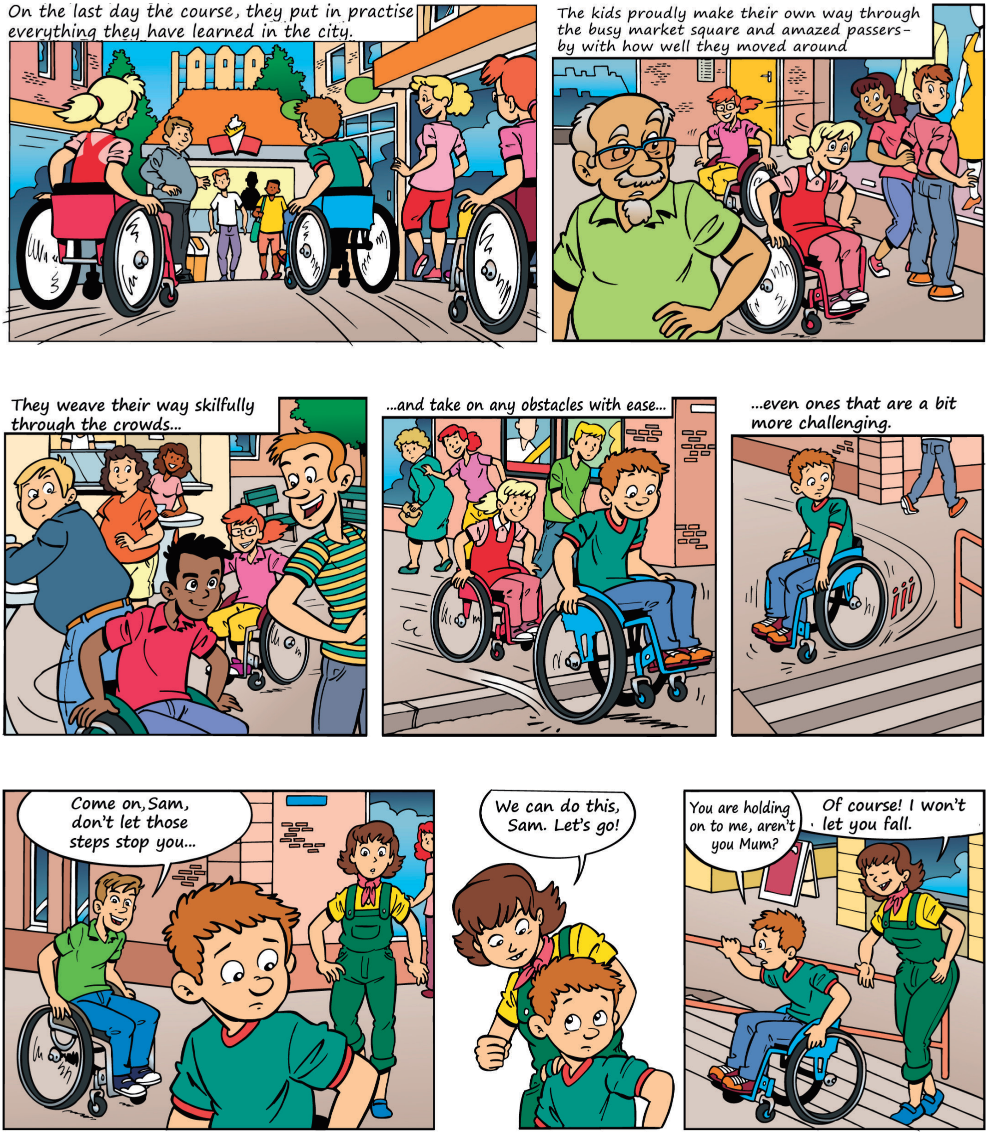
1. Weggemans RM, Backx FJG, Borghouts L, Chinapaw M, Hopman MTE, Koster A, et al. The 2017 Dutch Physical Activity Guidelines. Int J Behav Nutr Phys Act. 2018 Jun 25;15(1).
2. Warburton DER, Nicol CW, Bredin SSD. Health benefits of physical activity: The evidence. Vol. 174, CMAJ. Canadian Medical Association; 2006. p. 801–9.
3. Rimmer JA, Rowland JL. Physical activity for youth with disabilities: a critical need in an underserved population. Dev Neurorehabil [Internet]. 2008 [cited 2020 Apr 9];11(2):141–8. Available from: http://www.ncbi.nlm.nih.gov/pubmed/18415819
4. Burghard M, de Jong NB, Vlieger S, Takken T. 2017 Dutch Report Card+: Results from the first physical activity report card plus for Dutch youth with a chronic disease or disability. Front Pediatr [Internet]. 2018 [cited 2020 Apr 10];6:122. Available from: http://www.ncbi.nlm.nih.gov/pubmed/29761094
5. Bloemen MAT, van den Berg-Emons RJG, Tuijt M, Nooijen CFJ, Takken T, Backx FJG, et al. Physical activity in wheelchair-using youth with Spina Bifida: an observational study. J Neuroeng Rehabil [Internet]. 2019 Dec 14 [cited 2020 Apr 14];16(1):9. Available from: https://jneuroengrehab.biomedcentral.com/articles/10.1186/s12984-018-0464-x
6. Claridge EA, Bloemen MAT, Rook RA, Obeid J, Timmons BW, Takken T, et al. Physical activity and sedentary behaviour in children with Spina Bifida. Dev Med Child Neurol. 2019 Dec 1;61(12):1400–7.
7. Van Den Berg-Emons RJ, Bussmann JB, Stam HJ. Accelerometry-based activity spectrum in persons with chronic physical conditions. Arch Phys Med Rehabil [Internet]. 2010 Dec [cited 2020 Apr 9];91(12):1856–61. Available from: http://www.ncbi.nlm.nih.gov/pubmed/21112426
8. Ganz F, Hammam N, Pritchard L. Sedentary behavior and children with physical disabilities: a scoping review [Internet]. Disability and Rehabilitation. Taylor and Francis Ltd; 2020 [cited 2020 Apr 9]. p. 1–13. Available from: http://www.ncbi.nlm.nih.gov/pubmed/32116041
9. O'Brien TD, Noyes J, Spencer LH, Kubis H-P, Hastings RP, Whitaker R. Systematic review of physical activity and exercise interventions to improve health, fitness and well-being of children and young people who use wheelchairs. BMJ Open Sport Exerc Med [Internet]. 2016 Nov [cited 2020 Apr 10];2(1):e000109. Available from: http://www.ncbi.nlm.nih.gov/pubmed/27900176
10. 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 [Internet]. Vol. 59, Developmental Medicine and Child Neurology. Blackwell Publishing Ltd; 2017 [cited 2020 Apr 14]. p. 1004–10. Available from: http://www.ncbi.nlm.nih.gov/pubmed/28374442
11. Reedman S, Boyd RN, Sakzewski L. The efficacy of interventions to increase physical activity participation of children with cerebral palsy: a systematic review and meta-analysis. Dev Med Child Neurol [Internet]. 2017 Oct 1 [cited 2020 Apr 9];59(10):1011–8. Available from: http://www.ncbi.nlm.nih.gov/pubmed/28318009
12. Van Der Ploeg HP, Van Der Beek AJ, Van Der Woude LHV, Van Mechelen W. Physical activity for people with a disability: A conceptual model. Vol. 34, Sports Medicine. 2004. p. 639–49.
13. Bloemen MAT, 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 [Internet]. 2015 Feb 10 [cited 2020 Apr 9];15(1):11. Available from: http://www.ncbi.nlm.nih.gov/pubmed/25886148
14. Shields N, Synnot A. Perceived barriers and facilitators to participation in physical activity for children with disability: a qualitative study. BMC Pediatr [Internet]. 2016 Jan 19 [cited 2020 Apr 9];16(1):9. Available from: http://www.ncbi.nlm.nih.gov/pubmed/26786677
15. Zwinkels M, Verschuren O, de Groot JF, Backx FJG, Wittink H, Visser-Meily A, et al. Effects of High-Intensity Interval Training on Fitness and Health in Youth With Physical Disabilities. Pediatr Phys Ther [Internet]. 2019 Jan 1 [cited 2020 Apr 9];31(1):84–93. Available from: http://www.ncbi.nlm.nih.gov/pubmed/30507854
16. Lauglo R, Vik T, Lamvik T, Stensvold D, Finbråten A-K, Moholdt T. High-intensity interval training to improve fitness in children with cerebral palsy. BMJ Open Sport Exerc Med [Internet]. 2016 May 1 [cited 2021 Jan 19];2(1):e000111. Available from: http://dx.doi.org/10.1136/bmjsem-2016-000111
17. Bailey RC, Olson J, Pepper SL, Porszasz J, Barstow TJ, Cooper DM. The level and tempo of children's physical activities: An observational study. Med Sci Sports Exerc [Internet]. 1995 [cited 2020 Sep 8];27(7):1033–41. Available from: https://pubmed.ncbi.nlm.nih.gov/7564970/
18. Hicks AL, Martin Ginis KA, Pelletier CA, Ditor DS, Foulon B, Wolfe DL. The effects of exercise training on physical capacity, strength, body composition and functional performance among adults with spinal cord injury: A systematic review. Spinal Cord. 2011 Nov;49(11):1103–27.
19. Keeler L, Kirby RL, Parker K, McLean KD, Hayden JA. Effectiveness of the Wheelchair Skills Training Program: a systematic review and meta-analysis * [Internet]. Vol. 14, Disability and Rehabilitation: Assistive Technology. Taylor and Francis Ltd; 2019 [cited 2020 Apr 28]. p. 391–409. Available from: http://www.ncbi.nlm.nih.gov/pubmed/29616832
20. Kirby RL, de Groot S, Cowan RE. Relationship between wheelchair skills scores and peak aerobic exercise capacity of manual wheelchair users with spinal cord injury: a cross-sectional study. Disabil Rehabil. 2020 Jan 2;42(1):114–21.
21. Sawatzky B, Rushton PW, Denison I, McDonald R. Wheelchair skills training programme for children: A pilot study. Aust Occup Ther J [Internet]. 2012 Feb [cited 2020 Apr 9];59(1):2–9. Available from: http://www.ncbi.nlm.nih.gov/pubmed/22272877
22. https://wheelchairskillsteam.nl/.
23. Eddolls WTB, McNarry MA, Stratton G, Winn CON, Mackintosh KA. High-Intensity Interval Training Interventions in Children and Adolescents: A Systematic Review. Sports Med [Internet]. 2017 Nov 1 [cited 2020 Apr 9];47(11):2363–74. Available from: http://www.ncbi.nlm.nih.gov/pubmed/28643209
24. Graham HK, Harvey A, Rodda J, Nattrass GR, Pirpiris M. The Functional Mobility Scale (FMS). J Pediatr Orthop. 2004;24(5):514–20.
25. Chen WY, Lin YT, Chen Y, Chen KC, Kuo BIT, Tsao PC, et al. Reference equations for predicting standing height of children by using arm span or forearm length as an index. J Chinese Med Assoc. 2018 Jul 1;81(7):649–56.
26. Horemans H, Kooijmans H, van den Berg-Emons R, Bussmann H. The Activ8 activity monitor: Validation of posture and movement classification. J Rehabil Assist Technol Eng [Internet]. 2020

Jan [cited 2020 May 6];7:205566831989053. Available from: http://www.ncbi.nlm.nih.gov/pubmed/32206335

27. Leving MT, Horemans HLD, Vegter RJK, De Groot S, Bussmann JBJ, van der Woude LHV. Validity of consumer-grade activity monitor to identify manual wheelchair propulsion in standardized activities of daily living. PLoS One. 2018 Apr 1;13(4).
28. Claridge EA, Bloemen MAT, Rook RA, Obeid J, Timmons BW, Takken T, et al. Physical activity and sedentary behaviour in children with Spina Bifida. Dev Med Child Neurol [Internet]. 2019 Dec 1 [cited 2020 Apr 9];61(12):1400–7. Available from: http://www.ncbi.nlm.nih.gov/pubmed/31468507
29. Sol ME, de Groot JF, Zwinkels M, Visser-Meily JMA, Kruitwagen CLJJ, Verschuren O. Utrecht Pediatric Wheelchair Mobility Skills Test: Reliability, Validity, and Responsiveness in Youths Using a Manual Wheelchair. Phys Ther [Internet]. 2019 [cited 2020 Apr 9];99(8):1098–106. Available from: http://www.ncbi.nlm.nih.gov/pubmed/30939199
30. Sol ME, Verschuren O, de Groot L, de Groot JF, Fit-For-the-Future!-consortium. Development of a wheelchair mobility skills test for children and adolescents: combining evidence with clinical expertise. BMC Pediatr [Internet]. 2017 Feb 13 [cited 2020 Apr 9];17(1):51. Available from: http://www.ncbi.nlm.nih.gov/pubmed/28193204
31. Sol ME, Bloemen MAT, Verschuren O, Rushton PW, Visser-Meily JMA, de Groot JF. Wheelchair Mobility Confidence Scale for Dutch youth using a manual wheelchair. Disabil Rehabil Assist Technol [Internet]. 2019 Apr 23 [cited 2020 Apr 9];1–8. Available from: http://www.ncbi.nlm.nih.gov/pubmed/31012747
32. Bloemen MAT, de Groot JF, Backx FJG, Benner J, Kruitwagen CLJJ, Takken T. Wheelchair Shuttle Test for Assessing Aerobic Fitness in Youth With Spina Bifida: Validity and Reliability. Phys Ther [Internet]. 2017 [cited 2020 Apr 9];97(10):1020–9. Available from: http://www.ncbi.nlm.nih.gov/pubmed/29029556
33. 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 Jul 1;93(7):967–74.
34. Bongers BC, Rijks EBG, Harsevoort AGJ, 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 May 1;96(5):679–86.
35. 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 [Internet]. 2017 Jun 1 [cited 2020 Apr 9];98(6):1097–103. Available from: http://www.ncbi.nlm.nih.gov/pubmed/27633940
36. 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 Dec;55(12):1129–35.
37. Sullivan GM, Feinn R. Using Effect Size-or Why the P Value Is Not Enough. Available from: http://dx.doi.org/10.4300/JGME-D-12-00156.1

38. Bandura A, Freeman W, Lightsey R. Self-efficacy: The exercise of control [Internet]. New York; 1999 [cited 2020 Jun 9]. Available from: https://connect.springerpub.com/content/sgrjcp/13/2/158.full.pdf
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 [Internet]. 2011 Sep [cited 2020 Jun 26];25(7):597–606. Available from: https://pubmed.ncbi.nlm.nih.gov/21415263/
40. Raymond J, Davis GM, Fahey A, Climstein M, Sutton JR. Oxygen uptake and heart rate responses during arm vs combined arm/ electrically stimulated leg exercise in people with paraplegia. Spinal Cord. 1997;35(10):680–5.


On the last day the course, they put in practise everything they have learned in the city.
The kids proudly make their own way through the busy market square and amazed passers-by with how well they moved around
They weave their way skilfully through the crowds...
...and take on any obstacles with ease...
...even ones that are a bit more challenging.
Come on, Sam, don't let those steps stop you...
We can do this, Sam. Let's go!
You are holding on to me, aren't you Mum?
Of course! I won't let you fall.

Chapter 7

General discussion & summary

The main objectives of this thesis were (1) to develop outcome measures aimed at assessing wheelchair mobility skills (WMS) and confidence in wheelchair mobility (2) to validate an outcome measure aimed at objectively quantifying physical activity (PA) and (3) to evaluate the effect of a WMS training and exercise training on PA, physical fitness, WMS and confidence in wheelchair mobility in youth using a manual wheelchair.

MAIN FINDINGS

- The newly developed Utrecht Pediatric Wheelchair Mobility Skills Test (UP-WMST 2.0) is a reliable and valid outcome measure to assess WMS in youth using a manual wheelchair (**Chapter 2 and 3**).
- The Wheelchair Mobility Confidence Scale Dutch Youth (WheelCon-Mobility Dutch Youth) has shown evidence towards validity and internal consistency for the assessment of confidence in wheelchair mobility in Dutch youth using a manual wheelchair (**Chapter 4**).
- There is a low positive correlation between the WheelCon-Mobility Dutch Youth and the UP-WMST 2.0 (**Chapter 4**). As there can be a discrepancy between WMS and the confidence in wheelchair mobility, it is important to assess both the WMS and the confidence in wheelchair mobility. Interventions can be targeted at improving WMS, confidence in wheelchair mobility or both.
- An activity monitor (Activ8), shows good initial validity to detect 'active wheelchair use' in youth using a manual wheelchair, using a combination of a sensor at the wrist and wheel (**Chapter 5**).
- A combination of exercise and WMS training yielded significant positive long term effects on PA, WMS, confidence in wheelchair mobility and (an)aerobic performance in youth using a manual wheelchair (**Chapter 6**). There was no significant long term effect on cardiorespiratory fitness.

THEORETICAL CONSIDERATIONS

An important aim of pediatric rehabilitation is to optimize participation in physical activities for youth with a disability[1]. Over the last decades, multiple reviews[2–4] have reported on PA levels, including sedentary time, in youth with a disability and concluded that their PA levels were significantly lower than in typically developing peers. Other research has shown youth using a manual wheelchair to be less physically active than their ambulating peers with a disability[5–7]. One of the main findings of this thesis was the significant positive long term effect on increasing PA in youth using a manual wheelchair through a combined exercise and WMS training (**Chapter 6**)[8]. This was one of the first studies to report clinically relevant *long term* positive results on increasing PA in youth using a manual wheelchair[9,10]. Even with the positive results of the intervention, the overall level of PA in youth using a manual wheelchair remains low. This is not surprising as changing behavior is a complex problem where multiple factors can act as facilitators or barriers[11–13]. Qualitative research in youth with a disability described these facilitators and barriers towards PA using the framework of the International Classification of Functioning Disability and Health for Children and Youth (ICF-CY)[12,14] (figure 1). An example of possible facilitators and barriers are sufficient fitness, control of skills, motivation for PA (personal factors) and accessibility, parental beliefs and transport (environmental factors). These results are very relevant for clinical practice, where an extensive anamnesis and assessments using the ICF-CY framework could give an overview of which factors currently influence PA on the individual level and could be a target for an intervention. However, by using this approach a (long) list of individual facilitators or barriers could be identified in the ICF-CY. This summary of factors does not necessarily guide clinical decisions on which factor(s) to specifically target in an intervention. Moreover, often these factors are interdependent and interventions aiming to improve a single factor, for example physical fitness, do not lead to an increase in PA. Another limitation of using the ICF-CY for determining how to increase PA is that it does not give information about which factors are important to sustain PA in the long term. When aiming to increase and sustain PA, the PA behavior needs to change. Personal factors such as motivation, confidence and intention are essential determining factors of behavior[15–17] and should therefore play a central role in interventions when aiming to change and sustain PA. This lack of classification in the ICF-CY of personal factors has been recognized and reported on[18,19]. Last but not least, the ICF-CY currently does not have a language for activities

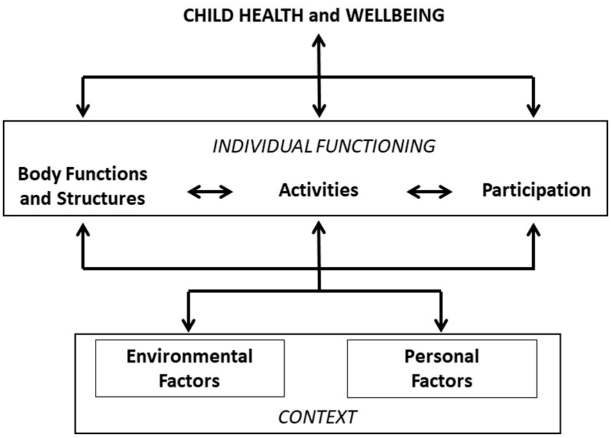


Figure 1. International Classification of Functioning Disability and Health for Children and Youth[14]

for people using a wheelchair or other assistive devices **(Chapter 1)**. Therefore, in my opinion, the ICF-CY lacks a common language on personal factors which are important to sustain PA behavior and gives insufficient direction to clinical practice on which combination of facilitators and barriers are of key importance to change and sustain PA in youth using a manual wheelchair.

A solution to this lack of language of personal factors in relation to PA might come from using the notion of 'Physical Literacy'. Physical Literacy is a perspective of looking at lifelong PA, where personal factors play a key role in determining PA behavior[20]. Over the last two decades, physical literacy has been implemented in physical education programs across the world[21–24], with the aim to increase lifelong PA in youth. I would like to explore, using the main findings of this thesis, whether this concept of physical literacy can give new directions for both rehabilitation care and research towards increasing long term PA in youth using a manual wheelchair.

1. What is physical literacy?

Physical literacy is a new emerging perspective on PA in health care[25] which originates from the field of physical education[26]. Physical literacy (figure 2, inner circle) can be described as:

" the motivation, confidence, physical competence, knowledge and understanding to value and take responsibility for engaging in physical activity for life"[20,27].

Physical literacy is a multidimensional viewpoint on PA, using a dynamic holistic approach consisting of four interrelated attributes that are of similar importance. These components are: physical (physical competence), affective (motivation & confidence), cognitive (knowledge & understanding) and PA behavior (value & engage)[27,28]. This theory can be seen as a combination of aspects from behavioral change theories[15–17] such as: motivation, intention, knowledge and attitude, and aspects important for the development of physical competence from motor learning theories[25,29]. Even though all aspects are important in physical literacy, the different attributes can become more or less important throughout life. For example, the cognitive attribute is less important in a young child as the role of family (social environment) will be more important at this period during the development[25]. Similarly, in adolescents the physical competence might become less important, while the motivation towards PA and the knowledge and understanding of the benefits of PA might become more important to value and engage in PA.

Physical literacy represents a unique lifelong journey for each individual and is inclusive to all[20]. The term 'literacy' refers to learning through effective interaction with the world[20].

To visualize this interaction, I have included social and physical environmental factors as an outer circle in the visual representation of physical literacy in figure 2.

Although the concept is inclusive to all, the practical application of the different attributes will be different for youth using a manual wheelchair compared to ambulatory peers. Figure 2 depicts the possible physical literacy journey of youth using a manual wheelchair, where the wheelchair should be seen as an extension of the body and therefore an important factor to consider in every attribute. I believe thinking along the line of physical literacy will be of additional value to health care professionals and researchers when aiming to increase PA in youth using a manual wheelchair. I will try to demonstrate the additional value through describing examples, using stories and data from our research (Box 1 and 2), where the interaction between the different attributes could help clinical decision making on which factors to address when aiming to increase PA in the long term for youth using a manual wheelchair.

2. How to assess physical literacy in youth using a manual wheelchair?

There has been controversy in literature on physical literacy as to whether one should assess physical literacy at all[25,30,31]. As Robinson states[30]: " Perhaps, in the very act of

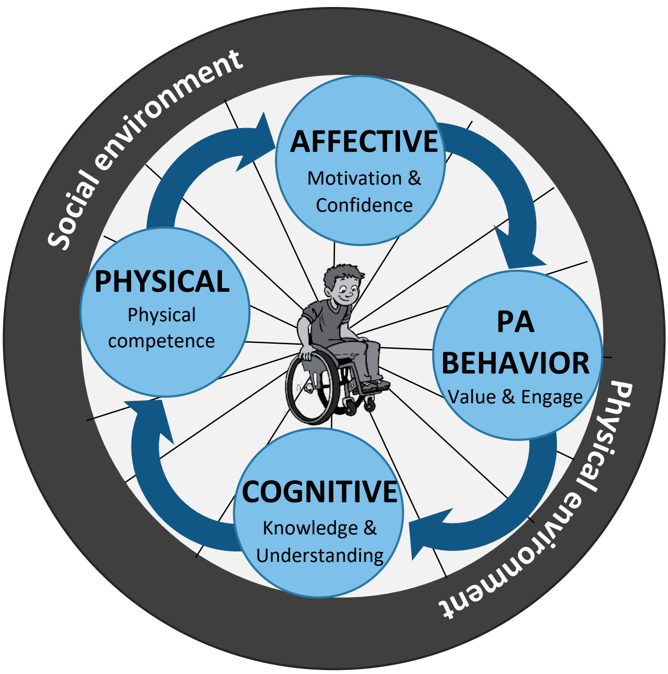


Figure 2. Visual representation of definition of physical literacy (inner circle) and the interaction with the physical and social environment (outer circle).

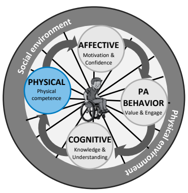
BOX 1: Sophie

Sophie is a sweet girl of ten years old with cerebral palsy (GMFCS 4) and a learning disability. She attends a special needs school in the Netherlands. Sophie arrives at her school by taxi (PA behavior), where her taxi driver is very helpful and pushes Sophie (social environment, PA behavior) to the entrance of the school. Here she slowly makes her way to the class room by herself (physical). Later that morning, Sophie's teacher (social environment) pushes her to the playground (against the advice of the physical therapist (social environment, PA behavior)), as the teacher feels there would not be much time left to play if Sophie would do this herself. At the accessible playground (physical environment), Sophie moves around a bit, but is also very happy to spend most of her break time watching the other children play (affective, PA behavior). At home, Sophie's mother (social environment) motivates her to propel her wheelchair by herself as much as possible, but due to time restraint choses to push her daughter when going outdoors (PA behavior).

Sophie participated in the combined wheelchair mobility skills and exercise training (**Chapter 6**) at her special needs school. She enjoyed the training sessions, especially the one where she got to practice her wheelchair mobility skills. She tried to make long powerful strokes and keep her hands in the air before she made the next stroke. Sophie also learned she can use furniture to push and pull herself forwards/backwards (physical). She really enjoys practicing this at home (affective) and through this learned to also use her trunk when making turns (cognitive, physical). She started to be more aware of the dimensions of her wheelchair by bumping into the cupboards and chairs at home (cognitive). While holding a wheelie is still too difficult for her, Sophie can now lift up her front caster when going over a low curb (physical). She uses this skill regularly to go over thresholds between rooms when she visits her neighbor friend's house (physical environment). At the start of the training, her level of PA was low (4.5% active wheelchair use/day, PA behavior), her physical competence was low (Shuttle Ride Test shuttles: 6, VO2peak: 21 ml/min/kg, Mean power MPST: 14 Watt, UP-WMST 2.0: 15 points). Sophie said that even though she had never done a lot of the items on the questionnaire (WheelCon-Mobility Dutch Youth: 130 points, affective), she was convinced she could do these by herself. There was a large discrepancy between her wheelchair mobility skills (low, physical) and her confidence in wheelchair mobility (high, affective).

After the training her PA level increased (6.0% active wheelchair use/day, PA behavior), while herphysical fitness and wheelchair mobility score remained the same (low, physical), her score on the WheelCon-Mobility reduced (affective). Having practiced some of the items and realizing she was not able to do a wheelie by herself, may have influenced her answers on the questionnaire. Sophie's mother (social environment) was very enthusiastic about the Wheelchair Mobility Skills training as she felt Sophie and she herself had learned a lot. Her mother made a comparison between teaching WMS and teaching her other daughter to cycle. As she could ride a bike herself, she knew how to teach this to her daughter and at what age she should start practicing this skill. With Sophie, she did not know which skills can be learned in a wheelchair and how and when you can practice this in everyday life. The mother was especially proud when Sophie had turned around to her and said: '*Stop mama, don't help me, I can do this myself'* (affective, physical).

measuring physical literacy, something is lost". The core of the problem is the holistic individual approach of physical literacy, which is lost with assessment of separate attributes. However, there is a need for physical literacy measurements to evaluate if interventions and programs aimed at increasing physical literacy are successful[32–35]. When talking about assessing physical literacy, one should keep in mind that physical literacy is about the interaction with the environment. It is for example not about an isolated skill, but about the ability to effectively and efficiently adapt one's movements as required by the environment[20]. Or to describe this in terms of the ICF-CY[14], it is not about one's capacity but about their performance in everyday life[36]. For youth using a manual wheelchair, wheelchair mobility is a pre-requisite for becoming physical active and hence an integral part of every attribute of physical literacy. I will therefore reflect per attribute on how the results of this thesis add to assessment of physical literacy in youth using a manual wheelchair and will place emphasis on what this means for assessing wheelchair mobility.



a. Physical Competence

Physical competence refers to an individual's ability to develop movement skills and patterns, and the capacity to experience a variety of movement intensities and durations[27]. Enhanced physical competence enables an individual to participate in a wide range of physical activities and settings[35]. Current assessments of physical competence in ambulatory youth include measures of physical fitness, such as the PACER test and measures of movement skills with an obstacle course[37]. For the assessment of physical fitness in youth using a manual wheelchair, several performance tests have been validated in the last decade, such as the Shuttle Ride Test and the Muscle Power Sprint Test[38–41].

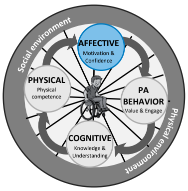
Current assessments of movement skills for typically developing youth include items like jump, kick and hop which are impossible to execute for youth using a manual wheelchair.

Wheelchair Mobility
The results of **Chapter 2 and 3** give an alternative method for assessing movement skills for youth using a manual wheelchair. The UP-WMST 2.0 is the first validated assessment tool for assessing WMS in youth using a manual wheelchair[42,43]. The UP-WMST 2.0 can be used in clinical practice, as it has demonstrated good validity, reliability and evidence towards responsiveness (**Chapter 3**). It can be used to assess if an intervention aimed at improving WMS is indicated and to document possible progress after WMS training. Furthermore, the UP-WMST 2.0 can be used in clinical decision making to decide on the appropriateness of the selected assistive device. A very low score on the UP-WMST 2.0 could be an indication that the wheelchair-user interaction is not appropriate to facilitate participation in physical activities, such as playing outdoors. Looking at the different attributes of physical literacy that influence PA in the example of Sophie, her physical competence could be argued to be a barrier towards PA. As her physical competence did not improve after the combined intervention this could be an argument for starting a different intervention aimed at improving physical competence. Another direction could be the re-evaluation of whether the settings of the manual wheelchair are optimal for participation in physical activities or if a manual wheelchair is the most appropriate device for Sophie to participate in physical activities outdoors.

The UP-WMST 2.0 does give valuable information about what a participant is able to do (capacity). It was developed to include basic and more complex WMS skills, as different skills can become important at different stages through childhood **(Chapter 2)**. This is one of the few studies reporting on WMS in youth and even though we included a relatively large heterogeneous sample in the development and assessment of the psychometric properties of the UP-WMST 2.0 (**Chapter 3**), questions remain regarding the assessment of WMS in all youth using a manual wheelchair. For example, our research included children from the ages of 5 to 18 years and questions remain regarding the assessment in pre-school children. Participants from our sample in **Chapter 6** started using a wheelchair from as young as 1.5 years old. Future research towards WMS in very young children could give information about if and how WMS training in this young population could lead to increased wheelchair mobility. As a comparison, power wheelchair mobility training is given in children as young as 20 months[44,45].

Another aspect of wheelchair mobility is the propulsion technique, i.e. movement pattern, push frequency and power output [46]. Even though learning to push the wheelchair with slow powerful strokes was part of the WMS training program (see example Sophie), this skill was not measured during the assessments in the intervention study reported in

Chapter 6. It remains therefor unclear whether WMS training can improve propulsion technique. Results of the combined exercise and WMS training study **(Chapter 6)** do indirectly indicate that participants improved their propulsion technique as they managed to reach on average a higher number of shuttles on the Shuttle Ride Test with the same cardiorespiratory effort. Future research should evaluate if it is possible to improve propulsion technique in youth using a manual wheelchair.



b. Affective: Motivation and confidence

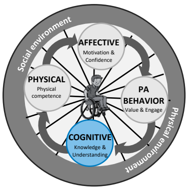
Motivation and confidence refers to an individual's enthusiasm for, enjoyment of and self-assurance in adopting PA as an integral part of life[27]. This 'fun' component is starting to gain more attention in pediatric rehabilitation, as one of the 'F-words' : Fun, Function, Family, Fitness, Future and Friendship[47]. These F-words are six key areas that are deemed as important for child development[47]. One important aim of the WMS training program (**Chapter 6, Chapter 3**) was to show/teach participants that using a manual wheelchair can also be fun. To create awareness of which WMS are possible and that these can be fun was the aim of a television episode on WMS for youth[48], showing which WMS skills youth need to perform to participate in daily life and that activities in a wheelchair such as skiing or performing WMS in skate park can be fun! This episode was broadcasted on Dutch national television as a side project of this thesis.

Regarding the assessment of the motivation (e.g. fun) and confidence towards PA of youth using a manual wheelchair, no valid or reliable measurement instruments are available. Two questionnaires assessing social cognitive determinants related to PA have been used as outcome measures in studies in youth with a disability[49–51]. However, neither of these questionnaires were specifically designed or validated for youth using a manual wheelchair.

Confidence in wheelchair mobility

The results of this thesis **(Chapter 4)** on the development of the Wheelchair Mobility Confidence Scale (WheelCon-Mobility) for Dutch Youth add to the assessment of the affective attribute of physical literacy in youth using a manual wheelchair. The WheelCon-Mobility shows evidence for internal consistency and validity in youth, aged 8-18 years, using a manual wheelchair. We reported on problems with self-reporting on confidence in a small percentage of children with learning disabilities who participated in one of our studies (**Chapter 6**). Secondary analyses of the results in **Chapter 4** on the correlation between the UP-WMST 2.0 and the WheelCon-mobility Dutch Youth, showed no significant difference in correlation between youth with or without a learning disability. However, clinicians should be aware of possible problems in the self-reporting on confidence in wheelchair mobility in youth with a learning disability and the assessment through a proxy questionnaire in parents/caregivers could give additional valuable information.

Another important implication for clinical practice, could be a possible discrepancy between the confidence in wheelchair mobility (WheelCon-Mobility) and the capacity to do a skill (UP-WMST 2.0), as was also the case in the example of Sophie (box 1). From this discrepancy it becomes clear that the assessment of confidence in wheelchair mobility is just as important as the assessment of wheelchair mobility skills as both the capacity and confidence dictate whether a WMS is actually performed in everyday life[52]. A logical next step for future research on WMS would be the development of an assessment tool to measure the actual performance of WMS in everyday life.



c. Cognitive: Knowledge and understanding

Knowledge and understanding includes the ability; to identify and express the essential qualities that influence movement, to understand the health benefits of an active lifestyle, and to appreciate appropriate safety features associated with PA in a variety of settings and physical environments[27]. This is somewhat similar to health literacy, where the ability to understand, to gain access to, and to use information plays an important role in promoting and maintaining good health[53]. A important new direction for future research aimed at increasing PA in youth using a manual wheelchair is to develop assessments and interventions aimed at increasing knowledge and understanding about PA in youth using a manual wheelchair. At the same time, current clinical practice could recognize the importance of developing knowledge and understanding of PA and incorporate questions about this topic in their anamnesis.

In the intervention study aimed at increasing PA in youth using a manual wheelchair (**Chapter 6),** we did not include a cognitive component in the assessment or intervention and can therefore not give any objective information on this attribute from the results of this thesis. There are however lessons to be learned from assessment of this attribute in typically developing children in combination with observations made by researchers and health care professionals during this thesis.

Knowledge & understanding PA in youth using manual wheelchair

Current assessment tools towards measuring physical literacy in typically developing youth[32,54] include questionnaires on the cognitive attribute of physical literacy. These tools should include questions related to a person's knowledge and understanding on both their physical abilities and their understanding of their own current state of health and wellness[30]. However, the current questionnaires are limited to the knowledge and understanding of the PA guidelines for their age group[55]. For youth using a manual wheelchair these questions are impossible to answer, as we do not know if the guidelines developed for ambulatory youth are also applicable to them. Propelling your wheelchair is physically straining and less energy efficient as walking[56] and therefore PA recommendations need to be adapted for youth using a manual wheelchair. Possible similar adaptations can be made as has been done in the adaption of PA guidelines for adults with a spinal cord injury [57].

Another important point was raised by several health care professionals from their experiences with the high intensity interval exercise training. A number of participants had to learn what maximal exertion felt like. Health care professionals needed to teach/ let participants experience, that maximal exertion was not a dangerous feeling that should be avoided but a state they should try to achieve during the exercise bouts. It could take a number of sessions and tremendous effort of the health care professionals to motivate participants to reach their maximal effort during the exercise bouts. The lack of understanding in some participants why physical exercise is important becomes clear from the response of one participant after the intervention: "*I don't recommend exercise training to others. The training makes you tired and you get muscle ache in your arms"*. These observation make it clear, that it is not only important to increase the knowledge and understanding towards PA, but also increase the knowledge and understanding towards physical fitness.

Knowledge & understanding wheelchair mobility

In addition to knowledge and understanding about their physical body which is similar to youth who are ambulatory, youth using a manual wheelchair also need to have knowledge and understanding about their wheelchair, which can be seen as an extension of their body. To illustrate this importance, I will now introduce Nora as example two.

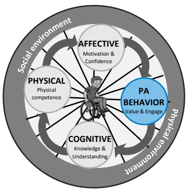
From the example of Nora, it becomes clear that an optimal wheelchair-user interface is an important prerequisite for performing WMS[56]. One astonishing observation I came across during assessments for this thesis, was the low tire pressure of almost every wheelchair which impacts the rolling resistance and everyday use of the wheelchair[46]. The tire pressure of more than three quarters of the participants (**Chapter 3-6**) was (far) below the advised pressure (as described on the tire). Moreover, the average weight of wheelchairs (excluding E-wheels) in the intervention study in this thesis was 19.4 kg (**Chapter 6**), where an average lightweight wheelchair for adult wheelchair users only weighs 12 kg[58]. This high wheelchair mass immediately negatively impacts rolling resistance of the wheelchair[46]. To optimize the wheelchair-user interface in youth using a manual wheelchair, the knowledge and understanding of wheelchair settings needs to improve in healthcare professionals to support youth, parents/caregivers and wheelchair suppliers in selecting a lightweight wheelchair[59]. Health care professionals can also play

BOX 2: Nora

Nora is a fourteen year old competitive girl (height 1.50m, weight 40 kg) with Spina Bifida (household ambulator). She uses a posterior walker for short distances indoors and a manual wheelchair for longer distances, sports and outdoors (PA behavior). Nora enjoys (affective) being active (7% active wheelchair use/day) and plays wheelchair basketball (1x/week), wheelchair tennis (1x/week) and uses her handbike regularly (PA behavior). Before commencing the combined intervention of wheelchair mobility skills and exercise training at her special needs school **(Chapter 6)**, Nora's physical competence was good (Shuttle Ride Test: shuttles 16, VO_2peak = 38ml/min/kg, UP-WMST 2.0 = 41 points (out of 51 points). The settings of Nora's wheelchair on the other hand hindered active independent wheelchair use. The tire pressure was far below optimum, and the wheelchair was heavy (24 kg). In addition, anti-tippers were set at 4 cm above the ground, and the hair of Maxi (her dog) had accumulated inside the casters of the front wheels. Several settings limited an active propulsion, such as center of pressure behind the axis of the rear wheel, handle bars extended (inviting people to push wheelchair) and a slouched sitting position. Nora is a bright girl, but does not seem to know wheelchair maintenance (tire pressure and cleaning) is important to keep her wheelchair rolling smoothly (cognitive). Nora also does not know anything about the wheelchair settings and options, e.g. how to fold away the anti-tippers. During the selection of the wheelchair, she only decided the color and her parents, physical/occupational therapist and wheelchair supplier (environment) made all the other decisions.

Nora does not enjoy being seen in public (social environment) in her wheelchair and is very adamant to not have any pictures or movies taken while she is in her wheelchair. If a photo needs to be taken, Nora would step out of her wheelchair and pose standing up. There was a large discrepancy between her wheelchair mobility scores on the UP-WMST (high, physical) and her confidence in wheelchair mobility (low, affective). During the first wheelchair mobility skills training session, her anti-tippers were removed and with some motivation of the teachers (social environment), she started practicing holding a wheelie. Her mother (social environment) objected at first when the anti-tippers were removed as this was considered far too dangerous and unnecessary. However, she quickly became amazed at all the skills Nora was capable of doing without the anti-tippers during the first session. She admitted this was an eye-opener as she did not know all these things were possible in a wheelchair. After the last wheelchair skills training session Nora's score on the UP-WMST 2.0 went up 5 points, while she had gained 20 points on the confidence in wheelchair mobility scale. Without being aware of her daughters assessments, Nora's mother said: *My daughter has gained so much confidence (affective), she really enjoys (affective) being in her wheelchair now and she tries to perform wheelies everywhere (8.5% active wheelchair use/day, PA behavior). I need to continuously take photos of her and post them on social media (social environment). I'm proud that she can do all of this, especially in this heavy wheelchair. I'm excited to see what she will be able to do in her new lightweight wheelchair. A lot of public places are not adapted (physical environment), so we are interested in a follow-up session once the new wheelchair arrives!*

an important role in educating a child and their parents/caregivers about the importance of wheelchair maintenance (tire pressure, cleaning front casters) and in helping to select a wheelchair with settings that stimulate independent active wheelchair use (such as light weight, seating angle, push bars etc). More information on wheelchair settings can for example be found on www.checkjezit.nl and www.wheelchairskillsteam.nl.

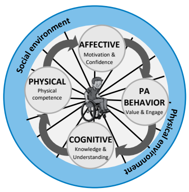


d. Physical activity behavior

Engagement in PA behavior for life refers to an individual taking personal responsibility for physical literacy by freely choosing to be active on a regular basis. This involves prioritizing and sustaining involvement in a range of meaningful and personally challenging activities, as an integral part of one's lifestyle[27]. Assessment of PA behavior in youth with and without a disability includes an integrated view of physical activity, sedentary behavior and sleep[55]. Current assessments of this attribute of physical literacy in typically developing youth includes assessment of average daily step count, self-reported sedentary time and self-reported number of days a week a child engages in moderate to vigorous PA[37].

PA in youth using a manual wheelchair

The results of (**Chapter 5**)add to the assessment of PA behavior in youth using a manual wheelchair, as this is the first validated objective activity monitor to assess 'active wheelchair use' in youth using a manual wheelchair. When compared to the assessment in typically developing youth, 'active wheelchair use' corresponds with assessment of average daily step count. When interpreting these results, one should be aware that the objective assessment of PA in people using a wheelchair is still in its infancy compared to the widely commercially available activity monitors in ambulatory persons. When looking at physical literacy and PA behavior in youth using a manual wheelchair, the most urgent step forward is the development of a commercially available activity monitor that can be used by the participant to inform them about how active they are on a daily basis. More knowledge and understanding about how physically active you are, can in itself lead to increased PA on a regular basis[60].



e. Environment

Even though the term 'literacy' refers to effective interaction with the world, current assessment tools of physical literacy give ample attention towards the environment. The environment incorporates a spectrum from the individual's most immediate environment, such as family and peers, to the general environment, such as cultural beliefs[61]. Environmental factors can be split into the social environment and the physical environment as is common in the ICF-CY[14].

Physical environment and wheelchair mobility

For youth with and without a disability, the physical environment can act as a facilitator or barrier towards becoming physically active[12,62]. For example, playgrounds in the Netherlands are often not inclusive for youth with a disability, limiting their opportunity to be physically active around their home[63]. Through increased wheelchair mobility, some obstacle in the physical environment, e.g. a curb, might no longer be perceived as a barrier to cross a street [12]. Or as explained in the example of Sophie, after the WMS training, the thresholds in her friend's house where no longer a barrier towards independent mobility.

Social environment and wheelchair mobility

The social environment plays an important role in the physical, social and psychological development of all youth, including those with a disability[64]. This was the reason for pediatric rehabilitation to change their approach of delivering health care from child-centered care to family-centered care[65]. One of the key factors to improve family-centered care is to empower parents/caregivers by providing information[66]. From remarks of parents involved in this thesis it became clear that youth and their parents are not sufficiently provided with information about what WMS are possible in a wheelchair and how settings of a wheelchair (e.g. anti-tippers) can facilitate or hinder wheelchair mobility. As mentioned by a parent: *"eye-opener, there is so much more possible in a wheelchair, than I previously thought!"*.

The parental beliefs about the wheelchair and awareness of WMS possibly influence the development of WMS in youth using a manual wheelchair, through overprotection or stimulation of independent wheelchair mobility. As mentioned by the mother of Sophie (example 1), we do not have any expectations on which WMS should or could be achieved at different stages in childhood. With no frame of reference, the social environment can possibly act overprotective or underestimate which WMS could be achieved. Overprotection and underestimation is more common in children with a disability than typically developing peers, and can cause lowered self-esteem and failure to reach their full potential[67,68].

To help build a frame of reference for parents/caregivers and health care professionals, more insight is needed about which WMS are appropriate for the child's age. Future research towards increasing wheelchair mobility in youth should include the beliefs towards a wheelchair and WMS of the immediate social environment (family and health care professionals) as this seems to be an area where there is a lot of room for improvement.

This section reported on valuable lessons for clinical practice and research regarding the assessment and attention for development of physical literacy in youth using a manual wheelchair. It described how wheelchair mobility is an important aspect to consider in every attribute of physical literacy for youth using a manual wheelchair.

3. Spinning the wheel of physical literacy

The overall level of PA in youth using a manual wheelchair is low[3,6,69]. Assessing the different attributes of physical literacy is important, but we also need to spin the wheel of physical literacy in youth using a manual wheelchair to have long term increases in PA. In **Chapter 6**, we found a long term significant increase in PA which could possibly be explained by increasing physical fitness (physical competence) through an exercise training, by increasing (confidence in) wheelchair mobility through a WMS training or by the combined effect of these training programs. Due to the design of the study, which evaluated the combined effect of exercise and WMS training, it was not possible to assess or compare the effects of individual training programs on long term increases in PA. However, when looking more closely at all the different attributes of physical literacy that were impacted by the WMS training program, it is possible that the WMS training program generated a larger long term increase in PA compared to the exercise training.

Wheelchair mobility Skills Training

Most parents/caregivers, youth and health care professionals were very enthusiastic about the WMS training sessions. These sessions addressed a gap in current pediatric rehabilitation, where insufficient attention was placed on improving wheelchair mobility. The success of the WMS training session can possibly be explained by looking at the complexity of all the interrelated attributes of physical literacy that started spinning in the WMS training program, which also occurred in the examples in box 1 and 2.

First of all, three of the training sessions were given by KJ-Projects, a wheelchair skills training foundation which is the current expert in giving WMS training in the Netherlands[70]. The trainers are wheelchair users, who act as a trainer and role model[71] for youth and their family to create awareness of the kind of WMS that are possible in a wheelchair and that being in a wheelchair can be fun (social environment, affective)! This enjoyment was reflected in the motivation to participate in training sessions by youth using a manual wheelchair, as reported by health care professionals (**Chapter 6**). By comparison, health care professionals reported on lower levels of motivation for the exercise training program. Secondly, youth trained in groups where they can experience learning new skills (physical) together with peers and act themselves as role models on how to acquire new WMS (social environment)[72]. As one participant commented: *"I practiced a lot, as we now do 'Fit For The Future' during break time on the school playground"(PA behavior).* Thirdly, the sense of accomplishment when acquiring a new difficult WMS (physical) almost automatically leads to increased confidence (affective)[52]. Fourthly and possibly the most important of all, was the inclusion of parents/caregivers in the WMS training sessions both in and outdoors. Here, parents/caregivers saw all the WMS their child could do and also learned how these WMS can be practiced in daily life (social environment)[73]. Some parents mentioned that they realized they helped their child too much, limiting the development of independent wheelchair mobility. At the same time participants and their parents/caregivers became aware of which WMS are necessary to learn to become independent in wheelchair mobility (cognitive) and may have noticed that the settings of their wheelchair hindered independent mobility (e.g. anti-tippers, wheelchair weight).

From this description above it becomes clear that the WMS training program has a broader effect than simply learning mobility skills in a wheelchair, as several attributes in the wheel of physical literacy started spinning. The measurement instruments developed in this thesis were able to assess some of these effects (WMS, confidence in wheelchair mobility and PA), but did not capture the complex interaction between attributes and with the social environment which in unique for every individual. The intervention reported in **Chapter 6** used a 'One size fit all' approach, given similar attention to the development of physical competence, motivation, confidence, knowledge and understanding towards PA behavior in all participants and their parents/caregivers. Future research towards physical literacy in youth using a manual wheelchair, should take into account the unique individual development of physical literacy. More or less emphasis on separate attributes or a combination of attributes in interaction with the social environment could possibly spin the wheel of physical literacy faster in youth using a manual wheelchair.

METHODOLOGICAL CONSIDERATIONS

In this thesis, practice based research was used as strategy to deliver practical relevance, while maintaining scientific rigor[74]. Practice based research uses questions derived from practice to develop knowledge that can directly support, transform or improve practices. In this thesis, questions from practice were formulated by KJ-Projects and physiotherapists, occupational therapists and PE teachers of De Hoogstraat Rehabilitation and De Trappenberg Rehabilitation. The main question was to assess if WMS training is an effective method to improve WMS in youth using a manual wheelchair.

Practice based research tries to address the gap between 'rigor' and ' relevance'. Bridging this gap has been a topic of scientific debate for over 60 years[75], as reaching highly relevant practice might have less academic rigor or vice versa. Practice based research, also called action research, takes place between the so called 'four contexts' (figure 3)[75]. Randomized control trials take place within context IV, contributing to theory with often limited practical value.

One of the strengths of using this practice based research strategy is that the results of this thesis generated highly relevant knowledge for clinical practice (context I.). The use of a heterogeneous sample which is representative for the population of wheelchair users in special needs schools in the Netherlands and the collaboration with health care professionals, youth and their parents/caregivers to develop measurement tools and intervention, generated knowledge that can be directly applied in special needs schools.

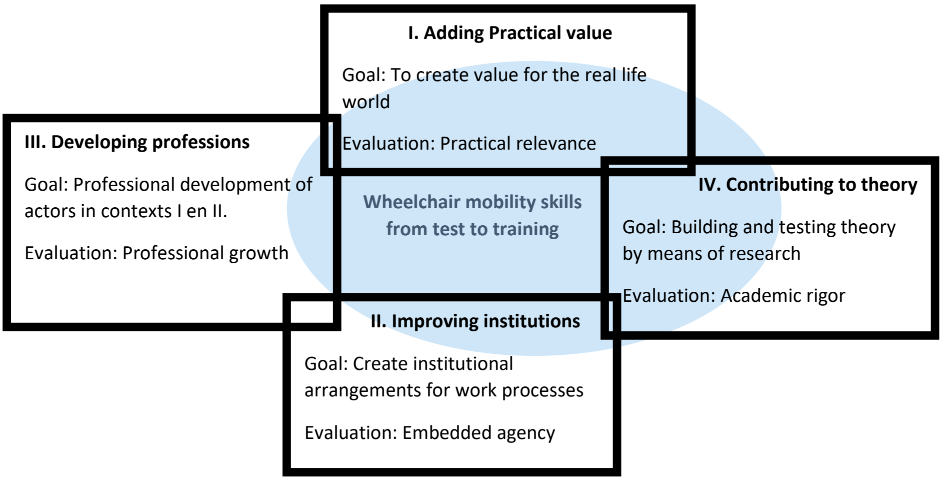


Figure 3. Positioning the research strategy of thesis 'wheelchair mobility skills from test to training' (blue circle) within model of action research[75].

This can be seen in the long list of clinical implications (see section below) that resulted from this thesis, of which many of them can be directly implemented in clinical practice. Moreover, during this thesis, physical therapists, occupational therapists and PE teachers of the involved rehabilitation centers/schools were trained and schooled in administering the UP-WMST 2.0, exercise training and WMS training protocol. Together with the use of exercise tests that are already commonly used in rehabilitation, these centers now have the tools and knowledge to continue with WMS and exercise trainings (context III. developing professions). After their participation in the intervention study, several special needs school, have implemented the WMS training program in their rehabilitation program for youth using a manual wheelchair (context II. improving institutions).

As mentioned above, the heterogeneity of the sample adds to the relevance of this thesis, but at the same time leads to large individual differences on the effect of the combined intervention in **Chapter 6**. This large variation in combination with a relatively small sample can give statistical challenges in detecting significant change. Even with these statistical challenges the results of the multi-level model analyses were highly significant ($P< 0.01$), contributing to the strength of the conclusion that the combined training appeared to have a positive long term results on PA, (an)aerobic performance, WMS and confidence in wheelchair mobility.

The balance between practical value (context I.) and academic rigor (context IV.) is an important point to consider when developing practice based research. One lesson that could be learned from the intervention study in **Chapter 6** about the balance between context I. and IV., is to improve the transparency in selecting participants. In our study, health care professionals at the local rehabilitation centers/schools played a role in contacting participants to participate in the research. From the graphs included as an appendix in **Chapter 6**, we can see that even though there was no statistical difference (possibly due to the large variation), one group (WMS before Exercise) had on average a lower physical fitness than the other group (Exercise before WMS) before commencing the training program. The group with lower fitness showed a higher increase after the combined training, than the group with a higher fitness. These results indicate a larger effect of WMS before exercise training compared to exercise before WMS training on physical fitness. However, one should be aware that the between groups difference in effect can also be caused by a statistical phenomenon called 'regression towards the mean' [76].

The difference between groups at the start of this study (**Chapter 6**), could possibly have occurred through selection bias, as one training protocol (WMS before or after exercise) was used per rehabilitation center/school. The difference between centers in which participants to contact could also lead to different samples per training protocol. Another explanation for the difference between the groups could be a difference in the level of attention WMS and physical fitness has received in the rehabilitation program at the centers/schools, leaving more or less room for improvement. As the collaboration with clinical practice is an inherent part of practice based research, a recommendation for future research is to gather extensive information about factors which could influence baseline outcomes or the effect of an intervention and that could differ between clinical practices.

When looking at the model of action research, another direction for future research would be to have more attention for professional development (context III.) and institutional organizations (context II.) when aiming to support, transform or improve practices. As described above in section 2c. (knowledge and understanding of the wheelchair), it becomes clear that many occupational therapists, physical therapists and wheelchair suppliers lack the appropriate knowledge and understanding of wheelchair settings and WMS training in youth using a manual wheelchair. Even though professional development has likely occurred in the health care professionals involved in the studies in this thesis, as they received training on how to assess and train WMS in youth using a manual wheelchair, this was not part of the assessments used in this thesis and a recommendation for future action research. In order to increase the general knowledge in physical therapist and occupational therapist regarding wheelchair mobility in youth using a manual wheelchair, educational institutions (context II.) should incorporate lessons regarding wheelchair mobility assessment, training and/or wheelchair settings in their education program[59] or this should be made available through a post graduate course. A first step to introducing this theme in the education program of health care professionals involved with youth using a manual wheelchair in the Netherlands, has been made by the Master of pediatric physical therapy from the HU university of Applied sciences Utrecht and Windesheim Master of physical education and sport pedagogy. Both educational institutions (context II.) have recently introduced WMS training and/or WMS assessment in their education program.

CLINICAL IMPLICATIONS

For all youth who receive a manual wheelchair and their family it is of utmost importance to create awareness of all the possibilities a wheelchair can offer to participate in physical activities. During the selection process of the first wheelchair, immediate attention and information about wheelchair settings and WMS that facilitate wheelchair mobility needs to be provided by health care professionals to parents/caregivers and child. Moreover, in all youth who uses a manual wheelchair, WMS and confidence in wheelchair mobility should be evaluated at different stages in childhood. Based on these evaluations, WMS training involving the parents/caregivers might be indicated.

Professional development

- Educational institutions of health care professionals, such as physical therapy and occupational therapy, should include an education program regarding wheelchair mobility. As health care professionals should have a basic level of WMS in order to be able to teach WMS to youth using a manual wheelchair. At the same time they should realize their limitation as ambulating person and acknowledge the important influence a role model (experienced wheelchair user) has to motivate participants (and their parents/caregivers) to learn WMS.
- Educational institutions of health care professionals, such as physical therapy and occupational therapy, should include an education program regarding wheelchair settings. Through improved knowledge and understanding of wheelchairs, health care professionals can play an important role in learning a child and their parents/caregivers about the importance of wheelchair maintenance (tire pressure, cleaning front casters) and wheelchair settings.

Assessment of wheelchair mobility

- As there can be a discrepancy between capacity and confidence, it is important to detect which aspect hinders performance of WMS in daily life and could possibly be improved through an intervention.
- The UP-WMST 2.0 evaluates the capacity to perform basic and some more complex WMS in youth using a manual wheelchair. A very low score on the UP-WMST 2.0 can indicate that the participant might experience problems with independent participation in wheelchair activities. A very high score indicates a participant has the capacity to perform basic and complex WMS as assessed in the UP-WMST 2.0. Participants with a high UP-WMST 2.0 might still experience problems with performing more complex WMS in daily life (for example, going down 3 steps) and could still benefit from a more advanced WMS assessment and training.
- To gather reliable results on multiple assessments of the UP-WMST 2.0, it is important to measure the tire pressure of both wheels and administer the UP-WMST 2.0 on the same surface, with the same instructions and instructor as advised in the UP-WMST 2.0 manual.

Wheelchair mobility skills training

- Wheelchair mobility skills training has a larger impact than just learning mobility skills in a wheelchair. It can also increase the confidence in wheelchair mobility, build enjoyment of moving around in a wheelchair and possibly change parental beliefs towards WMS and being in a wheelchair.
- Involvement of parents/caregivers in the WMS training of their child is important. This will create awareness in the parents/caregivers of what kind of WMS are possible in a wheelchair and how this can be taught/trained in everyday life.

- Role models and peer group training sessions increases the motivation to learn new WMS.
- In this thesis, the effect of WMS training and exercise training were evaluated in participants who use a wheelchair on a daily basis and attended schools for special education. Possible similar benefits can be found in youth using a manual wheelchair who attend regular education schools or use a wheelchair for long distances/sport.

Physical Activity and physical literacy

- The model of physical literacy depicted in figure 2 can be used by health care professionals in their clinical reasoning to give attention to possible important factors that can change and sustain PA in youth using a manual wheelchair. In addition to the usual focus in pediatric rehabilitation on ability (physical competence), this model makes it clear to also focus on motivation, confidence, knowledge and understanding towards PA and give attention towards the influence of the physical and social environment.
- A combination of WMS and Exercise training leads to a larger increase in PA in youth using a manual wheelchair, compared to the effect of a single training (WMS or exercise).

DIRECTIONS FOR FUTURE RESEARCH

The studies reported in this thesis are one of the first to address the gap in literature towards wheelchair mobility in youth using a manual wheelchair. As wheelchair mobility is a pre-requisite for being able to participate in physical activities, it is of critical importance to increase the knowledge in literature and clinical practice through further research on wheelchair mobility in youth using a manual wheelchair. Researchers should strive to collaborate internationally to reach larger sample sizes and generate more power of tests in this heterogeneous population. Further longitudinal research will improve the interpretation of measurement outcomes against the natural variability that can occur due to e.g. seasonable variations or the natural development of youth.

Professional development

- Future action research should be aimed at developing education programs on wheelchair mobility for health care professionals working with youth using a manual wheelchair.
- Future action research aimed at supporting and improving the pediatric rehabilitation care of youth using a manual wheelchair should include a transformation in institutional organizations (e.g. legislation, wheelchair suppliers, rehabilitation centers) to ascertain long term changes in clinical practice.

Wheelchair mobility skills training

- Future research towards WMS training in youth using a manual wheelchair, should be aimed at assessing the important role the social environment has on developing WMS. Moreover, future research should assess if WMS training of parents/caregivers can facilitate the development of wheelchair mobility in very young children.
- Future research should be aimed at evaluating the effect of WMS training on developing wheelchair mobility and improving WMS in pre-school children who use a manual wheelchair, youth using a manual wheelchair who attend regular education in the Netherlands and youth who only use a wheelchair for longer distances.
- Future research should be aimed at developing age appropriate WMS training goals/content. Through for example describing which WMS are necessary to independently participate in activities appropriate for the child's age (toddler, child, adolescent). This description can help build a frame of reference and awareness in parents/caregivers and health care providers on which WMS are recommended to independent perform at a certain age.

Assessment of wheelchair mobility

- Future research should focus on developing a measurement tool aimed at assessing the performance of WMS in daily life of youth using a manual wheelchair.
- Future research should be aimed at developing and evaluating WMS in very young (pre-school) manual wheelchair users.
- Future research towards assessing wheelchair mobility in youth should include an evaluation of propulsion method, such as propulsion technique and/or power output.

Physical Activity and physical literacy

- Future research towards physical literacy in youth using a manual wheelchair, should evaluate if it is possible to improve individual or a combination of attributes of physical literacy and whether these improvements lead to an increase and sustain PA behavior.
- When developing or evaluating new interventions aimed at increasing and sustaining PA in youth using a manual wheelchair, future research should take into account the individual unique development of physical literacy as 'one size does not fit all'. More or less emphasis on separate attributes or a combination of attributes in interaction with the social and physical environment might make it possible to spin the wheel of physical literacy faster in youth using a manual wheelchair.
- Future research should be aimed at developing measurement instruments for assessing the motivation, confidence, knowledge and understanding of wheelchair mobility in youth using a manual wheelchair.
- Future research should be aimed at developing PA guidelines for youth using a manual wheelchair.

- One of the most urgent steps forward is to develop a commercially available activity monitor that can be used by youth using a manual wheelchair to inform them and their parents/caregivers about their PA behavior on a daily basis.

REFERENCES

1. Hadders-Algra M, Maathuis K, eds. Kinderrevalidatie. In: *Edition: 5*. Koninklijke van Gorcum; 2015:3-27, 351-393, 417-435.
2. Carlon SL, Taylor NF, Dodd KJ, Shields N. Differences in habitual physical activity levels of young people with cerebral palsy and their typically developing peers: A systematic review. *Disabil Rehabil*. 2013;35(8):647-655. doi:10.3109/09638288.2012.715721
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(12):1856-1861. doi:10.1016/j.apmr.2010.08.018
4. Ganz F, Hammam N, Pritchard L. Sedentary behavior and children with physical disabilities: a scoping review. *Disabil Rehabil*. March 2020:1-13. doi:10.1080/09638288.2020.1723720
5. Gorter JW, Noorduyn SG, Obeid J, Timmons BW. Accelerometry: A Feasible Method to Quantify Physical Activity in Ambulatory and Nonambulatory Adolescents with Cerebral Palsy. *Int J Pediatr*. 2012;2012:1-6. doi:10.1155/2012/329284
6. Claridge EA, Bloemen MAT, Rook RA, et al. Physical activity and sedentary behaviour in children with Spina Bifida. *Dev Med Child Neurol*. 2019;61(12):1400-1407. doi:10.1111/dmcn.14333
7. Backx FJG, Nooijen CFJ, Bloemen MAT, et al. Physical activity in wheelchair-using youth with Spina Bifida: an observational study. *J Neuroeng Rehabil*. 2019;16(1):1-13. doi:10.1186/s12984-018-0464-x
8. Sol M, Verschuren O, Horemans H, et al. A Wheelchair Mobility Skills and Exercise training to improve physical activity in youth using a manual wheelchair. *Disabil Rehabil*.:(submitted).
9. 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;59(10):1004-1010. doi:10.1111/dmcn.13422
10. Reedman S, Boyd RN, Sakzewski L. The efficacy of interventions to increase physical activity participation of children with cerebral palsy: a systematic review and meta-analysis. *Dev Med Child Neurol*. 2017;59(10):1011-1018. doi:10.1111/dmcn.13413
11. Shields N, Synnot A. Perceived barriers and facilitators to participation in physical activity for children with disability: a qualitative study. *BMC Pediatr*. 2016;16(1):9. doi:10.1186/s12887-016-0544-7
12. Bloemen MAT, Verschuren O, van Mechelen C, 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(1):11. doi:10.1186/s12883-015-0265-9
13. Hesketh KR, Lakshman R, van Sluijs EMF. Barriers and facilitators to young children's physical activity and sedentary behaviour: a systematic review and synthesis of qualitative literature. *Obes Rev*. 2017;18(9):987-1017. doi:10.1111/obr.12562
14. World Health Organization. *International Classification of Functioning, Disability, and Health–Children and Youth Version*. Geneva: World Health Organization; 2011.
15. Hermsen S, Renes R, Frost J. Persuasive by design: a model and toolkit for designing evidence-based interventions. In: *Proceedings of CHI-Sparks, Creating the Difference*. ; 2014:74.

16. Bandura A. A social cognitive theory of personality. In: John LP& O, ed. *Handbook of Personality*. 2nd ed. New York: Guilford Publications; 1999:154-196.
17. Deci EL, Ryan RM. Self-determination theory. In: *Handbook of Theories of Social Psychology: Volume 1*. SAGE Publications Inc.; 2012:416-437. doi:10.4135/9781446249215.n21
18. Simeonsson RJ, Lollar D, Björck-Åkesson E, et al. ICF and ICF-CY lessons learned: Pandora's box of personal factors. *Disabil Rehabil*. 2014;36(25):2187-2194. doi:10.3109/09638288.2014.892638
19. Geyh S, Peter C, Müller R, et al. The Personal Factors of the International Classification of Functioning, Disability and Health in the literature–A systematic review and content analysis. *Disabil Rehabil*. 2011;33(13-14):1089-1102. doi:10.3109/09638288.2010.523104
20. Whitehead M. *Physical Literacy across the World*. Routledge; 2019. doi:10.4324/9780203702697
21. Canadian Heritage. *"Canadian Sport Policy (2012) Formative Evaluation and Thematic Review of Physical Literacy and LTAD.*; 2016. https://sirc.ca/wp-content/uploads/2019/12/formative_evaluation_thematic_review_physlit_ltad_2016.pdf.
22. Aspen Institute. *Physical Literacy: A Global Environmental Scan.*; 2015. https://www.aspeninstitute.org/wp-content/uploads/files/content/docs/pubs/GlobalScan.pdf.
23. Keegan RJ, Barnett LM, Dudley DA, et al. Defining physical literacy for application in Australia: A modified delphi method. *J Teach Phys Educ*. 2019;38(2):105-118. doi:10.1123/jtpe.2018-0264
24. Pot N, Hilvoorde I. A critical consideration of the use of physical literacy in the Netherlands. In: *ICSSPE Bulletin–Journal of Sport Science and Physical Education,*. ; :65.
25. Cairney J, Dudley D, Kwan M, Bulten R, Kriellaars D. Physical Literacy, Physical Activity and Health: Toward an Evidence-Informed Conceptual Model. *Sport Med*. 2019;49(3):371-383. doi:10.1007/s40279-019-01063-3
26. Standal O. Phenomenology and pedagogy in physical education. 2016. https://books.google.nl/books?hl=nl&lr=&id=50h-CwAAQBAJ&oi=fnd&pg=PP1&dq=Phenomenology+and+pedagogy+in+physical+education+standal+2015&ots=1T6P5rb9hm&sig=IuyB-BVh_tmZWRj9-k5hnW8Mr5g. Accessed December 18, 2020.
27. ParticipACTION. *Canada's Physical Literacy Consensus Statement: ParticipACTION; 2017*. https://www.participaction.com/en-ca/resources/physical-literacy.
28. Gunnell KE, Longmuir PE, Barnes JD, Belanger K, Tremblay MS. Refining the Canadian Assessment of Physical Literacy based on theory and factor analyses. *BMC Public Health*. 2018;18. doi:10.1186/s12889-018-5899-2
29. Stodden DF, Langendorfer SJ, Goodway JD, et al. A developmental perspective on the role of motor skill competence in physical activity: An emergent relationship. *Quest*. 2008;60(2):290-306. doi:10.1080/00336297.2008.10483582
30. Robinson DB, Randall L. Marking Physical Literacy or Missing the Mark on Physical Literacy? A Conceptual Critique of Canada's Physical Literacy Assessment Instruments. *Meas Phys Educ Exerc Sci*. 2017;21(1):40-55. doi:10.1080/1091367X.2016.1249793
31. Lundvall S. Physical literacy in the field of physical education–A challenge and a possibility. *J Sport Heal Sci*. 2015;4(2):113-118. doi:10.1016/j.jshs.2015.02.001

32. Cairney J, Clark HJ, James ME, Mitchell D, Dudley DA, Kriellaars D. The Preschool Physical Literacy Assessment Tool: Testing a New Physical Literacy Tool for the Early Years. *Front Pediatr*. 2018;6:138. doi:10.3389/fped.2018.00138
33. Francis CE, Longmuir PE, Boyer C, et al. The Canadian Assessment of Physical literacy: Development of a model of children's capacity for a healthy, active lifestyle through a Delphi process. *J Phys Act Heal*. 2016;13(2):214-222. doi:10.1123/jpah.2014-0597
34. Longmuir PE, Tremblay MS. Top 10 Research Questions Related to Physical Literacy. *Res Q Exerc Sport*. 2016;87(1):28-35. doi:10.1080/02701367.2016.1124671
35. Tremblay MS, Costas-Bradstreet C, Barnes JD, et al. Canada's Physical Literacy Consensus Statement: Process and outcome 11 Medical and Health Sciences 1117 Public Health and Health Services. *BMC Public Health*. 2018;18(S2):1034. doi:10.1186/s12889-018-5903-x
36. Holsbeeke L, Ketelaar M, Schoemaker MM, Gorter JW. Capacity, Capability, and Performance: Different Constructs or Three of a Kind? *Arch Phys Med Rehabil*. 2009;90(5):849-855. doi:10.1016/j.apmr.2008.11.015
37. Tremblay MS, Longmuir PE, Barnes JD, et al. Physical literacy levels of Canadian children aged 8-12 years: Descriptive and normative results from the RBC Learn to Play-CAPL project. *BMC Public Health*. 2018;18(Suppl 2). doi:10.1186/s12889-018-5891-x
38. Bongers BC, Rijks EBG, Harsevoort AGJ, 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(5):679-686. doi:10.2522/ptj.20150082
39. Verschuren O, Zwinkels M, Ketelaar M, Reijnders-van Son F, Takken T. Reproducibility and Validity of the 10-Meter Shuttle Ride Test in Wheelchair-Using Children and Adolescents With Cerebral Palsy. *Phys Ther*. 2013;93(7):967-974. doi:10.2522/ptj.20120513
40. Bloemen MAT, de Groot JF, Backx FJG, Benner J, Kruitwagen CLJJ, Takken T. Wheelchair Shuttle Test for Assessing Aerobic Fitness in Youth With Spina Bifida: Validity and Reliability. *Phys Ther*. 2017;97(10):1020-1029. doi:10.1093/ptj/pzx075
41. 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*. 2017;98(6):1097-1103. doi:10.1016/j.apmr.2016.08.469
42. Sol ME, Verschuren O, de Groot L, de Groot JF, Fit-For-the-Future!-consortium. Development of a wheelchair mobility skills test for children and adolescents: combining evidence with clinical expertise. *BMC Pediatr*. 2017;17(1):51. doi:10.1186/s12887-017-0809-9
43. Sol ME, de Groot JF, Zwinkels M, Visser-Meily JMA, Kruitwagen CLJJ, Verschuren O. Utrecht Pediatric Wheelchair Mobility Skills Test: Reliability, Validity, and Responsiveness in Youths Using a Manual Wheelchair. *Phys Ther*. 2019;99(8):1098-1106. doi:10.1093/ptj/pzz061
44. Field DA, Livingstone RW. Power mobility skill progression for children and adolescents: a systematic review of measures and their clinical application. *Dev Med Child Neurol*. 2018;60(10):997-1011. doi:10.1111/dmcn.13709

45. James D, Pfaff J, Jeffries LM. Modified Ride-on Cars as Early Mobility for Children with Mobility Limitations: A Scoping Review. *Phys Occup Ther Pediatr*. 2019;39(5):525-542. doi:10.1080/01942638.2018.1547808
46. Van Der Woude LHV, Veeger HEJ, Dallmeijer AJ, Janssen TWJ, Rozendaal LA. Biomechanics and physiology in active manual wheelchair propulsion. *Med Eng Phys*. 2001;23(10):713-733. doi:10.1016/S1350-4533(01)00083-2
47. Rosenbaum P, Gorter JW. The "F-words" in childhood disability: I swear this is how we should think. *Child Care Health Dev*. 2012;38(4):457-463. doi:10.1111/j.1365-2214.2011.01338.x
48. Klokhuis. Rolstoelskills. https://www.hetklokhuis.nl/tv-uitzending/4219/Rolstoelskills.
49. Te Velde SJ, Lankhorst K, Zwinkels M, et al. Associations of sport participation with self-perception, exercise self-efficacy and quality of life among children and adolescents with a physical disability or chronic disease-a cross-sectional study. *Sport Med–open*. 2018;4(1):38. doi:10.1186/s40798-018-0152-1
50. Van Wely L, Balemans ACJ, 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(1):40-49. doi:10.1016/j.jphys.2013.12.007
51. 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(5):448-455. doi:10.1111/j.1469-8749.2009.03609.x
52. Bandura A, Freeman W, Lightsey R. *Self-Efficacy: The Exercise of Control*. New York; 1999. https://connect.springerpub.com/content/sgrjcp/13/2/158.full.pdf. Accessed June 9, 2020.
53. Schulz PJ, Nakamoto K. Health literacy and patient empowerment in health communication: The importance of separating conjoined twins. *Patient Educ Couns*. 2013;90(1):4-11. doi:10.1016/j.pec.2012.09.006
54. Longmuir PE, Boyer C, Lloyd M, et al. The Canadian Assessment of Physical Literacy: Methods for children in grades 4 to 6 (8 to 12 years). *BMC Public Health*. 2015;15(1). doi:10.1186/s12889-015-2106-6
55. Tremblay MS, Carson V, Chaput JP, et al. Canadian 24-hour movement guidelines for children and youth: An integration of physical activity, sedentary behaviour, and sleep. *Appl Physiol Nutr Metab*. 2016;41(6):S311-S327. doi:10.1139/apnm-2016-0151
56. van der Woude LHV, de Groot S, Janssen TWJ. Manual wheelchairs: Research and innovation in rehabilitation, sports, daily life and health. *Med Eng Phys*. 2006;28(9):905-915. doi:10.1016/j.medengphy.2005.12.001
57. Martin Ginis KA, Van Der Scheer JW, Latimer-Cheung AE, et al. Evidence-based scientific exercise guidelines for adults with spinal cord injury: An update and a new guideline. *Spinal Cord*. 2018;56(4):308-321. doi:10.1038/s41393-017-0017-3
58. Sprigle S, Huang M. Impact of Mass and Weight Distribution on Manual Wheelchair Propulsion Torque. *Assist Technol*. 2015;27(4):226-235. doi:10.1080/10400435.2015.1039149
59. Kamalakannan S, Rushton PW, Giesbrecht E, et al. Wheelchair service provision education for healthcare professional students, healthcare personnel and educators across low- to high-resourced settings: a

scoping review protocol. *Disabil Rehabil Assist Technol*. December 2020:1-7. doi:10.1080/17483107.2020.1852325

60. Ridgers ND, McNarry MA, Mackintosh KA. Feasibility and Effectiveness of Using Wearable Activity Trackers in Youth: A Systematic Review. *JMIR mHealth uHealth*. 2016;4(4):e129. doi:10.2196/mhealth.6540
61. Bronfenbrenner U. Ecological systems theory. In: *Annals of Child Development*. ; 1989:185–246.
62. Davison KK, Lawson CT. Do attributes in the physical environment influence children's physical activity? A review of the literature. *Int J Behav Nutr Phys Act*. 2006;3(1):1-17. doi:10.1186/1479-5868-3-19
63. Van Melik R, Althuizen N. Inclusive Play Policies: Disabled Children And Their Access To Dutch Playgrounds. *Tijdschr voor Econ en Soc Geogr*. July 2020:tesg.12457. doi:10.1111/tesg.12457
64. Bronfenbrenner U. Environments in developmental perspective: Theoretical and operational models. In: *Measuring Environment across the Life Span: Emerging Methods and Concepts.* American Psychological Association; 2004:3-28. doi:10.1037/10317-001
65. Bamm EL, Rosenbaum P. Family-centered theory: Origins, development, barriers, and supports to implementation in rehabilitation medicine. *Arch Phys Med Rehabil*. 2008;89(8):1618-1624. doi:10.1016/j.apmr.2007.12.034
66. Alsem MW, Siebes RC, Gorter JW, Jongmans MJ, Nijhuis BGJ, Ketelaar M. Assessment of family needs in children with physical disabilities: development of a family needs inventory. *Child Care Health Dev*. 2014;40(4):498-506. doi:10.1111/cch.12093
67. Sanders KY. Overprotection and lowered expectations of persons with disabilities: The unforeseen consequences. *Work*. 2006;27(2):181-188. https://pubmed.ncbi.nlm.nih.gov/16971765/. Accessed August 21, 2020.
68. Clarke K, Cooper P, Creswell C. The Parental Overprotection Scale: Associations with child and parental anxiety. *J Affect Disord*. 2013;151(2):618-624. doi:10.1016/j.jad.2013.07.007
69. Bloemen MAT, van den Berg-Emons RJG, Tuijt M, et al. Physical activity in wheelchair-using youth with Spina Bifida: an observational study. *J Neuroeng Rehabil*. 2019;16(1):9. doi:10.1186/s12984-018-0464-x
70. https://wheelchairskillsteam.nl/.
71. Best KL, Miller WC, Huston G, Routhier F, Eng JJ. Pilot study of a peer-led wheelchair training program to improve self-efficacy using a manual wheelchair: A randomized controlled trial. *Arch Phys Med Rehabil*. 2016;97(1):37-44. doi:10.1016/j.apmr.2015.08.425
72. Worobey LA, Kirby RL, Heinemann AW, et al. Effectiveness of Group Wheelchair Skills Training for People With Spinal Cord Injury: A Randomized Controlled Trial. *Arch Phys Med Rehabil*. 2016;97(10):1777-1784.e3. doi:10.1016/j.apmr.2016.04.006
73. Kirby RL, Smith C, Billard JL, Irving JDH, Pitts JE, White RS. *Training a Parent in Wheelchair Skills to Improve Her Child's Wheelchair Skills: A Case Study.*; 1700.
74. Schuiling G, Kiewiet DJ. Action Research: Intertwining three exploratory processes to meet the competing demands of rigour and relevance. *Electron J Bus Res Methods*. 2016;14:111-124. www.ejbrm.com. Accessed December 18, 2020.

75. Schuiling G, Vermaak H. Four contexts of action research: Crossing boundaries for productive interplay. *Int J Action Res*. 2017;13(1):5-23. doi:10.3224/IJAR.V13I1.02
76. Barnett AG, van der Pols JC, Dobson AJ. Regression to the mean: What it is and how to deal with it. *Int J Epidemiol*. 2005;34(1):215-220. doi:10.1093/ije/dyh299

Nederlandse samenvatting

ACHTERGROND

Kinderen en jongeren met een beperkte loopfunctie door bijvoorbeeld Cerebrale Parese of Spina Bifida, maken in het dagelijks leven gebruik van een handbewogen rolstoel. In Nederland worden kinderen met een fysieke beperking, zoals bij Cerebrale Parese of Spina Bifida, vanaf het moment van diagnose doorgestuurd naar een kinderrevalidatie team om hun gezondheid en ontwikkeling te ondersteunen en stimuleren. Om hun participatie te bevorderen krijgen veel van deze kinderen al op jonge leeftijd een rolstoel om binnen en/of buitenshuis te gebruiken. Hoewel het oefenen en leren omgaan met hulpmiddelen (bijvoorbeeld een spalk of prothese) een belangrijk onderdeel is van een revalidatiebehandeling, is er in de kinderrevalidatie onvoldoende aandacht voor het zelfstandig leren rijden en manoeuvreren in een rolstoel. Dit moet veranderen, omdat het ontwikkelen van rolstoelvaardigheden de zelfredzaamheid kan vergroten en kinderen/jongeren dan meer mogelijkheden hebben om mee te kunnen doen in de maatschappij. Hierbij kan gedacht worden aan het buiten spelen of zelfstandig naar een vriendje gaan. Om kinderen en jongeren in dit proces te begeleiden en ondersteunen in het belangrijk om de rolstoelvaardigheden van kinderen in kaart te kunnen brengen. Zowel in praktijk als onderzoek is er tot op heden nog nauwelijks aandacht geweest voor het meten en trainen van rolstoelvaardigheid bij kinderen.

Het doel van dit proefschrift is om bij te dragen aan het verbeteren van rolstoelvaardigheid bij kinderen en jongeren in een handbewogen rolstoel. In dit proefschrift willen we 1) valide en betrouwbare meetinstrumenten ontwikkelen om de rolstoelvaardigheid en het zelfvertrouwen in het gebruik van de rolstoel in kaart te brengen; 2) de validiteit bepalen van een activiteiten monitor bij kinderen en jongeren in een rolstoel; 3) evalueren wat de effecten zijn van een combinatie van rolstoelvaardigheidstraining en fitheidstraining op de fysieke activiteit, fitheid, zelfvertrouwen en de rolstoelvaardigheid bij kinderen en jongeren in een handbewogen rolstoel.

WAT ZIJN DE UITKOMSTEN VAN HET ONDERZOEK?

Deel 1: Ontwikkeling van meetinstrumenten

Hoofdstuk 2 is een combinatie van kwalitatief en kwantitatief onderzoek. De huidige kennis uit de literatuur over rolstoelvaardigheidstesten bij volwassenen is gecombineerd met de mening van kinderen en jongeren, ouders en de klinische expertise van zorgprofessionals over welke rolstoelvaardigheden relevant zijn voor kinderen en jongeren in een handbewogen rolstoel. Op deze manier zijn 15 items, zoals voorwaarts rijden, slalom, verhoging op- en afrijden geselecteerd. Deze items vormen samen de nieuwe rolstoelvaardigheidstest voor kinderen en jongeren. Vervolgens is de haalbaarheid van het afnemen van de nieuw ontwikkelde rolstoelvaardigheidstest voor kinderen en jongeren geëvalueerd. In **hoofdstuk 3** zijn de psychometrische eigenschappen van deze rolstoelvaardigheidstest verder geëvalueerd. In de methode staat beschreven hoe de tijdscore per item omgezet kan worden naar een ordinale score, waarmee een totaal score voor de test kan worden berekend. De gevonden resultaten voor de betrouwbaarheid, validiteit en responsiviteit laten zien dat deze test goed te gebruiken is bij kinderen en jongeren om hun rolstoelvaardigheid te meten.

In het onderzoek beschreven in **hoofdstuk 4** is er een combinatie van kwalitatief en kwantitatief onderzoek gebruikt om een bestaande vragenlijst over het 'zelfvertrouwen in gebruik van de rolstoel' voor volwassenen aan te passen voor kinderen en jongeren. In een eerste fase is er een vertalingsproces doorlopen, waarbij de originele Canadese versie die ontwikkeld was vertaald is naar het Nederlands. Vervolgens zijn focusgroepen gehouden met kinderen, jongeren, ouders en zorgprofessionals met als onderwerpen; duidelijkheid over het begrip zelfvertrouwen voor kinderen en jongeren, en de geschiktheid van bestaande vragen voor de Nederlandse situatie. Op basis van deze informatie is de vragenlijst aangepast zodat deze geschikt is voor kinderen en jongeren in een handbewogen rolstoel in Nederland. In een laatste fase is de interne consistentie van de vragenlijst als goed beoordeeld. Ook zagen we een duidelijk verband tussen de uitkomsten op de zelfvertrouwen vragenlijst en de resultaten van de rolstoelvaardigheidstest, wat duidt op een goede construct validiteit. Dit was niet bij iedereen het geval. Een aantal deelnemers liet veel zelfvertrouwen zien bij een lage score op de rolstoelvaardigheidstest. Het is daarom belangrijk om zowel het zelfvertrouwen als de rolstoelvaardigheid te meten bij kinderen en jongeren in een handbewogen rolstoel.

Deel II Validiteit van een activiteiten monitor

In **hoofdstuk 5** rapporteren we dat de criterium validiteit van een activiteiten monitor (Activ8) voor het meten van 'actief rolstoel gebruik' bij kinderen en jongeren in een handbewogen rolstoel goed is. Deze activiteiten monitor is in eerder onderzoek gevalideerd bij volwassenen in een rolstoel, waarbij er 1 sensor op de pols werd geplaatst en 1 sensor dichtbij de as van het grote wiel. Door de combinatie van deze 2 sensoren is het mogelijk om actief rolstoel gebruik (rijden en manoeuvreren) te onderscheiden van bijvoorbeeld geduwd worden in een rolstoel (wielsensor is actief, polssensor is niet actief) of stilstaan. Hoewel de activiteiten monitor goed 'activiteit' kan detecteren, is deze niet goed om verschillende activiteiten in de rolstoel te onderscheiden. Zo is de activiteitenmonitor bijvoorbeeld niet in staat om het onderscheid te maken tussen snel rijden en langzaam rijden.

Deel III effect van interventie

In **hoofdstuk 6** beschrijven we de positieve lange termijn effecten van een combinatie van rolstoelvaardigheidstraining en fitheid training op de fysieke activiteit, aerobe en anaerobe prestatie, zelfvertrouwen in het gebruik van de rolstoel en rolstoelvaardigheid. Vanuit zes revalidatiecentra/mytylscholen in Nederland hebben 60 kinderen of jongeren meegedaan aan deze interventie studie, waarbij ze getraind hebben in groepjes van 4-6 deelnemers onder begeleiding van een kinderfysiotherapeut, ergotherapeut en/of docent lichamelijke opvoeding. De fitheidstraining bestond uit twee sessies per week waarin een high-intensity-interval trainingsprotocol werd uitgevoerd gedurende acht weken. De rolstoelvaardigheidstraining bestond ook uit twee sessies per week gedurende acht weken. Hierbij werden er drie sessies (start, midden, eind) gegeven door KJ-projects. KJ-projects is een bedrijf dat al meer dan 10 jaar ervaring heeft in het geven van rolstoelvaardigheidstraining bij kinderen en volwassenen. Ouders/verzorgers waren aanwezig bij deze sessies, zodat zij konden zien hoe vaardig hun kind al is, hoe nieuwe vaardigheden konden worden geoefend en welke vaardigheden in een rolstoel nog meer mogelijk waren. Het thuis oefenen (met ouders/verzorgers) werd ondersteund door middel van video opnames met gepersonaliseerde instructies voor het kind.

Uit de resultaten van de studie bleek dat de volgorde van trainen (rolstoelvaardigheidstraining voor of na fitheid training) geen effect had op de meeste uitkomstmaten, behalve voor zelfvertrouwen in het gebruik van de rolstoel. Er werden geen effecten van de training gevonden op de cardiorespiratoire fitheid (shuttle ride test ($VO_{2peak)}$), terwijl de aerobe prestatie (shuttle ride test (aantal trappen)) wel omhoog ging.

WAT ZIJN DE KLINISCHE IMPLICATIES EN RICHTINGEN VOOR VERVOLG ONDERZOEK?

Hoofdstuk 7, de algemene discussie, geeft een kort overzicht van de belangrijkste bevindingen van dit proefschrift. Vervolgens wordt er dieper ingegaan op deze resultaten en wordt bekeken of er nieuwe richtingen voor vervolg onderzoek en implicaties voor de praktijk kunnen ontstaan door de resultaten te bespreken vanuit een nieuw perspectief binnen de gezondheidszorg genaamd ‘Physical Literacy’. Dit perspectief op fysieke activiteit, gaat uit van de dynamische interactie tussen fysieke factoren (motorische capaciteit), affectieve factoren (het zelfvertrouwen en motivatie om te bewegen) en cognitieve factoren (kennis en begrip over het belang van bewegen) om te komen tot, en het behouden van, fysieke activiteit (leven lang bewegen). In dit hoofdstuk wordt besproken hoe dit concept toegepast kan worden op kinderen en jongeren in een handbewogen rolstoel en op welke manieren rolstoelvaardigheidstraining mogelijk invloed heeft op het ontwikkelen van Physical Literacy. De rolstoelvaardigheidstrainingen hadden bijvoorbeeld een positief effect op het vergroten van het zelfvertrouwen in de rolstoel, waardoor mogelijk de motivatie om zelf te bewegen in de rolstoel groter werd.

Voor de dagelijkse praktijk heeft dit proefschrift een aantal belangrijke implicaties opgeleverd; 1) het is belangrijk om van jongs af aan bewustwording van de mogelijkheden in een rolstoel (vaardigheid en fysieke activiteit) bij kinderen en hun ouders/verzorgers te ontwikkelen, 2) scholing van zorgprofessionals over rolstoelvaardigheid is belangrijk, zodat zij kinderen/jongeren en hun ouders beter kunnen ondersteunen in het selecteren van een (actieve) rolstoel, leren over onderhoud van de rolstoel (bandenspanning!) en oefenen van rolstoelvaardigheden, 3) het is belangrijk om zowel de rolstoelvaardigheid als het zelfvertrouwen in het gebruik van de rolstoel op verschillende momenten in de ontwikkeling te meten, om zo samen met kind en ouders/verzorgers te kunnen bepalen of rolstoelvaardigheidstraining is geïndiceerd.

In de aanbevelingen voor vervolg onderzoek wordt de noodzaak voor verder onderzoek in deze doelgroep, kinderen en jongeren in een handbewogen rolstoel, besproken met daarbij enkele methodologische overwegingen. Ook worden aanbevelingen gedaan over verder onderzoek naar; 1) de professionele ontwikkeling op het gebied van de rolstoel en rolstoelvaardigheid bij kinderfysiotherapeuten en ergotherapeuten, 2) het meten en trainen van rolstoelvaardigheid, 3) het verbeteren van de fysieke activiteit en/of physical literacy bij kinderen en jongeren in een handbewogen rolstoel.

Dankwoord

Ja, daar zijn we dan! Het schrijven van mijn dankwoord voor de afronding van mijn mooie en leerzame Phd-traject. Ik heb ontzettend veel geleerd en met heel veel plezier en enthousiasme aan de onderzoeken in dit proefschrift gewerkt. Ik voel mij echt bevoorrecht dat ik tijdens dit traject omringd ben geweest door betrokken collega's, een ondersteunend en motiverend promotie team en mijn lieve vrienden en familie. Naast dat ik nog mensen specifiek wil bedanken, wil ik allereerst alle **kinderen en jongeren en de ouders** bedanken die de afgelopen jaren meegedaan hebben aan interviews, focusgroepen, vragenlijsten en alle testen die onderdeel waren van de dataverzameling. Het waren vooral jullie ervaringen tijdens en na de rolstoelvaardigheidstrainingen die mij een enorme 'drive' gaven om dit onderwerp meer aandacht te geven in de praktijk, het onderwijs en de wetenschap.

Als eerste wil ik mijn promotieteam als '**team**' bedanken. Ik leg hier de nadruk op 'team', omdat ik mij als teamspeler heb gevoeld in onze gezamenlijke missie om hier een mooi en relevant proefschrift van te maken. De combinatie van jullie expertise en de persoonlijke betrokkenheid van jullie alle drie maakte ons tot een fijn promotie team. Jullie waren er altijd om mijn werk de goede richting op te begeleiden of om aan te geven dat het goed was om stapje terug te doen toen ik ziek werd door mijn zwangerschappen. Ik kon mij geen beter promotieteam wensen en wil jullie daarom als team en individueel bedanken voor jullie begeleiding.

Beste **Janke**, bedankt voor jouw fijne, goede en motiverende begeleiding. Vanaf onze eerste ontmoeting werd het mij al snel duidelijk dat wij dezelfde passie voor onderzoek hadden, waarin het altijd gaat over onderzoek waar je in de praktijk echt iets aan hebt. Daarnaast hebben wij op zoveel andere vlakken overlap: studeren aan de K.U. Leuven, getrouwd met een Engelstalige man en nu heeft mijn dochter ook nog dezelfde geboortedatum als jij. Ik heb zo ontzettend veel van jou geleerd over het doen van praktijkgericht onderzoek, maar vooral ook over de balans tussen werk en privé. Jouw ideeën voor het dissemineren van onderzoeksresultaten (zoals de klokhuisprijs), leken mij soms buiten mijn bereik, maar daar durfde ik dankzij jou motivering toch voor te gaan. Ik was dan ook echt in lichte paniek toen jij aangaf weg te gaan bij de Hogeschool Utrecht, maar kwam er gelukkig al snel achter dat jouw begeleiding op dezelfde manier doorging. Ik hoop nog lang met je te mogen samenwerken!

Beste **Olaf**, op mijn werk als kinderfysio bij de mytylschool Utrecht had ik al veel van mijn collega's gehoord over hun oud-collega Olaf. Jij had jezelf vanuit jouw baan als fysiotherapeut doorontwikkeld tot onderzoeker binnen de Hoogstraat, waarbij je een onderwerp vanuit de praktijk hebt onderzocht en de resultaten hebt laten terugvloeien in het dagelijks handelen van de kinderfysiotherapeut. Het combineren van praktijk en onderzoek was voor mij de reden om bij de Hoogstraat te gaan werken en ik had dan in jou direct het perfecte voorbeeld als co-promotor. Jouw expertise over het doen van onderzoek op mytylscholen heeft mij ontzettend geholpen bij de onderzoeksopzet en dataverzameling. Ook al was ons kantoor niet op dezelfde werkplek, jij was altijd snel en makkelijk bereikbaar om mee te denken aan een praktische oplossing voor een probleem waar ik mee zat. Ik wil jou bedanken voor jouw mogelijkheden om te denken in oplossingen, oog voor detail, corrigeren van mijn typfouten J, is het nou where of were? en tegelijk ook jouw focus voor het grotere verhaal als ik met mijn kop in de details vast zat en verdwaald was in het bos van de woorden, cijfers en statistiek. Ik hoop nog vaak met jou te mogen samenwerken.

Beste **Anne**, als promotor sloot jij als laatste aan bij mijn promotieteam en maakte het team compleet. Ik wil jou bedanken voor jouw begeleiding die mijn artikelen en onderzoek naar een hoger niveau hebben getild en nog belangrijker jou kritische blik waardoor altijd de klinische relevantie van het onderzoek op de voorgrond kwam. Ik denk dat er bij elke overleg telkens dezelfde vraag was, waarvan ik eerst een beetje van slag raakte en daarna inzag dat het beter en duidelijker omschreven moest worden. De 'waarom doe je dit' vraag. Waar het voor mijzelf vaak al logisch was waarom ik bepaalde keuzes had gemaakt, nodigde jij mij uit om te blijven denken over de onderbouwing van die keuzes. Ik wil jou bedanken voor je betrokkenheid zowel persoonlijk als professioneel, jouw enthousiasme voor dit onderwerp, jouw duidelijke feedback en de stimulatie die jij mij gaf om onderzoek te doen en te blijven doen. Ik hoop dat wij elkaar in de toekomst blijven ontmoeten en dat ik ook dan weer veel van jou kan leren.

Geachte **leden van de beoordelingscommissie**, prof. dr. C. Veenhof, prof. dr. R.H.H. Engelbert, prof. dr. M.J. Jongmans, prof. dr. W.L. van der Pol en prof. dr. A.I. Buizer, graag wil ik jullie bedanken voor de tijd en aandacht die jullie aan de beoordeling van mijn proefschrift hebben besteed. Ik vind het een eer dat jullie mijn proefschrift wilden lezen en beoordelen en kijken uit naar onze gedachtewisseling tijdens mijn verdediging.

Onderzoek doen bij kinderen en jongeren in een rolstoel op de scholen was een grote logistieke uitdaging. Ik wil daarom alle betrokken **therapeuten** van mytylschool De kleine Prins, Merem Revalidatie, mytylschool Roosendaal, Roessingh Revalidatie, Libranet en Heliomare bedanken en specifiek: Laura, Jorinde, Gert, Sanne, Marleen, Femke, Desirée, Bram, Allard, Gerben, Maaike, Marieke en Saskia. Ook wil ik alle **studenten** bedanken die geholpen hebben bij de dataverzameling op de mytylscholen en bij de verwerking en analyses van de onderzoeksgegevens!

Beste **Harriët**, bedankt voor de ontwikkeling die ik als junior onderzoeker naar Phd-student en nu ook als post-doc onderzoeker mag doormaken. Het is de manier waarop jij jouw rol als Lector vertolkt, dat ik een hele fijne samenwerking heb met jou en met alle collega onderzoekers. Jij hebt het mogelijk gemaakt dat wij een goede samenwerking hebben in de projecten, maar ook in het gevraagd en ongevraagd feedback geven op artikelen, onderzoeksvoorstellen of congres praatjes. Mede dankzij deze ondersteuning ben ik nu trots op de artikelen die in dit boekje staan. Bedankt voor het stimuleren van de peer-support gesprekken, koffie kletspraatjes, knuffels/kaartjes voor mijn kinderen, vertrouwen in mijn kunde als onderzoeker, en sinds COVID-19 de inspirerende wandelingen langs Rhijnauwen.

Lieve **(oud-)collega's van het LLG**: Manon, Marike, Imke, Eline, Janke O., Stefan, Tim, Kristel, Jacqueline N. Jacqueline O, Jan, Henri, Francois, Edwin, Michiel, Karlijn, Barbara, Ryan, Marlies, Martine, Han, Hannelies, Claudia en Sonja! Ik wil jullie allemaal bedanken voor de fijne samenwerking die wij als collega onderzoekers met elkaar hebben. Ik heb ontzettend veel geleerd van jullie op professioneel gebied, maar ook over hoe je werk en privé kan combineren of juist waar de uitdagingen daarin liggen J. Ik wil jullie bedanken voor jullie kritische feedback op mijn praatjes bij de peer-support groepen of KK overleggen, maar vooral ook voor de gezelligheid op meetdagen (Tim), congresbezoekjes, team'uitjes en de vele koffiekletspraatjes. Kristel, ik wil jou bedanken voor onze goede samenwerking in het afschrijven van het artikel in hoofdstuk 5.

Lieve **Marleen en Tess,** bedankt voor het delen van jullie ervaringen als ouder en kind. Tess, wat was je geweldig in de Klokhuisaflevering! Super mooi hoe jij hebt laten zien, hoe stoer het is om goede skills in je rolstoel te hebben. En Marleen, bellen met jou is altijd een leuke start met de 'Marleen' uitwisselingen. Bedankt voor het mooie voorwoord en ik wens je heel veel succes in je nieuwe carrière in de revalidatie wereld!

Lieve **Paranimfen** Manon, Marike en Lidwien. Of jullie er allemaal bij kunnen zijn tijdens mijn promotie is onzeker door de COVID-maatregelen, maar ik weet in ieder geval zeker dat ik zonder jullie nooit tot dit punt was gekomen. **Manon**, jouw promotieonderzoek was een van de aanleidingen om onderzoek te gaan doen naar rolstoelvaardigheden bij kinderen. We hebben daarom vanaf het begin gelijk veel samengewerkt, waarbij jouw expertise, motivatie en talent voor onderzoek mij ontzettend veel hebben geleerd. Ik ben vooral ook dankbaar voor de gezellige praatjes in de speeltuin met eerst jouw kinderen en daarna ook de mijne, en al die koffie'tjes die wij bij jou of mij thuis hebben gedronken of toch even wandelen om lekkere koffie te halen bij Cornelius. In onze overleggen lopen privé en werk onderwerpen vloeiend in elkaar over, waarbij wij beginnen over speelgoed van Lily en Faas dat leuk is voor Lara of Dain en eindigen in een leuke discussies over waar we naar toe willen met onze onderzoekslijnen of waar jij of ik zelf heen wil als onderzoeker. Ik hoop nog veel met jou samen te werken en koffie te drinken! **Marike**, zonder 'ie'. Samen met jou als collega docent en onderzoeker kon ik alle pieken en dalen in het doen van onderzoek, schrijven van artikels en combineren van onderzoek met de piekdrukte in het onderwijs delen. Omdat jouw Phd- traject ongeveer in dezelfde stadia verliep als het mijne, heb ik heel veel steun aan jou gehad om door te gaan waar het even minder snel/soepel ging dan verwacht en ook om de mooie prestaties te vieren. Zeker in het laatste jaar hebben wij ondanks het thuiswerken veel contact gehad via Teams of toch even live waar dat kon. Ik wil jou bedanken voor je lieve luisterende oor, kritische feedback, onze gedeelde ontdekkingstocht om het Phd-gebergte te beklimmen en de doorgestuurde qoutes zoals *"the only way to do it….. is to do it!"*. Ik kijk al uit naar jou Phd-feestje dat er vast snel aankomt! **Lidwien**, mijn kleine zusje. Jij bent in zoveel opzichten hetzelfde als ik en tegelijk toch ook weer heel anders. Ik vind het ontzettend fijn dat jij naast mij staat bij alle grote gebeurtenissen in mijn leven. Ik wil jou bedanken dat jij er altijd voor mij bent en de gezelligheid brengt in onze vakanties, familie uitjes digitaal of live en hopelijk nog op vele spelletjes avonden, omni-toernooi, skivakanties en noem maar op! Lara (en Dain) willen heel graag op vakantie met Aunty Lien (uit te spreken als anti-lien) en ook ik hoop nog heel veel mooie herinneringen met jou te maken. Jouw Phd-traject ging anders dan verwacht en gehoopt, maar des te meer bewonder ik jou doorzettingsvermogen om jou Phd nu verder af te maken. Ik heb er alle vertrouwen dat jij dit kunt en ben er altijd mocht je hulp nodig hebben of willen feesten.

Lieve **oud-collega's van Mytylschool De kleine prins/Utrecht**, jullie zijn met teveel om bij naam te noemen, maar verdienen zeker een groot dankwoord van mij. Ik wil jullie bedanken voor het mede-mogelijk maken van dit proefschrift. Jullie waren fantastische collega's om mee samen te werken en hebben jullie altijd heel flexibel opgesteld om werkdagen te ruilen/overnemen, zodat ik mijn onderzoek kon uitvoeren. En niet te vergeten de gezellige borrels bij Buurten! Jullie school mag echt een voorbeeld zijn voor andere`n, over hoe je rolstoelvaardigheid onderdeel kan maken van de zorg die jullie aanbieden. Ik wil wel 1 iemand specifiek bedanken en dat is Laura. Het was jouw werk over de rolstoelvaardigheidstest die de basis heeft gevormd voor de test beschreven in hoofdstuk 2 en 3. En het is ook onder meer jouw passie voor dit onderwerp waardoor de rolstoelvaardigheidstrainingen op de mytylschool zo succesvol draaien. Ik hoop dat wij nog lang blijven samenwerken en dat ons rolstoelvaardigheidswerk nog veel kinderen mag bereiken.

Lieve **mede phd-studenten** van mijn TULIPS 2017 groep en mentor **Prof. Louis Bont**, bedankt voor het delen van jullie eigen ervaringen in jullie onderzoek en het luisteren en meedenken met mijn phd-reis. Dit netwerk heeft mij ontzettend gestimuleerd om mijn kop uit het Phd zand te halen om vooruit te kijken naar hoe ik mijn onderzoek, praktijk en prive leven vorm wil geven in de toekomst.

En dan KJ-projects, iedereen van het Wheelchairskillsteam en specifiek natuurlijk **Kees-Jan**. Het was jouw ervaring in het geven van rolstoelvaardigheidstraining en de 'drive' om rolstoelvaardigheid op de kaart te zetten in Nederland dat wij zijn gaan samenwerken. Ik bewonder jouw passie en vechtlust om je hard te maken voor een onderwerp dat jij belangrijk vindt. Ik wil jou bedanken voor al jouw kennis en ervaring die jij gedeeld hebt met mij, studenten, fysiotherapeuten, ergotherapeuten, kinderen en hun ouders over rolstoelvaardigheid. Ik hoop dat wij samen onze plannen om een scholing te ontwikkelen voor therapeuten, ervaringsdeskundige rolstoelvaardigheidstrainers en/of ouders kunnen uitvoeren en op die manier nog meer kinderen en jongeren in een rolstoel en hun ouders kunnen bereiken.

Ik wil nog speciaal **Sam en Sarah** van het stripboek bedanken voor de illustraties die ik heb mogen delen in dit boek. Voor iedereen die na het zien van deze plaatjes interesse heeft gekregen in het stripboek kan je dit bestellen via samensarah.nl.

Lieve **familie, vrienden en vriendinnen**, bedankt voor jullie interesse in mijn onderzoek, maar vooral ook de gezellige momenten buiten het werk. Jullie zijn er altijd om samen mooie dingen te beleven en het werk even helemaal te vergeten. Ik wil nog specifiek **Kim** en **Tarunai** bedanken voor het vele oppassen, spelen met de kinderen en vooral ook de gezelligheid als de kinderen eindelijk slapen! Dear **Jenny, Brian, Mike, Sean and Lisa,** thank you for being a loving and kind family to me and my children. Lieve **papa en mama,** heel veel dank voor jullie ondersteuning aan mij en mijn gezin, de liefde en geloof in mijn kunnen als therapeut, onderzoeker en mens. Dankzij jullie heb ik mij zelf kunnen ontwikkelen tot de professional die ik nu ben, waarbij ik veel gebruik maak van de networking skills die ik geleerd heb van papa en van het creatieve oplossingsgericht denken van mama. Lieve **Jeroen, Lieke, Karlien, Poini, Lidwien, Mike, nichtjes en neefjes,** ik ben zo blij met onze gezellige ietwat drukke familie, hopelijk maken we nog veel mooie herinneringen samen.

Lara and Dain, being with you two is the most precious and wonderful experience of my life. I hope you two will keep on causing lots of trouble and laughter together! My dear sweet **Phelie**, thank you for all your support, proof reading of all my articles multiple times, your love, kind words and faith in me as a professional and as a mother. Ik hou van jou!

About the author

CURRICULUM VITAE

Marleen Sol was born in Breda, the Netherlands on August 8th, 1985. She studied 'Rehabilitation science and Physiotherapy' at the KU Leuven in Belgium. During her studies she volunteered in sport ski camps for children with a disability (To Walk Again). Here she experienced the joy and growth in confidence when children with a disability learned to ski down the slopes of Austrian Alps. Through this experience she started getting interested in promoting physical activity in children with a disability. In her master Rehabilitation Science and Physiotherapy, she specialized in pediatrics and started to have the ambition to improve the quality of care for children with a disability through research. In 2011 she completed her thesis on the clinical and neurological determinants of upper limb movement pathology in children with hemiplegic cerebral palsy and graduated Magna Cum Laude.

In 2011 she started to gain experience as a pediatric physical therapist at Rehabilitation Center De Hoogstraat in the school for special education (Mytylschool De kleine prins Utrecht) where she worked for nine years. She immediately got the opportunity to participate in a research project as part of 'LondenTIMES 2012', a project in which eight (ex)-patients with a sports ambition of De Hoogstraat went to visit the Paralympics 2012. After working for four years as a pediatric physical therapist, her interest in promoting physical activity in children with a disability and her ambition to become a researcher came together in a junior research position in the research project called 'Fit For the Future' from the HU University of Applied Sciences and the Center of Excellence for Rehabilitation Science (a collaboration between the University Medical Center Utrecht and De Hoogstraat Rehabilitation). While continuing to work as a pediatric physical therapist, she started her Phd on wheelchair mobility skills in youth using a manual wheelchair as part of the 'Fit for the Future' project in 2015. In the same year, she also volunteered to be the cofounder of a sports club for children with a disability in Utrecht, called Only Friends Utrecht. Here, she continue to work as a volunteer in the board and as a sports instructor. During her Phd she was part of the TULIPS Phd Program 2017-2019 (Training Upcoming Leaders in Pediatric Science) and started working as a lecturer at the Master Program Physical Therapy of the HU University of Applied Science in 2019. Marleen is currently living in Utrecht with her husband Phelie Maguire and their two children, Lara and Dain.

INTERNATIONAL PUBLICATIONS

Sol ME, Verschuren O, de Groot L, de Groot JF, Fit-For-the-Future!-consortium. Development of a wheelchair mobility skills test for children and adolescents: combining evidence with clinical expertise. BMC Pediatr. 2017;17(1):51.

Sol ME, de Groot JF, Zwinkels M, Visser-Meily JMA, Kruitwagen CLJJ, Verschuren O. Utrecht Pediatric Wheelchair Mobility Skills Test: Reliability, Validity, and Responsiveness in Youths Using a Manual Wheelchair. *Phys Ther*. 2019;99(8):1098-1106.

Sol ME, Bloemen MAT, Verschuren O, Rushton PW, Visser-Meily JMA, de Groot JF. Wheelchair Mobility Confidence Scale for Dutch youth using a manual wheelchair. Disabil Rehabil Assist Technol 2019 Apr 23;1–8.

Lankhorst K, **Sol ME**, van den Berg-Emons RJG, Horemans H, de Groot J. The criterion validity of the Activ8 to measure physical activity in youth using a wheelchair. (*Submitted)*.

Sol ME, Verschuren O, Horemans H, Westers P, Visser-Meily J.M.A, Fit-for-the-Future Consortium, De Groot J.F. The effects of wheelchair mobility skills and exercise training on physical activity, fitness, and confidence in youth using a manual wheelchair. *Accepted for publication in Disability and Rehabilitation.*

INTERNATIONAL PRESENTATIONS

Sol ME, Verschuren O, de Groot L, de Groot JF, Fit-For-the-Future!-consortium. Development of a wheelchair mobility skills test for children and adolescents: combining evidence with clinical expertise. Oral Presentation at the World Conference Physical Therapy 2017

Bloemen M, van der Klooster KJ, **Sol M**, van der Krogt M. Let's Ride….. Wheelchair skills training to stimulate participation. Mini-symposium European Academy for childhood disability 2017

Sol ME, De Groot JF, Verschuren O, Visser-Meily JMA. *Let's Ride….Effectiveness of wheelchair skills training in manual wheelchair-using children.* Poster presentation at the Spina Bifida World Conference 2017

Sol ME, Bloemen MAT, Verschuren O, Rushton PW, Visser-Meily JMA, de Groot JF. Wheelchair Mobility Confidence Scale for Dutch youth using a manual wheelchair. Part of the *Let's Ride….. study. Manon Bloemen on behalf of Marleen Sol, Oral Presentation at the* World Conference Physical Therapy 2019

Sol ME, Verschuren O, Horemans H, Westers P, Visser-Meily J.M.A, Fit-for-the-Future Consortium, De Groot J.F. The effects of wheelchair mobility skills and exercise training on physical activity, fitness, and confidence in youth using a manual wheelchair. Oral Presentation at the World Conference Physical Therapy 2021

De Groot L, van der Klooster KJ on behalf of **Marleen Sol,** Workshop wheelchair mobility skills at the European Pediatric Physical Therapy Congress 2019

FIVE MOST IMPORTANT NATIONAL PUBLICATIONS, PRESENTATIONS & AWARDS.

Sol ME, De Groot JF, Verschuren O, Visser-Meily JMA. Klokhuisaflevering: Rolstoelskills, NTR, 06-11-2019. Winner klokhuis wetenschapsprijs 2018

Sol ME, Verschuren O, de Groot JF, Van der Klooster KJ, Klem M, Visser-Meily JMA. Rolstoelvaardigheid bij kinderen, Let's Ride… Op 't randje balanceren kan je leren. NTR 2017 nummer 4, reprinted with permission by BOSK Magazine 2017; nummer 2.

Sol ME, De Groot JF, Verschuren O, Visser-Meily JMA . Workshop and oral presentation: Wheelchair mobility skills training and evaluation in children and Youth. Co-Organizer Symposium Fit For the Future, Let's Ride 2016. Winner most relevant research award.

Sol ME, De Groot JF, Verschuren O, Visser-Meily JMA. Effect van rolstoelvaardigheid training bij kinderen in een handbewogen rolstoel. Oral Presentation, Fit For the Future – symposium for parents and children 2016.

Sol ME, de Groot JF, Gaasbeek M, Verschuren O. Poster pitch: Effect van rolstoelvaardigheid training bij kinderen in een handbewogen rolstoel at the Jaarcongres Nederlandse Vereniging voor Fysiotherapie Kinderen 2015. Winner Poster award.

UMC Utrecht

Universiteit Utrecht

ISBN 978-94-6423-294-3