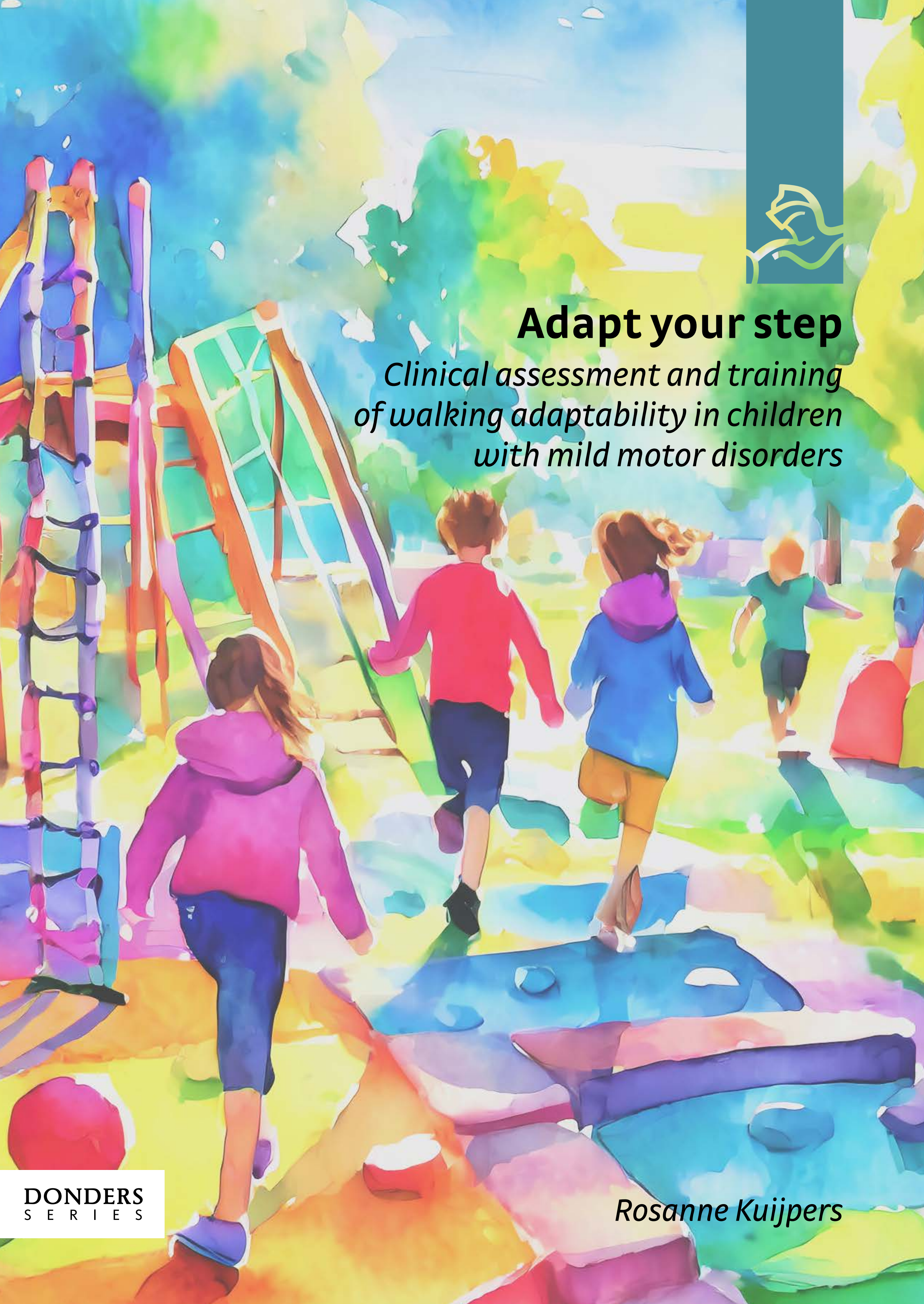

Adapt your step

Clinical assessment and training of walking adaptability in children with mild motor disorders

Rosanne Kuijpers

DONDERS SERIES

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Colofon

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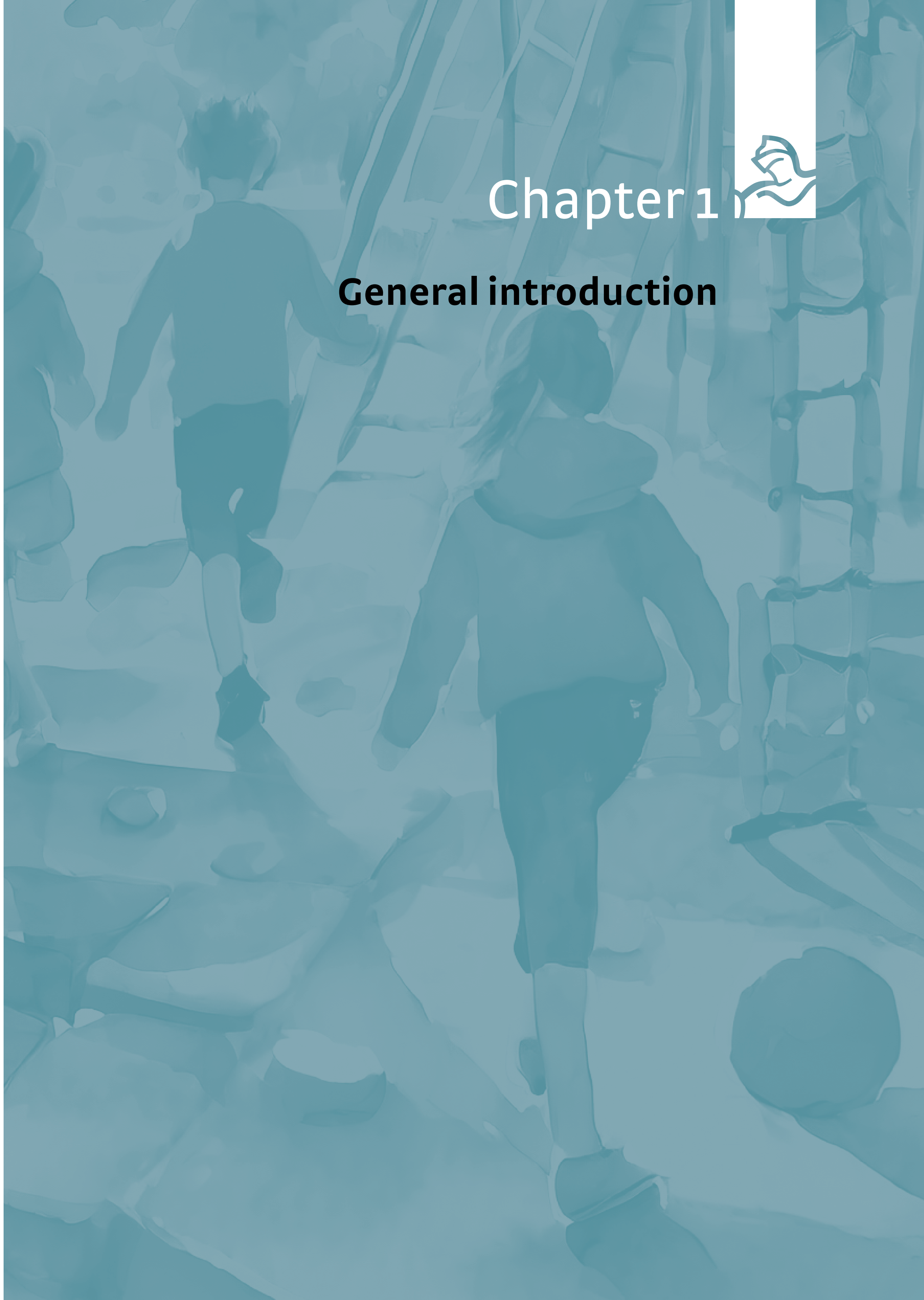

Chapter 1

General introduction

Human movement is a complex phenomenon, which emerges from interactions between the individual, the task and the environment[1]. The individual generates a movement – intentionally or unintentionally – to perform a task or spontaneous movement within a certain environment[1], such as lifting a cup from the table, walking to the supermarket or avoiding an approaching ball. Within the individual, movements occur from the interaction between several processes, including processes related to action, perception, and cognition. Movement is commonly characterized in the context of accomplishing a specific action (e.g., walking)[1]. Perception – the organization, interpretation, and conscious experience of sensory information[2] – is essential for action and vice versa. Cognitive processes such as attention or motivation, and emotional aspects of motor control such as fear of movement also contribute to the achievement of movement goals[1]. An everyday-example of the interaction between action, perception, and cognition is when a child walks on the street and wants to cross (i.e., action), hears a car approaching (i.e., perception), and is conscious to wait until the car has passed before crossing the street (i.e., cognition). After crossing the street the child has to step up onto the pavement which provides new input for action, perception, and cognition, a process that continues all day in many tasks and spontaneous movements.

This example shows that walking in daily life is more than walking on a 'red carpet', especially in the lives of children. Avoiding other children while playing in the schoolyard, accelerating to cross the street between two approaching cars, and avoiding toys on the ground are essential skills for safe ambulation in everyday life. This so-called skill of walking adaptability is often affected in children with motor disorders, which is not always recognized in those with relatively mild impairments. Therefore, more attention is needed for walking adaptability in clinical practice as well as in research, which will be illustrated by the case description in the boxes throughout this General introduction and the General discussion.

In my work as a pediatric exercise therapist in primary health care and at regular and special primary schools, I used to see many children with motor disorders. I helped these children to improve their fine and gross motor skills in a playful way. Later in their learning process, I often joined them in daily life situations (school and gym classes, in the schoolyard, during sports) to help them transfer their learned skills into daily life. I liked to contribute to children's motor development and saw what the therapy could do for children and their parents, but the scientific evidence behind it was often lacking. The opportunity to contribute to a research project on walking adaptability in children with motor disorders gave me the chance to bring clinical practice and scientific research together.

I will give a case example to illustrate the problems children with mild motor disorders may face in daily life situations. The case is based on the stories of children who participated in the research projects, but does not concern one specific participant.

Max is a 9-year-old boy who attends a regular primary school. As most children of his age, he enjoys the breaks the most, when he can play outside in the schoolyard with his classmates. He prefers to play on the climbing frame or turn the rope when they do rope skipping. When they play tag in the schoolyard, Max cannot turn quickly enough to avoid being tagged. He is a bit slow and clumsy in his movements and he often trips and falls. Max likes the gym

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classes, but when they choose teams to play football or basketball, he remains among the last children to be chosen. He finds it hard to pass the ball while running, to quickly turn, and to run and keep an overview over the game. Sometimes he stumbles over the ball making him feeling embarrassed for his classmates.

This chapter starts with an explanation of the theoretical background of walking adaptability in general. This is followed by a description of two motor disorders: Developmental Coordination Disorder and Cerebral Palsy, including a description of the difficulties children with these disorders experience with walking adaptability. Then, a description of the state of the art in measurement and training of walking adaptability is given. Finally, the aims of the studies included, and the outline of this thesis are provided.

Walking adaptability

The neural control of walking can be explained by a tripartite model (see Figure 1) of which all three parts are necessary for safe ambulation in daily life. The model comprises stepping, postural equilibrium, and walking adaptability[3]. First, the central nervous system generates the basic stepping pattern of rhythmic reciprocal limb movements to propel the body forward while supporting the body against gravity[3]. Second, it has to maintain the body upright in space by keeping the center of mass within the constantly moving base of support (control of equilibrium). Third, the central nervous system has capabilities for locomotor control to adapt the basic stepping pattern to environmental circumstances or changes in a task or movement goal[3,4]. The development of walking in children reflects a hierarchy within this tripartite model. In the second week of their life, more than 50% of the newborns can already make stepping movements[5]. Around the age of 15 months, the balance control has developed sufficiently to learn to walk independently. In the first months hereafter, the child learns to walk faster, with increasing step length and with a narrower step width[6], indicating the continuing development of postural balance. Children's walking speed keeps increasing until the age of 4 or 5 years[7,8]. The ability to adapt walking to environmental or task demands (e.g., obstacle avoidance) continues to develop into adolescence, specifically regarding the maintenance of medial-lateral stability and the finetuning of foot placement around an obstacle[9]. The current thesis focuses on proactive (feedforward) walking adaptability, which adaptations occur in response to visual stimuli, for example when walking in a cluttered environment or accelerating gait to avoid being tagged during play[4].

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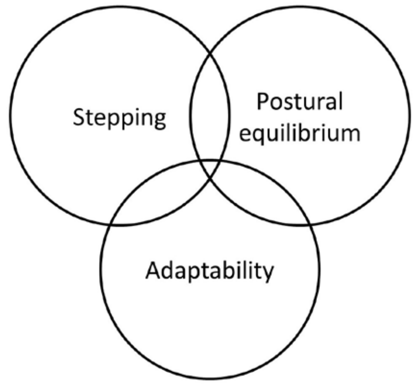

Figure 1. Neural control model of functional walking. Copied from: Balasubramanian et al. 2014[3] (reprinted with permission from Chitra L.K. Balasubramanian).

Walking adaptability in Developmental Coordination Disorder and Cerebral Palsy

Children with mild motor disorders can typically walk on a smooth path or in a quiet hallway without problems (the 'red carpet'), but encounter difficulties adapting their gait to a more challenging task or environment. In this thesis I will focus on two of the most common neurodevelopmental disorders in children: Developmental Coordination Disorder (DCD) and Cerebral Palsy (CP)[10]. DCD has a prevalence of 5 to 6% in school-aged children[11]; CP has an incidence of 1.5 to 3 per 1.000 live births[12].

DCD

Children with DCD cannot coordinate their movements in the same way as typically developing (TD) children, resulting in clumsy, inaccurate or slow movements[13]. They also have difficulties learning a new motor task, and problems with automatization of movements and performing dual tasks[13]. This results in both gross and fine motor problems. The problems persist into adolescence[14] and even into adulthood[15]. The exact etiology of DCD is still unknown, but recent neuroimaging studies indicate underactivation in multiple brain regions involved in two important systems for motor learning: the network involved in internal modeling and the mirror neuron system (essential for observational learning and motor imagery)[16]. However, further research is needed to examine the specific effects of these patterns of underactivation on the coordination problems of children with DCD.

Four main hypotheses have been proposed to explain DCD in terms of the following deficits: visuospatial, procedural learning, executive functioning, and internal modeling[13]. The visuospatial deficit involves difficulties with identifying objects and their localization in space, and is closely connected with action systems in motor learning[17]. However, this hypothesis cannot explain the motor problems of all children with DCD, as it is only present in a subgroup of children with DCD[18]. Procedural learning concerns the development of implicit memory for cognitive and motor routines and skills[19]. Children with DCD initially have difficulties with procedural learning compared to TD children, but they are able to learn, consolidate, automate, and transfer new skills after proper training[20] which weakens this hypothesis. Executive functioning – a set of mental processes that help an individual concentrate and pay attention such as reasoning, problem solving, and planning[21] – play a role in the problems of children with DCD, but they do not specifically explain the cause of problems in DCD as executive functioning problems are also observed in children with other neurodevelopmental disorders[22,23].

A fourth – and currently dominant – hypothesis concerns an internal modeling deficit, which involves the inability to anticipate the consequences of actions and to make rapid online corrections. Such an internal model is based on recent motor experiences and involves making a prediction of the *expected* sensory feedback from the generated motor plan. This prediction is then compared by the central nervous system to the *actual* sensory feedback when executing the planned movement, which permits discrepancies to be sensed and corrected[24,25]. Online correction of a movement thus occurs when the initial movement plan was not accurate enough due to an incorrect internal model or to changes in the task or environment[13]. The internal modeling deficit has an impact on motor learning[13], which is consistent with the performance deficits of children with DCD[26].

CP

In contrast to DCD, for which no clear neural cause has yet been found, CP is a permanent disorder caused by non-progressive disturbances in the fetal or infant brain[12]. CP can be caused by aberrant cortical development during pregnancy (e.g. because of genetic factors, an infection caught by the mother, or an injury to the unborn baby's head) or damage to the brain during or short after birth (e.g. an oxygen deficiency during a complicated birth, a stroke, or an infection of the brain)[27]. The problems with the development of movement and posture can be linked to the damaged location of the brain. Alongside their motor problems, children with CP often show disturbances of sensation, perception, cognition, communication, and behavior[12]. While CP is a non-progressive disorder, it is often accompanied by progressive musculoskeletal problems (such as scoliosis, contractures, and hip dislocation) caused by insufficient variation in posture and movement[28]. Children with CP have problems with muscle tone and strength and sensory information processing[29]. This leads to difficulties with the coordination of the lower as well as the upper extremities. Limited arm movements caused by spasticity can reduce stability during walking[30]. Besides, children face difficulties with the musculoskeletal and sensory components of postural control, and anticipatory and reactive postural adjustments[29].

The severity of CP can be classified using the Gross Motor Function Classification System (GMFCS), which is displayed in Figure 2[31]. Children in GMFCS level I can walk independently and walk the stairs without handrails. They can perform gross motor skills such as running and jumping, but their speed, balance, and coordination are affected. Children in GMFCS level II have difficulties with walking long distances, walking on uneven terrain, and walking in crowded areas. They can walk the stairs but only with use of handrails and they have minimal ability to perform gross motor skills such as running and jumping. Children in GMFCS levels III, IV and V are increasingly dependent on devices for mobility, such as crutches, a walker, or a wheelchair. With increasing GMFCS level, head and trunk balance decrease and likewise the ability to sit independently[31]. This thesis focuses on children with CP in GMFCS levels I and II – mild CP –, which represent around 60% of the total population of children with CP[32].

Daily life situations

As a result of their impairment, both children with DCD and mild CP have a poorer physical capacity[11,33], are less physically active[11,34], and trip and fall more frequently[35,36] compared to TD children. They face challenges in daily life, for example during sports, play and school activities[37,38]. It is not hard to imagine them having difficulties playing tag in a crowded schoolyard with running children, rolling balls, different terrain (grass, sand, tiles), and surrounding trees or plants. These limitations also have negative social consequences: being part of a group and being able to take part in group activities are essential for the development of children. Children with DCD and mild CP often have a more negative perceived athletic competence[11,39] and lower quality of life[40,41] compared to TD children.

Figure 2. Schematic explanation of GMFCS levels. Copied from: Bill Reid, Kate Willoughby, Adrienne Harvey and Kerr Graham, The Royal Children's Hospital Melbourne (reprinted with permission from Kerr Graham).

State of the art: walking adaptability measurements and training

As outlined above, walking adaptability is an essential skill to navigate through our physical and social environment. Children with DCD and mild CP have shown to perform poorer on walking adaptability tasks compared to TD children[42-45], while a secondary motor or cognitive task aggravates this performance deficit[45-47]. The majority of these studies on walking adaptability in children with DCD and mild CP were performed using a 3D motion capture system in a movement laboratory[42-47], which is expensive, time-consuming, and not motivating for children. However, no clinical measurement tools exist for measuring walking adaptability.

Several clinical measurement tools are available to assess a range of gross motor skills in children with mild motor disorders, such as the Movement Assessment Battery for Children[48]

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and Buininks-Oseretsky Test of Motor Proficiency[49] for children with DCD, and the Gross Motor Function Measure-Challenge Module[50] and Test of Gross Motor Development[51] for children with mild CP. Also, less extensive tests are available evaluating running speed (Muscle Power Sprint Test) or running agility (Ten Times Five Meter Sprint Test)[52]. While these tests evaluate aspects related to walking adaptability (e.g., walking on a line or balance beam, stepping over a balance beam, running through pylons, slide between points), none of the tests assess the speed-accuracy trade-off involved in coordinating precise foot placement in complex and dynamic daily life environments.

For children with DCD, only a small number of studies focused on gait training. One study examined the effect of task-oriented training which was focused on gross motor skills including walking (e.g., turning, sudden stops, walking with dual tasks or walking in a crowded environment). The study showed a positive effect of the training on overall motor proficiency[53]. Another study examined the effect of (dynamic) balance training (e.g., walking with raised heels, balancing a ball while walking), and found an improvement on static and dynamic balance[54]. However, neither study evaluated the effects of training on walking adaptability skills.

Despite his disorder, Max is functioning well enough to be able to participate in regular education. However, he has difficulty to keep up with his classmates in different activities like playing in the schoolyard and physical education classes. He particularly faces difficulties with activities in which he has to quickly adapt his movements to the environment or task, such as in football, basketball or playing tag.

Last year, Max went to a physiotherapy practice where he successfully improved his walking endurance. This year, he received therapy at school where he learned to better kick and throw a ball, run faster, and dribble with a ball while running. He also practiced these skills during play in the schoolyard and physical education classes together with his therapist, but it was difficult for him to focus on these motor skills in the rush of the game.

Max would like to further improve his motor skills so he can better keep up with classmates. Specifically adapting his movements to the environment or task is still difficult for Max. Also his parents would like a suitable intervention for Max to give him more confidence in more complex walking skills in interaction with his classmates.

In children with mild CP, gait training is very common, but the majority of studies focused on walking speed, both as the training goal and as the outcome measure[55,56]. Different kinds of gait training (overground training, treadmill training with and without bodyweight support, with and without virtual reality (VR), with and without incline) show a positive effect on walking speed and endurance[55,56], and some show a positive effect on walking, running and jumping skills[56]. Treadmill training might be more effective than overground training for improving walking speed and endurance, which is possibly explained by the intensity and increased stepping repetition of treadmill training[56]. To date, specific training targeting walking adaptability (e.g., target stepping, obstacle avoidance) has not yet been performed in children with DCD and mild CP.

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Development of new technologies contributed to the possibility to train walking adaptability in a safe, controlled and challenging way in clinical practice. The C-mill (Motek Medical, Houten, the Netherlands) is an example of such a technology. It is a treadmill device with augmented reality which can evoke gait adjustments. The treadmill belt can be augmented with projected visual context such as targets and obstacles which are placed based on foot placement. Therefore, it is a suitable device to train walking adaptability. For different adult populations such as people with a stroke or Parkinson's disease, C-mill training has been proven to be an effective way to improve walking adaptability[57-59]. We hypothesized that for children with mild motor problems, C-mill training might be beneficial as well. Using the C-mill allowed me to design a training protocol in accordance with practical recommendations for motor skills training in children with DCD and CP, which state that training should be focused on daily activities, be task-specific, and meet goals that are relevant in daily life and chosen by the child[11,60]. A variable instead of a repetitive practice structure should be applied[61], and children should learn implicitly with an external focus of attention[16]. Moreover, interventions should be enjoyable and motivating for the child[60], and children should receive augmented (i.e., extrinsic) feedback when learning new motor skills[16,60]. Augmented feedback is defined as information on execution or outcome of the movement[62], and has been proven to be valuable in motor learning of walking skills in children with CP[63]. Finally, the use of augmented reality might improve children's training motivation as they can play games on the treadmill in different environments (e.g., forest, beach or city).

Taken together, it can be concluded that measuring walking adaptability with an overground task would have added value in clinical practice. Moreover, walking adaptability training should be part of motor skill learning programs.

Aim and outline of this thesis

The overarching aim of this thesis was to develop and evaluate clinical assessment and training of walking adaptability in children with mild motor disorders (DCD and mild CP). The ultimate goal is to enable identification of children who have problems with walking adaptability and to help them improve this skill by means of training, to foster engaging in daily activities together with other children and to improve participation.

Together with my supervisory team, I co-developed the Walking Adaptability Ladder test for Kids (WAL-K), which is a new clinical test for assessing walking adaptability in children. Establishing the psychometric properties is a necessary step towards its clinical use for identifying children who have difficulties with walking adaptability and for evaluating effects of treatment. To this end, the aim of **Chapter 2** was to assess the reliability and validity of the WAL-K in children with typical motor development and in children with DCD.

In daily life, children encounter many situations in which they have to perform dual tasks while walking. Therefore, it is important to examine whether dual tasking has a disproportionate effect on walking adaptability in children with mild motor disorders compared to children with a typical motor development. In **Chapter 3**, using an observational design, the aim was to assess the effects of a visuo-motor and cognitive dual task on walking adaptability in children with and without DCD.

For children with impaired walking adaptability, dedicated training may help improve this skill, with potential beneficial effects on daily life activities. Therefore, the aim of **Chapter 4** was to examine the effect of a treadmill training program using the C-mill with respect to walking adaptability in children with DCD. To evaluate training effects, I used the WAL-K and walking adaptability tasks on a treadmill with and without secondary tasks. In addition, parents' perception of the treadmill training was evaluated.

As children with CP also experience problems with walking adaptability, in **Chapter 5** I aimed to evaluate the reliability and validity of the WAL-K in children with mild CP (GMFCS levels I and II). The aim of **Chapter 6** was to evaluate the effect of treadmill training using the C-mill in children with mild CP with respect to walking adaptability, using the WAL-K as the primary outcome. Walking speed, functional muscle strength, perceived athletic competence, and health-related quality of life were evaluated as secondary outcomes. An additional aim was to identify potential factors influencing the effect of the training. Parents' perception of the training was also evaluated.

In **Chapter 7**, the research conducted in this thesis is summarized and discussed. The theoretical, clinical, and educational implications of the findings are outlined, and methodological considerations and directions for future research are discussed.

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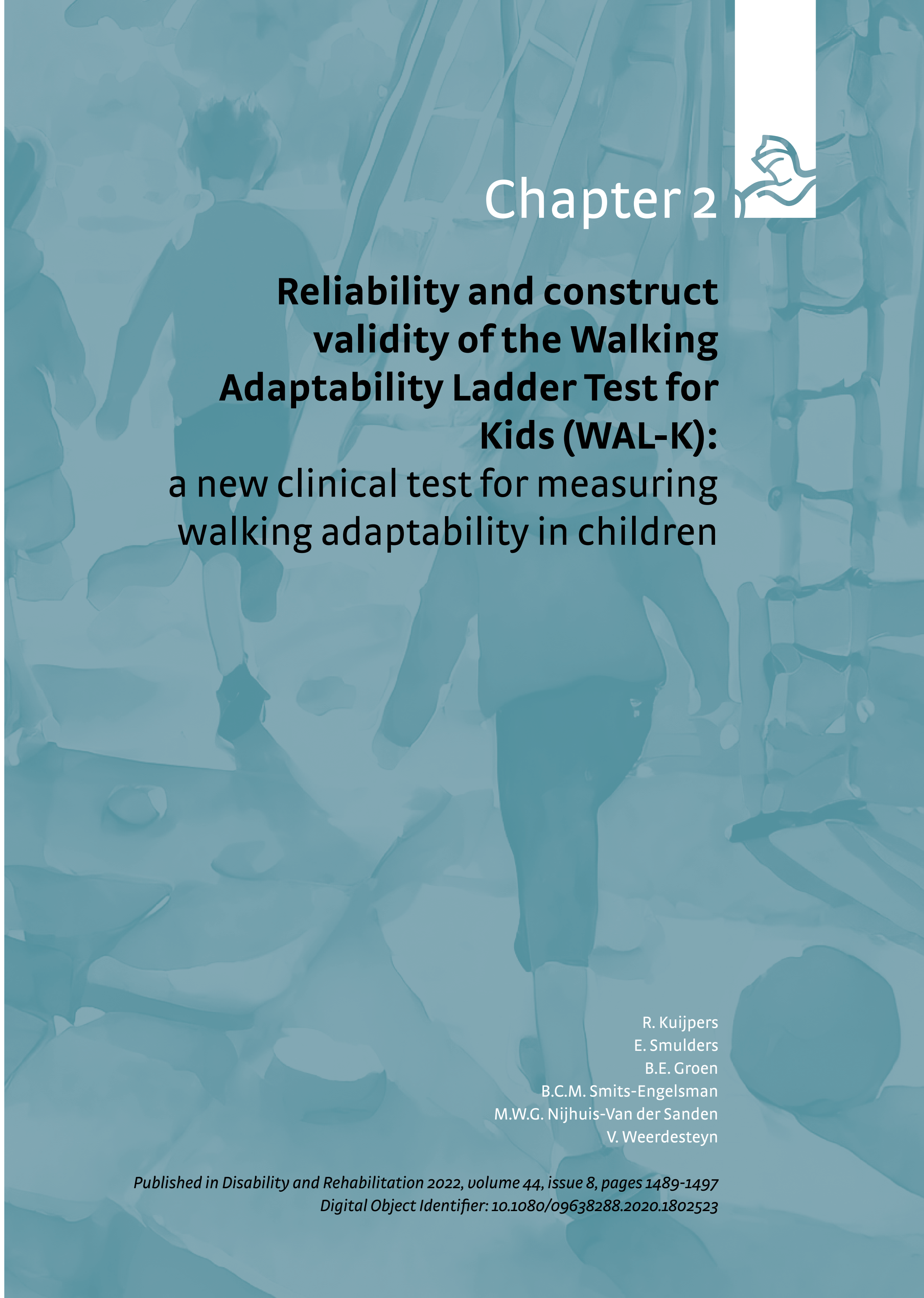

Chapter 2

Reliability and construct validity of the Walking Adaptability Ladder Test for Kids (WAL-K): a new clinical test for measuring walking adaptability in children

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Abstract

Purpose

Walking adaptability is essential for children to participate in daily life, but no objective measurement tools exist. We determined psychometric properties of the Walking Adaptability Ladder test for Kids (WAL-K) in 6-12 year old children.

Materials and methods

In total, 122 typically developing (TD) children and 26 children with Developmental Coordination Disorder (DCD) completed the single and double run conditions of the WAL-K. Intra-rater, inter-rater and test-retest reliability were determined by ICCs and Smallest Detectable Change (SDC) in 53 TD children. Construct validity was determined by comparing WAL-K scores between 69 TD and all DCD children and correlating these scores with age and MABC-2 scores.

Results

ICCs for reliability varied between 0.76 and 0.99. Compared to the first test performance, WAL-K scores were lower (i.e., better) at retest. SDCs for test-retest reliability varied between 20.8 and 26.1% of the mean scores. WAL-K scores were significantly higher (i.e., worse) in DCD children compared to TD children ($p<0.001$). Significant negative correlations were found with MABC-2 (-0.52 and -0.60) and age (-0.61 and -0.68).

Conclusions

The WAL-K shows to be a valid, reliable and easy-to-use tool for measuring walking adaptability in children. Adding an extra practice trial may reduce the observed learning effect.

Background

Gross motor skills are important for children to fully participate in activities of daily living (ADL), such as activities during playtime in the schoolyard and sports. Walking activities in daily life of children typically involve frequent and rapid modifications of the walking pattern to reach intended goals while handling the demands of the environment (e.g., to avoid stumbling over objects or colliding with playmates)[1,2]. This skill is commonly referred to as walking adaptability[3]. Children with Developmental Coordination Disorder (DCD) show greater variability and asymmetry of spatial-temporal measures of gait compared to typically developing (TD) children[4,5]. They also walk with shorter stride length and duration[6] and have an increased step width and increased medio-lateral velocity of the centre of mass, both during undisturbed walking[7] and in lab-based tests concerning obstacle crossing or walking on irregular terrain[8–10]. Due to these gait deviations, children with DCD experience difficulties with age-appropriate walking activities in daily life[11] and they trip and fall more often[12,13]. As a result, these children participate less in physical activities[14] and might therefore develop psychosocial or physical problems[14–16].

Children with DCD typically struggle with timing and coordination of movements, which also pertains to the spatial and temporal control of the walking pattern[11]. At present, a prominent hypothesis on the basis of motor control deficits in DCD concerns deficits in the forward modeling of movement. The ability of the nervous system to predict the future location of moving limbs using a forward model underlies the ability to make rapid online corrections during movement[11]. A deficit in the forward modeling of movement limits both the speed and accuracy of online error correction[11,17,18].

In this context, assessment of the adaptability of walking appears highly relevant for evaluating a child's ability to participate in age-appropriate ADLs. Commonly used tests and questionnaires for evaluating gross motor skills in children are the Movement Assessment Battery for Children (MABC-2)[19], the Bruininks-Oseretsky Test of Motor Proficiency (BOT-2)[20], the Körperkoordinationstest für Kinder (KTK)21, the Developmental Coordination Disorder Questionnaire (DCDQ)[22], and the Motor Observation Scale for Teachers (MOQ-T)[23]. Although these tests cover many important motor skills needed for children's ADL, items that specifically evaluate walking adaptability are not included.

For children, less commonly used tests exist that examine some form of walking adaptability[24,25]. However, these tests are either performed at comfortable walking speed or involve adaptation of the walking pattern, but lack the combination of adaptation of walking speed and walking pattern. For elderly people and adults with movement disorders, some more tests are available that specifically focus on walking adaptability, or contain walking adaptability items[26–29]. Yet, these tests are not sufficiently suitable for measuring walking adaptability in children, because of their known ceiling effects in healthy children[30] as well as in children with mild motor impairments[31,32]. The apparent ceiling effect may be explained by these tests being conducted at comfortable speed, thus being too easy for children who typically perform many activities of daily living at running speeds. Tasks that involve higher speeds in combination with adaptation of the walking pattern may better reflect the difficulties that children encounter during sports or play in daily life, which also pertains to children with mild motor impairments. Hence, there is a need for a walking adaptability

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test for children that specifically focuses on the combination of modifications to speed and accuracy during walking.

To fill this void, we developed the Walking Adaptability Ladder test for Kids (WAL-K), which was inspired by the agility ladder that is often used in sports training. The ladder of the WAL-K has stepping targets that successively decrease in size, which forces a child to continually adapt step length and cadence to the targets. In addition, as a result of the imposed foot placement adaptations, the forward velocity of the center of mass has to be adjusted from step to step to control dynamic stability[33]. This makes the WAL-K much more challenging than an equally spaced agility ladder. The WAL-K taps into the specific problem areas of children with DCD for evaluating walking adaptability: it requires the transformation of visual information of the targets into a forward model for stepping, online error corrections to fine-tune foot placement, and continual adaptations of both step length and walking speed.

A first study on the reliability (n=34) and validity (n=125) of a short (4 m) version of the test, the so-called Ladder Agility Test (LAT), was conducted in typically developing (TD) children of 7-10 years old[34]. It showed good test-retest reliability (ICC>0.81) and strong internal consistency (α=0.85). Yet, it was observed that in the 4m LAT, the relatively small difference in target size (decrease from 44 to 28 centimeters) still imposed a rather limited challenge to speed and step length adaptations. In the WAL-K, we therefore extended the step ladder to 10m with 19 targets and enlarged the difference in target size between the first and last target to 36 centimeters (64 to 28 centimeters). The longer 10m WAL-K also forces children to adapt their steps for a much longer period, which enhances the probability of errors in subsequent steps or adaptation of the walking speed and enhances the sensitivity of the measurement. Therefore, we expect the WAL-K allows for a better evaluation of whether a child really masters the skill to adapt walking. Hence, by lengthening the ladder and thus increasing the required change in foot placement, we expected that a difference in walking adaptability between TD children and children with DCD would become highly evident. Besides, in contrast to the 4m version of the test (in which children who made more than three errors started an extra trial and the outcome was based on the mean of three instead of two trials), we calculated all mistakes without a maximum (the aim was as few as possible) while completing the WAL-K.

The aim of this study was to determine intra-rater, inter-rater and test-retest reliability of the WAL-K in children aged 6 to 12 years old. As we plan to use the WAL-K for identifying children with walking adaptability problems, a second important aim was to determine its construct validity. Intraclass correlations (ICCs) >0.75 will indicate a reliable clinical utility of the WAL-K[35]. The construct validity of the WAL-K would be supported by significant differences between TD children and children with DCD and by strong correlations (r=0.60-0.79) between WAL-K scores and age in TD children. We also expected significant, yet somewhat lower correlations between the WAL-K and an existing motor test (MABC-2) in TD children, as both tests evaluate motor skills, but capture different constructs.

Methods

This study had an observational design. The flow of enrolment, assessments and the use of the samples to answer the different research questions is displayed in Figure 1.

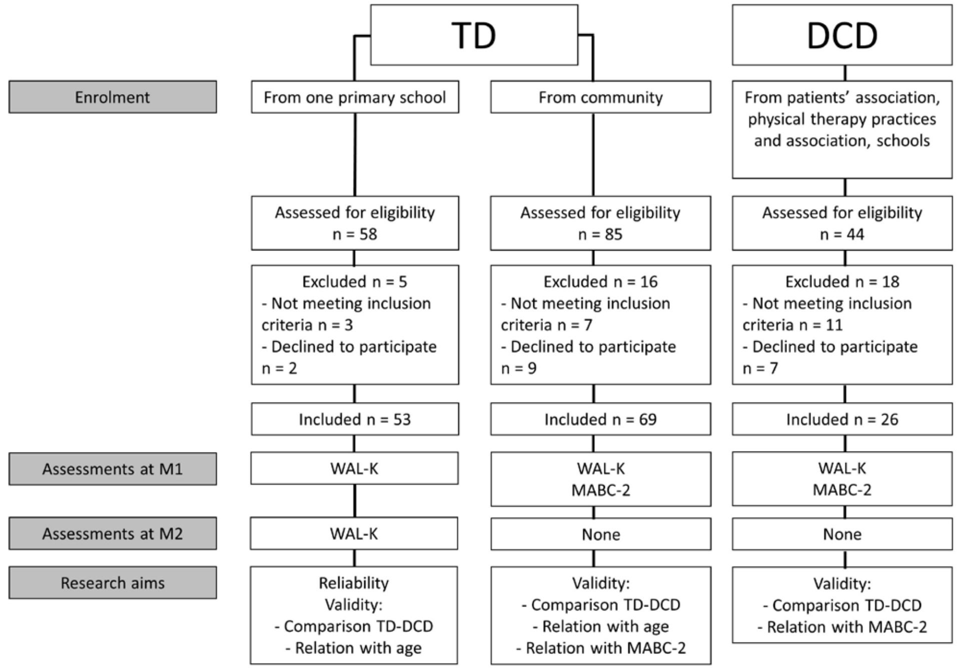

Figure 1. Flow diagram of enrolment, assessments and the use of the samples to answer the different research questions.

Participants

One recruitment wave of TD participants, for testing reliability, took place at a regular primary school in Oploo, a small town in the southern part of The Netherlands. The teachers handed out information letters about the study to the parents, and after having received consent from the parents, the teachers verified that the children were 6 to 12 years old and presented with typical motor development. Fifty-eight TD children from the school were assessed for eligibility by the research team. For the measurement of construct validity we recruited TD children from the community: colleagues and family of the researchers were informed about the study, and researchers posted a recruitment notice at local schools. When parents were interested in the study, they directly contacted the researcher. This recruitment wave resulted in 85 TD children being assessed for eligibility. Children with DCD were recruited through the patients' association, physical therapy practices and association, and schools. A total of 44 children with DCD were assessed for eligibility.

Inclusion criteria for the TD children were: aged 6 to 12 years old, attending a regular primary school, and having a typical motor development, as indicated by the teacher and/or parents. Children with DCD had to have a diagnosis of DCD from a medical practitioner, or had to be referred to a therapist because of motor performance problems interfering with daily life and/or academic achievement as indicated by parents completing the DCDQ. In that case, the therapist examined the motor performance problems using the MABC-2 based on the research criteria for DCD[36] and checked if the motor impairment could not be explained by mental retardation (IQ>70), visual problems or neurological problems as indicated by parents. Exclusion criteria for both TD and DCD children were temporary complaints influencing walking ability, or neurological, orthopaedic, cardiovascular or visual problems.

The parents or legal representatives of all participants gave written informed consent before participating in the study. The study was approved by the regional Medical Ethical Committee Arnhem-Nijmegen (2017-3465).

Measurements

Walking Adaptability Ladder Test for Kids (WAL-K)

For the WAL-K, a 10-meter ladder (Medipreventiecentrum B.V., Hengelo, The Netherlands) was used with 19 rectangular targets successively decreasing in size by 2 centimeters each, resulting in target sizes ranging from 64 to 28 centimeters. The ladder lies flat on the ground. All targets have a width of 42 centimeters. The transverse beams of the ladder are yellow plastic bars of 4 millimeters thick, the sides of the ladder are red ribbons. A cone was placed fifty centimeters from the end of the ladder (Figure 2 and Supplemental Material). The child was instructed to walk in the ladder, turn around the cone and walk back in the ladder, as fast and accurately as possible. Due to the successive decrease of the target sizes (and increase on the way back), children have to continually adapt their walking pattern, as well as their walking speed.

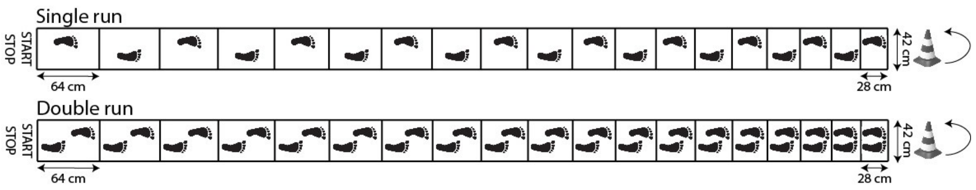

Figure 2. Schematic set-up of the WAL-K.

The WAL-K was completed in two different conditions: while stepping once (single run) and stepping twice (double run) in each target. First, the researcher demonstrated the single run and after one practice trial the child performed the task twice. Thereafter, the same procedure was followed for the double run. When a child did not understand the instructions, extra instruction or encouragement was given during the practice trial. Participants were filmed from the waist downwards for post-hoc scoring. All children were tested in a hallway by two experienced physical therapists (RK and CS) who were trained to administer the test. It took about 10 min to administer the WAL-K. The second measurement for the test-retest reliability was performed in 53 TD children after one week. Exactly the same procedure was followed by the same tester at the same location.

Outcomes of the WAL-K were: total time to complete the ladder back and forth (in seconds), and the total number of mistakes (touching a bar, the wrong number of steps in a target, or missing a target). For each trial, we determined a combination score of time and mistakes (0.5 s penalty for each mistake). The trial with the best WAL-K combination score of the two attempts per task was used as outcome measure for statistical analysis, further referred to as WAL-K score.

For determining reliability of the WAL-K, four independent trained raters (IC, LH, EM, and JP) scored the videos of 53 TD children with regards to completion time and number of mistakes. The videos were equally at random divided among the raters. For determining intra-rater reliability, each rater scored the videos of measurement 1 (M1) twice for 13 or 14 children, with one week in between and without insight in their previous scores. Each rater also scored the videos of the same children of measurement 2 (M2), without insight in the scores of M1, for determining test-retest reliability. For determining inter-rater reliability, each rater scored the M1 videos of another 13 or 14 children. The order of the movies did not change between the first and second scoring. Children were filmed from the waist downwards. Children who were recruited from the primary school were tested in a quiet hallway at their school; children who were recruited from community and the children with DCD were tested in a quiet hallway at the hospital.

MABC-2

Motor performance was assessed in 69 TD children and all children with DCD (n=26) by RK using the Dutch edition of the MABC-2[19]. This test contains three different age bands (3-6 years, 7-10 years, and 11-16 years), each consisting of eight items. These items are divided into three subsets: manual dexterity (3 items), aiming and catching (2 items), and balance (3 items). Raw scores are converted into standard scores, ranging from 1 to 19. The MABC-2 has excellent test-retest reliability (ICC=0.97) and internal consistency (α=0.90) for the total score in children with DCD[37]. It took about 30 min to administer the MABC-2 and children were tested in a room at the hospital.

Statistical analysis

Reliability

Intra-rater, inter-rater and test-retest reliability were calculated in 53 TD children using the Intraclass Correlation Coefficient (ICC) estimates and their 95% confidence intervals, based on a single-rating, absolute agreement, two-way random-effects model, ICC model 2,1[38]. This model takes into account systematic differences between raters or measurement moments[39]. Paired samples t-tests and Bland Altman analyses were performed to investigate absolute differences between M1 and M2

With regard to the test-retest reliability, the Smallest Detectable Change (SDC) was determined, which is the smallest change that can be detected by the instrument, beyond measurement error[40]. Within a child, a change in WAL-K score above the SDC can be considered as a real change when evaluating improvement on walking adaptability after an intervention[40]. The following formula was used: SDC=1.96*SDd, where SDd is the standard deviation of the difference values between M1 and M2[40]. The percentage of SDC of the mean was calculated using the mean of M1. A percentage of SDC of <30% was considered as acceptable and <10% as excellent[41].

Construct validity

Only for comparing WAL-K with MABC-2, the scores on the WAL-K had to be adjusted for the age of the child, because the standard scores on the MABC-2 are adjusted for age as well. Therefore, a linear regression (Figure 3) was used with WAL-K score as dependent variable and age as independent variable. The age-adjusted WAL-K score at M1 was compared to the total standard score and the three subscores on the MABC-2 for both TD and DCD children and the TD and DCD children together, using a Spearman's rank correlation coefficient. From the linear regression analysis, also the correlation between age and WAL-K score at M1 in TD children was determined. The WAL-K score at M1 was compared between TD children (unadjusted score of first rater) and children with DCD, using an ANCOVA with age and gender (because of the typical overrepresentation of boys in the DCD population) as a covariate.

All analyses were performed in SPSS version 25 and alpha was set at 0.05. For the ICCs, the following cut off values were used: ICC under 0.5 poor reliability; between 0.5 and 0.75 moderate; between 0.75 and 0.9 good, above 0.9 excellent[35]. For the correlation coefficients, the following cut off values were used: 0.00-0.19 very weak; 0.20-0.39 weak; 0.40-0.59 moderate; 0.60-0.79 strong; 0.80-1.00 very strong[42].

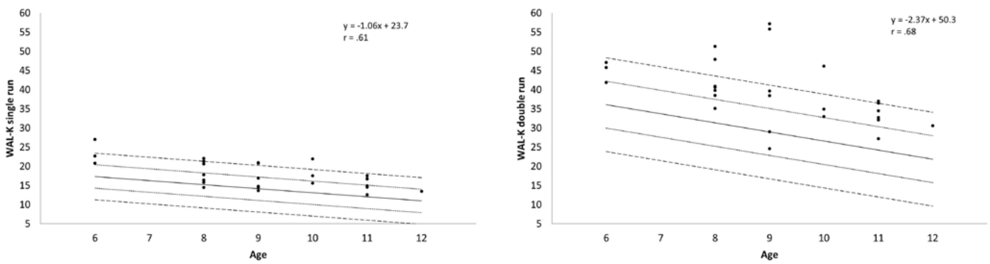

Figure 3. Data of children with DCD for the single run and double run. The lines are based on the data of the TD children (n=122): mean (straight line) with + and -1 SD (dotted line) and + and -2 SD (striped line).

Results

Of the 143 eligible TD children, 21 children did not participate because they declined (n=11) or were excluded because they did not meet the inclusion criteria because of orthopaedic or visual problems or temporary complaints influencing walking ability (n=10). Of the 44 eligible children with DCD, eighteen did not participate because they declined (n=7) or were excluded because they did not meet the inclusion criteria (too high scores on the MABC-2 (n=4); no limitations in daily life as indicated by the DCDQ (n=2); visual and/or orthopaedic problems (n=3); neurological problems (n=2)). The final sample consisted of 122 TD children and 26 children with DCD (Figure 1). The characteristics of all participants are presented in Table 1.

Table 1. Characteristics of participants and comparison between groups (Chi square test for gender and independent samples t-test for age)

		TD		DCD		Comparison between groups	
		N	%	N	%	X^2/t	p
Gender	Boys	52	42.6	18	69.2	X^2=6.09	.01
	Girls	70	57.4	8	30.8		
Age	6	14	11.5	3	11.5		
	7	16	13.1	0	0		
	8	16	13.1	7	26.9		
	9	30	24.6	6	23.1		
	10	21	17.2	3	11.5		
	11	15	12.3	6	23.1		
	12	10	8.2	1	3.8		
Average age (SD)		8.9 (1.8)		9.1 (1.7)		t=-.40	.69

TD=typically developing; DCD=Developmental Coordination Disorder; SD=standard deviation

All children were able to complete WAL-K at M1 and, when applicable, M2. Children had no problems with or complaints about the test and the raters observed no falls or trips. The complete test procedure could be performed within ten minutes. Some children needed extra instruction, which occurred more often in younger children and in the children with DCD. One video was missing due to technical problems; therefore, this trial was excluded from data analysis.

Reliability

Results of the reliability analyses in 53 TD children are presented in Table 2. The ICCs for intra-rater and inter-rater reliabilities were 0.99 (95% CI 0.98-0.99) and 0.99 (95% CI 0.97-0.99) for the single run, and 0.99 (95% CI 0.98-0.99) and 0.98 (95% CI 0.97-0.99) for the double run. The ICCs for test-retest reliability were 0.76 (95% CI 0.62-0.85) for the single run, and 0.78 (95% CI 0.31-0.91) for the double run.

Table 2. Intra-, inter-rater and test-retest reliability of the WAL-K (n=53)

	Intra-rater reliability			Inter-rater reliability			Test-retest reliability			
	Mean R1 (SD)	Mean R2 (SD)	ICC (95% CI)	Mean R1 (SD)	Mean R2 (SD)	ICC (95% CI)	Mean M1 (SD)	Mean M2 (SD)	ICC (95% CI)	SDC (%)
single run	13.4 (2.7)	13.4 (2.7)	.99* (.98-.99)	13.4 (2.7)	13.4 (2.8)	.99* (.97-.99)	13.4 (2.7)	13.9 (2.6)	.76* (.62-.85)	3.5 (26.1)
double run	28.9 (5.6)	28.8 (5.7)	.99* (.98-.99)	28.9 (5.6)	28.9 (5.9)	.98* (.97-.99)	28.9 (5.6)	26.3 (5.7)	.78* (.31-.91)	6.0 (20.8)

* p<.001; R1=rating 1; R2=rating 2; M1=measurement 1; M2=measurement 2; SD=Standard Deviation; ICC=Intraclass Correlation Coefficient; CI=95% Confidence Interval; SDC (%)=Smallest Detectable Change at 95% confidence level and % of the mean

Paired samples t-tests and Bland Altman plots (Figure 4) revealed a significant difference between M1 and M2 concerning the double run ($p<0.001$), with lower scores (better performance) on M2 (mean M1: 28.9, mean M2: 26.3). Significant differences were observed in both completion time (-1.0 s, $p=0.04$) and mistakes (-3.2 mistakes, $p<0.001$). For the single run no significant differences between M1 and M2 existed ($p=0.13$).

The SDCs were 3.5 points (26.1% of the mean) for the single run and 6.0 points (20.8% of the mean) for the double run.

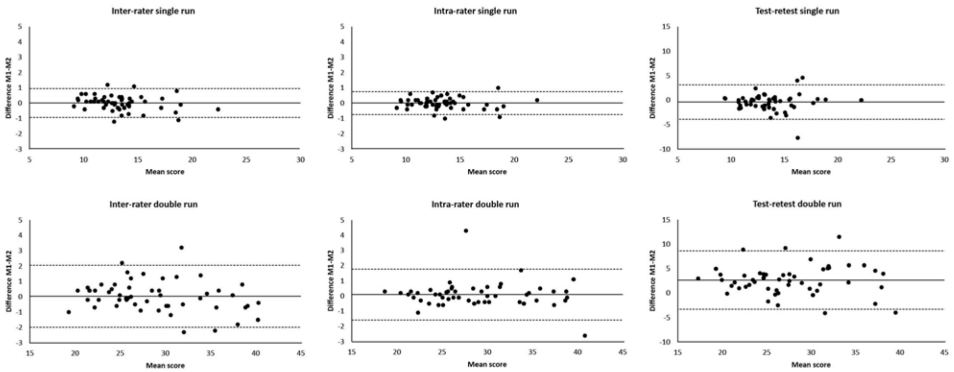

Figure 4. Bland-Altman plots for intra-rater, inter-rater, and test-retest reliability concerning single and double run of the WAL-K in TD children.

Construct validity

A significant, negative correlation was found between age and performance in TD children on the WAL-K for both the single run ($r=-0.61$, $p<0.001$) and the double run ($r=-0.68$, $p<0.001$), with older children performing better (Figure 3). The correlation (Spearman's rho) with age for the children with DCD was -0.54 ($p=0.01$) for the single run and -0.62 ($p=0.001$) for the double run.

A significant difference existed between TD children and children with DCD for both the single ($F(1, 144)=480.09$, $p<0.001$) and double run ($F(1, 144)=860.77$, $p<0.001$) with no significant effect of gender (single run $p=0.43$; double run $p=0.89$). TD children showed a better performance (i.e., lower WAL-K score) than children with DCD (Figure 3 and Table 3). For the single run, seven children with DCD (27%) performed between 1 and 2 SD above the mean of the TD children, and WAL-K scores of six children with DCD (23%) exceeded 2 SD above the mean. For the double run, fourteen children with DCD (54%) performed between 1 and 2 SD above the mean of the TD children, and seven children with DCD (27%) scored higher than 2 SD above the mean.

Table 3. Descriptive statistics in mean (SD) and comparison between groups (independent samples t-test)

		TD	DCD	Comparison between groups		
				t	Effect size	p
WAL-K Single run	Total score	14.2 (3.1)	17.8 (3.6)	-5.25	1.07	<.001
	Completion time	13.4 (2.6)	16.5 (2.8)	-5.58	1.15	<.001
	Mistakes	1.6 (2.8)	2.5 (4.4)	-.97	0.24	.34
WAL-K Double run	Total score	29.1 (6.2)	39.2 (8.3)	-7.06	1.38	<.001
	Completion time	24.2 (4.8)	31.0 (7.9)	-4.29	1.04	<.001
	Mistakes	9.9 (7.9)	16.5 (12.9)	-2.52	0.62	.02
MABC-2 Standard score	Total score	10.0 (2.7)	4.5 (2.2)	8.93	2.23	<.001
	Manual dexterity	9.8 (2.8)	4.7 (2.6)	7.87	1.89	<.001
	Aiming & catching	9.4 (2.7)	6.6 (3.3)	4.12	0.93	<.001
	Balance	10.3 (2.7)	6.5 (2.7)	6.12	1.41	<.001

TD=typically developing; DCD=Developmental Coordination Disorder; SD=standard deviation; WAL-K=Walking Adaptability Ladder test for Kids; MABC-2=Movement Assessment Battery for Children version 2; Total WAL-K score=completion time (s)+0.5*mistakes; Effect size=Cohen's *d*

The linear regression for normalizing the WAL-K scores for age resulted in the following formula for the single run: age-adjusted score=WAL-K score+(1.06*age), and for the double run: age-adjusted score=WAL-K score+(2.37*age). There were significant moderately strong negative correlations between the total score on the MABC-2 and the age-adjusted scores on the WAL-K for the TD and DCD children together (single run, rho=-0.52; double run, rho=-0.60, Table 4). For the TD and DCD groups separately, not all correlations were significant. Weak negative correlations were observed between the total score of the MABC-2 and the WAL-K single run in TD children, while a moderately strong negative correlation between the MABC-2 and the WAL-K double run was found in the DCD children (Table 4). Average scores on the WAL-K and MABC-2 are presented in Table 3.

Table 4. Spearman's rank correlations of age-adjusted WAL-K scores and MABC-2 standard scores

Age-adjusted WAL-K score	MABC-2 standard score	TD+DCD (n = 95)		TD (n = 69)		DCD (n = 26)	
		rho	p	rho	p	rho	p
Single run	Total score	-.52	<.001	-.34	.01	-.38	.07
	Manual dexterity	-.49	<.001	-.28	.02	-.41	.05
	Aiming and catching	-.38	<.001	-.26	.03	-.27	.19
	Balance	-.33	.001	-.10	.40	-.31	.12
Double run	Total score	-.60	<.001	-.19	.13	-.52	.01
	Manual dexterity	-.54	<.001	-.15	.23	-.42	.04
	Aiming and catching	-.34	<.001	-.15	.21	-.17	.41
	Balance	-.44	<.001	-.04	.72	-.50	.01

WAL-K=Walking Adaptability Ladder test for Kids; MABC-2=Movement Assessment Battery for Children version 2; TD=typically developing; DCD=Developmental Coordination Disorder

Discussion

The aim of the current study was to determine the reliability and construct validity of the 10m WAL-K for assessing walking adaptability in children aged 6-12 years old. Excellent intra- and interrater reliabilities were found, as well as good test-retest reliability. The strong correlation with age and the significant differences in WAL-K scores between TD and DCD indicate a good construct validity of the WAL-K.

Our results show excellent intra-rater and inter-rater reliability (ICC>0.9) for the WAL-K, with ICCs in the same order of magnitude as those reported for commonly used gross motor skills tests such as the MABC-2 and BOT-2[43]. In contrast to the live scoring procedures used in these tests, we chose to apply a post-hoc scoring procedure for the WAL-K based on video recordings, which may have contributed to its excellent reliability. In pilot testing it was observed that some children made a substantial number of mistakes, which were difficult to count correctly during live scoring. Using video recordings, the number of mistakes can more easily be measured after the test. Since we used a regular mobile phone to record the videos and the scoring generally took only five minutes, the post-hoc offline video scoring procedure does not appear to impede the clinical utility of the WAL-K. The scoring using video recordings is therefore recommended for clinical use to ensure test reliability. It must be mentioned, though, that inter-rater differences in clinical practice may also be related to differences in instruction and motivation of the testers, and this potential source of variability was not accounted for in our study.

Test-retest reliability for the WAL-K was found to be good: ICC single run=0.76; ICC double run=0.78. These values are comparable to the test-retest reliability of the MABC-2[43], but somewhat lower than for the BOT-2[43], which both have excellent test-retest reliability. Our results for test retest reliability are consistent with the findings of Smits-Engelsman et al., who evaluated the shorter version (4m) of the WAL-K (single run=0.87, double run=0.81)[34]. Similar to their findings[34] a systematic improvement in double run scores was found between M1 and M2, which reflects a learning effect on the test (see Bland-Altman plots in Figure 4). The large confidence interval for the ICC on the double run (0.31 – -0.91) and the rather high SDC values (3.5 points, 26.1% of the mean for the single run; 6.0 points, 20.8% of the mean for the double run) can at least partly be attributed to this learning effect. A learning effect is more often observed in motor tests, as practice allows children to improve on complex motor tasks by forming context-specific memories[44]. In addition, children are more familiar with the test at M2[25]. To reduce this learning effect and to narrow the confidence interval for the ICC on the double run, it would be beneficial to add (at least) one more practice trial to the testing procedure in future studies. This is expected to decrease the SDC and thus improve the utility of the WAL-K as a measurement tool for evaluating the effects of interventions aimed at improving walking adaptability. The SDC should again be calculated after adding one more practice trial and should be calculated in children with DCD, since this would be the target population for an intervention aimed at improving walking adaptability.

Our results provide evidence for good construct validity of the WAL-K. As hypothesized, the TD children performed significantly better on the WAL-K than children with DCD, which shows the discriminative validity of the 10m version of the WAL-K. The difference was most prominent for the double run, with 81% of the children with DCD scoring above the mean +1 SD of the TD children, whereas this was true for 50% of the children with DCD during the single run. Furthermore, we found a strong association between age and WAL-K scores, which demonstrates that the motor skill tested by the WAL-K develops as children grow older, at least within the age range (6 to 12 years) tested in this study. Indeed, we found no ceiling effect, as the scores kept decreasing up to the age of twelve years, particularly in the double run (i.e., the curve in Figure 3 did not stabilize). Compared to the previously reported results of the 4m version of the test, we found stronger correlations between WAL-K scores and age (4m version: r=-0.33 for single run and r=-0.25 for double run; 10m version: r=-0.61 for single run and r=-0.68 for double run). Hence, it appears that the two changes that we implemented compared to the version as reported by Smits-Engelsman et al. (i.e., increasing the length from 4 to 10m, and allowing children to make >3 mistakes) have positively influenced the construct validity of the test.

In line with our hypothesis, the weak to moderate correlations between the WAL-K and the MABC-2 show that the children's motor skill levels are also reflected in their WAL-K scores. However, we did not expect strong correlations, because the WAL-K tests a very specific motor skill, while the MABC-2 tests a much broader set of motor skills. Somewhat surprisingly, the balance subscore of the MABC-2 which includes the gross motor skill items standing on one leg, jumping/hopping, and walking on a line did not yield better correlations with the WAL-K than did the manual dexterity and ball skills subscores of the MABC-2. This might be due to the presumed deficit in the forward modelling of movement, which is a generalized problem which affects both fine and gross motor skills[17]. This may also explain why, in the subgroup analysis, we found moderate correlations between the MABC-2 and the more complex WAL-K double run in the DCD group, whereas only weak correlations were observed in the TD group. It must be mentioned, though, that the DCD subgroup was relatively small and these results should thus be interpreted with caution. Overall, the correlation coefficients of the WAL-K with the MABC-2 in the subgroup analysis were lower than for the combined TD and DCD groups. This can be explained by the limited ranges of scores on the WAL-K and MABC-2 in the separate groups. Compared to the first study on the 4m LAT, the observed correlations with the MABC-2 in the TD children are comparable.

In our study, data of participants of different samples were combined to assess both reliability and construct validity, which may be seen as a limitation. Yet, we felt that combining these data was justified by the applied standardization of the measurement procedures, which was the same across the three samples. Combining these data increased the sample size of the TD children, which allowed for a more reliable estimate of changes in WAL-K scores across the age range tested. The inclusion of TD children scoring below the 16th percentile on the MABC-2 can be considered a limitation, which may have reduced the observed differences in WAL-K scores between TD children and children with DCD. Therefore, the inclusion of these TD children does not affect our conclusion about the differences in performance between TD children and children with DCD. Another limitation may be that our testing protocol did not specifically include instructions on the amount and way of encouragement given to the children. Although this is unlikely to have affected our present results (our two testers were trained together for conducting the measurements), the amount and way of encouragement need to be added explicitly to the testing protocol for minimizing interrater differences in clinical practice. We recommended to give a maximum of two clues during each of the two trials.

2

Future research on the WAL-K should focus on the responsiveness of the test in TD children, taking into account the recommendations regarding one more practice trial to reduce the effects of learning on the task. It also remains to be investigated whether the WAL-K is reliable and responsive in populations with motor problems, such as DCD and Cerebral Palsy. Additionally, age-related reference values of a larger sample of TD children should be determined before the WAL-K can be used as a diagnostic tool for identifying walking adaptability problems.

In conclusion, the newly developed WAL-K shows to be a reliable and valid measurement tool for measuring walking adaptability in children. It is inexpensive and easy to use and takes little time to perform. The WAL-K can easily be used in clinical practice and research for assessment of a child's walking adaptability.

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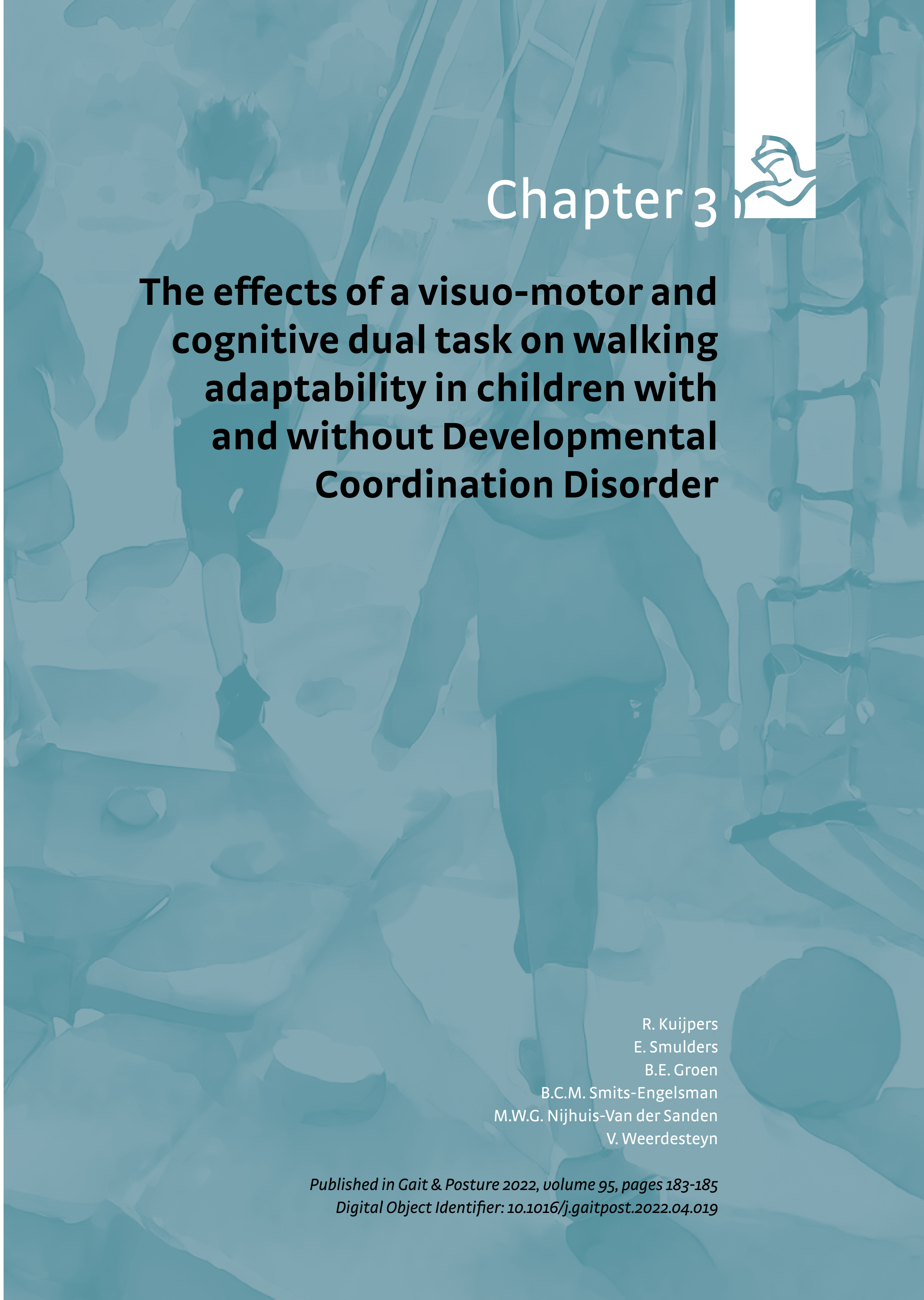
Supplemental material

Supplemental material contains videos of the WAL-K single run and double run, which are available on: https://www.tandfonline.com/doi/suppl/10.1080/09638288.2020.1802523?scroll=top&role=tab



Chapter 3

The effects of a visuo-motor and cognitive dual task on walking adaptability in children with and without Developmental Coordination Disorder

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Abstract

Background
Children with Developmental Coordination Disorder (DCD-C) have motor coordination deficits which lead to difficulties in sports and play activities that require adaptations of the walking pattern. Sports and play often involve performing dual tasks, which affects performance in DCD-C more than in typically developing children (TD-C). So far, testing the impact of dual tasking on walking adaptability in DCD-C has received little scientific attention.

Research question
We tested the hypothesis that 6-12 year old DCD-C will show lower levels of walking adaptability than TD-C, and that due to problems with automatization this difference will increase when they are forced to divide their attention between tasks when a concurrent visuo-motor or cognitive task is added.

Methods
Twenty-six DCD-C and sixty-nine TD-C were included in this cross-sectional study. They performed a challenging walking adaptability (WA) task on a treadmill as a single, a visuo-motor dual and a cognitive dual task at a pace of 3.5 km/h. Repeated measures ANCOVAs were performed with condition (single/dual task) as within-subjects factor, group (TD/DCD) as between-subjects factor, and age as covariate.

Results
DCD-C performed poorer on the WA task than TD-C. The group differences increased when a concurrent visuo-motor task was added, but not when adding a concurrent cognitive task. A significant effect of age was found with younger children performing worse on all tasks.
Significance: The results highlight the problems DCD-C have with walking adaptability and dual tasks, which capacities are essential for full participation in sports and play activities. Future research should investigate whether DCD-C may benefit from task-specific walking adaptability training.

Significance
The results highlight the problems DCD-C have with walking adaptability and dual tasks, which capacities are essential for full participation in sports and play activities. Future research should investigate whether DCD-C may benefit from task-specific walking adaptability training.

Background

Five to six percent of school-aged children have Developmental Coordination Disorder (DCD)[1]. Children with DCD (DCD-C) have motor coordination deficits leading to a higher risk to trip and fall[2]; and to difficulties in daily activities that require adaptations of the walking pattern[3,4] which often involve performing dual tasks. Previous studies have demonstrated that DCD-C have more problems with dual tasking than typically developing children (TD-C), which is likely due to their automatization deficit[5,6]. Hence, DCD-C need to allocate more attention to perform a single task than TD-C, and as a result they have less resources left for the concurrent task[7,8]. These deficits may particularly compound walking adaptability, as making gait adaptations with a concurrent motor or cognitive task requires repeated switching of the attentional focus in a time-critical manner. Yet, the impact of dual tasking on walking adaptability in DCD-C has received little scientific interest[9]. Here, we tested the hypothesis that 6-12 year old DCD-C show lower levels of walking adaptability than TD-C; and that the aforementioned automatization deficit will further impact their performance when the addition of a concurrent visuo-motor or cognitive task forces them to divide their attention. Larger dual-task costs were expected for the visuo-motor than the cognitive task, and most prominently in DCD-C, because attention had to be divided over shared visuo-motor resources[8,10].

Methods

Participants
Twenty-six DCD-C (as defined by the DSM-V criteria1) and sixty-nine TD-C were included, all 6-12 years without neurological, orthopedic or cardiovascular disorders, or visual problems. All participants' parents gave written informed consent. The study was approved by the regional Medical Ethics Committee (NL59150.091.16).

Procedure
Walking adaptability (WA) tasks were performed on the C-mill, a treadmill with embedded force plates on which stepping stones or obstacles were projected to evoke gait adjustments[11]. Children first practiced undisturbed treadmill walking. They then performed the WA tasks at a pace of 3.5 km/h in the following order: first as a single task (WA^{single}), second while concurrently performing a secondary visuo-motor task (WA^{motor}), and third with a secondary cognitive task ($WA^{cognitive}$). All tasks were developed for the purpose of this study. In the dual-task conditions, children were instructed to perform both tasks as well as they could. The WAsingle involved continuously stepping -as accurately as possible- on white stepping stones projected on the treadmill belt according to the individual walking pattern (as registered by the C-mill during walking without projected context). Ten percent of these stepping stones randomly changed into obstacles (red stripes projected across the stepping stone) two steps before expected foot landing. After one minute of practice, the task was recorded for four minutes. The secondary visuo-motor task involved stabilizing a tennis ball on a racket. The ball was attached to the racket with a string to prevent it from falling on the treadmill. When the ball fell off the racket, the researcher placed it back. The number of falling balls was registered. The secondary cognitive task comprised listening to a music piece interspersed with sounds (n=34) such as a doorbell, a phone, and a rooster. The children had to say 'yes' when they heard the rooster

(n=13). The number of omitted/wrong answers was registered. The secondary tasks were also performed as a single task while standing still for two minutes, directly before the WA dual task. The primary outcome was WA success rate during 4 min. A step was considered successful when the measured center of pressure at midstance lay within the stepping stone or outside the obstacle.

Statistical analysis

Data were analysed with SPSS V25.0 (SPSS Inc, Armonk, NY). We performed two separate repeated measures ANCOVAs (WA^{single} versus WA^{motor}; WA^{single} versus $WA^{cognitive}$) with condition (single/dual task) as within-subjects factor, group (TD/DCD) as between-subjects factor, and age (in months) as covariate (alpha=0.025 to correct for repeated testing). For the secondary visuo-motor and cognitive tasks, differences in performance (error rate per 2 min) between the single and dual task were tested with a Wilcoxon signed-rank test. To evaluate the interaction between task (single/dual) and group, we compared differences in error rate of the single and dual task between the groups (TD/DCD) with a Mann Whitney U-test. Alpha was set at.05.

Results

The results are displayed in Table 1 and Fig. 1; individual scores are shown in the appendix. Seven TD-C (10%) and four DCD-C (15%), 6-9 years old, did not perform the WA^{motor} because they felt insecure. For the $WA^{cognitive}$, data of one child with DCD was missing because he developed motion sickness. All children were well able to perform the measurements without concentration problems or getting tired. WA^{motor} success rates were lower than WA^{single} (*task*, $F(1,81)=35.12$, $p<.001$, $\eta_p^2=.30$) and DCD-C performed poorer than TD-C (*group*, $F(1,81)=38.39$, $p<.001$, $\eta_p^2=.32$). The difference between the groups was magnified when adding a secondary visuo-motor task (*group*task*, $F(1,81)=8.84$, $p=.004$, $\eta_p^2=.10$). Success rates improved with age (*age*, $F(1,81)=45.47$, $p<.001$, $\eta_p^2=.36$). WA^{single} and $WA^{cognitive}$ success rates did not differ (*task*, $F(1,91)=0.37$, $p=.544$, $\eta_p^2=.004$), but DCD-C performed poorer than TD-C (*group*, $F(1,91)=26.18$, $p<.001$, $\eta_p^2=.22$). No group by task interaction was found (*group*task*, $F(1,91)=0.33$, $p=.567$, $\eta_p^2=.004$). Success rates again improved with age (*age*, $F(1,91)=39.37$, $p<.001$, $\eta_p^2=.30$). Under dual-task conditions, we observed higher error rates on the secondary visuo-motor (TD: z=-5.8, p<.001; DCD: z=-4.1, p<.001) and cognitive tasks (TD: z=-4.7, p<.001; DCD: z=-3.9, p<.001) compared to single task conditions. DCD-C showed larger decrements in secondary task performance than TD-C (visuo-motor: U=188.0, z=-5.0, p<.001; cognitive: U=562.0, z=-2.7, p=.008).

Table 1. Descriptive statistics and comparison between groups (Independent samples T-test, Chi-Square test for gender, Mann Whitney U test for error rates)

	TD (n = 69)	DCD (n = 26)	*p*-value
Group characteristics, mean (SD)			
Age	8.9 (1.9)	9.1 (1.7)	.937
Gender	25 boys (36%) 44 girls (64%)	18 boys (69%) 8 girls (31%)	.004
Length (m)	1.39 (0.12)	1.42 (0.13)	.409
Weight (kg)	32.4 (8.2)	36.6 (11.8)	.051
BMI (kg/m2)	16.5 (2.0)	17.9 (3.1)	.009
MABC-2 total standard score	10.0 (2.7)	4.5 (2.2)	<.001
Walking adaptability success rate, mean (SD)			
WA^{single}	87.5 (13.1)	73.2 (17.9)	.001
WA^{motor}	82.5 (16.3)	60.6 (23.0)	<.001
$WA^{cognitive}$	86.2 (14.4)	73.0 (16.9)	<.001
Secondary task performance, median (IQR)			
Visuo-motor task error rate (errors/2 min)			
Single task	0.0 (0.0)	0.0 (1.0)	.005
Dual task	1.5 (4.0)	7.5 (6.8)	<.001
Difference between single and dual task	-1.5 (4.0)	-7.0 (5.0)	<.001
Cognitive task error rate (errors/2 min)			
Single task	0.0 (0.0)	0.0 (0.0)	.093
Dual task	0.0 (1.0)	1.0 (1.3)	.022
Difference between single and dual task	0.0 (1.0)	-1.0 (1.3)	.008

TD=typically developing; DCD=Developmental Coordination Disorder; SD=standard deviation; IQR=interquartile range

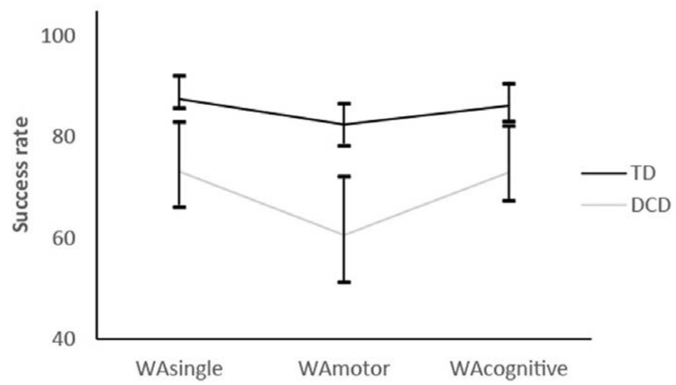

Figure 1. Mean scores with 95% CI per group per task.

Discussion

This study confirmed our hypothesis that walking adaptability is affected in 6-12-year-old DCD-C, with a disproportionate decrement in both WA and secondary task performance when a concurrent visuo-motor task was added. The concurrent cognitive task did not have a greater impact on walking adaptability in DCD-C compared to TD-C, yet both secondary tasks in DCD-C showed larger decrements under dual-task conditions. The observation that dual-task costs were more pronounced for WA^{motor} may be due to this task recruiting resources from the same (visuo-motor) domain, while the $WA^{cognitive}$ recruits resources from different domains (visuo-motor and auditive)[8,10]. Our findings are consistent with previous dual task studies that included walking tasks[9,12,13], yet our novel standardized assessment on the C-mill imposed time-critical step adjustments to targets and obstacles to further challenge the children's dual-task walking capacity. The fixed walking speed forced the children to divide their attention over the WA and secondary task to keep up performance; this allowed us to test automatization of walking adaptability at the children's maximum capacity. Particularly in the WA^{motor} we found no ceiling effects in success rates even in older TD-C. The substantially lower success rates in DCD-C highlight the difficulties they may experience while dual-tasking in daily activities that require adaptations of the walking pattern to the dynamic environment. A limitation is that some younger children, mostly DCD-C, did not perform the WA^{motor} because they felt too insecure, which may also be due to this task recruiting resources from the same (visuo-motor) domain which makes it more challenging. Therefore, the reported difference in dual-task effects between the groups may be an underestimation. Future research may focus on training of walking adaptability in children with motor problems, as mastering this skill is important for participation in daily activities[5,14]. Better automatization of walking adaptability may increase the capacity for dual tasking important in daily-life activities[5].

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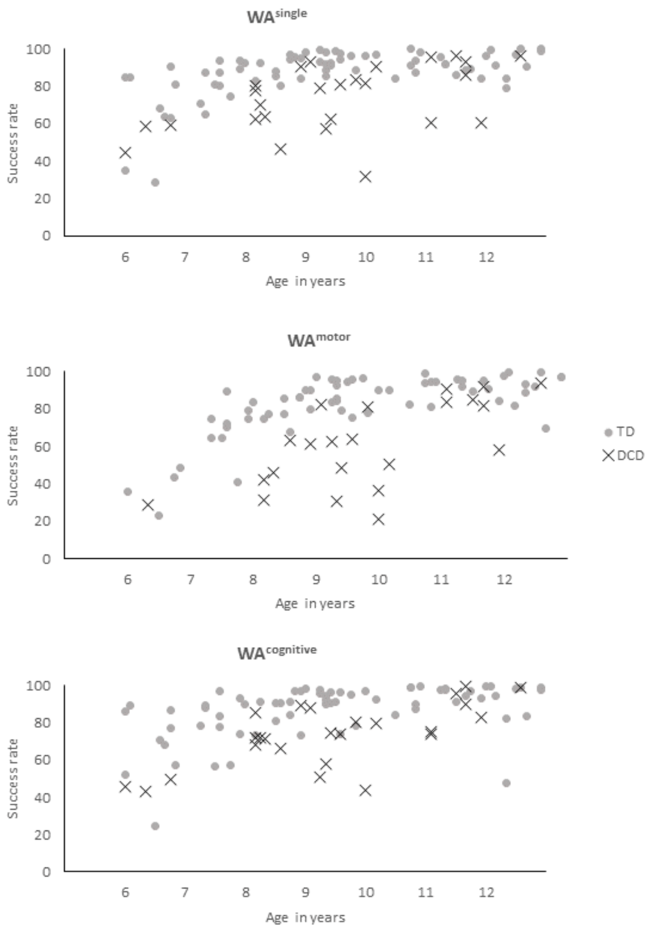
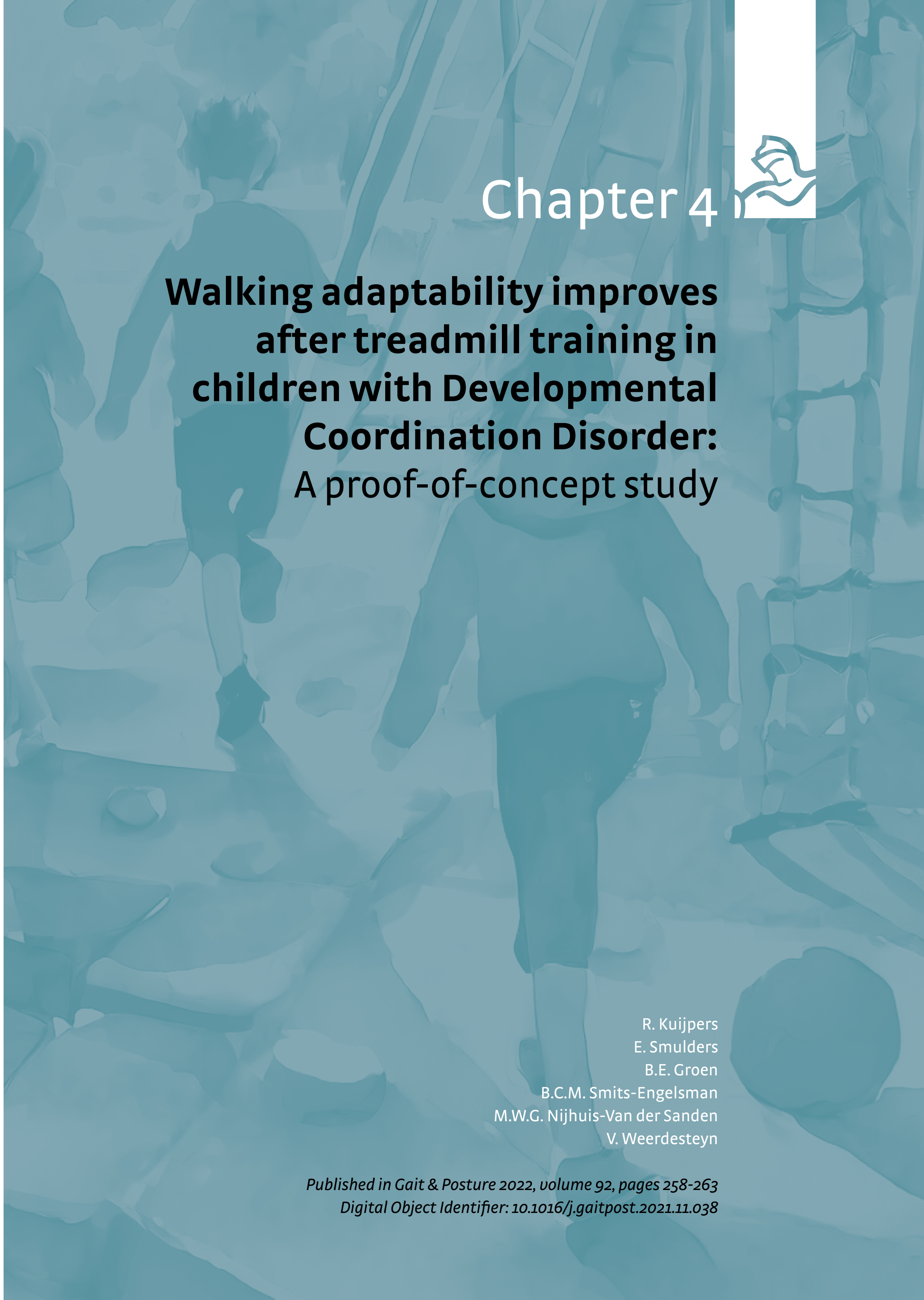
3

Supplemental material

Appendix I. Data of TD children and children with DCD on the three tasks.



WA^single^=Walking Adaptability single task; WA^motor^=WA task combined with visuo-motor task; WA^cognitive^=WA task combined with cognitive task; TD=Typically Developing children; DCD=children with Developmental Coordination Disorder.



Chapter 4

Walking adaptability improves after treadmill training in children with Developmental Coordination Disorder: A proof-of-concept study

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Abstract

Background
Children with Developmental Coordination Disorder (DCD) have motor coordination deficits leading to difficulties in sports and play that require adaptations of the walking pattern. Children with DCD indeed demonstrate poorer walking adaptability (WA) compared to typically developing children, but it remains elusive whether WA can be improved by training.

Research question
Does augmented-reality treadmill training lead to improvements in WA in children with DCD?

Methods
Seventeen children with DCD were included in this proof-of-concept intervention study. They received a six-session training on the C-mill, a treadmill on which gait adjustments can be evoked by projected visual context. The effect of the training was evaluated before (M1), directly after training (M2) and after 6 months follow-up (M3) using the WAL-K (single and double run) and WA-tasks on the C-mill (as a single and with concurrent visuo-motor and cognitive task). In addition, parents completed a questionnaire on their perception of the training. Linear Mixed Model analyses were performed to assess the differences in WAL-K scores and success rates on the WA-tasks between M1-M2 and M1-M3.

Results
Children significantly improved on the WAL-K double run and on all three WA-tasks between M1-M2 and M1-M3. Children did not improve on the WAL-K single run. Parents found the training useful and fun for their child and indicated that their child fell less frequently.

Significance
The results show that C-mill training had positive and task-specific effects on WA in children with DCD, which effects generalized to an overground task and were retained at 6 months follow-up. This may help children with DCD to better participate in daily activities. Future research should include a control group to examine the effectiveness of the training program compared to receiving no training and may also examine the effect of the training on participation in daily life.

Background

Children with Developmental Coordination Disorder (DCD) are impaired in both gross and fine motor coordination[1]; the estimated prevalence of DCD is 5-6% in school aged children[2]. Children with DCD experience difficulties in daily activities such as sports, play, and school activities, and parents often report their children to be clumsy and to fall frequently[3]. As a result of their motor problems, children with DCD participate less in physical activities compared to their peers, especially in team sports[2]. This may lead to psychosocial problems –such as social anxiety or depression– or physical health problems –such as obesity or decreased physical fitness[2].

4

A prominent hypothesis concerning motor problems of children with DCD is a deficit in the internal modeling of movement (i.e., predictive control)[4]. The central nervous system uses an internal model to predict the future location of moving limbs enabling rapid online corrections during movement. A deficit in predictive control limits both the speed and accuracy of online error correction in gross and fine motor activities in children with DCD[4-8]. Predictive control is also essential during walking, when frequent and rapid modifications of the walking pattern have to be made to handle the demands of the environment like puddles, obstacles or uneven surfaces[9]. This so-called walking adaptability is highly relevant in children's daily activities such as avoiding objects or playmates during sports or playtime. Children with DCD perform worse on walking adaptability tasks compared to typically developing (TD) children[10-12], which performance deficit is further aggravated when a secondary motor or cognitive task is added[12-14]. Yet, while recent studies have demonstrated that predictive control of the upper extremity can be improved by training[15,16], such studies related to walking adaptability in children with DCD are still lacking.

The aim of this proof-of-concept study was to examine the effect of augmented reality treadmill training on walking adaptability in children with DCD. Children practiced on the C-mill (Motek Medical, Culemborg, the Netherlands), in which the treadmill belt was augmented with visual context (targets/obstacles) to provoke walking adaptations. The characteristics of training tasks were aligned with the specific motor control deficits in children with DCD, i.e., "tasks that demand precision (both spatial and temporal), advanced planning, or that stress the system in a way that requires some adaptation/adjustment at a perceptual-motor level to maintain stability"[6]. We hypothesized that C-mill training would yield significant improvements in walking adaptability tasks on the treadmill, and that these task-specific training effects would also generalize to an overground walking adaptability task. In interpreting the results of this uncontrolled study, we will also discuss our findings in light of reference values of TD children[12,17].

Methods

Participants
Twenty-seven children with DCD, 6-12 years old, were recruited from the patient database of Tolbrug Specialized Rehabilitation, patients' association, physical therapy practices, and schools. Children needed to have a formal diagnosis of DCD from a medical practitioner, or had to meet the DSM-V criteria as tested by the researcher (RK)[17,18]. In addition, the children

had to perform below the median age-adjusted reference walking adaptability scores of TD children[12,17]. Exclusion criteria were temporary complaints influencing walking ability, or neurological, orthopedic, cardiovascular, or visual problems. Parents gave written informed consent prior to participation. The study was approved by the regional Medical Ethical Committee Arnhem-Nijmegen (2016-2885).

Study protocol

The children received six 30-minute sessions of C-mill training (180 min) in three weeks which is comparable to other studies which found improvements of motor skills in children with DCD after interventions of 120 and 200 min, respectively[19-21]. At baseline, descriptive data were collected regarding age, gender, height, weight, and current sport activities. Data of the most recent assessment of the MABC-2 were obtained from the child's physical therapist. If this assessment was more than 6 months ago, the MABC-2 was reassessed[22]. Walking adaptability assessments were conducted at baseline (M1), after the training period (M2), and after a follow-up period of 6 months (M3). All measurements and training sessions were performed by a specialized pediatric exercise therapist (RK) at Tolbrug Specialized Rehabilitation in 's-Hertogenbosch or Radboud university medical center in Nijmegen, The Netherlands between September 2017 and December 2018.

C-mill training

Training sessions (see Table 1) were performed at comfortable walking speed and participants were instructed not to use the handrails. The type and duration of the exercises were standardized for all participants, but the level of difficulty was adjusted to the individual performance level of the child, based on the therapist's clinical judgment.

The training protocol was designed in line with the international clinical practice recommendations on DCD, which state that training of children with DCD should be activity- or participation-oriented, focused on daily activities, and meeting goals that are relevant in daily life[2]. C-mill training also meets the need of children with DCD for receiving augmented (i.e., extrinsic) feedback when learning novel and complex motor skills[6]. In addition, the interactive and gaming-like training opportunities of the C-mill were deemed particularly engaging and motivating for children.

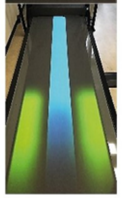
Walking adaptability measurements

To evaluate training effects, we conducted the Walking Adaptability Ladder test for kids (WAL-K) and walking adaptability tasks (WA-tasks) on the C-mill. The measurements were performed during separate study visits in the fixed below-mentioned order.

Walking Adaptability Ladder test for Kids (WAL-K)

Children performed the WAL-K, a 10-meter agility ladder with 19 targets successively decreasing in size from 64 to 28 cm[17]. The child was instructed to walk back and forth, while stepping into the targets as fast and accurately as possible. The WAL-K was completed in two conditions: while stepping with one (single run) and both feet (double run) in each target. WAL-K score was calculated as completion time plus a 0.5 s penalty for each mistake (touching a bar, the wrong number of steps in a target, or missing a target). The WAL-K has been shown reliable and valid for measuring walking adaptability in children[17].

Table 1. Training protocol

	Exercise		Duration (min)
1	Determination of comfortable walking speed (only in training sessions 1 and 2)		2
2	Warming up at comfortable walking speed		3
3	Target stepping: with variation in step length, step width, and symmetry. Difficulty was increased by increasing the irregularity between steps.		5
4	Obstacle avoidance: projection of unilateral or bilateral obstacles in front of the left or right foot, with differing available response times and sizes. Visual and auditory feedback on performance was given. Difficulty was increased by increasing the sizes and available response times.		5
5	Slalom walking: projection of a slalom walking path with cones in the corners. Visual feedback on performance was given. Difficulty was increased by decreasing the width of the path and increasing the frequency of corners.*		2.5
6	Tandem walking: projection of a narrow walking path. Visual feedback on performance was given. Difficulty was increased by decreasing the width of the path.*		2.5
7	Fun and functional forest game: projection of targets (footballs or stars) and obstacles (e.g. rabbit, tree, squirrel) in which children received points for successful target hits, but lost points when hitting obstacles. Visual and auditory feedback on performance was given. Children of 6-9 years old performed the game at an easy level; children of 10-12 years old performed the game at a medium level.		4

The comfortable walking speed of training 2 was used in training 3 to 6. Between all exercises, children had a 1-minute break. The order of exercises 3 to 6 was different in every session. *The slalom and tandem exercises were alternated: the slalom was performed in sessions 1, 3 and 5; the tandem was performed in sessions 2, 4 and 6.

WA-tasks on the C-mill

The velocity of the treadmill was set at 3.5 km/h for all C-mill measurements. Following 2-minute familiarization of walking on the treadmill, the WA-tasks were performed. White stepping stones were projected across the entire treadmill relative to foot placement, based on the center of pressure position at mid stance. Approximately 6 stepping stones were visible at any time ahead of the participant, depending on step length. The child was instructed to step on the stepping stones as accurately as possible. Of these stepping stones, 10% randomly changed into an obstacle two steps before expected foot landing, as indicated by the stepping stone changing to red-white color. The child was instructed to step beside the obstacle and then onto the next stepping stone. The child first practiced the task for one minute, then it was recorded for four minutes (≥20 obstacles). Importantly, this task was not included in the training sessions. The children performed the WA-task as a single task (WA^{single}), and while concurrently performing a secondary visuo-motor (WA^{motor}) or cognitive task (WA^{cognitive}). A 2-minute break was allowed between each task.

The secondary motor task involved stabilizing a tennis ball on a racket. The ball was attached to the racket with a string to prevent it from falling on the treadmill. When the ball fell off the racket, the researcher placed it back. The number of ball drops was counted. The secondary cognitive task involved listening to a piece of music interspersed with sounds (n=34), such as a doorbell, a ringing phone, and a rooster. The child had to indicate the sound of the rooster (n=13) by saying 'yes'. The number of omitted/wrong answers was registered. Prior to performing the WA^{motor} and WA^{cognitive}, motor and cognitive single task performance was recorded while standing still for two minutes.

WA-task performance was calculated as the weighted score for hitting the stepping stones and avoiding the obstacles. A step was considered successful when the measured center of pressure at mid stance lay within the stepping stone or outside the boundaries of the obstacle. For the secondary visuo-motor and cognitive tasks, error rates per 2 min were determined.

Questionnaires

At M2 and M3, parents completed a short survey on their perception of the C-mill training using a 5-point Likert scale. At M2, parents rated how much their child experienced the training as fun, difficult, and useful, how they perceived the quality of the supervision, and how likely they would recommend the training to others. At M3, parents rated their child's activity level, falls, responsiveness to the environment during walking, participation in play and sports activities, and self-perception on his/her motor skills in the 6 months after the training. There was also room for additional comments.

Statistical analysis

To assess the differences in WAL-K scores (total scores, completion time, and mistakes) and WA success rates (WA^{single}, WA^{motor} and WA^{cognitive}) between M1-M2 and M1-M3, Linear Mixed Model (LMM) analyses were performed. Data were normally distributed. Dummy variables for M2 and M3 were included in the model as fixed effects. An unstructured covariance type was used because of the uneven time periods between the measurements. Restricted maximum likelihood was used to estimate the parameters.

For the error rates of the secondary tasks on the C-mill, the differences between M1-M2 and M1-M3 were analyzed using a Friedman test (with post hoc Wilcoxon signed rank test), as these data were not normally distributed. Results of the surveys at M2 and M3 were presented descriptively. All analyses were performed using IBM SPSS Statistics v.25 (IBM Corp., Armonk, NY), and alpha was set at 0.05.

Results

Of the 27 recruited children, 6 children were excluded because they performed above the median age-adjusted reference walking adaptability scores of TD children[12], and 4 children declined participation prior to the start of the training. The remaining 17 children (13 boys, 4 girls) with DCD had a mean age of 8.8 (±1.7) (Table 2) and all completed the six training sessions. Two 6-year-old children and one 8-year-old child did not perform the WA^{motor} because they felt unconfident. For the WA^{cognitive}, data of one 10-year-old boy was missing at M1 because he developed motion sickness due to the moving projections on the C-mill. One 10-year-old child was lost to follow-up at M3.

Table 2. Descriptive statistics

Group characteristics, mean (SD)			
Gender	13 boys (76%), 4 girls (24%)		
Age (years)	8.8 (1.7)		
Height (cm)	140 (12)		
Weight (kg)	35.8 (9.5)		
Sports (min/week)	109 (84)		
MABC-2 manual dexterity standard score	4.4 (2.4)		
MABC-2 ball skills standard score	6.5 (3.5)		
MABC-2 balance standard score	6.2 (3.0)		
MABC-2 total standard score	4.4 (2.3)		
	M1 (n = 17)	M2 (n = 17)	M3 (n = 16)
WAL-K, mean (SD, range)			
Single run total score (s)	18.7 (3.6, 13.7-27.0)	19.2 (3.8, 14.2-28.1)	18.2 (3.1, 14.2-25.0)
Single run completion time (s)	17.2 (3.0, 13.7-22.5)	18.0 (3.1, 14.2-24.1)	17.6 (3.0, 13.7-23.0)
Single run mistakes (n)	3.0 (4.8, 0.0-15.0)	2.2 (2.4, 0.0-8.0)	1.3 (1.7, 0.0-6.0)
Double run total score (s)	40.4 (6.9, 29.0-57.2)	37.1 (7.7, 27.5-56.6)	36.5 (7.9, 25.9-51.5)
Double run completion time (s)	32.4 (8.8, 16.8-51.7)	29.9 (6.5, 21.0-45.6)	33.2 (6.3, 25.4-48.5)
Double run mistakes (n)	15.9 (13.0, 0.0-46.0)	14.3 (12.2, 0.0-40.0)	6.8 (6.0, 0.0-20.0)
WA success rate, mean (SD, range)			
WA^{single} (%)	69.3 (18.0, 31.6-95.9)	85.9 (14.1, 39.1-98.8)	91.1 (11.8, 51.9-100.0)
WA^{motor} (%)*	54.7 (22.4, 21.1-91.7)	71.8 (20.2, 30.1-94.4)	80.3 (14.3, 35.8-99.3)
WA^{cognitive} (%)#	67.2 (17.0, 43.2-99.1)	86.2 (12.3, 54.0-99.0)	90.2 (9.7, 58.8-99.7)
Secondary task performance, median (IQR)			
Visuo-motor task error rate (errors/2 min)			
Single task	1.0 (3.0)	0.0 (1.0)	0.0 (2.0)
Dual task	10.0 (5.9)	7.5 (6.4)	7.8 (8.4)
Cognitive task error rate (errors/2 min)			
Single task	0.0 (0.0)	0.0 (0.0)	0.0 (0.0)
Dual task	1.5 (1.4)	0.0 (0.5)	0.0 (0.4)

MABC-2=Movement Assessment Battery for Children, version 2; SS=standard score; WAL-K=Walking Adaptability Ladder test for Kids; WA=Walking Adaptability task; *data of 3 children were missing at all measurement moments; #data of one child was missing at M1.

WAL-K

Double run WAL-K scores significantly decreased (i.e., improved) between M1-M2 (-8%) and M1-M3 (-9%; Table 3 and Figure 1). Between M1-M2, no significant difference in completion time and mistakes was found. Between M1-M3, children made significantly fewer mistakes (-58%), but no significant difference in completion time was found. Five children (29%) improved more than the previously reported Smallest Detectable Change (SDC) of 6.0 s between M1-M2[17]. Eight children (50%) improved more than 6.0 s between M1-M317. No significant differences were found between M1-M2 and between M1-M3 for the WAL-K single run.

Table 3. Results Linear Mixed Model

	Comparison M1-M2 F (df)	p	Comparison M1-M3 F (df)	p
WAL-K				
Single run total score	.70 (1,16.0)	.415	.65 (1,14.6)	.434
Completion time	1.3 (1,16.0)	.267	.327 (1,15.7)	.576
Mistakes	.596 (1,16.0)	.452	4.0 (1,16.1)	.063
Double run total score	4.6 (1,16.0)	.049	5.0 (1,16.1)	.040
Completion time	2.4 (1,16.0)	.140	.083 (1,16.0)	.777
Mistakes	.327 (1,16.0)	.576	7.4 (1,16.1)	.015
WA success rate				
WA^{single}	23.2 (1,16.0)	<.001	39.1 (1,16.0)	<.001
WA^{motor*}	11.4 (1,13.0)	.005	25.3 (1,13.1)	<.001
$WA^{cognitive\#}$	17.0 (1,15.0)	.001	40.0 (1,15.0)	<.001

WAL-K=Walking Adaptability Ladder test for Kids; WA=Walking Adaptability; *data of 3 children were missing at all measurement moments; #data of one child was missing at M1.

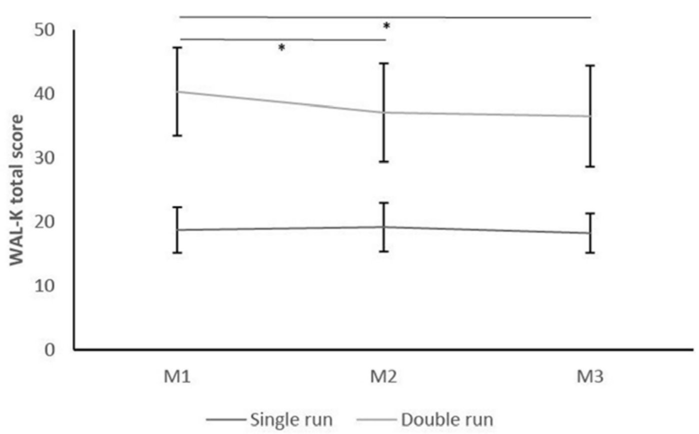


Figure 1. Mean scores with SDs for the WAL-K total scores. *p<.05; indications of significance only count for the double run.

WA-tasks on the C-mill

Success rates of all WA-tasks had significantly improved between M1-M2 (+24-31%) and M1-M3 (+32-47%; Table 3 and Figure 2). Children made significantly fewer mistakes on the secondary task during $WA^{cognitive}$ (X^2=11.61, p=.003) between M1-M2 (-67%; Z=-2.89, p=.004) and M1-M3 (-56%; Z=-2.35, p=.019). No significant change was seen in secondary task performance for WA^{motor} (X^2=2.23, p=.328).

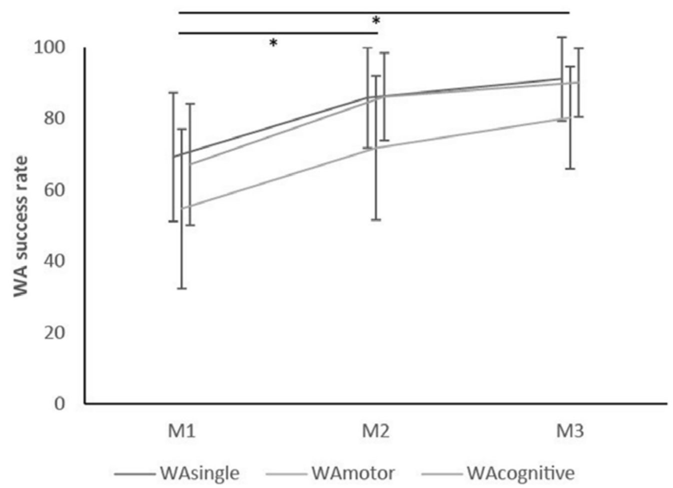


Figure 2. Mean scores with SDs for the success rates of the WA-tasks. *p<.01; indications of significance count for all three tasks.

Evaluation training

Results of the surveys are displayed in Table 4. At M2, parents were in general positive about the fun their child experienced, the usefulness of the training, and the supervision, while the perceived difficulty of the training was most often rated as neutral. The majority of parents (93%) would recommend the training to others.

At M3, 73% of the parents indicated that their child fell less frequently. About half of the parents indicated that their child better responded to the environment during walking and had a more positive self-perception on his/her motor skills, while the other half of the parents was neutral about these statements. The results concerning physical activity and participation were more variable.

Discussion

The aim of this proof-of-concept study was to examine the effect of C-mill augmented reality treadmill training on walking adaptability in children with DCD. We found improvements on treadmill-based walking adaptability tasks, and we observed generalization of training effects to an overground walking adaptability task (WAL-K double run). These effects were seen immediately after training and were retained after 6 months follow-up.

Table 4. Results of the survey on the parents' perception of the training and its utility

		Totally disagree	Disagree	Neutral	Agree	Totally agree
M2 (n=14)	My child experienced the training as fun			2 (14%)	3 (21%)	9 (64%)
	My child experienced the training as difficult	1 (7%)	4 (29%)	8 (57%)	1 (7%)	
	My child experienced the training as useful			3 (21%)	4 (29%)	7 (50%)
	Supervision during the training was good				2 (14%)	12 (86%)
	I would recommend the training to others			1 (7%)	4 (29%)	9 (64%)
M3 (n=15)	In the 6 months after the training, my child...					
	... was more active		3 (20%)	5 (33%)	3 (20%)	4 (27%)
	... fell less			4 (27%)	8 (53%)	3 (20%)
	... better responded to the environment during walking			8 (53%)	5 (33%)	2 (13%)
	... participated more in play and sports activities		1 (7%)	9 (60%)	4 (27%)	1 (7%)
	... had a more positive self-perception on his/her motor skills			7 (47%)	5 (33%)	3 (20%)

The improved success rates on all three WA-tasks on the C-mill confirm the expected task-specific effects of the C-mill training program on walking adaptability. These effects correspond with earlier research that demonstrated the importance of task-specificity of training for improving motor skills in children with DCD2. Compared to the mean scores of 69 TD children in a previous study (mean age 8.9 years, SD 1.9), the walking adaptability success rates of the children with DCD at baseline were on average 1.3-1.7 SDs lower, yet at the follow-up assessment, the performance of the children with DCD had advanced to values of 0.13 below to 0.28 SDs above the mean scores of TD children[12]. As the particular WA-tasks were not included in the training, these findings may suggest a training-induced normalization of walking adaptability performance in children with DCD. Yet, as the context on the C-mill during training and assessments was similar, it is likely that the improved walking adaptability success rates can at least partly be attributed to familiarization with these types of treadmill-based tasks.

In addition to the improved success rates following training and after follow-up, we also observed improvements in secondary task performance while performing the WA-task, albeit only significantly for the cognitive task. These results indicate that after training, children needed to allocate less attentional resources to the WA-task –while also achieving better success rates– and had more capacity left to focus on the secondary task, possibly due to improved automatization of walking adaptability skills.

Importantly, the effects of training were not only observed on the C-mill-based WA-tasks, but also generalized to walking adaptability overground. This transfer to an overground task is particularly relevant, since children with DCD are known to have difficulties with the transfer of training effects to daily life situations[2]. At baseline, average double run WAL-K scores were 2.1 SDs below the age-adjusted reference values of TD children, whereas after follow-up, this difference was reduced to 1.4 SDs below reference values[17]. These improvements on the WAL-K double run were not only significant, but exceeded the previously reported SDC value of 6 s in half of the children with DCD at the follow-up measurement[17]. Hence, this confirms the presence of a clinically relevant effect of training on overground walking adaptability in children with DCD, yet they did not advance to the performance level of TD children[17]. The transfer to daily life was also supported by the results of the questionnaire; the majority of parents reported fewer falls, and half of the parents responded that their child better attended to the environment. It may be of interest for future studies to explore whether adding overground walking adaptability exercises during training may further improve the generalization of the training effects.

In contrast to the observed improvements on the WAL-K double run, no significant effects were found on the single run, whereas the baseline performance deficit on the single run (2.0 SDs below reference values of TD children) was in the same order of magnitude to that on the double run. As the observed improvement on the double run total score after follow-up was largely due to a reduction in the number of mistakes (i.e., accuracy) without significant changes in completion time, it may be suggested that the lower number of mistakes on the single run (3.0 vs. 15.9 on the double run) left relatively little room for improvement in task accuracy. This is partly in line with the results of previous research, showing that following Wii balance training, task accuracy in a dynamic balance game was more sensitive to change than speed20.

Our training was designed to challenge the children's predictive control by provoking rapid and unexpected changes of the walking pattern. Although our outcome measures did not exclusively assess predictive control deficits, the results generally support an improvement in predictive control as the underlying mechanism. It is known that problems with predictive control are being magnified during dual tasks[6], hence the reduction in dual task costs that we found in the WA^{motor} and $WA^{cognitive}$ fits with the suggested improvement in predictive control. In addition, the generalization of the training effect to the untrained, overground context of the WAL-K double run and the retention after 6 months follow-up are in line with this hypothesis. However, with the current results we cannot provide conclusive evidence on whether children improved their predictive control after walking adaptability training, so this warrants further research.

This proof-of-concept study comes with some limitations. The lack of a control group precludes identifying to what extent the observed improvements may be explained by learning effects on the task itself. It must be mentioned, though, that the improvements in WAL-K double run scores (3.3 at M2 and 3.9 s at M3) exceed the previously reported test-retest difference in TD children of 2.6 s[17]. Therefore, we believe that children showed a real improvement on the WAL-K after training. A second limitation concerns the relatively short intervention period of only 180 min. Although this may not be intensive enough to fully achieve the potential benefits of C-mill training in children with DCD, we deliberately chose this limited training period because the largest gains are generally achieved in the first sessions of an intervention[23] and previous studies also found improved motor skills after short interventions (120 and 200 min) in children with DCD[19-21]. It remains an open question whether a longer training period or a blended C-mill and overground training program might result in larger gains.

In conclusion, C-mill training was well-received by the children and their parents and it appears promising for improving walking adaptability in children with DCD, which skill is highly relevant in daily life activities. The current study gives directions for possible measurement tools and effect sizes, which information is needed for conducting a definite trial on the efficacy of C-mill training for improving walking adaptability in children with DCD. Based on the present results, we recommend using the double run of the WAL-K as the primary outcome measure for such a trial.

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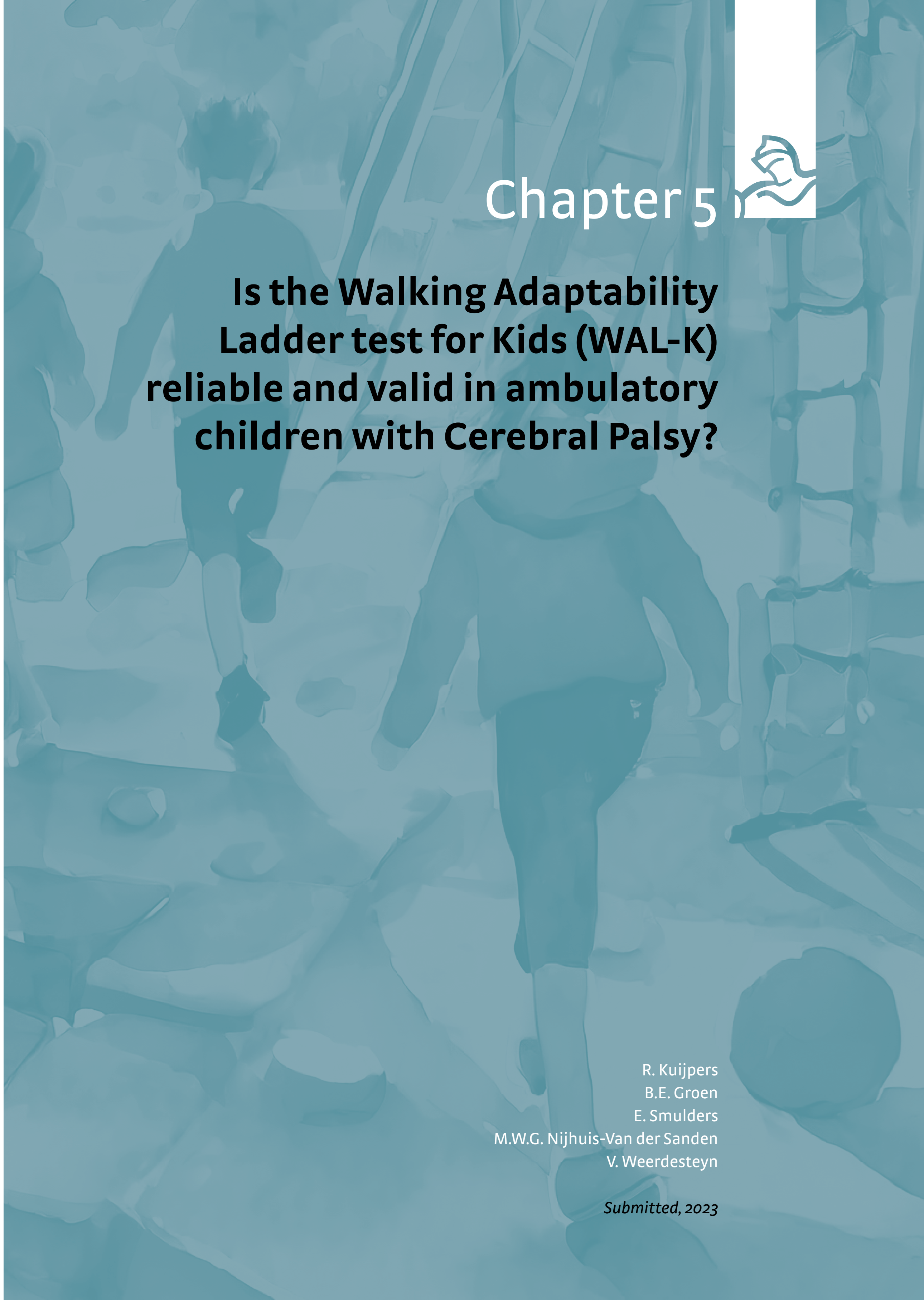

Chapter 5

Is the Walking Adaptability Ladder test for Kids (WAL-K) reliable and valid in ambulatory children with Cerebral Palsy?

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Submitted, 2023

Abstract

Purpose
Walking adaptability is essential for children to participate in daily life. We studied whether the Walking Adaptability Ladder test for Kids (WAL-K) is reliable and valid for assessing walking adaptability in 6-12 year old ambulatory children with Cerebral Palsy (CP).

Materials and methods
Thirty-six children with CP (26 GMFCS level I, 10 GMFCS level II) completed the single and double run of the WAL-K. Intra- and inter-rater reliability were determined by Intraclass Correlation Coefficients (ICCs). Construct validity was determined by comparing WAL-K scores between 122 typically developing (TD) and CP children taking age into account, comparing WAL-K scores between CP children in GMFCS levels I and II, and correlating WAL-K scores with scores of the 10 times 5 meter Sprint Test (10x5mST).

Results
ICCs for reliability varied between 0.997 and 1.000. WAL-K scores were significantly higher (i.e., worse) in CP children compared to TD children ($p<0.001$), and in children in GMFCS level II compared to GMFCS level I ($p=.001$). Significant positive correlations were found between the WAL-K and 10x5mST (single run $r=.89$, double run $r=.84$).

Conclusions
The WAL-K shows to be a promising reliable, valid, and easy-to-use tool for assessing walking adaptability in children with CP. Responsiveness to change has yet to be evaluated.

Introduction

Cerebral Palsy (CP) is a permanent neurological disorder that impacts the development of motor function and posture, and is caused by non-progressive pre-, peri-, or post-natal damage of the developing brain[1]. The estimated incidence of CP ranges between 1.5 and 3.0 per 1000 live births[2-5]. The disorder leads to physical problems that occur lifelong and already start in early childhood, such as impaired muscle tone, strength and coordination, sensory problems, and musculoskeletal problems such as scoliosis, contractures, and dislocation of the hip[6-8]. This results in difficulties performing daily activities at school, in sports, and in play[9], which vary greatly with severity of the disorder[1]. The mobility level of children with CP can be described using the Gross Motor Function Classification System (GMFCS) which ranges from level I to V[10]. Children in GMFCS levels I and II (mild CP, around 60% of the CP population[11]) are able to walk independently and often follow regular education and sports activities, but their speed, balance and coordination during walking are limited[10]. They often trip and fall[12] and cannot keep up with peers, which leads to reduced participation in sports and school activities[9,13] and decreased quality of life[14].

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In literature, walking adaptability is defined as the ability to adapt the walking pattern to the demands of the environment[15], such as avoiding obstacles in the schoolyard, or reacting to running children or rolling balls in gym classes. This is an important skill for children for participation in daily activities[16-18]. When walking on a slope or uneven ground and when crossing obstacles, children with mild CP were found to adapt their walking speed, spatiotemporal parameters, joint angles, and toe clearance to a lesser extent than typically developing (TD) children do[19]. These findings of impaired walking adaptability, as obtained using lab-based 3D motion capture[19], point at a relevant domain of motor functioning that is not commonly assessed in clinical practice. This is mainly due to the lack of a validated clinical test to asses walking adaptability in children with CP. A few tests exist that assess performance of more advanced walking tasks in children with mild CP[20], such as the Gross Motor Function Measure-Challenge Module (e.g. sideways crossover on line; run to line, pick up object, run back; running through pylons)[21], and the Test of Gross Motor Development-Second Edition (e.g. run; slide from one point to another)[22]. Yet, none of these tests assess the capacity to accurately adapt foot placement with concurrent demands regarding walking speed, which speed-accuracy trade-off is essential for walking adaptability in daily life. Interestingly, previous studies that tested the speed-accuracy trade-off of arm movements in people with CP demonstrated proportionally larger increments in movement times with greater accuracy demands as compared to typically developing peers[23,24]. This observation suggests that assessing CP-related impairments in walking adaptability may be facilitated by imposing accuracy demands regarding foot placement.

The recently developed Walking Adaptability Ladder test for Kids (WAL-K) is an easy-to-use and inexpensive test which measures children's maximum capacity to adapt both accuracy and speed during walking[25]. In the WAL-K, children have to continually adapt step length and cadence to incrementally larger or smaller targets[25]. The WAL-K has excellent intra- and inter-rater reliability (ICC 0.98-0.99) and good test-retest reliability (ICC 0.76-0.78) in TD children. Its construct validity was shown by moderate correlations with age and Movement Assessment Battery for Children (MABC-2) scores, and its discriminative ability between TD children and children with Developmental Coordination Disorder (DCD)[25]. Yet, before the WAL-K may be used to measure walking adaptability in ambulant children with CP, its psychometric properties have to be determined in this target population.

The first aim of this study was to determine the intra- and inter-rater reliability of the WAL-K in children with mild CP aged 6 to 12 years. For reliable clinical use of the WAL-K, we defined minimum ICC values of 0.75[26]. The second aim was to determine the construct validity of the WAL-K in children with mild CP, which would be supported by significant differences in WAL-K scores between children with mild CP and a cohort of TD children from a previous study of our group[25], and between children in GMFCS levels I and II. In addition, we hypothesized (at least) moderate correlations (r = -0.40 – -0.59) between WAL-K-scores and an existing running agility test (10 times 5 meter sprint test, 10x5mST)[20].

Materials and methods

Participants

Between September 2019 and January 2022, children with CP were recruited from a special school (Maartenschool) and outpatient rehabilitation center (Sint Maartenskliniek) in Nijmegen, The Netherlands. The physiotherapist handed out the information letter to the parents, and the parents contacted the researcher if they wanted their child to participate in the study.

Inclusion criteria for the children with CP were an age between 6 and 12 years old and a diagnosis of CP (either spastic, ataxic, or dyskinetic) classified with GMFCS level I or II from a medical practitioner. Exclusion criteria were Botulinum Toxin injections during the past 3 months, surgery of the lower extremity during the past year, temporary complaints influencing walking ability (e.g. sprained ankle), and visual problems. Children needed to be able to perform the tests according to the instructions.

Parents or legal representatives of all participants gave written informed consent before participating in the study, and children aged 12 years also provided written informed consent themselves. The study was exempt from medical ethical review by the CMO Arnhem/Nijmegen (2017-3465 and 2020-6504), as it was not subject to the Medical Research Involving Human Subjects Act (WMO). All study procedures were conducted in accordance with the Declaration of Helsinki.

Measurements

Before testing, data about age, gender, height, and weight of the children were collected and CP characteristics (GMFCS level, type) were extracted from the electronic patient files by the physiotherapist. All children were tested once in a quiet hallway at their school or in the rehabilitation center by an experienced researcher (MP, RK, MN, LV or JW). All assessors were trained during 1.5 hours by a researcher with extensive experience in administering the tests. The training entailed reading the test protocol, testing each other and receiving feedback from the researcher. The assessors were also supervised by the researcher the first time they tested a child. Children wore their usual clothing, shoes, and orthoses if applicable. The tests were performed in the following fixed order.

Walking Adaptability Ladder test for Kids (WAL-K)

The WAL-K, as described by Kuijpers et al (2022)[25] (see figure 1), uses a 10-meter agility ladder with 19 targets successively decreasing in size. Children were instructed to walk in the ladder, turn around a cone and walk back in the ladder, as accurately and as fast as possible. Children completed both the single run (stepping once in each target) and double run (stepping twice in each target) four times: two practice trials and two recorded trials. When a child did not understand the instructions, extra instruction or encouragement was given during the practice trial. For post-hoc scoring, participants were filmed from the waist downwards using a regular mobile phone while the researcher walked next to the child.

Total time to complete the ladder (in seconds) and total number of mistakes (touching a bar, the wrong number of steps in a target, or missing a target) were scored. For both recorded trials, a total score of completion time plus 0.5 seconds penalty for each mistake was calculated and the best total score of the two attempts was used for analysis.

For determining reliability of the WAL-K, independent raters scored the videos with regards to completion time and number of mistakes. The raters were trained during 1.5 hours by a researcher with extensive experience in scoring the videos. The raters read the scoring protocol, scored eight videos (four single run, four double run) together, and the raters then scored eight different videos (four single run, four double run) themselves. They received feedback on their scores from the researcher and each other to align their way of scoring. For inter-rater reliability, two raters (MP and RK for the first 20 participants and BG and RK for the rest of the participants) both scored the videos of measurement 1 (M1) independently. For intra-rater reliability, MP (for the first 20 participants) and RK (for the rest of the participants) scored the videos of the measurement twice with at least one week in between and without having insight in the previous scoring (rating 1 [R1] and rating 2 [R2]). For 9 of the 36 participants (25%), the assessor who walked next to the child during completion of the WAL-K was the same individual as the rater.

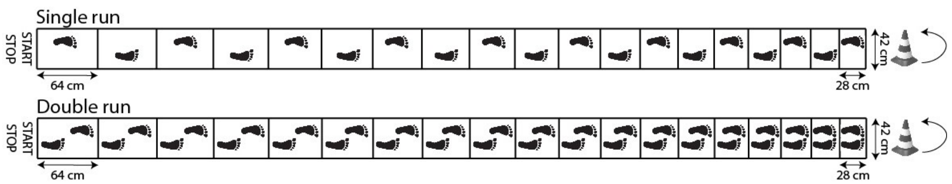

Figure 1. Schematic set-up of the Walking Adaptability Ladder test for Kids (WAL-K). Targets decrease in size from 64 to 28 cm and have a width of 42 cm. Children were instructed to walk in the ladder, turn around a cone and walk back in the ladder, as accurately and as fast as possible. Children completed both the single run (stepping once in each target) and double run (stepping twice in each target) four times: two practice trials and two recorded trials.

10 times 5 meter Sprint Test (10x5mST)

The 10x5mST is a valid and reliable high-level gross motor assessment tool to test speed of walking and turning in ambulant school-aged children with CP (GMFCS level I and II)[20,27]. It can discriminate between children in GMFCS level I and II[27].

Children were instructed to run as fast as possible between two taped lines on the floor with a distance of 5 meters in between, which had to be covered 10 times. They had to place at least one foot on or over the line, otherwise the trial was aborted and the participant performed an additional trial. Two correct trials were performed with 2 minutes of rest in between. During the test, children were verbally encouraged to run as fast as they could. The fastest trial was used for analysis.

Statistical analysis

Reliability

We determined Intraclass Correlation Coefficient (ICC) estimates (model 2,1) and their 95% confidence intervals[28] to evaluate intra- and inter-rater reliability[29]. The following cut off values were used: ICC under 0.5 poor reliability; between 0.5 and 0.75 moderate; between 0.75 and 0.9 good, above 0.9 excellent[26]. Paired samples t-tests and Bland Altman analyses with 95% limits of agreement (LoA) were performed to investigate absolute differences between the two ratings/raters.

Construct validity

To test the hypotheses that children with CP show poorer performance compared to TD children and that performance improves with motor development (i.e., age), we compared the WAL-K scores (total score, completion time and mistakes of the first rating of RK) of the children with CP to reference data collected from 122 TD children (52 boys, 70 girls; mean age 9 ± 2[25]), using an ANCOVA with age as covariate. To test the hypothesis that children in GMFCS level I perform better than those in GMFCS level II, a similar analysis was used within the group of children with CP to compare the scores between children in GMFCS levels I and II. In addition, Pearson correlation coefficients were determined between the WAL-K scores and the 10x5mST scores. The following cut off values were used: 0.00-0.19 very weak; 0.20-0.39 weak; 0.40-0.59 moderate; 0.60-0.79 strong; 0.80-1.00 very strong[30].

All analyses were performed in SPSS version 25 and alpha was set at 0.05.

Results

Of the 38 included children with CP, data of two children were excluded from analysis. One 8-year-old boy could not concentrate well enough to execute the task properly, and one 11-year-old girl could not follow the instructions to perform the task as fast as possible. Hence, data of 36 children were included in the analysis. The characteristics of the participants are presented in table 1.

Table 1. Descriptive statistics in mean (SD) or number (%) for the total group of children with Cerebral Palsy, and children in Gross Motor Function Classification System (GMFCS) level I and II

	Total group (n = 36)	**GMFCS I (n = 26, 72%)**	**GMFCS II (n = 10, 28%)**
Age (years)	8.9 (1.8)	9.0 (1.7)	8.8 (2.3)
Height (cm)	139.7 (12.6)	142.1 (11.8)	133.6 (12.2)
Weight (kg)	34.8 (11.6)	36.6 (12.0)	31.3 (9.9)
Gender	23 boys (64%)	16 boys (62%)	7 boys (70%)
	13 girls (36%)	10 girls (38%)	3 girls (30%)
Type	29 unilateral (81%)	24 unilateral (92%)	5 unilateral (50%)
	7 bilateral (19%)	2 bilateral (8%)	5 bilateral (50%)
Orthosis use	5 participants (14%)	2 participants (8%)	3 participants (30%)
10x5mST (s)	30.2 (6.1)	28.6 (5.2)	33.9 (6.9)

10x5mST = 10 times 5 meter Sprint Test

Reliability

The ICCs for intra- and inter-rater reliabilities were .999 (95% CI .998-1.000) and .997 (95% CI .995-.999), respectively, for the single run, and 1.000 (95% CI .999-1.000) and .997 (95% CI .994-.998) for the double run (see table 2). Paired samples t-tests revealed no significant systematic differences between the two ratings/raters for the single or double run (p = .13-.89). Bland Altman plots are presented in figure 2, showing LoA of -1.14–1.20 in single run WAL-K score for intra-rater reliability and -1.78–2.05 for inter-rater reliability. LoA in double run WAL-K score were -1.20–1.17 for intra-rater reliability and -2.66–3.47 for inter-rater reliability. The ranges of LoA are relatively small compared to the scale of the WAL-K values, indicating a good agreement between the ratings/raters in a majority of participants.

Table 2. Intra- and inter-rater reliability of the Walking Adaptability Ladder test for Kids (WAL-K)

	Intra-rater reliability				Inter-rater reliability			
	Mean R1 (SD)	Mean R2 (SD)	ICC (95% CI)	Paired samples t-test	Mean R1 (SD)	Mean R2 (SD)	ICC (95% CI)	Paired samples t-test
Single run (s)	25.9 (13.3)	25.8 (13.4)	.999* (.998-1.000)	t = .28; p = .78	25.9 (13.3)	25.7 (13.4)	.997* (.995-.999)	t = .84; p = .41
Double run (s)	48.7 (19.9)	48.8 (19.8)	1.000* (.999-.1.000)	t = -.14; p = .89	48.7 (19.9)	48.3 (19.6)	.997* (.994-.998)	t = 1.57; p = .13

* p < .001; R1 = rating 1; R2 = rating 2; ICC = Intraclass Correlation Coefficient; CI = 95% Confidence Interval; SDC (%) = smallest detectable change at 95% confidence level and % of the mean.

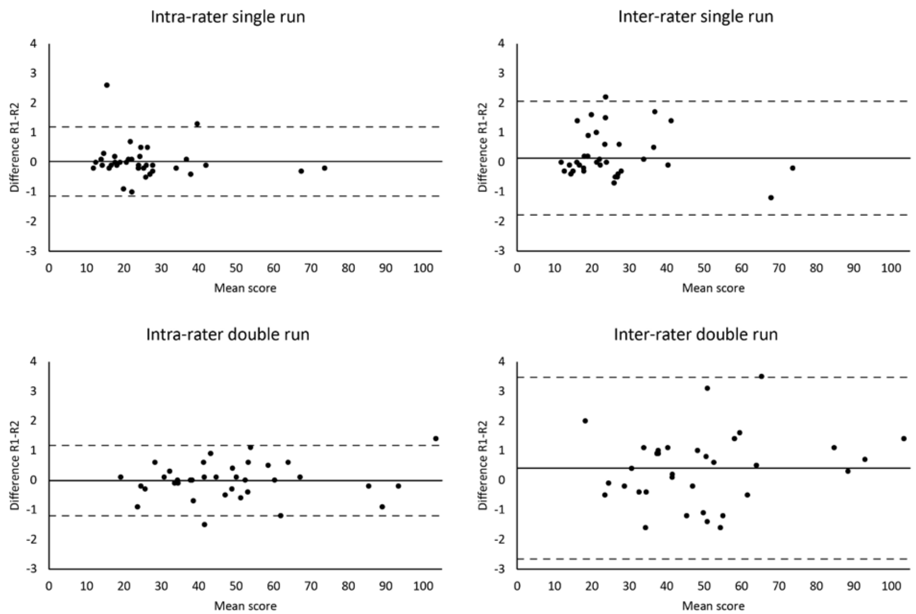

Figure 2. Bland-Altman plots for intra- and inter-rater reliability concerning the single and double run of the Walking Adaptability Ladder test for Kids (WAL-K) in children with Cerebral Palsy. The horizontal axis represents the mean of the two ratings/raters and the vertical axis represents the difference between them. The solid line shows the mean difference (systematic error, which is close to zero) and the dashed lines show the 95% limits of agreement (LoA; random error).

Construct validity

Children with CP showed a significantly poorer performance (i.e. higher WAL-K score) than our previously measured cohort of TD children25 (see table 3), on the single (F(1, 155) = 82.00, p < .001) as well as the double run (F(1, 155) = 101.17, p < .001). For the single run, we found a significant interaction between group and age (F(1, 155) = 5.56, p = .020); it was not significant for the double run (F(1, 155) = 2.54, p = .113). Post hoc analyses yielded significant differences between children with CP and TD children in both completion time (single run, F(1,155) = 86.20, p < .001; double run, F(1,155) = 118.36, p < .001) and number of mistakes (single run, F(1,155) = 25.21, p < .001; double run, F(1,155) = 4.11, p = .044). Completion time contributed strongly to the total WAL-K score (≥84.2%, see table 3) in children with CP as well as TD children. The single run showed a small but significant shift towards a smaller relative contribution of completion time in the children with CP compared to TD children (91.9 vs 95.1%, p=.029), whereas the double run yielded non-significant between-group differences in relative contributions of completion time and mistakes. For the single run, WAL-K scores of four children with CP (11%) were between 1 and 2 SD above the mean of the TD children (i.e. worse than TD children), and WAL-K scores of 22 children with CP (61%) exceeded 2 SD above the mean. For the double run, WAL-K scores of three children with CP (8%) were between 1 and 2 SD above the mean of the TD children, and WAL-K scores of 21 children with CP (58%) exceeded 2 SD above the mean (see figure 3).

As in the children with CP no significant correlation was observed between age and WAL-K score for the single (r = .08, p = .631) or double run (r = -.07, p = .706), we did not include age as a covariate in the GMFCS level I versus level II analysis. We here used an independent t-test. Children in GMFCS level II scored significantly higher (i.e., worse) than children in GMFCS level I, on both the single (t(10.23) = -2.528, p = .029) and double run (t(34) = -3.622, p < .001) (see figure 3). Significant differences existed between children in GMFCS level II and I in completion time (single run, (F(1, 32) = 9.32, p = .005); double run, (F(1, 32) = 7.21, p = .011) and number of mistakes (single run, (F(1, 32) = 14.71, p = .001); double run, (F(1, 32) = 16.05, p < .001).

A significant and very strong positive correlation was found between WAL-K score and 10x5mST score for both the single run (r = .89, p < .001), and the double run (r = .84, p = .003).

Table 3. Walking Adaptability Ladder test for Kids (WAL-K) scores divided in completion time and mistakes for children with Cerebral Palsy (CP) (R1) and typically developing (TD) children[25]

	CP, mean (SD)		TD, mean (SD)	
	Single run	Double run	Single run	Double run
Absolute values				
Total score (s)	25.9 (13.3)*	48.7 (19.9)*	14.2 (3.1)	29.1 (6.2)
Completion time (s)	23.3 (10.9)*	42.2 (16.9)*	13.4 (2.6)	24.2 (4.8)
Mistakes (number)	5.1 (6.2)*	13.0 (10.7)*	1.6 (2.8)	9.9 (7.9)
Relative values				
Completion time (%)	91.9 (7.1)*	87.4 (8.1)	95.1 (8.0)	84.2 (10.8)
Mistakes (%)	8.1 (7.1)*	12.6 (8.1)	4.9 (7.9)	15.8 (10.7)

Relative values indicate the contribution of time and mistakes to the total WAL-K score. * indicates a significant difference (p<.05) between TD and CP.

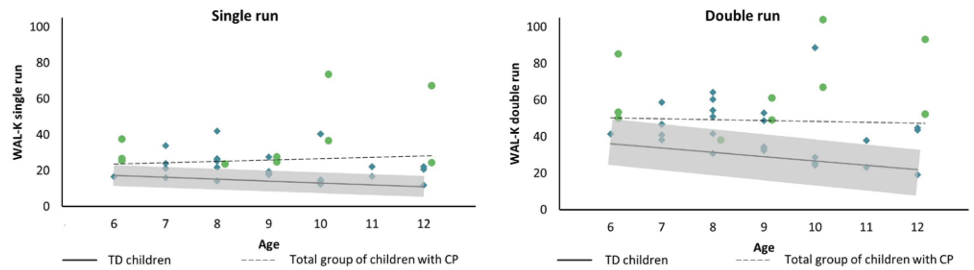

Figure 3. Relationship of Walking Adaptability Ladder test for Kids (WAL-K) scores with age. Individual data of children with Cerebral Palsy (CP) are shown for the single run and double run (lower score is a better performance): blue diamonds for Gross Motor Function Classification System (GMFCS) level I, green dots for GMFCS level II. Black solid line indicates the regression line of typically developing (TD) children (n=122), with the grey area showing ±2SD[25]. Black striped line indicates the regression line of children with CP (n=36).

Discussion

The aim of the current study was to determine the intra- and inter-rater reliability and construct validity of the WAL-K in children with mild CP aged 6 to 12 years. Excellent intra- and inter-rater reliabilities were found. A good construct validity of the WAL-K was indicated by the significantly poorer WAL-K scores in children with CP compared to TD children, in children in GMFCS level II compared to children in GMFCS level I, and by the very strong correlations between WAL-K and 10x5mST scores.

The intra- and inter-rater reliabilities were excellent (ICC > 0.99) and are comparable to the previously reported ICCs (0.98-0.99) for the WAL-K in TD children[25]. This excellent reliability may have been facilitated by the offline (i.e. post-hoc) scoring using the video recordings. We deliberately chose to perform offline scoring based on pilot measurements in TD children revealing that it was difficult to score mistakes online, i.e., during execution of the task, while also attending to the child. We expect that online scoring may be even more difficult in children with CP given their larger number of mistakes and greater attendance needed. We expect that offline scoring is well feasible in clinical practice, as a regular mobile device can be used for video recording and scoring takes little additional time (approximately 5-10 minutes). Our results demonstrated that WAL-K scores of the children with CP were nearly two-fold higher (i.e. poorer) than those previously obtained in a cohort of TD children[25]. The discriminative ability of the WAL-K is further supported by the significantly poorer WAL-K scores in children with CP in GMFCS level II compared to children in GMFCS level I. These findings are in agreement with observations from lab-based walking adaptability studies[19] that demonstrated that children with CP have severe difficulties adjusting their gait pattern to environmental demands and constraints. The present results add to those of our previous study in children with DCD[25] and support the application of the WAL-K as a new clinical test for distinguishing between children with and without motor disorders.

Whereas one may have expected the greater difficulty of the double run to further enhance the discriminative ability compared to the single run, we observed roughly similar proportions of age-adjusted individual scores that exceeded mean+2SD of the TD cohort (61 vs 58% for single and double run, respectively). In addition, the relative contribution of completion time to the total WAL-K score (i.e., speed-accuracy trade-off) was similar for TD children and children with CP, albeit with a marginal shift (3%) towards a proportionally larger number of mistakes in the children with CP for the single run. Previous studies on goal-directed arm movements also found significant differences between TD children and children with CP, but in these studies, proportionally larger differences were found in movement times between TD children and children with CP when accuracy demands increased[23,24]. The finding that group differences were not magnified in the double run may suggest that accuracy demands did not greatly differ between the two conditions.

The relationship between WAL-K scores and age which was found in an earlier study in TD children[25] appeared to be absent in children with CP, as illustrated by the weak and non-significant correlations. In children with CP, the heterogeneity in physical abilities probably bears a stronger relationship with WAL-K score than age. This can be illustrated by the WAL-K scores of two children of 10 and 12 years old in GMFCS level II which deviated considerably from the rest of the children with CP (see figure 3). These two children (both with a spastic motor type and bilaterally affected) were more severely affected than the other children with GMFCS level II, which reflects the large variability in mobility function that is most prominent within GMFCS level II at school (50m) and community distances (500m)[31]. Yet, due to the presently included small subgroup of children in GMFCS level II (n=10), no firm conclusions can be drawn about the relationship with age. This may be further examined in a larger group of children with a more balanced distribution among ages and GMFCS levels.

The correlation between the WAL-K and 10x5mST scores were higher than we hypothesized based on the expected accuracy demands of the WAL-K compared to the 10x5mST. Probably, the degree of overall motor impairment, as indicated by GMFCS level, is an important determinant of performance on both the WAL-K (testing walking adaptability) and 10x5mST (testing agility). Indeed, similar to the present findings, highly significant differences in 10x5mST scores have previously been demonstrated between children in GMFCS levels I and II[27]. The relationship with overall motor impairment may therefore explain the high correlation between the two tests.

The current study comes with some limitations. Most importantly, we did not study the test-retest reliability of the WAL-K in children with CP, and therefore it is not yet possible to interpret changes in WAL-K scores after treatment[32]. In our previous study in TD children, we observed a small but significant systematic difference between test and re-test, probably due to a learning effect on the task. Based on these observations, we decided to add an extra practice trial to the test protocol. It remains to be tested whether this indeed reduces potential learning effects on re-test WAL-K scores. In addition, the smallest detectable change needs to be determined in children with CP. In TD children, SDCs were 3.5s for the single run and 6.0s for the double run[25], but these may differ in children with CP. To support the clinical usability of the WAL-K, the responsiveness to change, including test-retest reliability, smallest detectable change, and minimum clinically important difference in children with CP needs further study. Second, the sample size of the study did not meet the advised number of 50 participants for reliability and validity studies[32]. Nevertheless, we believe that the present results strongly support the reliability and validity of the WAL-K in children with CP, in line with our previous findings in children with DCD and TD children[25]. However, the sample comprised mainly unilaterally affected children, so the results are possibly less representative for bilaterally affected children. Dussault-Picard et al. (2022) stated that the absence of stratification on type of CP "may overlook relevant information such as the extent of adaptations required to cope with daily locomotion according to severity of functional impairments"[19]. Therefore, future research should stratify by type of CP to study whether this influences performance on the WAL-K, preferably including at least include 50 children and with even distribution of GMFCS levels I and II.

Lastly, two children were not able to perform the test following the instructions, possibly caused by problems with executive functioning such as attention, planning, and working memory (prevalence of more than 50% in children with CP)[33]. Especially during the first assessments of the walking adaptability with the WAL-K (similar to a first stage of motor learning) there is a high load on executive functioning[34]. With future use of the WAL-K for research or clinical practice, researchers and therapists should be aware that the level of executive functioning may impact the child's ability to properly understand the instructions, and accordingly judge the validity of the obtained score.

In conclusion, the WAL-K seems to be a promising reliable and valid measurement tool for measuring walking adaptability in children with CP and is an addition to existing walking tests. It is the first measurement tool that specifically assesses walking adaptability in children with CP which is an essential skill in daily life activities, and gives additional information for clinical reasoning. The WAL-K can be used in clinical practice and research to identify children with CP who have problems with walking adaptability.

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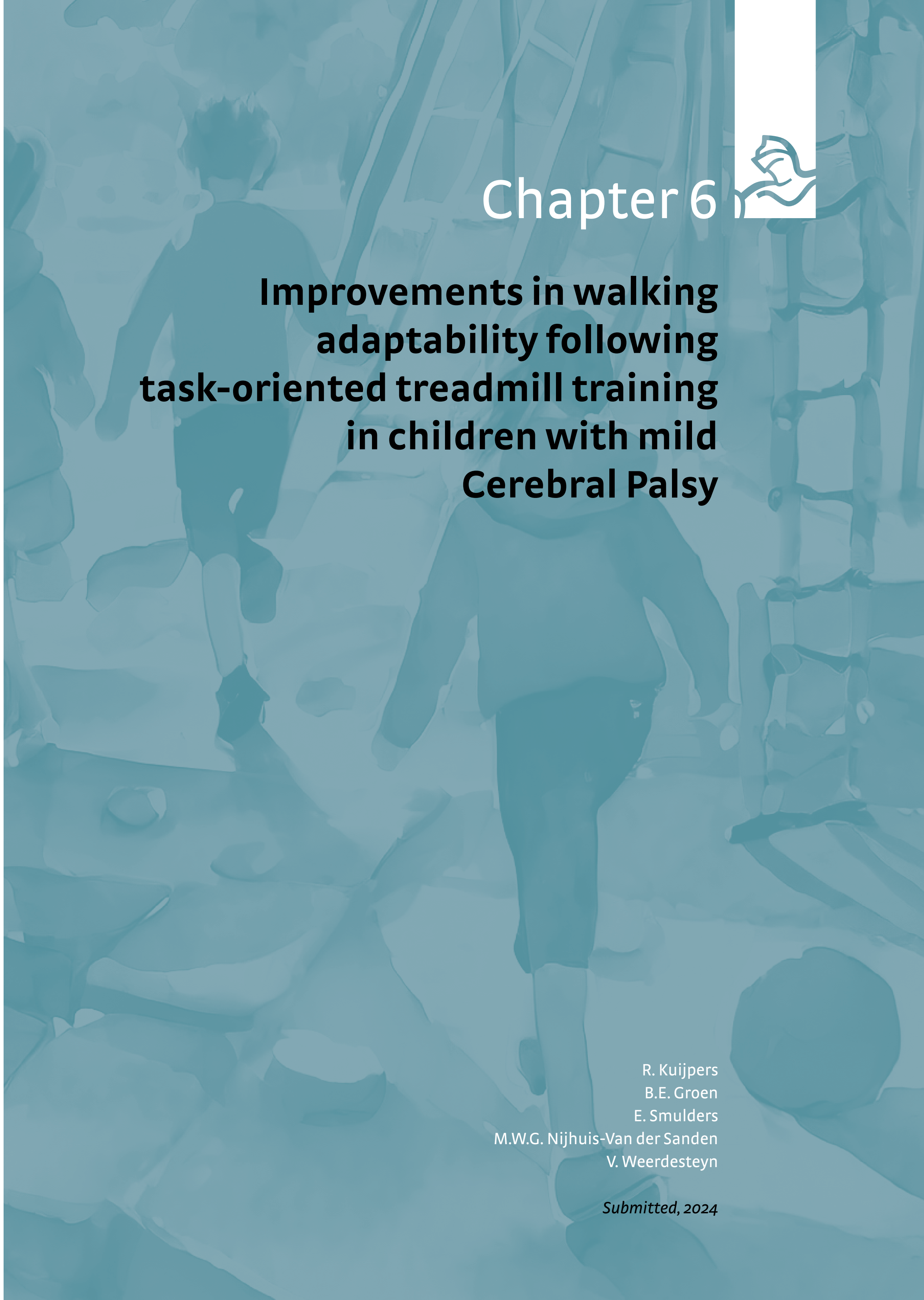

Chapter 6

Improvements in walking adaptability following task-oriented treadmill training in children with mild Cerebral Palsy

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Abstract

Purpose
Children with Cerebral Palsy (CP) can have difficulties with walking adaptability (WA), which is important for participation in daily life. The purpose was to examine the effect of task-oriented treadmill training on WA, walking speed, muscle strength, quality of life, and perceived athletic competence in children with CP. The second aim was to define possible factors influencing the training effect.

Materials and methods
Fourteen children with mild CP received a ten-sessions training on a treadmill, the C-mill. Gait adjustments were evoked by visual context projected on the treadmill belt. The effects of training were evaluated using the Walking Adaptability Ladder test for Kids (WAL-K; single and double run) as primary outcome measure, and the 10 Meter Walking Test, Functional Strength Measurement, KIDSCREEN, and Self-Perception Profile for Children as secondary outcome measures. Measurements were administered before (M1), directly after training (M2) and after 3 months follow-up (M3). The parents of the participating children completed a questionnaire on their perception of the training. The differences in scores between M1-M2 and M1-M3 were analyzed using Linear Mixed Models.

Results
Children significantly improved on the WAL-K double run between M1-M2 and M1-M3. They improved on treadmill-based WA-tasks between M1-M2, while scores remained stable between M2-M3. A poorer baseline WAL-K score and younger age were associated with greater improvement following training. Walking speed, muscle strength, quality of life, and perceived athletic competence did not change. Parents found the training useful and fun for their child and indicated that their child fell less frequently.

Conclusions
Task-oriented treadmill training had positive effects on WA in children with mild CP, which were retained after 3 months follow-up.

Introduction
Cerebral palsy (CP) is a neurological disorder which affects movement and posture, but may also impact other domains such as cognitive function. It is caused by non-progressive damage in the developing brain before, during, or shortly after birth[1]. The estimated incidence of CP is 1.5 to 3.0 per 1000 live births[1]. Children with CP have impaired muscle strength, tone and coordination; sensory problems[2]; and they may experience associated musculoskeletal problems (e.g., scoliosis, contractures, dislocation of the hip[3]. The impairments of children with CP differ by severity[1]. Children classified with Gross Motor Function Classification System (GMFCS) levels I and II (mild CP) can walk independently[4], but their coordination, balance and walking speed are limited[5]. They are often able to participate in regular education, sports and play[4], but compared to typically developing children they experience more problems in these daily activities[6]. Children with mild CP trip and fall frequently[7] and they show lower participation in physical activities compared to their peers[8]. This may lead to a decreased health-related quality of life[9] and less positively perceived athletic competence[10].

Children with CP commonly receive gait training, overground or on a treadmill, to improve their mobility. These types of training seem to be effective to improve walking speed, endurance, gross motor function, spatiotemporal variables, and functional mobility[11]. Treadmill training tends to show stronger effects in improving walking speed and endurance than overground training, possibly due to the increased stepping repetition and intensity of treadmill training[11]. Besides, virtual reality can magnify the effects of gait training and increase patient engagement[11]. Thus far, the focus of gait training interventions has mainly been on steady-state walking. Yet, participating in regular physical education and sports with other children also requires the ability to adapt to the environmental context[12], for example when avoiding playmates or objects during playtime or physical education classes. This so-called walking adaptability is defined as the ability to make adaptations to the walking pattern to meet task goals and environmental demands[13]. Children with CP can have an impaired dynamic stability during walking adaptability tasks such as walking on uneven ground or when crossing obstacles[14]. The impaired stability is expressed in making more use of their arms[5] and having an increased toe clearance[5,12,15,16], step width[15,16], and trunk and pelvis movements[16] compared to TD children. However, to our knowledge, there is a general lack of research on walking adaptability training and related outcome measures in children with CP.

Therefore, the primary aim of this study was to examine the effect of task-oriented treadmill training with augmented reality on walking adaptability, walking speed, functional muscle strength, health-related quality of life, and perceived athletic competence in children with mild CP (GMFCS-level I and II). We hypothesized that training-induced gains in walking adaptability on the treadmill would generalize to an overground task, as such generalization is a prerequisite for potential benefits in daily life walking conditions. The second aim was to define possible factors –related to the severity of symptoms and the motor control deficit– influencing the effect of the training.

Materials and methods

Participants
Children and adolescents with CP –hereafter mentioned 'children'– of 6-17 years old were invited via a special school (Maartenschool, Nijmegen, the Netherlands), patient database of rehabilitation center Sint Maartenskliniek (Nijmegen, the Netherlands), patients' association CP Nederland, local physical therapy practices, sports clubs for children with disabilities, a private Facebook group of parents of children with CP, and CP-net, a national network of health professionals working with children with CP.

Participants needed to have a formal diagnosis of CP, classified with GMFCS-level I or II, and a request for help regarding walking difficulties. Exclusion criteria were Botulinum Toxin injections in the lower extremities in the past 3 months, surgery to the lower extremities in the past two years, epilepsy, severe vision or cognitive impairments, and temporary complaints influencing walking (e.g., sprained ankle). Parents and participants above 12 years old gave written informed consent prior to participation.

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Design and protocol amendments
Originally, this study was set up as a randomized controlled trial (ICTRP: NL8154) with a waiting list control group design. The original protocol involved randomization of participants into the intervention group (intervention started immediately) or to the waiting list control group (intervention started after 5 weeks). Measurements took place before the start of the intervention (M1), after the intervention (M2) and three months after the end of the intervention (M3). The waiting list control group had an extra measurement at the beginning of the waiting period (M0). The starting date was September 1st 2019. All measurements and training sessions were performed at the rehabilitation center Sint Maartenskliniek in Nijmegen, the Netherlands, until December 2021. The study was approved by the regional Medical Ethical Committee Arnhem-Nijmegen (2018-4223).

The COVID-19 pandemic, however, forced us to deviate from our original study protocol. Shortly after the first children had been included, the pandemic hit, which severely impacted recruitment, not only during the lockdowns (i.e., 6 months lab closure in 2020), but also in the aftermath. The pandemic put a considerable strain on the life of families with children with disabilities due to closed schools and rehabilitation centers, which was not limited to the period of the lockdowns but also had its effect long after the lockdowns. Parents were reserved to let their child participate because he/she fell behind at school and/or in rehabilitation programs. Due to the difficult and slow inclusion of participants we implemented the following changes to our study, including a change in the design:

- We prolonged the study duration by 15 months until March 2023.
- To avoid losing any eligible children, we were compelled to abandon the original RCT design. When parents decided on participation of their child, many of them were not willing to wait for 5 weeks to start with the training due to planning issues at home. Therefore, we decided to place children in the waiting list control group not based on randomization, but based on availability and willingness of their parents. As we did not expect many parents opting for the waiting list control group, we also decided to evaluate the intervention using a pre-post analysis.

- We accepted a variable range of 3-6 weeks for scheduling the training sessions upon request of the parents instead of the previously-defined period of 5 weeks.

As a result, in the remainder of the paper we analyze and report pre- versus post-training results collapsed across the intervention and waiting-list control groups.

Intervention: C-mill training
Participants received ten 45-minute sessions of task-oriented C-mill training (total training time 450 min) during 3-6 weeks (tuned to parents' preferences) which is comparable to other studies which found improvements of steady-state walking (e.g., walking speed) in children with CP after interventions of 420-540 min[17-19].

The C-mill is a treadmill with embedded force plates. Visual context (targets/obstacles) can be projected on the treadmill belt to provoke walking adaptations. The training protocol was designed based on the protocol that we previously used in children with Developmental Coordination Disorder[20] and on the protocol of Timmermans et al. (2016) who performed C-mill training in stroke patients[21]. Training sessions were performed at the participant's comfortable treadmill walking speed, as measured at M0 or M1, and re-evaluated in training sessions 3 and 6. Participants were instructed not to use the handrails. Three physical therapists experienced in using the C-mill guided the training sessions; a participant was always trained by the same therapist.

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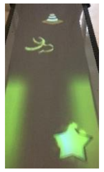
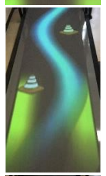
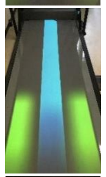
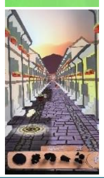
The type and duration of the exercises were standardized with exercise blocks focusing on target stepping, obstacle avoidance, walking in a slalom or tandem track, and accelerating and decelerating (see table 1). All participants of 6-9 years old and participants of 10-13 years old with GMFCS-level II started the exercises at level 1; participants of 10-13 years old with GMFCS-level I and participants of 14-17 years old started the exercises at level 2 (see table 1). For the rest of the training, the level of difficulty was adjusted to the individual performance level of the participant, based on the following predefined cut-off values21. When a participant had a success rate of ≥90% (80% for the obstacles), the level was increased in the next session; when a participant had a success rate of 70-90% (60-80% for the obstacles), the level was kept the same in the next session; when a participant had a success rate of <70% (60% for the obstacles), the level was decreased in the next session. The therapist could always overrule the decision to in- or decrease the level in case the therapist felt that the level based on the cut-off values did not match to the performance level of the child[21]. If this was the case, the therapist made a note on the case report form.

Results for each task were extracted from the C-mill software (see table 1). Performance of each task was scored on a 10-point scale using the following formula, adapted from Timmermans et al. (2019): level*factor*performance(%)/100, in which the factor was determined by 10 divided by the number of possible levels for the specific task21. For example, a child who had a success rate of 85% on the stepping stone task in level 4, had a total score of 4.25 (4*(10/8)*85/100). A higher score represented a better performance. In addition, the comfortable walking speed in sessions 1, 3 and 6 was also reported.

Table 1. Training protocol

	Exercise		Duration (min)	# of levels	Variation between levels	Success rate
1	Determination of comfortable walking speed (only in sessions 3 and 6).		2			n/a
2	Warming up at comfortable speed.		3			n/a
3	Target stepping with random obstacles: continuous projection of targets of which a certain % suddenly changed into an obstacle.		4	8	Increasing irregularity in step length/ width/ symmetry, increasing number of obstacles, decreasing available response time for obstacles	Mean of % correct hits of targets and % correct avoidances of obstacles
4	Fun and functional game (forest, beach or stars): projection of targets (footballs or stars) and obstacles (e.g., rabbit, starfish, banana) in which children received points for successful target hits, and lost points when hitting obstacles. Visual and auditory feedback on performance was given.		4	3	Increasing number of targets and obstacles	Mean of % correctly hit targets and % correctly avoided obstacles
5	Obstacle avoidance: projection of unilateral or bilateral obstacles in front of the left or right foot. Visual and auditory feedback on performance was given.		4	6	Decreasing available response time, increasing size	% correctly avoided obstacles
6	Slalom walking: projection of a slalom walking path with cones in the corners. Visual feedback on performance was given.*		2.5	5	Increasing the frequency of corners	% correctly placed steps (on the path)
7	Tandem walking: projection of a narrow walking path. Visual feedback on performance was given.*		2.5	5	Decreasing the width of the path	% correctly placed steps (on the path)
8	Accelerating/decelerating: projection of a green area which moves to the front of the belt and back. Visual feedback on performance was given.		2.5	5	Increasing the speed in which the green area moves	% correctly placed steps (in the green area)
9	Fun and functional game with front screen (pizza, bin or football): no projection on the belt, but a game shown at the screen in front of the treadmill. Children had to collect ingredients for a pizza, collect waste with a bin or bounce back a football.		4	5	Decreasing the time between the ingredients/ waste/ bounces	n/a

Between all exercises, children had a 2-minute break. The order of exercises was different in every session. *The slalom and tandem walking exercises were alternated: the slalom was performed in the odd sessions; the tandem was performed in the even sessions.

Measurements

At baseline, descriptive data were collected from the child regarding age, gender, and current sport and therapy activities of the child. Height and weight were measured; GMFCS level and type of CP were extracted from the electronic patient file. Below we outline the measurements that were conducted; see figure 1 for an overview at which time points (M0, M1, M2 and M3) these measurements were performed. Measurements were conducted by one experienced researcher in the following order: Walking Adaptability Ladder Test for Kids, 10 Meter Walk Test, Functional Strength Measurement, and walking adaptability tasks on the GRAIL. It took 2-2.5 hours to conduct the tests. The KIDSCREEN and Self-Perception Profile for Children were completed online before the start of the measurement.

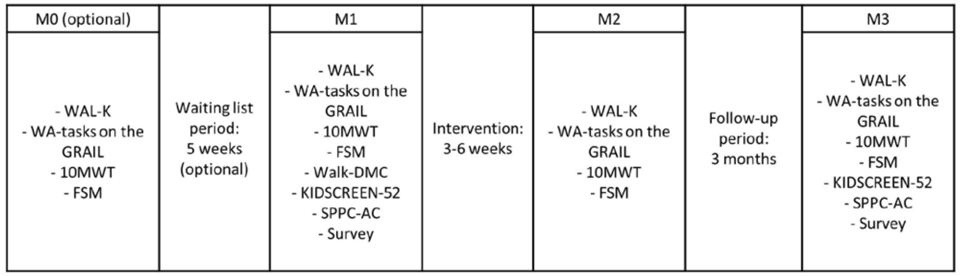


Figure 1. Overview on measurements performed at M0, M1, M2 and M3.

Primary outcome – Walking Adaptability Ladder Test for Kids (WAL-K)

The WAL-K consists of a 10m agility ladder, lying flat on the ground, with 19 targets decreasing in size[22]. The participant was instructed to walk back and forth in the ladder while stepping into the targets as accurately and fast as possible. Participants performed the WAL-K in two conditions in the following fixed order: the single run (stepping with one foot in each target) and the double run (stepping with both feet in each target). The researcher first demonstrated the task once, then the participant performed two practice trials and finally two trials were recorded. WAL-K score (s) was calculated as completion time + 0.5 sec * number of mistakes (touching a bar, the wrong number of steps in a target, or missing a target). The best score of the two recorded trials was used in the analyses. The WAL-K is a reliable and valid tool for measuring walking adaptability in children[22].

Secondary outcomes

Walking adaptability tasks on the GRAIL

Obstacle avoidance and target stepping tasks were performed using the Gait-Real time Analysis Interactive Lab (GRAIL) (Motek Medical, Houten, the Netherlands). The GRAIL comprises an instrumented dual-belt treadmill with two embedded force plates, a ten-camera motion capture system (Vicon, Oxford, UK), and a 180-degree semi-cylindrical screen for the projection of synchronized 3-dimensional environments. Using the Vicon model (version 2.8.1), 18 reflective markers were placed on the lower body of the participant for collecting motion data. An additional marker was placed on the anterior side of the shoe at the distal second tarsal. Marker positions were tracked at 100 Hz to assess foot placement with respect to the projected obstacles and targets. Projections on the treadmill were matched to treadmill speed using D-flow software (Motek Medical).

After (safety) instructions and putting on a safety harness, the participant's comfortable walking speed was determined during the first GRAIL measurement (M0 or M1). Thereafter, participants walked 2 minutes in a neutral virtual reality environment at their comfortable walking speed to get used to walking on the treadmill

Participants performed three walking adaptability tasks on the GRAIL at comfortable walking speed (see figure 2). For the *obstacle avoidance task*, the participant was instructed to avoid unilateral red-white colored obstacles projected on the treadmill with an available response time of ~0.25s, ~0.5s or ~0.7s (equal probability in random order). Obstacles were placed at the anticipated location of the following foot placement. The task started with 20 practice trials, followed by 30 experimental trials which were used for analysis.

For the *target stepping task*, the participant was instructed to step as accurately as possible on projected white targets (shoe size) with an available response time of ~0.7s. Targets were placed at +60%, 0% or -60% of step length or at +25%, 0% or -25% of step width. The task started with 10 practice trials, followed by 30 experimental trials which were used for analysis.

The third task was a *combination task* in which the aforementioned obstacles and targets were combined. The task consisted of 30 experimental trials without practice trials.

A custom-made MATLAB script was used to (post-hoc) determine stepping accuracy, defined as the percentage of the foot surface area that was placed correctly (i.e., outside the obstacles and within the targets). Mean stepping accuracy over all projections in each walking adaptability task was determined for both legs together for each participant. A higher score (%) on these tasks represents a better stepping accuracy.

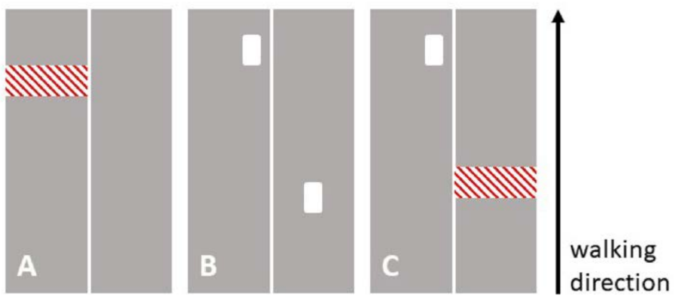


Figure 2. Schematic set-up in top view of the walking adaptability tasks on the GRAIL.

10 Meter Walk Test (10MWT)
The 10MWT was used to determine walking speed. The participant was instructed to walk 10 meters overground between two lines, three times at comfortable walking speed and three times at maximum walking speed (without running). The average completion time (m/s) of the three attempts was used for analysis. The 10MWT is a reliable test for determining walking speed in children[23].

Functional Strength Measurement (FSM)
Functional muscle strength was measured using the 4 lower extremity items of the FSM: standing long jump (cm), lateral step up, sit to stand, and stair climbing. For the lateral step up, for unilaterally affected participants the results of the most affected leg were reported, for bilaterally affected participants the results of the non-preferred leg were reported. Scores were defined as the number of repetitions in 30s, except for the standing long jump for which the distance in cm was the outcome measure. The FSM is a reliable and valid tool to measure functional strength in children with CP[24].

KIDSCREEN-52
Health-related quality of life was assessed using the Dutch version of the KIDSCREEN-52. This is a valid and reliable questionnaire for children[25]. We reported results of the domains physical well-being, psychological well-being, moods and emotions, self-perception, and social acceptance, as the other domains were considered less relevant for the focus of our study. Other studies also used selected domains of the KIDSCREEN, among which is one study of the developer of the KIDSCREEN[26,27].

Answers are given on a 5-point Likert scale. Scores were summed and transferred to percentile scores per domain (ranging from 0-100) using the KIDSCREEN-52 scoring protocol[28]. A higher score represents a better health-related quality of life. Both participants and their parents completed the questionnaire at M1 and M3.

Self-Perception Profile for Children, domain Athletic Competence
Perceived athletic competence was assessed using the Dutch version of the Athletic Competence domain of Harter's Self-Perception Profile for Children (SPPC-AC)[29]. The questionnaire is reliable and valid for assessing perceived athletic competence in children with physical disabilities[30]. It consists of 6 items of athletic skills which are scored on a 4-point Likert scale. Additionally, participants were asked to indicate the importance of each skill on a 4-point Likert scale. For both competence and importance, scores can range from 6-24 with a higher score representing a more positive perceived athletic competence. Participants completed the questionnaire at M1 and M3.

Survey to evaluate parents' views on the training and the period after the training
At the end of M2 and M3, parents completed a survey on their perception of the C-mill training using a 5-point Likert scale. At M2, parents rated how much their child experienced the training as fun, difficult, fatiguing, and useful, how they perceived the quality of the supervision, if the sessions were easy to fit in parents' planning, and how likely they would recommend the training to others. At M3, parents rated their child's activity level, falls, responsiveness to the environment during walking, participation in play and sports activities, and self-perception on his/her motor skills in the 3 months after the training. They were also asked whether they observed changes in their child's daily activities. There was also room for additional comments.

Exploratory outcome: walk-DMC
Muscle coordination patterns were measured at M1 to explore whether the severity of coordination problems was associated with the effect of the training. During two minutes of unperturbed walking on the GRAIL we collected surface EMG-signals from bilateral rectus femoris, vastus lateralis, medial hamstring (semitendinosus), medial gastrocnemius, and

tibialis anterior. EMG electrodes (30x24mm; Kendall, Mansfield, MA) were placed according to the Seniam guidelines[31].

The dynamic motor control index during walking (walk-DMC) is a summary measure of the complexity of muscle activation and is derived from a synergy analysis of the EMG signals[32]. A generalized MATLAB script (R2020b) published by MacWilliams et. al. (2021) was used to compute walk-DMC scores from the surface EMG signals[33]. The EMG signals were bandpass filtered from 10 Hz to 1 KHz by the preamplifier and the data was sampled at 1000 Hz. The EMG signals were further pre-processed and filtered using the MATLAB script[33].

The average walk-DMC score of TD children is 100 with a standard deviation of 10. A higher walk-DMC score corresponds to having better motor control. For children with mild CP (GMFCS levels I and II) the walk-DMC score ranges from 40.2 to 91.3[32,34,35]. The walk-DMC score of the most affected side (or non-preferred side for bilaterally affected participants) was used for statistical analysis.

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Statistical analysis
Descriptive statistics were used to describe the baseline data. To evaluate pre- versus post-training differences, Linear Mixed Model (LMM) analyses were used to compare M1-M2 and M1-M3 scores for the WAL-K, walking adaptability tasks on the GRAIL, 10MWT, and FSM. Dummy variables for M2 and M3 were included in the model as fixed effects. Note that these analyses were conducted collapsed across groups, as explained under Design and protocol amendments. An unstructured covariance type was used because of the different time periods between the measurements. Restricted maximum likelihood was used to estimate the parameters. To assess differences between M1-M3 for the KIDSCREEN-52 and SPPC-AC, a paired samples t-test was used.

To assess potential relationships between the effect of the training and each of the pre-defined factors of interest (age, GMFCS level, type of CP, waiting list control or intervention group, baseline WAL-K score, and walk-DMC score), we determined Pearson correlation coefficients between these factors and the delta scores of M1-M2 and M1-M3 on the WAL-K double run.

As exploratory outcome measure, comfortable walking speed during the training sessions was compared using paired samples t-tests (session 1 with session 3 and session 1 with session 6). For these analyses, alpha was set at 0.025 to correct for multiple testing. Progress during the training sessions was visualized in graphs with lines for each participant per task.

All analyses were performed using IBM SPSS Statistics v.25 (IBM Corp., Armonk, NY), and alpha was set at 0.05. Results of the surveys were presented descriptively.

Results

Of the 74 invited children, 16 children did not meet the inclusion criteria and 39 children (67%) ultimately chose not to participate (see figure 3), mainly because parents found the burden too high (n=18) or experienced planning issues (n=12). Of the remaining 19 children (13 boys, 6 girls, mean age 9.0, SD 2.4), three children withdrew between the first measurement and the start of the intervention, as due to COVID-19 restrictions, they had to wait a very long time before study activities could be resumed. One child had an unexpected surgery after the first measurement and one child broke a leg during the period of training, which was unrelated to the intervention. Eventually, 14 children completed the intervention. Participants completed on average 8.6 training sessions. Group characteristics of the participants are shown in table 2.

Of the eight children in the waiting list control group (3 were randomized, 5 were placed voluntarily), four children completed the study. Of the eleven children who were allocated to the intervention group, ten children followed the intervention (see figure 3). For two participants, the follow-up period was six instead of three months, because of COVID-19-related lab closure.

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Table 2. Group characteristics, mean (SD)

Gender	8 boys (57%), 6 girls (43%)
Age (years)	9.2 (2.2)
Height (cm)	144 (14)
Weight (kg)	38.5 (15.0)
GMFCS level	12 level I (86%), 2 level II (14%)
Type	12 unilateral (86%), 2 bilateral (14%)
Sports (min/week)	98 (76)
Walk-DMC most affected leg	74.9 (16.4)

SD = standard deviation; Walk-DMC = dynamic motor control index during walking.

Measurements
Primary outcome – WAL-K
Double run WAL-K scores significantly decreased (i.e., improved) between M1-M2 (mean difference = -4.9s, 95% CI = -7.7 – -2.1) and M1-M3 (mean difference = -6.9s, 95% CI = -11.0 – -2.8; see table 3 and 4 and figure 4). Between M1-M2, 5 children (36%) improved more than the smallest detectable change of 6.0s on the double run, as previously reported for a cohort of typically developing children22. Between M1-M3, 4 children (29%) improved more than the smallest detectable change on the double run.

Between M1-M2, completion time significantly decreased (mean difference = -3.7s, 95% CI = -6.1 – -1.2) and a decrease albeit not significant was observed in number of mistakes (mean difference = -2.5, 95% CI = -5.1 – 0.1) on the double run. Between M1-M3, completion time (mean difference = -5.0s, 95% CI = -8.0 – -2.1) and number of mistakes (mean difference = -3.4, 95% CI = -5.6 – -1.1) on the double run significantly decreased. No significant differences for WAL-K single run scores were found between M1-M2 and M1-M3. For completeness, the results of the WAL-K including the M0 scores of the waiting-list control group are shown in appendix 1.

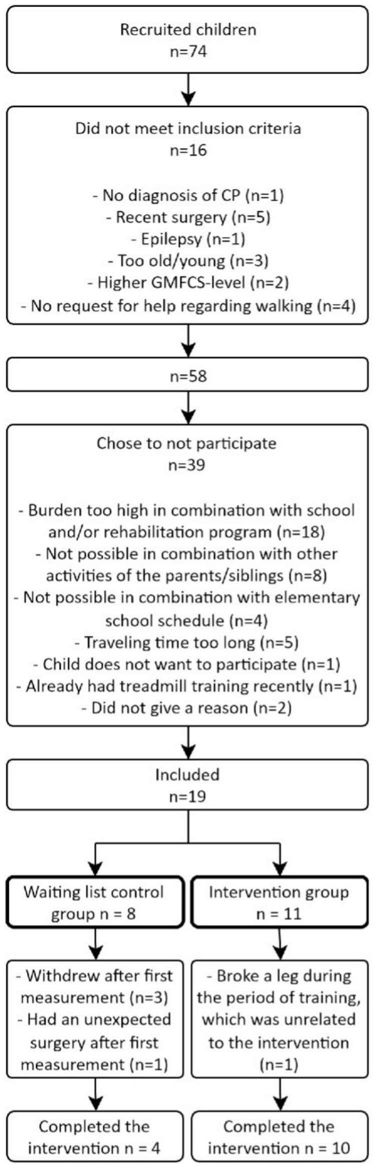

Figure 3. Flow chart of recruitment and inclusion.

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Table 3. Descriptive statistics

	M1	M2	M3
WAL-K, mean (SD)			
Single run total score (s)	19.9 (6.4)	18.4 (4.4)	18.4 (5.7)
Single run completion time (s)	18.5 (5.2)	17.7 (4.1)	18.0 (5.5)
Single run mistakes (n)	2.7 (3.2)	1.5 (2.3)	0.9 (1.5)
Double run total score (s)	38.1 (12.6)	33.2 (9.1)	31.2 (10.2)
Double run completion time (s)	35.2 (11.4)	31.6 (8.5)	29.9 (9.4)
Double run mistakes (n)	5.8 (5.5)	3.3 (2.5)	2.6 (3.0)
*WA tasks on the GRAIL, mean (SD)**			
Obstacle task success rate (%)	89.3 (12.4)	93.8 (7.2)	93.9 (7.2)
Target task success rate (%)	59.2 (16.7)	65.6 (10.6)	62.3 (14.2)
Combination task success rate (%)	73.1 (14.3)	78.8 (9.2)	79.8 (6.1)
10MWT, mean (SD)			
Comfortable (s)	7.8 (0.8)	7.5 (1.0)	7.9 (1.0)
Maximal (s)	5.1 (0.6)	4.9 (0.6)	4.9 (0.5)
KIDSCREEN-52 percentile score, mean (SD)			
Child			
Physical well-being	75.4 (26.2)		57.6 (34.5)
Psychological well-being	62.8 (26.3)		62.9 (28.3)
Moods & emotions	58.9 (31.0)		53.9 (27.9)
Self-perception	64.3 (22.8)		68.0 (22.8)
Social acceptance	59.2 (37.4)		46.4 (35.6)
Parent			
Physical well-being	55.2 (32.0)		58.3 (38.8)
Psychological well-being	62.3 (31.3)		61.0 (34.9)
Moods & emotions	39.9 (31.0)		44.2 (29.7)
Self-perception	59.0 (21.9)		66.5 (31.4)
Social acceptance	44.7 (28.8)		58.6 (37.9)
SPPC-AC, mean (SD)			
Boys			
Competence	16.9 (2.8)		16.4 (2.7)
Importance	17.3 (2.5)		18.6 (2.5)
Girls			
Competence	17.6 (2.5)		16.0 (1.6)
Importance	16.8 (3.0)		15.8 (2.9)

SD = standard deviation; WAL-K = Walking Adaptability Ladder Test for Kids; WA = walking adaptability; 10MWT = 10 Meter Walk Test; SPPC-AC = Self-Perception Profile for Children, domain Athletic Competence.

Table 4. Results Linear Mixed Model

	Comparison M1-M2		Comparison M1-M3	
	F (df)	p	F (df)	p
WAL-K				
Single run total score	4.30 (1,13.0)	.058	3.21 (1,13.1)	.096
Completion time	2.57 (1,13.0)	.133	.77 (1,12.0)	.397
Mistakes	2.39 (1,13.0)	.146	6.27 (1,13.3)	.026
Double run total score	14.48 (1,13.0)	.002	16.79 (1,10.5)	.002
Completion time	10.07 (1,13.0)	.007	14.73 (1,13.0)	.002
Mistakes	4.42 (1,13.0)	.056	10.53 (1,13.1)	.006
*WA tasks on the GRAIL success rate**				
Obstacles (%)	4.92 (1,13.2)	.045	2.98 (1,7.3)	.126
Targets (%)	4.63 (1,12.4)	.052	.95 (1,6.0)	.368
Combination task (%)	3.4 (1,11.9)	.090	3.78 (1,4.7)	.114
10MWT				
Comfortable	2.31 (1,13.0)	.153	.044 (1,12.8)	.837
Maximal	4.34 (1,13.0)	.058	2.74 (1,12.7)	.122
FSM#				
Standing long jump	.15 (1,13.0)	.708	1.55 (1,12.7)	.236
Lateral step up most affected leg	1.91 (1,13.0)	.190	4.26 (1,13.3)	.059
Sit to stand	.42 (1,13.0)	.528	4.53 (1,10.6)	.058
Stair climbing	.31 (1,10.8)	.587	3.03 (1,10.4)	.111

WAL-K = Walking Adaptability Ladder test for Kids; WA = walking adaptability; 10MWT = 10 Meter Walk Test; FSM = Functional Strength Measurement.
* Data were missing for six measurements (3 times M2, 3 times M3) of 6 different children because of technical issues with the GRAIL. # Data of the FSM stair climbing of four measurements (divided over 3 participants) were missing.

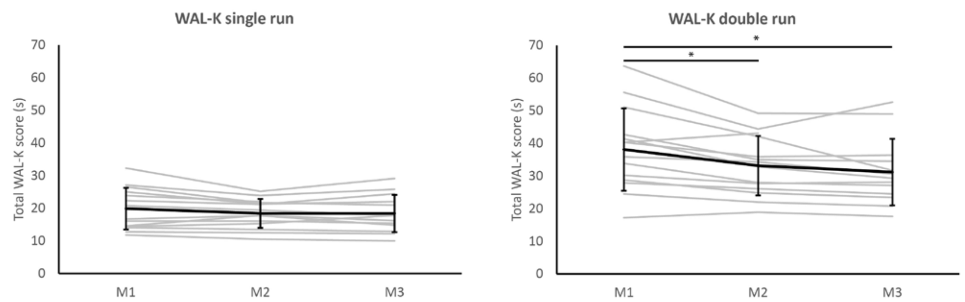

Figure 4. Individual scores (grey) with mean scores and SDs (black) of the WAL-K scores. *p <.01.

Secondary outcomes

Stepping accuracy on the GRAIL obstacle avoiding task significantly increased (i.e., improved) between M1-M2 (mean difference = 2.9%, 95% CI = 0.3 – 6.1, see table 3 and 4). We observed higher stepping accuracy on the target task between M1-M2 (mean difference = 6.4%, 95% CI = 0.5 – 13.4, p = .052) as well as on the combination task (mean difference = 5.5%, 95% CI = 2.9 – 13.9, p = .090), although differences were not significant. No significant differences between M1-M3 were found.

For the 10MWT and FSM no significant differences were found between M1-M2 and M1-M3 (see table 3 and 4). In addition, no significant differences between M1-M3 were found for the KIDSCREEN-52 and SPPC-AC.

Thirteen parents completed the survey regarding their views on the training at M2; twelve parents completed the survey regarding their views on the period after the training at M3. Results of the surveys are displayed in table 5. At M2, parents were generally positive about the fun their child experienced, the usefulness of the training, the supervision, and the possibility to fit the training in their planning at home. The training was not perceived as difficult, and the results concerning whether the training was fatiguing were mixed. Most parents (85%) would recommend the training to others. Additionally, parents commented positively on the possibility to practice different situations such as obstacle avoidance compared to regular gait training. Parents liked the fun games that allowed children to earn points, but advised to develop more games to choose from.

At M3, most parents (67%) indicated that their child fell less frequently, whereas 25% of the parents did not notice any difference. Half of the parents (50%) indicated that their child better responded to the environment during walking and had a more positive self-perception on his/her motor skills, while the rest of the parents did not notice any difference. Half of the parents (50%) was neutral about whether their child participated more in play and sports, while 25% agreed and 25% disagreed.

Baseline WAL-K score (r = .943, p < .001) and age (r = -.570, p = .042) significantly correlated with the WAL-K double run delta scores between M1-M2, while walk-DMC score did not (r = -.05, p = .876). A poorer baseline WAL-K score and younger age were associated with greater improvement following training. The numbers of participants with GMFCS level II (n=2) and those who were bilaterally affected (n=2) were considered too low for formal analysis.

During the training sessions, comfortable walking speed on the C-mill significantly increased between session 1 and 3 (t=-6.34, p<.001, mean difference = 0.18 m/s 95% CI = 0.24 – 0.12) and between session 1 and 6 (t=-9.42, p<.001, mean difference = 0.29 m/s, 95% CI = 0.36 – 0.23) (see appendix 2). Difficulty levels of the C-mill tasks increased in the first 5 to 6 training sessions and remained relatively stable thereafter (see appendix 2).

Table 5. Survey to evaluate parents' views on the training and the period after the training

		Totally disagree	Disagree	Neutral	Agree	Totally agree
M2 (n = 13)	My child experienced the training as…					
	… fun	1			5	7
	… difficult	3	9	1		
	… fatiguing	3	2	2	4	2
	… useful			2	7	4
	Supervision during the training was appropriate				2	11
	It was possible to fit the training in our planning at home		1	2	8	2
	I would recommend the training to others		1	1	4	7
M3 (n = 12)	In the 3 months after the training, my child…					
	… was more active			11	1	
	… fell less frequently		1	3	5	3
	… better responded to the environment during walking		1	5	6	
	… participated more in play and sports activities		3	6	3	
	… had a more positive self-perception on his/her motor skills		2	4	6	

Discussion

The primary aim of this study was to examine the effect of task-oriented treadmill training with augmented reality on walking adaptability in children with mild CP, as measured using the WAL-K. We found significant improvements on the WAL-K (double run) when we compared pre-intervention scores with those post training. These effects were retained after 3 months follow-up. A similar pattern of results was observed for the obstacle avoidance task on the GRAIL, albeit only significant between pre and post intervention (M1-M2). No significant differences were observed regarding walking speed, muscle strength, quality of life, and perceived athletic competence. Poorer baseline scores on the double run of the WAL-K and younger age were found to correlate with greater post-intervention gains on WAL-K scores.

In this first study on walking adaptability training in children with CP, an important finding was that improvements could be observed in a test condition (overground WAL-K) that was distinctly different from the walking adaptability tasks included in the training program (i.e., treadmill based). This apparent generalization of training effects from the C-mill training environment to an overground task is particularly relevant, as children with CP are known to have difficulty with transfer of a learned skill from virtual to daily-life situations[36]. The transfer to daily life situations also seems to be supported by a majority of parents reporting fewer falls. These findings are promising given the relevance of walking adaptability for participation in daily life – in particular for children with mild CP who often participate in physical activities (e.g., sports and play) with able-bodied peers.

While the walking adaptability tasks on the GRAIL (i.e., secondary outcomes) showed a pattern of results that was similar to that of the WAL-K, significant differences could only be demonstrated for the obstacle avoidance task between M1-M2. This was not in line with our expectations, as we had anticipated stronger effects on the treadmill-based WA assessments because of their greater resemblance to the tasks included in the training program. It may be that the WA tasks on the GRAIL suffered from ceiling effects, thus limiting the room for improvement following training. Indeed, a mean obstacle avoidance baseline score of 89.3% may hint at such a ceiling effect. As this task included obstacles of different levels of difficulty to allow each child a certain degree of success for keeping motivation[37], we also looked into the scores of the more difficult trials (available response times of ~0.25s), because the accuracy rates decrease with increasing difficulty[38-40]. Yet, this post-hoc analysis did not yield significant differences between M1-M2 and M1-M3 either. Furthermore, the baseline scores of the target stepping task and the combined obstacle-target task were substantially lower, thus leaving more room for improvement, yet no significant differences were observed in these outcomes.

Alternatively, the (fixed) treadmill speed at which the WA tasks were performed may have impacted the sensitivity to change. At baseline, we determined the individual's comfortable walking speed on the GRAIL and used this fixed speed throughout the treadmill-based WA assessments. Of note, this walking speed was on average 0.5 m/s lower compared to the baseline overground comfortable walking speed, as measured using the 10MWT. This observation is in line with previous research indicating that typically developing children also had a lower comfortable walking speed in a single session of treadmill walking compared to overground walking (mean difference: 0.21 m/s)[41]. As the CP children progressed in the training program, their treadmill-based comfortable walking speed significantly increased from 0.8 m/s in the first training session to 1.1 m/s in the last session. The latter speed is rather similar to comfortable walking speed on the 10MWT at M2 (1.0 m/s), which shows that children gradually learned to walk at their comfortable overground walking speed on a treadmill. A higher walking speed seems to have a beneficial influence on interlimb coordination and coordinative stability in children with CP[42], the low walking speed used during the WA assessments on the GRAIL may have obscured potentially relevant training-induced differences.

In contrast to the significant increase in treadmill-based walking speed from training session 1 to 6 (+0.29 m/s, appendix 2), no significant differences in comfortable overground walking speed were observed, as measured with the 10MWT. This indicates the context-specific nature of the observed gains in treadmill-based walking speed. As comfortable walking speed typically decreases in the presence of visual constraints (e.g., stepping targets[43]), we here argue that the similar treadmill-based speed that could be maintained in the final training sessions (i.e., while making gait adaptations) compared to the regular comfortable overground walking speed also testifies to improvements in walking adaptability in our group of children with CP. Children showed progress across training sessions in walking speed as well as level of difficulty, indicating an improvement in walking adaptability.

Greater post-intervention gains on WAL-K double run scores were found to correlate with younger age and poorer baseline scores on the WAL-K double run but not with walk-DMC scores. WAL-K scores are associated with age in children with CP[44], so the effect of age is probably based on collinearity with baseline score. Our results are largely in line with the results of Shuman et al. (2018) who found that children with greater impairments in walking pre-treatment had the best outcomes after orthopedic surgery, botox injection or physical therapy[45]. They also found, in contrast with our results, that children with lower walk-DMC scores (more impaired motor control) had worse treatment outcomes compared to children with higher walk-DMC scores (less impaired motor control)[45]. More research is needed on the relationship between walk-DMC and functional gait outcomes pre- and post-treatment.

The lack of a control group is a major limitation of the current study. We started our study just prior to the onset of the COVID-19 pandemic and this had a profound and lasting impact on the rate of participant inclusion. Even after the lockdown-related restrictions had been lifted, many parents indicated to be too overburdened to consider participation of their child. The majority of parents who decided not to participate with their child, indicated that this was due to the high burden of the study (long-lasting intervention and extensive measurements), and planning issues at home because of parents' work, the child's school schedule, and sports activities of the child and siblings. These planning issues were further aggravated in the aftermath of the COVID-19 pandemic. Due to the low rate of inclusion in the trial and the reluctance expressed by parents of children allocated to the waiting-list control group, we made the decision to depart from the original controlled study design. We chose to conduct a pre versus post training comparison as a second best option. Yet, this comes with the limitation that we cannot exclude a possible systematic difference due to repeated WAL-K test administration influencing the observed pre versus post training differences. Conclusions regarding training-induced gains are therefore preliminary and require confirmation in a controlled trial. The small sample size and overrepresentation of unilaterally affected children and children in GMFCS level I might limit the generalizability of the findings.

Based on our experience conducting this study, there are some considerations that deserve mention for enhancing the feasibility of future studies. Ideally, the study is designed in co-creation with the target population, taking into account their preferences and constraints regarding amount, duration and location of training sessions and measurements. As the C-mill is available in a limited number of physiotherapy practices, the burden of travelling to these locations may be reduced by including a smaller number of C-mill training sessions alternated with overground training sessions with a local physiotherapist. This combination might also improve the transfer of learned skills[46]. Another consideration regarding the measurements is that administering the WAL-K is much easier and less time-consuming than measurements on the GRAIL, while it also appears to be more sensitive to change. Using only the WAL-K for evaluating effects of training could reduce the burden of our time-consuming measurement protocol. Yet, the test-retest reliability and smallest detectable change of the WAL-K in children with CP should be further investigated to determine whether an improvement after training can be attributed to the effect of the training.

In conclusion, task-oriented treadmill training with augmented reality seems to be promising for improving walking adaptability in children with CP and the training was well-received by the children and their parents. A poorer baseline WAL-K score and younger age were associated with greater improvement following training. Walking adaptability is a highly relevant skill in daily life activities, so more attention should be paid to this skill in future research. The current study gives directions for possible measurement tools and their effect sizes to evaluate walking adaptability training in children with CP.

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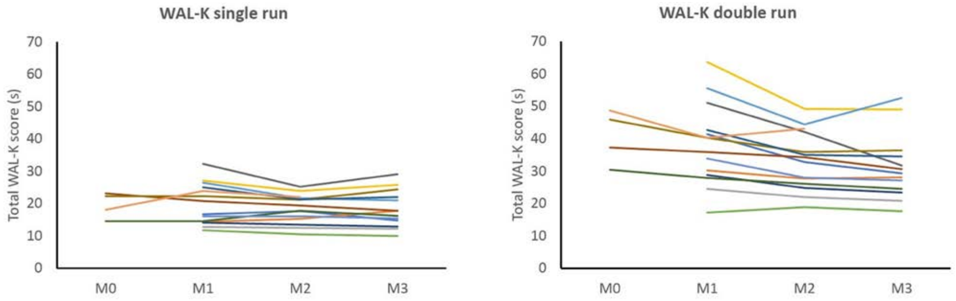
6

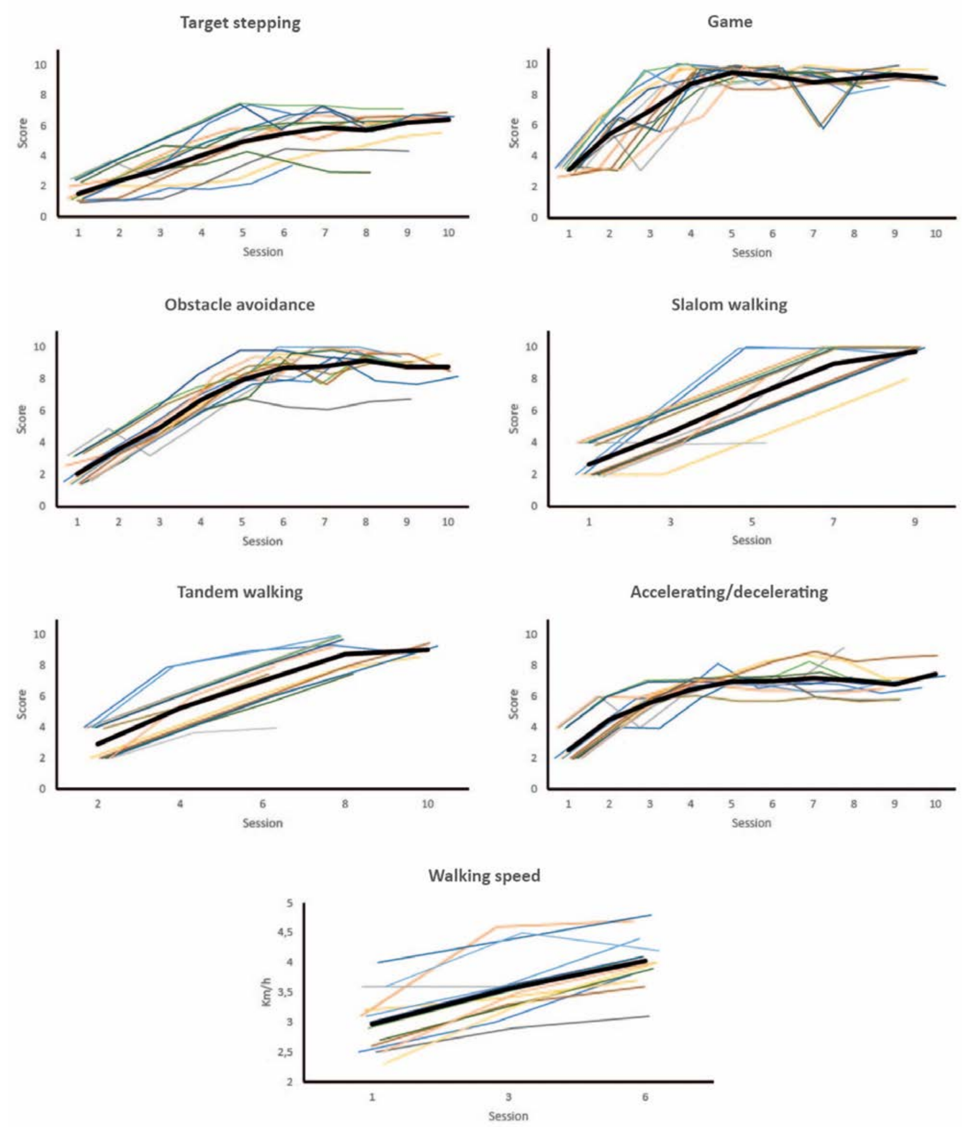
Supplemental material

Appendix I. Graphs of single and double run WAL-K scores at M0, M1, M2 and M3

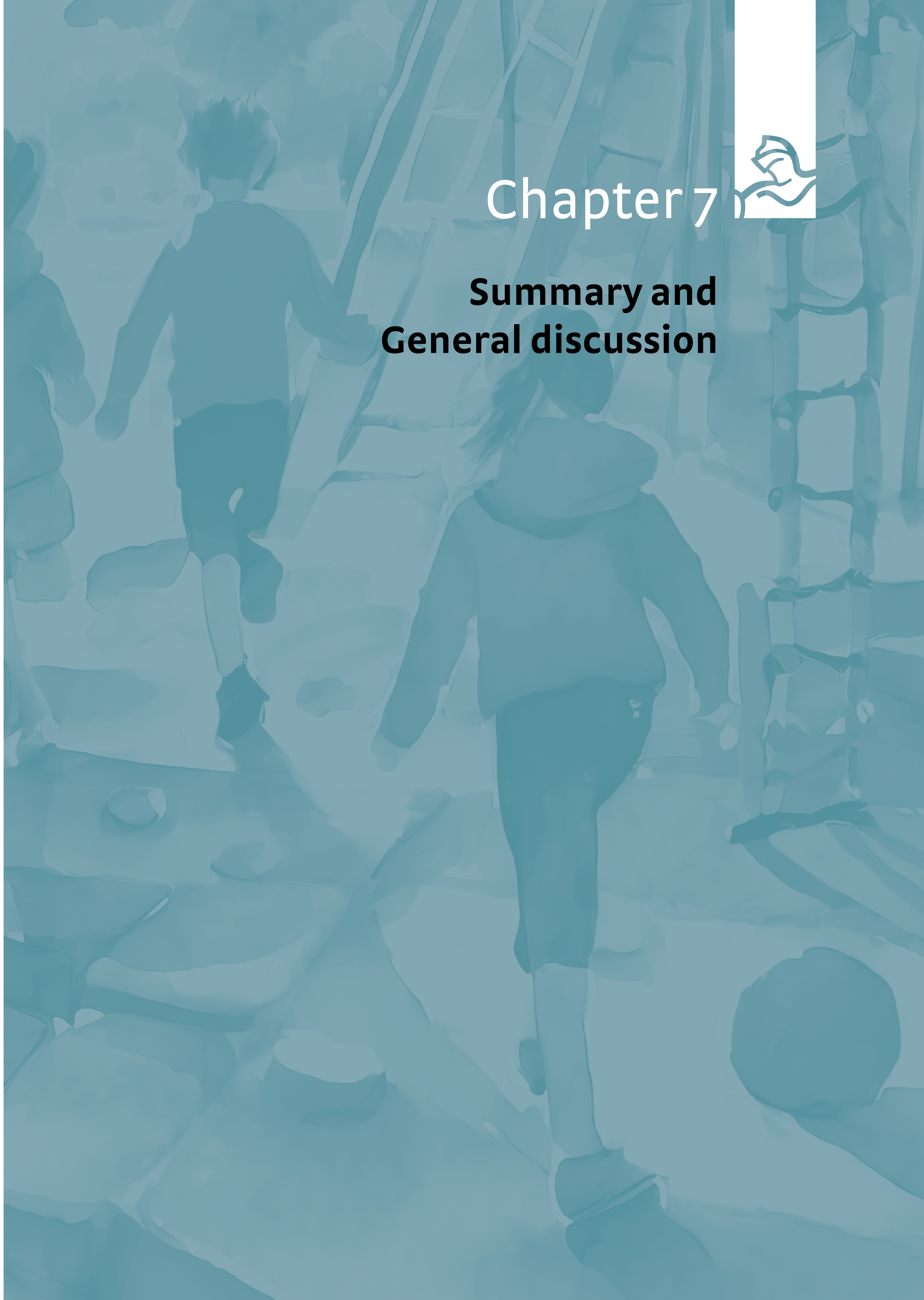


M0 = beginning of the waiting period; M1 = pre-intervention; M2 = post-intervention; M3 = after 3 months follow-up. Each line represents a participant.

Appendix II. Graphs of the six tasks and walking speed during the training sessions



Performance of each task was scored on a 10-point scale using the following formula, adapted from Timmermans et al. (2019): level*factor*performance(%)/100, in which the factor was determined by 10 divided by the number of possible levels for the specific task[21].



Chapter 7

Summary and General discussion

The final chapter of this thesis starts with a summary of the main findings, followed by a discussion on assessment of walking adaptability using the WAL-K, training walking adaptability on a treadmill, clinical and educational implications, methodological considerations, and directions for future research.

Summary

Children with mild motor disorders, such as DCD and mild CP, often show difficulties with walking adaptability, i.e., the ability to modify the walking pattern to reach intended goals while handling the demands of the environment (e.g., to avoid stumbling over a ball or colliding with playmates). Walking adaptability is essential for children to participate in daily activities, such as sports, play and school activities. However, no objective measurement tools for walking adaptability currently exist, and interventions only focus at regular walking or balance, rather than walking adaptability. Therefore, the overarching aim of this thesis was to develop and evaluate clinical assessment and training of walking adaptability in children with mild motor disorders. The ultimate goal was to enable identification of children who have problems with walking adaptability, and to improve their walking adaptability by training, to allow them to better participate in daily activities together with other children.

Our group developed the Walking Adaptability Ladder test for Kids (WAL-K) as a new measurement tool for walking adaptability. **Chapter 2** of this thesis describes the psychometric properties of the WAL-K in 6-12 year old children. In total, 122 typically developing (TD) children and 26 children with Developmental Coordination Disorder (DCD) completed the single and double run conditions of the WAL-K. Intra-rater, inter-rater and test-retest reliability were determined by intraclass correlation coefficients (ICCs) and Smallest Detectable Change (SDC) in 53 TD children. Construct validity was determined by comparing WAL-K scores between 69 TD and all DCD children and correlating WAL-K scores with age and with scores on the Movement Assessment Battery for Children, version 2. The ICCs for reliability varied between 0.76 and 0.99. Compared to the first test performance, WAL-K scores were significantly lower (i.e., better) at retest. SDCs for test-retest reliability varied between 20.8 and 26.1% of the mean scores. WAL-K scores were significantly higher (i.e., worse) in DCD children compared to TD children ($p < 0.001$). Significant negative correlations were found with the Movement Assessment Battery for Children (-0.52 and -0.60) and age (-0.61 and -0.68). The WAL-K showed to be a valid, reliable and easy-to-use tool for measuring walking adaptability in children. Adding an extra practice trial may reduce the observed learning effect.

In daily life, children encounter many situations in which they have to perform dual tasks while walking, such as sports and play. Therefore, in **Chapter 3** I test the hypothesis that 6-12 year old children with DCD would show lower levels of walking adaptability than TD children, and that due to problems with automatization this difference would increase when they are forced to divide their attention between tasks when a concurrent visuo-motor or cognitive task is added. Twenty-six children with DCD and sixty-nine TD children were included in this cross-sectional study. They performed a challenging walking adaptability task on a treadmill as a single, a visuo-motor dual and a cognitive dual task at a pace of 3.5 km/h. Repeated measures ANCOVAs were performed with condition (single/dual task) as within-subjects factor, group (TD/DCD) as between-subjects factor, and age as covariate. Children with DCD performed significantly poorer on the walking adaptability task than TD children. The group differences significantly increased when a concurrent visuo-motor task was added, but not when adding a concurrent cognitive task. A significant effect of age was found with younger children performing worse on all tasks. These results highlighted the problems children with DCD have with walking adaptability and dual tasks, which capacities are essential for full participation in sports and play activities.

As children with DCD were found to have problems with walking adaptability and dual tasks, **Chapter 4** aims to describe whether augmented-reality treadmill training leads to improvements in walking adaptability in 6-12 year old children with DCD. Seventeen children with DCD were included in this proof-of-concept intervention study. They received a six-session training on the C-mill, a treadmill on which gait adjustments can be evoked by projected visual context. The effect of the training was evaluated before (M1), directly after training (M2), and after 6 months follow-up (M3) using the WAL-K (single and double run) and walking adaptability tasks on the C-mill (as a single and with concurrent visuo-motor and cognitive task). In addition, parents completed a questionnaire on their perception of the training. Linear Mixed Model analyses were performed to assess the differences in WAL-K scores and success rates on the walking adaptability tasks between M1-M2 and M1-M3. Children significantly improved on the WAL-K double run and on all three walking adaptability tasks between M1-M2 and M1-M3. Children did not improve on the WAL-K single run. Parents found the training useful and fun for their child and indicated that their child fell less frequently. The results showed that C-mill training had positive and task-specific effects on walking adaptability in children with DCD, which effects generalized to an overground task and were retained at 6 months follow-up. This may help children with DCD to better participate in daily activities.

Children with Cerebral Palsy (CP) experience similar problems with walking adaptability as children with DCD, and I reasoned that implementing the WAL-K as a clinical test would be of value in this group of children as well. Therefore, in **Chapter 5** the psychometric properties of the WAL-K are determined in 6-12 year old children with mild CP (GMFCS levels I and II). In total, 36 children with CP completed the single and double run conditions of the WAL-K. Intra-rater and inter-rater reliability were determined by ICCs. Construct validity was determined by correlating the WAL-K scores with scores of a sprint test (10x5mST), comparing WAL-K scores between 122 TD and all CP children, and comparing the WAL-K scores between children with CP in GMFCS levels I and II. ICCs for reliability varied between 0.997 and 1.000. Significant positive correlations were found with the 10x5mST ($r=.89$ and $.84$). WAL-K scores were significantly higher (i.e., worse) in CP children compared to TD children ($p<0.001$), and in children with GMFCS level II compared to GMFCS level I ($p=.001$). The WAL-K showed to be a reliable, valid, and easy-to-use tool for measuring walking adaptability in children with mild CP.

Given the problems that children with mild CP have with walking adaptability, **Chapter 6** aims to examine the effect of augmented reality treadmill training on walking adaptability in 6-17 year old children with mild CP. Fourteen children with CP (GMFCS levels I and II) received a ten-session training on a treadmill on which gait adjustments can be evoked by projected visual context. The effect of the training was evaluated before (M1), directly after training (M2), and after 3 months follow-up (M3) using the WAL-K (single and double run) as primary outcome measure. Parents completed a questionnaire on their perception of the training. Linear Mixed Model analyses were performed to assess the differences in WAL-K scores between M1-M2 and M1-M3. Children significantly improved on the WAL-K double run between M1-M2 and M1-M3, but not on the WAL-K single run. Parents found the training useful and fun for their child and indicated that their child fell less frequently. Treadmill training had positive effects on walking adaptability in children with CP, which were retained after 3 months follow-up. This may help children with CP to better participate in daily activities.

Max was asked by his rehabilitation physician whether he was interested to participate in a training for a research project with the aim to improve his walking adaptability. His rehabilitation physician expected that the treadmill training which was given in this study could be beneficial for Max to improve his ability to adapt his movements to the environment or task and to give him more confidence in complex walking skills. His parents talked about the study with Max, and together they decided to participate to try to further improve his walking skills.

Max followed nine training sessions. He really enjoyed training on the treadmill, especially the games with which he could earn points. He noticed that he improved on the exercises during the training which gave him confidence in his skills on the treadmill but also during play in the schoolyard and physical education classes. After he finished the training, Max and his parents noticed that he fell less and he could better adapt his walking to the task or environment, for example when walking on an irregular pavement or walking on a line during physical education. Sometimes, Max still has difficulty with walking or running in a crowded environment or when he has to perform multiple tasks simultaneously, especially when he is tired. Yet, he is glad that his walking skills have improved and that he is better able to play together with his classmates.

General discussion

The topics that will be addressed in this General discussion are reliability, validity and responsiveness of the WAL-K, as a new clinical test for assessing walking adaptability; potential efficacy and feasibility of training walking adaptability on a treadmill; clinical and educational implications; methodological considerations; and directions for future research. When I mention children with (mild) CP, I specifically refer to children with CP in GMFCS levels I and II. When I mention children with mild motor disorders, I refer to children with DCD and children with mild CP.

This thesis provides evidence that walking adaptability can be objectively measured using the newly developed WAL-K in TD children and children with mild motor disorders such as DCD and mild CP. Furthermore, the results suggest that children with DCD and mild CP can benefit from treadmill training using augmented reality to improve their walking adaptability, potentially leading to enhanced participation in daily activities. These findings have important clinical implications for the assessment and treatment of walking adaptability in children with mild motor disorders.

Assessment of walking adaptability using the WAL-K

As described in Chapters 2 and 5, the WAL-K was found to be a reliable and valid instrument for measuring walking adaptability in TD children and children with mild motor disorders. Lab-based experimental studies have demonstrated that step adjustment is difficult for children with mild motor disorders, specifically, the assessment of step adjustments under high time pressure is something which could not be measured with other clinical measurement tools[1].

To discuss implications of the findings of the studies related to psychometric properties of the WAL-K, this paragraph is structured based on the COSMIN criteria for methodological quality of a new measurement tool: reliability, validity, and responsiveness[2].

Reliability

The work presented in this thesis demonstrates excellent intra- and inter-rater reliability of the WAL-K in TD children and children with CP. The excellent inter-rater reliability indicates that the WAL-K can be scored reliably by multiple therapists or researchers. In pilot experiments during development of the WAL-K, we noticed that scoring the number of mistakes during task execution (i.e., online) was challenging, particularly when a child made a lot of mistakes. We therefore decided to make video recordings and to score afterwards (i.e., offline), which reduced the measurement error. Offline (or post-hoc) scoring gives raters the possibility to play the video in slow motion or rewind the video when in doubt whether a mistake was made. While this approach requires additional time (approximately 5-10 minutes) to analyze a child's performance, it enhances the accuracy of assessment.

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In TD children, the test-retest reliability of the WAL-K was good. The random error was larger for the double run compared to the single run, which may be attributed to the higher variation in scores on the double run. The systematic error (i.e., systematic difference between test and re-test) on the double run is likely due to a learning effect between the two measurements. Based on these results in TD children, we suggested to add an extra practice trial to the protocol to further improve the test-retest reliability by reducing the learning effect on the WAL-K. The addition of an extra practice trial was applied in the study on reliability and validity of the WAL-K in children with CP (Chapters 5 and 6). Originally, I had planned to also study the test-retest reliability in children with CP, because compared to TD children, they show decreased motor learning capacities with higher variability (i.e., standard deviations)[3,4] which may affect the SDC.

Yet, due to the recruitment difficulties in the group of CP children, as also eluded to in Chapter 6, the number of included children (n=9) was deemed insufficient to evaluate test-retest reliability in Chapter 5. Therefore, no conclusions can yet be drawn regarding test-retest reliability and SDC of the WAL-K in children with mild motor disorders. Therefore, the SDC remains to be determined in a sample of children with mild motor disorders, preferably with a wide range of severity of motor problems.

Validity

The results of Chapters 2 and 5 indicated a good validity of the WAL-K, as demonstrated by its ability to distinguish between TD children and children with mild motor disorders. Additionally, the correlations between WAL-K scores and age further support the validity. Regarding the discriminative validity of the WAL-K, children with DCD showed a substantially poorer performance on the single and double run as compared to TD children: 50% of the children with DCD scored 1 or more standard deviations (SDs) above the mean of TD children on the single run, while this was 81% on the double run. Children with DCD thus showed greater difficulties on the double run, whereas this was not the case in the children with CP: 72% scored 1 or more SDs above the mean of TD children on the single run, and this was 66% on the double run. For the children with CP, this might be due to the fact that 81% of the children in the sample was unilaterally affected and therefore able to compensate for their affected leg in the double run by stepping first with their less affected leg. This was confirmed by observations during task execution.

Age moderately to strongly correlated with WAL-K scores in TD children and children with mild motor disorders. Previous studies in children that used tasks involving a speed-accuracy trade-off showed that movement time (under constant accuracy demands) decreases with increasing age and is dependent on motor maturation5. As motor maturation is delayed in children with DCD and CP, this may also (partly) explain the observed significant differences in WAL-K scores between TD children and children with DCD and CP in Chapters 2 and 5.

A factor that might have influenced the validity of the WAL-K in children with DCD and CP is executive functioning, which is a set of mental processes that facilitate focus and attention when reasoning, problem solving, and planning[6]. Almost all children with DCD7 and more than 50% of children with CP have executive functioning problems[8] which are most prominent in the domains of attention, planning, and working memory[4,8]. Particularly in the first stage of motor learning, like when a child performs the WAL-K for the first time, there is a high load on working memory which is needed to hold and process incoming information (e.g., verbal instructions, visual feedback on the performance, and sensory feedback resulting from the motor performance)[9]. Consequently, deficits in executive functions might influence the performance on the WAL-K in children with DCD and CP. In the current thesis, executive functioning was not explicitly tested, so no conclusions can be drawn about the impact of these factors on WAL-K performance. For now, I recommend that therapists and researchers pay close attention to whether the child understands the instructions properly while administering the WAL-K. If a child does not perform the test according to the instructions, it should be marked as 'failed', in line with the instructions for other motor tests such as the Movement Assessment Battery for Children, Bruininks-Oseretsky Test of Motor Proficiency, and Gross Motor Function Measure[10-12].

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Responsiveness

In the absence of an existing gold standard for assessing walking adaptability, I decided to use the WAL-K for evaluating training-related improvements in walking adaptability in Chapters 4 and 6, despite a lack of information on the test-retest reliability and responsiveness of the WAL-K in children with mild motor disorders. The results of this thesis indicate that the WAL-K is responsive to change, as in both training studies, significant improvements on the WAL-K double run were observed post intervention, in parallel with improvements on treadmill-based adaptability tests. It remains to be determined, though, whether these changes reflected training-induced improvements in walking ability or merely indicated a learning effect on the test itself, as observed between test and retest in the TD children in Chapter 2. In future studies, responsiveness of the WAL-K can also be studied through the collection of longitudinal data[13] which might answer the question whether motor maturation is reflected in better WAL-K scores as a child grows older.

Training walking adaptability on a treadmill

Chapters 4 and 6 were the first studies to examine the potential efficacy of walking adaptability training in children with mild motor disorders. Improvements on treadmill-based walking adaptability tasks were found, yet despite the tasks being different from those used in the C-mill training, the improved success rates may partially be attributed to children's familiarity with the treadmill-based walking adaptability tasks. Importantly, I also found a generalization of the training effects to the overground WAL-K (double run), which effects were retained after follow-up. The observed transfer to an overground task is particularly relevant as children with DCD and CP often struggle to apply their learned skills to everyday situations[15,16]. This observation was also supported by parents reporting improvements in their child's walking ability in daily life after completion of the training.

While significant improvements were observed in the WAL-K double run, no significant effects of the training were found on the single run. Children with DCD as well as children with CP made relatively few mistakes on the single run before the start of the training, leaving limited room for improvement on this task. In contrast, the double run gave more room for improvement: children with DCD made many mistakes before the start of the training compared to TD children, and children with CP needed considerably more time to complete the task compared to TD children. Based on these results, preference would be given to the WAL-K double run for testing effects of an intervention on walking adaptability compared to the single run.

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Concerning the observed improvements on the WAL-K following training, conclusions should be drawn with caution, as the studies in Chapters 4 and 6 did not include a control group. While the training study in children with CP was designed as an RCT with a waiting-list control group, we were compelled to change the study to a pre-post design due to the slow inclusion rate (which was greatly impacted by the COVID-19 pandemic, as elaborated on in Chapter 6). The lack of a control group is the most important methodological limitation of the current thesis. It precludes drawing definitive conclusions on whether the improvement children showed after training was actually caused by the training. Alternatively, the improvement may have been due to a leaning effect on the WAL-K, as was observed in the study in Chapter 2 on the test-retest reliability in TD children. If so, I expect this to only partly explain the observed improvements following training, as the pre to post changes in WAL-K scores were about twice as large as the systematic test-retest differences in TD children. In addition, as recommended in Chapter 2, an additional practice trial for the WAL-K was implemented in the training studies in Chapters 4 and 6 to (supposedly) reduce such systematic test-retest effects. Yet, the impact of adding an extra practice trial on the systematic test-retest differences remains to be studied, both in TD children and in children with mild motor disorders. Besides, the natural development of children's motor skills could also yield improvements in WAL-K scores at post-intervention and retention, although this is unlikely given the age of the children and the relatively short time between the measurements; it is not to be expected that children in this specific age range show substantial gains in motor maturation in the time frame of three to six weeks.

In general, the results of the current thesis suggest that C-mill training yielded positive effects on walking adaptability in children with DCD and CP. The solid grounding of the training protocol in contemporary models of motor learning might have contributed to these positive results. Although DCD and CP are different diagnoses caused by different deficits (coordination and internal modeling deficits in DCD and muscle tone/strength/coordination and sensory deficits in CP), the same underlying motor learning principles which are represented in C-mill training might have had their (combined) influence on the effect of the training. The potential working ingredients we derived from motor learning models are task-specific training, implicit learning with an external focus of attention, augmented feedback, motivation, and variable practice.

- *Task-specific training*
  Task-oriented approaches are focused on the performance of a specific movement task, such as handwriting, catching a ball, or tying shoelaces, to help improve the specific skill[17] and such approaches have been proven effective in children with DCD and CP[15,18]. Within task-specific training, either a repetitive or variable practice structure can be applied[19]. Repetitive practice leads to improved performance during the acquisition phase of motor learning, while variable practice enhances transfer and retention[20]. In the training studies presented in this thesis, a task-oriented approach with variable practice was applied as children performed different walking adaptability exercises on the C-mill at different levels. Children were able to transfer their learned skills to a different task –as indicated by improvement on the WAL-K– and the effect was retained after a follow-up period. In addition, Bonney et al. (2017) showed that improvement on a task trained in a virtual reality environment can be transferred to functional activities[19]. This finding is in line with the results of Chapters 4 and 6 of this thesis in which children show a transfer of walking adaptability skills from an augmented reality treadmill environment to an overground task.

- *Implicit learning with an external focus of attention*
  Human movement is largely based on implicit knowledge of task execution[21]: people can drive a bike or car and walk through a room while holding a cup of hot tea, without the ability to explain in detail how they perform these complex movements in terms of muscle activity, joint angles, and balance. An external focus of attention during motor learning is supposed to enhance implicit learning[22]. With an external focus, attention is paid to the effect of the movement in the environment (e.g., earning points when stepping on a star in a C-mill task) instead of the movement itself when focusing internally (e.g., fully extending your knee). It is hypothesized that implicit learning using an external focus of attention would be more beneficial for children with reduced working memory capacity[9] such as children with DCD and CP[4,8], as working memory involvement is a prerequisite for explicit learning[23]. According to Beek and Roerdink (2022), walking on a treadmill with projected visual context such as the C-mill, which was used for treadmill training in Chapters 4 and 6 of this thesis, is an example of implicit learning with an external focus[21]. Adapting a movement to the demands of the projected context makes use of implicit learning to execute a movement in a certain way to be successful.

- *Augmented feedback*
  Augmented (i.e., extrinsic) feedback is defined as additional information on execution or outcome of a movement[24]. The C-mill provides augmented feedback, for example by means of a projected check mark with a corresponding sound following a successful trial or a cross with a corresponding sound following a mistake. Also, points are given for successful trials which are displayed on the treadmill. Augmented feedback was shown to be beneficial for motor learning in children with DCD[25] and in children with CP[26]. It has also been advised

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to give children with motor disorders some degree of autonomy in determining the frequency and timing of feedback[26]. Giving feedback after each trial –which was done in the C-mill training in this thesis– might lead to dependency of the feedback provided[26]. In future research on C-mill training in children with motor disorders, more attention may be paid to the way feedback is given in terms of focus (on performance or results), specificity, timing, frequency, and level of guidance according to the model of extrinsic feedback from Schoenmaker et al. (2022)[26].

- *Motivation*
  Throughout the intervention, children were generally well-motivated and enjoyed the training sessions. They liked the C-mill exercises, and the gaming element further improved their enjoyment. However, sometimes children were less motivated during a session to perform the exercises according to the instructions, for example when the weather was good and they preferred to go swimming with a friend. More autonomy over the training conditions might further improve their motivation, as it is known from previous research that autonomy over certain aspects of the practice conditions improves motivation and even motor learning[27,28]. In Chapters 4 and 6, children already had some autonomy over the exercises, as twice in every sessions, they were given the choice which game out of three options they wanted to perform. While the order of the exercises was predefined, giving children the possibility to determine the order themselves could have further empowered them with a sense of autonomy.

- *Variable practice*
  It is hypothesized that through variable practice –adaptation of movements to variable circumstances– children deduce general principles for effectively performing a task across different exercises[29]. Ambulant children with CP seem to benefit more from variable practice of gross motor skills compared to repetitive practice[30]. To date, only one study examined the difference between variable and repetitive practice on improving static balance in children with DCD, and found no difference between the types of training on motor learning[31]. In the C-mill training, I chose to use variable practice of skills. Children performed different exercises in a session, and followed their own learning curve per exercise, so for example, they performed the obstacle avoidance exercise on level 3 and the stepping stone exercise on level 5 in the same session. Apart from the potential positive effect of variable practice on motor learning, children and parents found the different exercises and levels motivating to finish the training sessions.

Along with the motor learning principles that I applied in designing the C-mill training protocol, incorporating dual tasking might be a valuable addition. The results in Chapter 3 showed that adding a secondary visuo-motor task magnified the differences in walking adaptability between TD children and children with DCD, thus reflecting poorer automatization of the primary motor task in the latter group. Whether training with dual-task conditions may help improve the automaticity of walking adaptability remains for further research, as I am unaware of any previous studies that have specifically examined the additive effects of dual tasking on training-induced gains.

Feasibility
Despite the desirability of walking adaptability training as expressed by the parents and children in the target groups of DCD and CP, and the benefits experienced by the children who participated in the training studies, inclusion of study participants proved to be hugely challenging. In conversations with parents who declined to participate with their child, various reasons were given. The most frequently-mentioned reason was the considerable burden for the child and family: children have to go to school, sports and rehabilitation programs, their parents have to go to work, and siblings also have school and sports schedules which parents have to take into account. Consequently, scheduling the training sessions and measurements within the context of their daily lives proved to be difficult. Also the distance to the training location was often reported as a reason to not participate.

The influence of the aforementioned factors dramatically increased during the COVID-19 pandemic, which was also recognized in other studies[32]. Despite extension of the study period and additional funding, it remained very challenging to recruit children and parents to participate in the study. The COVID-19 pandemic had direct as well as indirect effects on the inclusion. The direct impact was that people were overloaded because their normal routines and schedules were turned upside down due to closure of the complete society. Not only participants and their parents, but also professionals and management staff involved in the study recruitment and logistics were too overburdened to go the extra length. Indirectly, the pandemic hindered building personal relationships with stakeholders in my network (parents, therapists, rehabilitation physicians, the patients' association), as online meetings are simply less effective than in-person meetings. These personal relationships are often highly beneficial to create goodwill towards involvement in a study. I was skillful in motivating people to help me with my study, but the pandemic put a strain on my inventiveness and willingness to keep contacting people. Sometimes it was frustrating that the study did not go as it was planned, but the consequences of the pandemic were force majeure.

To decrease the burden of participation for the child and family, several solutions might considered. The impact of the travelling distance to a C-mill training location (note that only a few local physiotherapy practices in the Netherlands possess a C-mill) could be decreased by alternating C-mill training with overground training at home, at a local physiotherapy practice or in a fitness center. Addition of overground training to the protocol could be beneficial to improve the transfer of the learned skills to daily life situations[1]. Besides, in overground training it is possible to include vertical step adaptation, which cannot be trained on the C-mill. It might also be beneficial for parents and children to give them the option to follow such a multimodal training during a holiday camp with a short and intensive training period. Holiday rehabilitation camps have been proven to be effective to improve child-chosen goals in children with DCD[33] and functional mobility goals in children with CP[34]. The effectiveness of a multimodal training to improve walking adaptability should be investigated in future research. However, as mentioned before, the C-mill training protocol aligns well with contemporary models of motor learning, so this should be maintained as a basis for the mentioned hybrid solutions.

Alternatively, new rehabilitation technology may become available for enabling walking adaptability training without the dependency on a C-mill, perhaps even at home without support of a therapist. This would be a solution for the growing strain on our healthcare system because of increasing demand and costs, and decreasing availability of healthcare personnel. The HoloLens (Microsoft Corporation, Redmond, WA, USA) is a promising innovation for training walking adaptability, making use of mixed reality (i.e., enabling an individual to perceive the real, physical world and objects as well as convincing and interactive virtual objects[35]). Research has shown that obstacle avoidance could potentially be studied and maybe also trained using the HoloLens[36]. Besides, the HoloLens has excellent sensitivity, specificity, accuracy, and precision for step detection in children with CP in GMFCS levels I to III[37]. More research is needed to study whether tailored training applications using the HoloLens (or similar technologies) provide a viable option for practicing walking adaptability in clinical populations such as children with mild motor disorders.

Despite parents' restraints on study participation, the adherence to the training sessions was high: 100% in the children with DCD and on average 86% in the children with CP. This adherence is high compared to other gait interventions in children with DCD (around 80%[38,39]) and children with CP (74-100%[40]). It might point at parents' and children's satisfaction with the training, which was also supported in the surveys parents completed after the training: 89% of all parents would recommend the training to others. Besides, many parents expressed a willingness to complete the intervention in hopes of seeing considerable improvement of their child's walking adaptability.

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Clinical implications

The most important message I want to convey with this thesis is that walking adaptability warrants much more attention in clinical practice and clinical research in children with mild motor disorders. As stated in all chapters of this thesis, walking adaptability is an essential skill for children to participate in daily activities, but many children with mild motor disorders have difficulties with walking adaptability and therefore, cannot participate in daily activities as they would want to. Especially in children with DCD, walking problems –not even specified to the skill of walking adaptability– need more attention. These problems often remain unnoticed in clinical practice where more attention is paid to fine motor problems, while a considerable amount (14-18%) of children with DCD has gross motor problems[41].

The WAL-K can be used in clinical practice as a reliable and valid measurement tool to assess walking adaptability in children with DCD and CP. The lack of a ceiling effect in children with mild motor disorders further supports the diagnostic value of the WAL-K. Evaluative use of the WAL-K is subject to further research. When it is used in clinical practice for evaluation purposes, clinicians should be aware of a possible learning effect on the task and a the current lack of SDC and MCID values for interpreting individual clinical differences.

Based on the results of Chapters 2 and 4, it may be questioned whether both the single and double run of the WAL-K should be maintained in the test protocol for diagnostic purposes. The Pearson correlation between the single and double run scores was 0.66 ($p<.001$) in TD children, 0.65 ($p<.001$) in children with DCD, and 0.91 ($p<.001$) in children with CP. Based on these correlations, it can be argued that in children with CP, testing only the single or double run may possibly give sufficient information on their walking adaptability. However, the studied samples are too small to give a clear recommendation about the difference between the single and double run. Future research could test this difference in larger samples. Yet, with the knowledge of this thesis, it is recommended to keep testing both the single and double run, as it does not cost a lot of effort and time to test both and it can give valuable information –both quantitative and qualitative– for clinical reasoning.

Although the work in this thesis does not provide conclusive evidence for the efficacy of C-mill training to improve walking adaptability in children with mild motor disorders, the results are promising. The positive results correspond with my personal observations during the training sessions: almost all children took advantage of the training in their own way. Some children improved more than others, but (almost) all children and their parents indicated that they improved their walking ability. All parents of the children with DCD (100%) and 85% of the parents of the children with CP would recommend the training to others, which further supports that children can benefit from the training. The most important step to definitive scientific evidence for C-mill training in the population of children with mild motor disorders is the addition of a control group to prove whether C-mill training is more effective to improve walking adaptability in children with mild motor disorders compared to usual care. However, for the time being, I would certainly recommend to already provide walking adaptability training using the C-mill –in case it is available– for children with mild motor impairments who want to improve their walking adaptability.

Educational implications

Creating more awareness for walking adaptability in clinical practice should already start in the education of clinicians, such as physio-/exercise therapists and rehabilitation physicians. In their educational curriculum, introduction of the model of Balasubramanian et al. (2014)[42] could provide a solid conceptual framework. As explained in the General introduction, this model on the neural control of walking comprises stepping, postural equilibrium, and walking adaptability. This would help allocate the much-needed attention to walking adaptability assessment and training in clinical education. The principle of variable practice has a place in education, for example for pediatric physiotherapists, but this is hardly ever applied to the skill of walking.

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To teach the future generation of physiotherapists and/or exercise therapists the importance of walking adaptability in children's and other people's daily life, I propose the following concrete activities:

- Organize a course in which students learn how to measure and train walking adaptability in patient populations with visits to rehabilitation centers which have devices such as the C-mill. I would design this course around a case description of an existing pediatric or adult patient with walking adaptability problems and let students test and train this patient during the visit to the rehabilitation center. In this way, students will be involved in the complete process of clinical reasoning around walking adaptability problems

- Organize internships in clinical practices or rehabilitation centers where students collect reference data of the WAL-K in different healthy populations and in patient populations to be able to compare a person's walking adaptability to reference data. In this way, students get familiar with the construct of walking adaptability and gain experience in performing research on this topic.

Methodological considerations

As already elaborated on in the paragraph 'Training walking adaptability on a treadmill', the most important methodological limitation is the lack of a control group in both training studies. A second methodological limitation is the lack of blinding of the tester and therapist in the training studies, as I performed all measurements and training sessions for the training studies myself. Several factors can be identified that minimized the potential effects of the lack of blinding on the results and conclusions. First, the training protocol and instructions for exercises and tests were standardized. Second, scoring of outcomes left little room for interpretation. The scores of the treadmill-based tasks were objectively determined by software and the video-based WAL-K scoring yielded highly consistent outcomes within and between raters. To further minimize a potential effect of the lack of blinding, I did not look up the pre-intervention scores before conducting and scoring a post-intervention measurement. However, an (unconscious) effect of this lack of blinding cannot be ruled out completely.

Concerning the training studies, in particular in children with CP, parents' views on the feasibility in terms of the duration and amount of measurements and training sessions could have been further explored in advance. This might have improved the feasibility of the study for parents and, thereby, the number of children that could have been included. Yet, no one could have anticipated the COVID-19 pandemic and the considerable strain it put on all of our lives. While I recommend collecting the parents' (and other stakeholders') views for informing future study designs, I believe it would not have solved the recruitment difficulties that I experienced under the given circumstances.

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Directions for future research

Based on the implications in this discussion, the following recommendations are given for future research on walking adaptability in children with mild motor disorders.

- Concerning the test-retest reliability, SDC and MCID of the WAL-K, I suggest to collect data of a sample of children with mild motor disorders with a broad range across severity of motor problems. This can be done by involving physiotherapists (and students) who treat children with CP and DCD in private practices as well as in rehabilitation centers. They could make video recordings of two performances with one week in between. When knowing the SDC and MCID of the WAL-K in children with mild motor disorders, a change in WAL-K scores after an intervention can be properly interpreted by the therapist or researcher.

- I suggest to sample data from TD children to be able to compute growth curves of neurotypical development of walking adaptability across childhood. The educational trajectories from students physiotherapy or exercise therapy can be used to achieve this goal, as the universities for applied sciences have an important role to educate scientists in practice and can play a facilitating role in data management. Ideally, a national database would be set up so data of a large group of TD children can be used.

- It may be of interest to study whether adding a dual task condition to the WAL-K may further increase its sensitivity in identifying walking adaptability difficulties, as compared to the present single task performance. According to Balasubramanian et al. (2014), motor and cognitive dual tasking are two essential domains of walking adaptability[42]. Examples are crossing the street between two approaching cars or playing football with classmates. The performance of dual tasks is related to the executive functioning domains of attention and working memory[9]. A dual task paradigm can be used to assess the level of automaticity of a movement, and if a movement is being executed automatically, less cognitive resources (e.g., attention and working memory) are needed to control the movement[43]. Assessing dual tasks during walking adaptability might thus give a more complete view on the construct of walking adaptability as in daily life, situations in which walking adaptability is needed often come together with performing dual tasks[42]. As I demonstrated Chapter 3 of this thesis, adding a dual task condition to the walking adaptability task on the C-mill magnified the differences between TD children and children with DCD. Therefore, adding a dual task trial to the existing protocol of the WAL-K is of interest, but the secondary task needs to be selected with care, as dual task performance is highly dependent on the nature and difficulty of the task[44].

- Additional research is needed for obtaining more conclusive evidence regarding the efficacy of walking adaptability training in children with mild motor disorders. The study design (e.g., amount, duration and location of measurements) and content of the intervention (e.g., duration, intensity, C-mill training or a hybrid solution) should be determined in co-creation with parents and other stakeholders. It goes without saying that the preferences of parents should always be considered in light of the scientific aims of the study.

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Nederlandse samenvatting

Kinderen met milde motorische aandoeningen, zoals DCD (Developmental Coordination Disorder) en milde CP (Cerebrale Parese), hebben vaak moeite met het loopaanpassingsvermogen. Het loopaanpassingsvermogen is het aanpassen van het looppatroon om beoogde doelen te bereiken waarbij tegelijkertijd rekening wordt gehouden met de eisen die de omgeving stelt (bijvoorbeeld het voorkomen van struikelen over een bal of botsen met klasgenootjes). Loopaanpassingsvermogen is essentieel voor kinderen om deel te nemen aan dagelijkse activiteiten zoals sport, spel en schoolactiviteiten. Er bestaan echter geen objectieve meetinstrumenten voor loopaanpassingsvermogen en interventies richten zich alleen op het onverstoorde lopen of de balans, in plaats van op loopaanpassingsvermogen. Daarom was het overkoepelende doel van dit proefschrift om klinische beoordeling en training van het loopaanpassingsvermogen bij kinderen met milde motorische aandoeningen te ontwikkelen en te evalueren. Het uiteindelijke doel was om kinderen te kunnen identificeren die problemen hebben met het loopaanpassingsvermogen en hun loopaanpassingsvermogen te verbeteren door middel van training, zodat ze beter kunnen deelnemen aan dagelijkse activiteiten samen met andere kinderen.

De stapladdertest (in het Engels de Walking Adaptability Ladder test for Kids, WAL-K) is een nieuw ontwikkeld meetinstrument voor loopaanpassingsvermogen. In **hoofdstuk 2** van dit proefschrift zijn de psychometrische eigenschappen van de WAL-K onderzocht bij kinderen van 6 tot 12 jaar. In totaal hebben 122 kinderen met een reguliere motorische ontwikkeling en 26 kinderen met Developmental Coordination Disorder (DCD) de twee taken van de WAL-K uitgevoerd: met één stap in elk vak en met twee stappen in elk vak. Intra- en interbeoordelaarsbetrouwbaarheid en test-hertestbetrouwbaarheid werden bepaald door middel van correlatiecoëfficiënten en het kleinste verschil in score dat kan worden onderscheiden van meetfouten bij 53 kinderen met een reguliere motorische ontwikkeling. De constructvaliditeit werd bepaald door WAL-K-scores te vergelijken tussen 69 kinderen met een reguliere motorische ontwikkeling en alle kinderen met DCD, en door WAL-K-scores te correleren met leeftijd en scores op de Movement Assessment Battery for Children, versie 2 (MABC-2). De correlatiecoëfficiënten voor betrouwbaarheid varieerden tussen 0.76 en 0.99. In vergelijking met de eerste test waren WAL-K-scores significant lager (beter) bij hertest. Het kleinste verschil in score dat kan worden onderscheiden van meetfouten varieerde tussen 20.8% en 26.1% van de gemiddelde scores. WAL-K-scores waren significant hoger (slechter) bij kinderen met DCD in vergelijking met kinderen met een reguliere motorische ontwikkeling ($p<0.001$). Er werden significante negatieve correlaties gevonden met de MABC-2 (-0.52 en -0.60) en leeftijd (-0.61 en -0.68). De WAL-K bleek een valide, betrouwbaar en gemakkelijk te gebruiken instrument te zijn om loopaanpassingsvermogen bij kinderen te meten. Het toevoegen van een extra oefenpoging kan het waargenomen leereffect verminderen.

In het dagelijks leven komen kinderen vaak situaties tegen waarin ze dubbeltaken moeten uitvoeren tijdens het lopen, zoals bij sporten en tijdens spel. Daarom testten we in **hoofdstuk 3** de hypothese dat kinderen van 6 tot 12 jaar met DCD lagere niveaus van loopaanpassingsvermogen hebben dan kinderen met een reguliere ontwikkeling en dat dit verschil toeneemt wanneer ze gedwongen worden om hun aandacht te verdelen tussen taken wanneer gelijktijdig een visuo-motorische of cognitieve taak wordt toegevoegd. Zesentwintig kinderen met DCD en 69 kinderen met een reguliere motorische ontwikkeling namen deel aan deze cross-sectionele studie. Ze voerden een uitdagende taak voor loopaanpassingsvermogen uit op een loopband als enkeltaak, als visuo-motorische dubbeltaak en als cognitieve dubbeltaak met een snelheid van 3.5 km/u. Een ANCOVA voor herhaalde metingen werd uitgevoerd met conditie (enkel-/dubbeltaak) als within-subjects factor, groep (reguliere ontwikkeling/DCD) als between-subjects factor, en leeftijd als covariaat. Kinderen met DCD presteerden significant slechter op de taak voor loopaanpassingsvermogen dan kinderen met een reguliere motorische ontwikkeling. De groepsverschillen namen significant toe wanneer een gelijktijdige visuo-motorische taak werd toegevoegd, maar niet wanneer een gelijktijdige cognitieve taak werd toegevoegd. Er werd een significant effect van de leeftijd gevonden waarbij jongere kinderen slechter presteerden op alle taken. Deze resultaten benadrukken de problemen die kinderen met DCD hebben met loopaanpassingsvermogen en dubbeltaken, die essentieel zijn om volledig te kunnen deelnemen aan sport- en spelactiviteiten.

Aangezien kinderen met DCD problemen bleken te hebben met loopaanpassingsvermogen en dubbeltaken, had **hoofdstuk 4** als doel om te onderzoeken of augmented-reality loopbandtraining leidt tot verbeteringen in loopaanpassingsvermogen bij kinderen van 6 tot 12 jaar met DCD. Zeventien kinderen met DCD namen deel aan deze interventiestudie. Ze kregen een training van 6 sessies op de C-mill, een loopband waarop loopaanpassingen kunnen worden uitgelokt door geprojecteerde visuele context. Het effect van de training werd beoordeeld vóór (M1), direct na de training (M2) en na zes maanden follow-up (M3) met behulp van de twee taken van de WAL-K (één en twee stappen in elk vak) en taken voor loopaanpassingsvermogen op de C-mill (als enkele en met gelijktijdige visuo-motorische en cognitieve taak). Daarnaast vulden ouders een vragenlijst in over hun perceptie van de training. Lineair Mixed Model analyses werden uitgevoerd om de verschillen in WAL-K-scores en succespercentages op de taken voor loopaanpassingsvermogen tussen M1-M2 en M1-M3 te bepalen. Kinderen verbeterden significant op de taak van de WAL-K met twee stappen in elk vak en op alle taken voor loopaanpassingsvermogen op de C-mill tussen M1-M2 en M1-M3. Kinderen verbeterden niet op de taak van de WAL-K met één stap in elk vak. Ouders vonden de training nuttig en leuk voor hun kind en gaven aan dat hun kind minder vaak viel. De resultaten toonden aan dat C-mill training positieve en taakspecifieke effecten had op loopaanpassingsvermogen bij kinderen met DCD. De effecten werden gegeneraliseerd naar een taak op de grond (de WAL-K) en werden behouden na zes maanden. Deze training zou kinderen met DCD kunnen helpen om beter deel te nemen aan dagelijkse activiteiten.

Vanwege de vergelijkbare problemen met loopaanpassingsvermogen van kinderen met Cerebrale Parese (CP) als kinderen met DCD, zou het waardevol zijn om de WAL-K ook als klinische test bij deze groep kinderen te kunnen implementeren. Daarom werden in **hoofdstuk 5** de psychometrische eigenschappen van de WAL-K onderzocht bij kinderen van 6 tot 12 jaar met milde CP (GMFCS-niveau I en II). In totaal voerden 36 kinderen met CP de twee taken van de WAL-K uit: met één stap in elk vak en met twee stappen in elk vak. Intra- en interbeoordelaarsbetrouwbaarheid werden bepaald door correlatiecoëfficiënten. De constructvaliditeit werd bepaald door de WAL-K-scores te correleren met scores van een sprinttest (10x5mST), door de WAL-K-scores te vergelijken tussen 122 kinderen met een reguliere ontwikkeling en alle kinderen met CP, en door de WAL-K-scores te vergelijken tussen kinderen met CP in GMFCS-niveau I en II. Correlatiecoëfficiënten voor betrouwbaarheid varieerden tussen 0.997 en 1.000. Er waren significante positieve correlaties gevonden met de 10x5mST (r = .89 en .84). WAL-K-scores waren significant hoger (slechter) bij kinderen met CP in vergelijking met kinderen met een reguliere ontwikkeling (p <0.001) en bij kinderen met GMFCS-niveau II vergeleken met GMFCS-niveau I (p = .001). De WAL-K bleek een betrouwbaar, valide en gemakkelijk te gebruiken instrument te zijn voor het meten van loopaanpassingsvermogen bij kinderen met milde CP.

Aangezien kinderen met milde CP problemen bleken te hebben met loopaanpassingsvermogen, was **hoofdstuk 6** gericht op het onderzoeken van het effect van augmented-reality loopbandtraining op het loopaanpassingsvermogen bij kinderen van 6 tot 17 jaar met milde CP. Veertien kinderen met CP (GMFCS-niveau I en II) kregen een training van 10 sessies op een loopband waarbij loopaanpassingen kunnen worden uitgelokt door geprojecteerde visuele context. Het effect van de training werd beoordeeld vóór (M1), direct na de training (M2) en na drie maanden follow-up (M3) met behulp van de twee taken van de WAL-K (één en twee stappen in elk vak) als primaire uitkomstmaat. Ouders vulden een vragenlijst in over hun perceptie van de training. Linear Mixed Model analyses werden uitgevoerd om de verschillen in WAL-K scores tussen M1-M2 en M1-M3 te beoordelen. Kinderen verbeterden significant op de taak van de WAL-K met twee stappen in elk vak tussen M1-M2 en M1-M3, maar niet op de taak van de WAL-K met één stap in elk vak. Ouders vonden de training nuttig en leuk voor hun kind en gaven aan dat hun kind minder vaak viel. Loopbandtraining had positieve effecten op het loopaanpassingsvermogen bij kinderen met CP, die behouden werden na drie maanden. Deze training zou kinderen met CP kunnen helpen beter deel te nemen aan dagelijkse activiteiten.

In **hoofdstuk 7** werden de belangrijkste bevindingen van dit proefschrift bediscussieerd. We vonden dat het loopaanpassingsvermogen objectief gemeten kan worden met behulp van de nieuw ontwikkelde WAL-K bij kinderen met een reguliere motorische ontwikkeling en kinderen met milde motorische aandoeningen zoals DCD en CP. De resultaten geven aan dat kinderen met DCD en milde CP baat kunnen hebben bij loopbandtraining met behulp van augmented reality om hun loopaanpassingsvermogen te verbeteren. Dit kan leiden tot betere mogelijkheden om aan dagelijkse activiteiten deel te nemen. De bevindingen uit dit proefschrift geven belangrijke klinische implicaties voor de beoordeling en behandeling van loopaanpassingsvermogen bij kinderen met milde motorische aandoeningen. De belangrijkste boodschap die ik wil uitgedragen met dit proefschrift is dat er zowel in de klinische praktijk als het klinisch onderzoek bij kinderen met milde motorische aandoeningen meer aandacht moet komen voor het loopaanpassingsvermogen.

Research data management

General information about data collection
This research followed the applicable laws and ethical guidelines for medical scientific research. Research Data Management was conducted according to the FAIR principles. The paragraphs below specify in detail how this was achieved.

Ethics
This thesis is based on the results of human studies, which were conducted in accordance with the principles of the Declaration of Helsinki. Written informed consent was given by all participants. All studies were approved by the regional Medical Ethical Committee Arnhem-Nijmegen (chapter 2 – dossier number 2017-3465; chapter 3 – dossier number NL59150.091.16; chapter 4 – dossier number 2016–2885; chapter 5 – dossier numbers 2017-3465 and 2020-6504; chapter 6 – dossier number 2018-4223).

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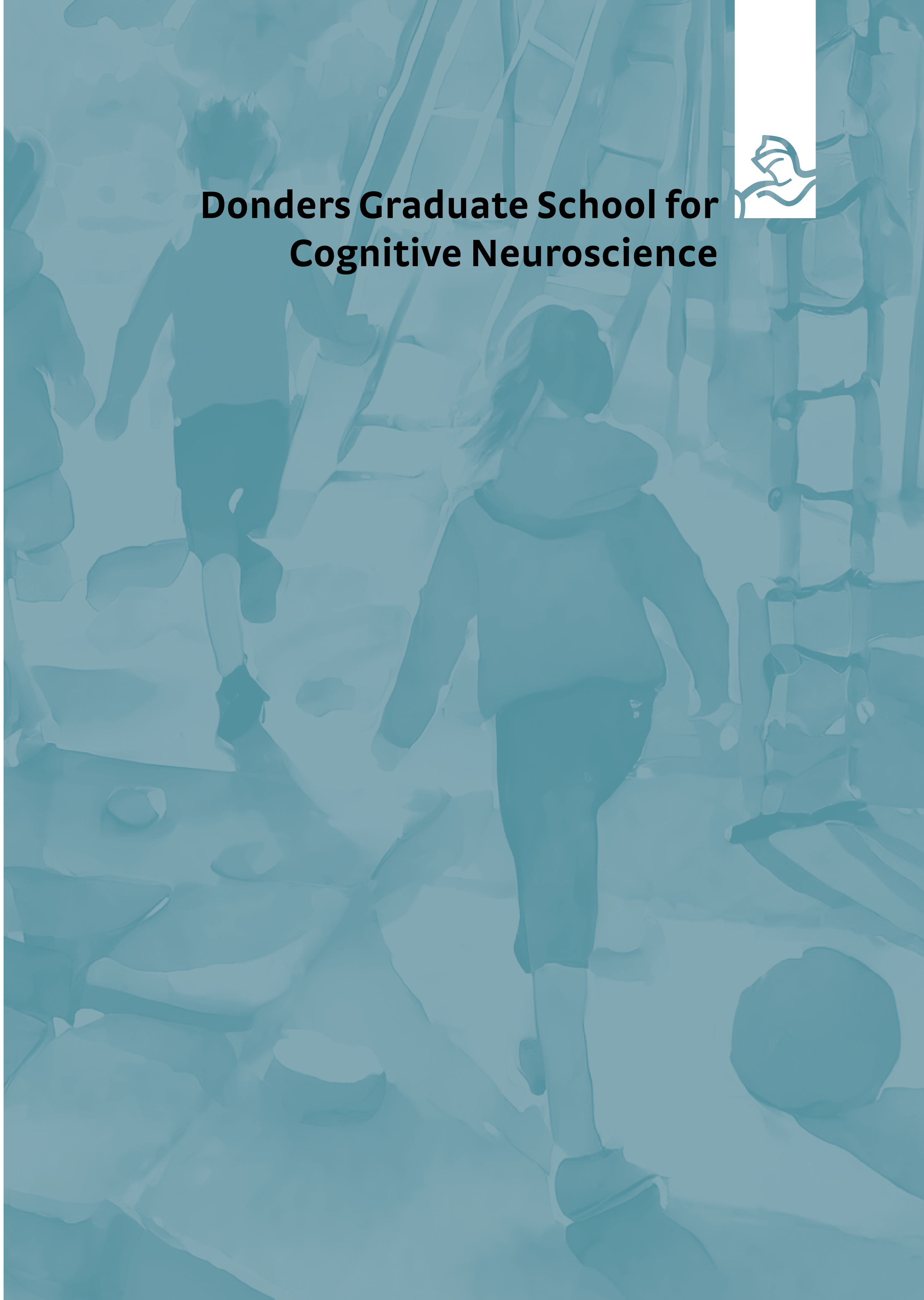
FAIR principles
Findable
Data of chapters 2, 3 and 4 were stored on the server of the Rehabilitation department of the Radboudumc: Q:\Research\063 C-Mill DCD. Data of chapters 5 and 6 were stored on the server of the research department at the Sint Maartenskliniek: V:\research_reva_studies\817_CMill_training_ CP. The paper CRF files and informed consent forms were stored in the department's archive (for chapters 2, 3 and 4 in the Radboudumc and for chapters 5 and 6 in the Sint Maartenskliniek).

Accessible
All data will be stored for 15 years after the completion of the studies. Data are accessible upon reasonable request by contacting the corresponding author.

Interoperable
Documentation was added to the data sets to make the data interpretable. The documentation contains links to publications, references to the location of the data sets and description of the data sets. The data were stored in the following file formats: *.mat (MATLAB, Mathworks, USA), *.csv (Microsoft Office Excel), and *sav (SPSS, IBM Corp., USA).

Reusable
While obtaining informed consent, individual participants had to indicate whether they would allow reuse of their data during a period of 15 years. Data obtained from participants that indicated that their data could be reused, is available for reuse during this time period.

Privacy
The privacy of the participants in this thesis has been warranted using encrypted and unique individual subject codes. The encryption key was stored separately from the research data and was only accessible to members of the project who needed access to it because of their role within the project.



Donders Graduate School for Cognitive Neuroscience

For a successful research Institute, it is vital to train the next generation of young scientists. To achieve this goal, the Donders Institute for Brain, Cognition and Behaviour established the Donders Graduate School for Cognitive Neuroscience (DGCN), which was officially recognised as a national graduate school in 2009. The Graduate School covers training at both Master's and PhD level and provides an excellent educational context fully aligned with the research programme of the Donders Institute.

The school successfully attracts highly talented national and international students in biology, physics, psycholinguistics, psychology, behavioral science, medicine and related disciplines. Selective admission and assessment centers guarantee the enrolment of the best and most motivated students.

The DGCN tracks the career of PhD graduates carefully. More than 50% of PhD alumni show a continuation in academia with postdoc positions at top institutes worldwide, e.g. Stanford University, University of Oxford, University of Cambridge, UCL London, MPI Leipzig, Hanyang University in South Korea, NTNU Norway, University of Illinois, North Western University, Northeastern University in Boston, ETH Zürich, University of Vienna etc.. Positions outside academia spread among the following sectors: specialists in a medical environment, mainly in genetics, geriatrics, psychiatry and neurology. Specialists in a psychological environment, e.g. as specialist in neuropsychology, psychological diagnostics or therapy. Positions in higher education as coordinators or lecturers. A smaller percentage enters business as research consultants, analysts or head of research and development. Fewer graduates stay in a research environment as lab coordinators, technical support or policy advisors. Upcoming possibilities are positions in the IT sector and management position in pharmaceutical industry. In general, the PhDs graduates almost invariably continue with high-quality positions that play an important role in our knowledge economy.

For more information on the DGCN as well as past and upcoming defenses please visit:
http://www.ru.nl/donders/graduate-school/phd/

About the author

Rosanne Kuijpers was born on January 27th 1990 in Boxmeer. She grew up with her parents and two sisters in Oploo, a small town in Noord-Brabant.

After graduating from secondary school in 2008 at the Elzendaal College in Boxmeer, she started the bachelor Exercise Therapy at the University of Applied Sciences Utrecht. In 2012, she obtained her bachelor degree and started working as an exercise therapist at a special elementary school in Rotterdam. In the same year, she started the post-bachelor study Pediatric Exercise Therapy at the University of Applied Sciences Utrecht, from which she graduated in 2015. From 2012 until 2021, she worked as a pediatric exercise therapist at different elementary schools, in healthcare institutions, and private practices.

In 2012, Rosanne also started the master Clinical Health Sciences at Utrecht University. During her masters, she became interested in scientific research in pediatric clinical populations. She did an internship on the long-term effects of congenital heart diseases in children at the Wilhelmina Children's Hospital in Utrecht and wrote her master thesis about determinants of physical activity in children at elementary school. Rosanne graduated in 2015 and started in 2016 as a research assistant at the department of Rehabilitation of the Radboudumc in Nijmegen on a project about walking adaptability in children with Developmental Coordination Disorder. In 2019, she started as a junior researcher at the Sint Maartenskliniek in Nijmegen on a project about walking adaptability in children with Cerebral Palsy.

Currently, Rosanne is working as program manager Healthcare Improvement at NEO Huisartsenzorg, the regional organization for general practitioners in Nijmegen and surroundings. She is engaged in improving the regional collaboration between general practitioners and other healthcare providers.

List of publications

International scientific publications

Kuijpers R, Smulders E, Groen BE, Smits-Engelsman BCM, Nijhuis-Van der Sanden MWG, Weerdesteyn V. Reliability and construct validity of the Walking Adaptability Ladder Test for Kids (WAL-K): a new clinical test for measuring walking adaptability in children. Disability and Rehabilitation. 2022;44(8):1489-1497.

Kuijpers R, Smulders E, Groen BE, Smits-Engelsman BCM, Nijhuis-Van der Sanden MWG, Weerdesteyn V. Walking adaptability improves after treadmill training in children with Developmental Coordination Disorder: A proof-of-concept study. Gait & Posture. 2022;92:258-263.

Kuijpers R, Smulders E, Groen BE, Smits-Engelsman BCM, Nijhuis-Van der Sanden MWG, Weerdesteyn V. The effects of a visuo-motor and cognitive dual task on walking adaptability in children with and without Developmental Coordination Disorder. Gait & Posture. 2022;95:183-185.

National publication

Kuijpers R, Smulders E, Weerdesteyn V. Functionele loopvaardigheid van kinderen met Developmental Coordination Disorder (DCD). Nederlands Tijdschrift voor Oefentherapie. 2019.

Conference abstracts

Kuijpers R, Smulders E, Weerdesteyn V. The speed agility ladder: A new clinical test for measuring gait adaptability in children. ISPGR world conference, Fort Lauderdale, Florida, 2017 (poster presentation).

Kuijpers R, Smulders E, Groen BE, Weerdesteyn V, Nijhuis-van der Sanden MWG. Task-oriented treadmill training improves gait adaptability in children with Developmental Coordination Disorder. ESMAC international conference, Amsterdam, 2019 (oral presentation).

Kuijpers R, Smulders E, Groen BE, Weerdesteyn V, Nijhuis-van der Sanden MWG. Gait adaptability can be improved by task-oriented treadmill training in children with developmental coordination disorder. EUPPT conference, Utrecht, 2019 (oral presentation).

Kuijpers R, Smulders E, Groen BE, Weerdesteyn V, Nijhuis-van der Sanden MWG. Comparison of functional gait between children with developmental coordination disorder and typically developing children. EUPPT conference, Utrecht, 2019 (poster presentation).

Kuijpers R, Smulders E, Groen BE, Weerdesteyn V, Nijhuis-van der Sanden MWG. Task-oriented treadmill training improves gait adaptability in children with Developmental Coordination Disorder. Donders Discussions, Nijmegen, 2019 (oral presentation).

Kuijpers R, Smulders E, Groen BE, Weerdesteyn V, Nijhuis-van der Sanden MWG. Functional gait in children with Developmental Coordination Disorder compared to typically developing children. ISPGR world conference, Edinburgh, 2019 (poster presentation).

Kuijpers R, Smulders E, Groen BE, Weerdesteyn V, Nijhuis-van der Sanden MWG. Task-oriented treadmill training improves gait adaptability in children with Developmental Coordination Disorder. ESMAC international conference, Amsterdam, 2019 (oral presentation).

Kuijpers R, Groen BE, Smulders E, Nijhuis-van der Sanden MWG, Weerdesteyn V. Task-oriented treadmill training to improve walking adaptability in children with Cerebral Palsy. ESMAC international conference, virtual meeting, 2021 (poster presentation).

Kuijpers R, Smulders E, Groen BE, Weerdesteyn V, Nijhuis-van der Sanden MWG. Walking adaptability can be improved by task-oriented treadmill training in children with Developmental Coordination Disorderer. EACD conference, virtual meeting, 2021 (oral presentation).

Van Gelder H, Snik D, Höweler S, Groen B, **Kuijpers R**. Behandeling van loopproblemen bij mensen met Cerebrale Parese. LEC symposium, Nijmegen, 2022 (oral presentation).

Kuijpers R, Groen BE, Smulders E, Nijhuis-van der Sanden MWG, Weerdesteyn V. Het meten van het loopaanpassingsvermogen bij kinderen. Symposium Kinderoefentherapie, Utrecht, 2023 (oral presentation).

Name PhD Student: Rosanne M.H. Kuijpers
Department: Research (Sint Maartenskliniek)
Graduate School: Donders Graduate School

Training activities	Year	Hours
Courses and workshops		
- BROK	2018	42
- Mindfulness-based stress reduction, Radboudumc	2018	56
- How to write a medical scientific paper, Radboud University	2018	9
- EMG course, Society for Movement Analysis Laboratories in the Low Lands (SMALLL) conference	2018	10
- Matlab programming for data acquisition and analysis, Radboud University	2019	84
- Movement Science in Rehabilitation, Radboud University	2019	84
- Donders Graduate School Day	2019, 2020	14
- GRAIL course, Motek Forcelink	2019	28
- Projectmanagement voor promovendi, Radboud University	2019	56
- Donders Scientific Integrity course	2020	7
- Scientific writing for PhD candidates, Radboud University	2020	84
- Education in a nutshell, Radboud University	2021	28
- Re-registration BROK	2021	5
- The next step in my career, Radboud University	2021	23
- Presentatietraining Spies & Spreken	2023	8
(Inter)national symposia and conferences		
- International Society of Posture and Gait Research (ISPGR) world conference, poster presentation	2017, 2019	64
- Society for Movement Analysis Laboratories in the Low Lands (SMALLL) conference, poster presentation	2017, 2018	16
- Johanna Kinderfonds (JKF) en Kinderrevalidatie Fonds Adriaanstichting (KFA) national conference, attendee	2018	8
- European Society of Movement Analysis for Adults and Children (ESMAC) international conference, oral presentation	2019, 2021	56
- Donders Discussions, oral presentation	2019	16
- European Pediatric Physiotherapy (EUPPT) conference, oral and poster presentation	2019	8
- European Academy of Childhood Disability (EACD) conference, oral presentation	2021	24
- Loopexpertisecentrum (LEC) conference, oral presentation	2022	8
- Symposium Kinderoefentherapie, oral presentation	2023	8
Other		
- Writing Week Department Rehabilitation, Radboudumc	2018	56
- Personal Development Week Department Rehabilitation, Radboudumc	2018	37
- Donders Graduate School Introduction Day	2019, 2020	7
- Research Lunches Department Rehabilitation, Radboudumc	2019, 2020	56
- Kinderrevalidatie Research Meetings, Radboudumc	2019, 2020	16
- Research Content Meeting (ReCoMe), Sint Maartenskliniek	2019-2023	50
Teaching activities		
Lecturing		
- Minor 'Moving questions', Bachelor Biomedical Sciences, Radboud University	2021, 2023	16
- Organization of Meet the PhD	2021	8
Supervision of internships		
- Supervision of Master student Biomedical Engineering (University of Twente)	2019	84
- Supervision of Master student Biomedical Sciences (RU)	2020	84
- Supervision of Master student Doctor – Clinical Scientist (Maastricht University)	2021	84
- Supervision of Master student Biomedical Sciences (RU)	2021	84
- Supervision of Master student Biomedical Sciences (RU)	2021	84

Dankwoord

Na 5 jaar werken ligt het er dan, mijn proefschrift, mijn 'boekje'. Een afsluiting van een mooie en leerzame periode waarin ik mezelf nog beter heb leren kennen, en tegelijkertijd het begin van nieuwe uitdagingen en avonturen. Ik wil mijn dank uitspreken aan alle mensen die, op welke wijze dan ook, een bijdrage hebben geleverd aan de totstandkoming van dit proefschrift. Enkele mensen wil ik hier specifiek noemen.

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Vivian, alweer 7,5 jaar geleden nam je me aan als onderzoeksassistent, en sinds die tijd heb ik ontzettend veel van je geleerd. Ik bewonder je doorzettingsvermogen, je gedetailleerde inhoudelijke kennis en je professionaliteit. Je gaat altijd voor het beste resultaat zonder scherpte te verliezen, zelfs midden in de nacht. Ik ben blij dat ik onderdeel mocht zijn van de onderzoeksgroepen in het Radboudumc en de Sint Maartenskliniek (SMK) waar zoveel mooie projecten lopen.

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De kinderfysiotherapeuten en kinderrevalidatieartsen van de SMK wil ik bedanken voor het werven van kinderen en het helpen met pilottests. In het bijzonder wil ik Bram en Margit bedanken voor jullie enthousiasme en flexibiliteit bij het geven van C-mill training, daar hebben jullie me erg mee geholpen.

De oud-collega's van de afdeling Revalidatie van het Radboudumc wil ik bedanken voor de goede start die jullie me hebben gegeven, de gezelligheid (in de vorm van borrels, de persoonlijke ontwikkelingsweek, 'so you think you can calibrate', congresbezoekjes) en de leerzame labmeetings en journal clubs.

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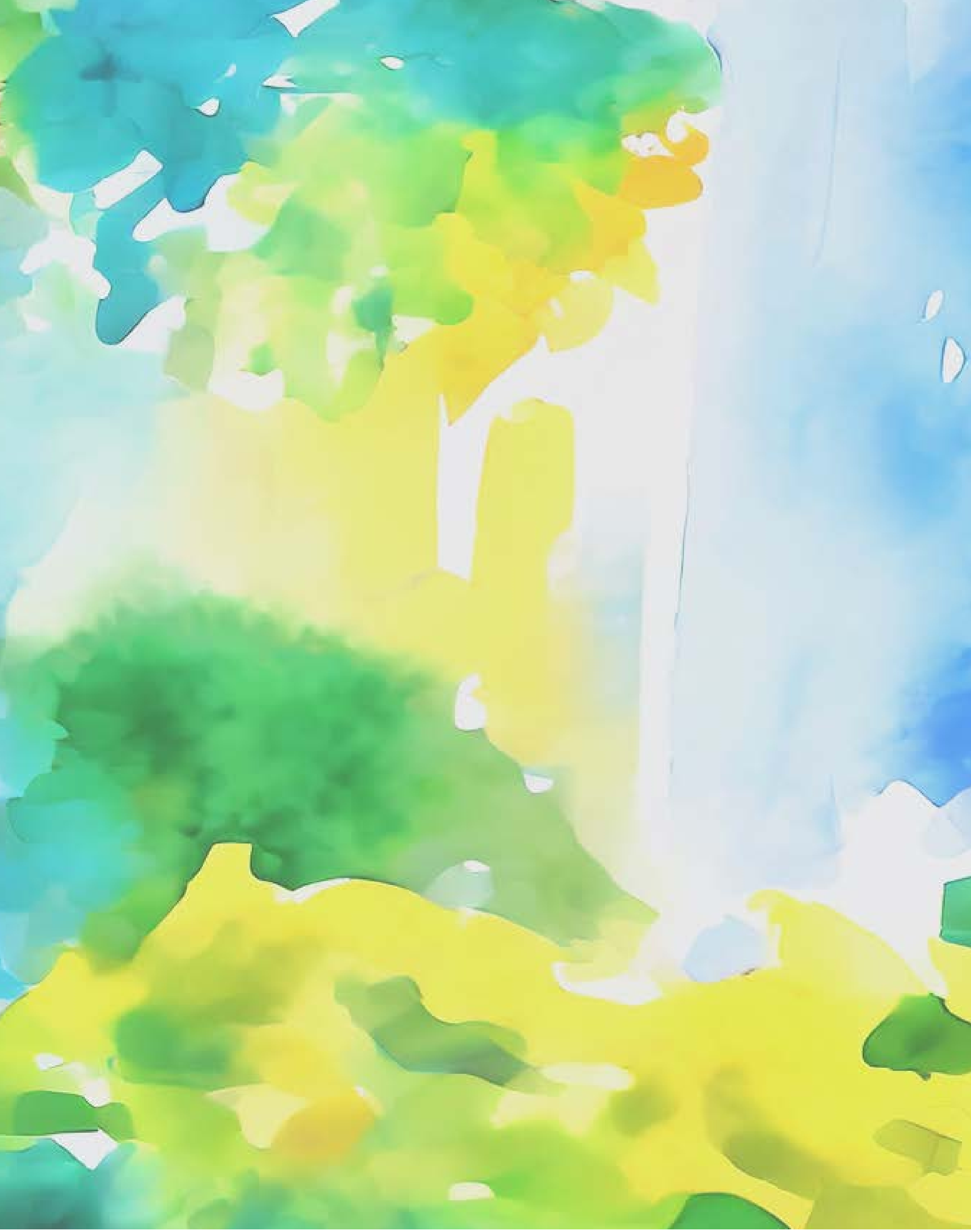

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