fitkids thesis

fitkids

Creating possibilities for youth with chronic medical conditions

UMC Utrecht Brain Center

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fitkids

Creating possibilities for youth with
chronic medical conditions

COLOFON

FITKIDS EXERCISE THERAPY PROGRAM

Creating possibilities for youth with chronic medical conditions

ISBN: 978-94-028-1695-2

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Cover design by Peter van Limbeek

Lay-out design by Joppe Klein, persoonlijkproefschrift.nl

Printed by Ipskamp Printing, proefschriften.net

The work presented in this thesis is supported by the Johan Cruyff Foundation, the Rabobank Foundation and the Dutch Foundation Innovation Alliance (SIA RAAK) project numbers 2011-13-035P and SIA RAAK PRO-4-03.

The printing of this thesis was financially sponsored by:

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FITKIDS EXERCISE THERAPY PROGRAM

Creating possibilities for youth with chronic medical conditions

FITNESS- EN OEFENPROGRAMMA FITKIDS

Creëren van mogelijkheden voor kinderen en adolescenten
met een chronische ziekte of beperking

(met een samenvatting in het Nederlands)

PROEFSCHRIFT

ter verkrijging van de graad van doctor
aan de Universiteit Utrecht,
op gezag van de rector magnificus, prof. dr. H.R.B.M. Kummeling,
ingevolge het besluit van het
college voor promoties
in het openbaar te verdedigen
op donderdag 14 november 2019
des ochtends te 10.30 uur

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TABLE OF CONTENTS

CHAPTER 1

General introduction

YOUTH WITH CHRONIC MEDICAL CONDITIONS

Prevalence

Recently published data show that in the Netherlands over 1.3 million children and adolescents, between 0 and 25 years of age, grow up with a chronic medical condition (CMC), which equals to more than 1 in 4 children.[1] These growing numbers are consistent with other countries[2,3] and these young people are therefore an increasingly important group of healthcare users. In literature, a wide variability in prevalence estimates is reported depending on the definition of CMC used. For the estimate of prevalence presented here, a disease or condition is considered to be a chronic condition if: (1) the diagnosis is based on medical scientific knowledge and can be established using reproducible and valid methods or instruments according to professional standards, (2) it has been present for longer than 3 to 6 months or it has occurred 3 times or more during the past year, and/or (3) the condition requires long-term medicine use, treatments and/or special equipment.[1]

Population

The population of children and adolescents with a CMC is highly heterogeneous and these chronic conditions can be classified into diseases (somatic and psychological) and disabilities (physical and intellectual).[1] Somatic diseases can be life-threatening (e.g., cancer), can cause recurrent complaints (e.g., asthma, cystic fibrosis, epilepsy), or may have a progressive character (e.g., rheumatism). Examples of psychological diseases are anxiety and mood disorders, autism spectrum disorders and attention deficit hyperactivity disorder (ADHD). Examples of physical disabilities are congenital heart defects, cerebral palsy, spina bifida and visual impairment. Intellectual disabilities include for example Down syndrome. In the Netherlands, asthma is the most common CMC in children and young people between 0-25 years of age (4.6%), followed by anxiety and mood disorders (4.1%) and ADHD (3.6%).[1]

Evidence shows that many children and adolescents with CMCs have reduced fitness levels compared to their typically developing peers.[4-7] Additionally, many studies have demonstrated the associations between insufficient physical activity, low physical fitness and a higher prevalence of risk factors which lead to cardiovascular and metabolic diseases.[8,9,10] At the same time, a high level of physical fitness in childhood and adolescence is associated with lower risks of future overweight, fatness, and metabolic syndrome.[11] Research also supports the view that physical fitness benefits children's cognitive functioning[12] and health-related quality of life (HRQoL).[13,14] The current understanding of the relationship between physical activity, physical fitness and health can be visualized using the model from Bouchard et al (Figure 1). The model illustrates that physical activity can influence fitness and health and that the relationships are also reciprocal.[15] Additionally, factors such as personal attributes are known to influence physical fitness, physical activity and health.

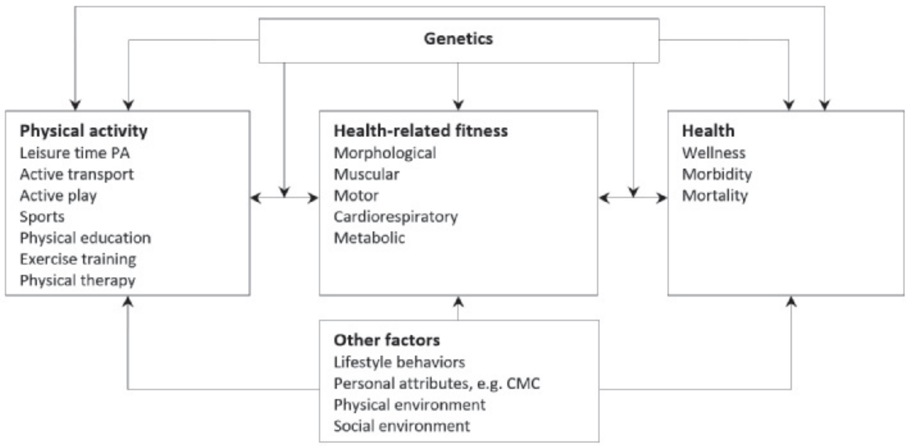

Figure 1. Model of Bouchard and Shepard; linking physical activity, health-related fitness and health.

PHYSICAL ACTIVITY

Physical activity is defined as: 'any bodily movement produced by skeletal muscles that results in an increase in metabolic rate over resting energy expenditure'.[16] Recently published data shows that physical activity is a strong predictor of aerobic fitness in youth with CMCs.[17] Public-health guidelines suggest that typically developing children and adolescents between 6 to 17 years of age should be active for at least 60 minutes per day, predominantly in the form of moderate- or vigorous-intensity physical activity. In addition to the 60 minutes daily physical activity, children and adolescents should include muscle-strengthening and bone-strengthening activities on at least three days a week.[18-21] Recent national survey data points out that only 47% of the children and 27% of the adolescents with CMCs meet these guidelines in the Netherlands. The survey also points out that participation in sports highly contributes to overall physical activity in children (19 min/day) and adolescents (39 min/day) with CMCs.[18] Although the percentages of children and adolescents with CMCs meeting the regular exercise recommendations are comparable with those found in the typically developing population, promoting participation in physical activity in youth with CMCs is particularly valuable as they are at higher risk for developing low activity levels.[22]

PHYSICAL FITNESS

Physical fitness is 'the ability to carry out daily tasks with vigour and alertness, without undue fatigue and with ample energy to engage in leisure time pursuits and to meet the above-average physical stresses encountered in emergency situations'.[23] Physical fitness involves both health-related and performance-related components. Health-related fitness is 'a state characterized by an ability to perform daily activities with vigour, and traits and capacities that are associated with low risk of premature development of the hypokinetic diseases' (i.e., those associated with physical inactivity). Performance-related fitness refers to 'the abilities associated with adequate athletic performance, and encompasses components such as isometric strength, power, speed-agility, balance and arm-eye co-ordination'.[24] Bouchard & Shephard[15] have presented a comprehensive definition of health-related physical fitness, composed of five major components which has been used in several studies.[25-27] The components defined are: (1) morphological components (e.g., body composition, flexibility), (2) muscular components (e.g., power or explosive strength, muscular endurance), (3) motor components (e.g., agility, coordination), (4) cardiorespiratory components (e.g., endurance, submaximal exercise capacity, cardiac function) and (5) metabolic components (e.g., glucose tolerance, lipid metabolism). Exercise training programs in children with CMCs have shown to be effective in improving different components of physical fitness.[6,28,29] Exercise is a subset of physical activity that is planned, structured, and repetitive and has the final or an intermediate objective to improve or maintain one or more components of physical fitness.[16]

FITKIDS INTERVENTION

Fitkids is an exercise therapy program implemented in nearly 165 physical therapy practices in the Netherlands.[30] The Fitkids intervention consist of exercise training and physical activity counseling for children and adolescents with CMCs between 6-18 yr. The Fitkids intervention focuses on improving physical fitness, HRQoL and participation in physical activity. Outcome measures of the intervention are defined according to the body functions and structures and activities levels of the International Classification of Functioning, Disability, and Health (ICF).[31] At the body functions level, aerobic fitness, anaerobic fitness and muscle strength are included as outcome measures, whereas at the level of activity, walking capacity is included and at the level of participation HRQoL and participation in physical activity are included. The main components of physical activity counseling incorporated in the Fitkids intervention are: (1) increasing awareness of parents and children on the benefits of regular physical activity for children with CMCs, (2) providing information on the possibilities of increasing physical activity in daily life and (3) finding a suitable sport to participate in after the Fitkids program.

FITT factors

The basic framework for developing exercise training programs is the FITT-principle. FITT stands for frequency, intensity, time and type of exercise.[32] In preparing the FITT factors of the Fitkids intervention, it was of priority that the exercise recommendations for aerobic exercise for youth with CMCs were taken into consideration[32-34] as well as the busy schedules of families with children with CMCs[35] and, the applicability of the intervention within primary pediatric physical therapy practice. For example, a qualitative research study indicated that parents of children with CMCs prefer exercise training on a weekly basis because of the number of commitments and demands they already have in everyday life.[36] In contrast, a training frequency of at least twice a week is recommended to improve aerobic fitness in youth with chronic conditions.[33]

Concerning the FITT factors of the Fitkids intervention, the following selections were made. Children train 2 times a week within a 3-month period followed by a reduced frequency of once a week in month 4-6. The training sessions, supervised by an experienced physical therapist, are divided in 40 minutes of aerobic training using indoor exercise equipment (i.e., treadmill, stationary bike, rowing machine, cross-trainer), and 10 minutes of both warming up and cooling down. Indoor exercise equipment stimulates the movements of aerobic exercises such as walking, jogging and rowing. These activities are, in accordance with the recommendations, rhythmic in nature and include large muscle groups. The warm-up and cool down are based on unstructured anaerobic physical activity. To fit within the disparate settings of physical therapy practices, physical therapists are free to choose any indoor recreational activity during this part of the training, for instance simple competitive games, ball games, or circuit training. During the warm-up and cool down socialization and having fun are key factors of the Fitkids training. The Fitkids intervention does not include extensive heart rate monitoring, as it is also not included in daily life activities. However, during the exercise training sessions, moderate exercise intensity is monitored using subjective signs such as an increased breathing frequency, increased sweating and facial flushing.

Through the use of indoor exercise equipment, the Fitkids intervention discriminates itself from other training programs based on more functional activities.[28,37] Main reason for choosing this configuration is that group activities are offered alternately with individual activities making the program easily accessible for the heterogenous group of patients visiting the pediatric physical therapy practice. Fitkids also discriminates itself by including a heterogenous population were many other programs focus on homogenous groups such as pediatric obesity[38] or children with CP.[37]

It was hypothesized that the exercise training program Fitkids would improve physical fitness and HRQoL in children and adolescents with CMCs. To test this hypothesis, we evaluated the program under controlled conditions and determined the effects on aerobic fitness, anaerobic fitness, muscle strength, walking capacity and HRQoL. This study is outlined in **chapter 3.** Prior to this study, we first described the population of children and adolescents participating in Fitkids with respect to demographics, medical diagnosis and initial fitness levels. This study, which is outline in **chapter 2**, also aimed to specify the exercise tests used in pediatric physical therapy practices to determine the initial aerobic fitness of children participating in Fitkids. The results of this study have been used to further refine the Fitkids intervention and to prepare the intervention for effectiveness testing.

MEASURING PHYSICAL FITNESS

The assessment of physical fitness is recommended prior to initiation of any exercise training to establish a baseline measure for aerobic capacity, but also to provide individuals with safe training recommendations.[39] Traditionally, exercise testing has focus on the cardiopulmonary system by measuring aerobic exercise capacity,[40] which has become synonymous with physical fitness or aerobic fitness in recent years. A cardiopulmonary exercise test (CPET) can be used to accurately determine peak oxygen uptake (VO_{2peak}), which is considered to be the gold standard for physical fitness by the World Health Organization.[41] CPET consists in applying a gradually increasing intensity exercise until exhaustion or until the appearance of limiting symptoms or signs. During CPET oxygen uptake (VO_2), carbon dioxide exhalation (VCO_2) and minute ventilation (VE) are measured. The CPET provides joint data analysis that allows complete assessment of the cardiovascular, respiratory, muscular and metabolic systems during exertion.[42] Comprehensive assessment of aerobic fitness in children, especially those with a CMC, is an important part of pediatric physical therapy practice. Because of the absence of special equipment needed for respiratory gas analysis in most pediatric physical therapy practices, there is a growing interest in methods in which physical fitness is estimated using predicting equations from functional outcomes during exercise tests.[43] These tests do not provide diagnostic or prognostic information, but offer an indication concerning a child's physical fitness. For many years, the Bruce treadmill protocol has been the most frequently used protocol in children and adolescents using a treadmill for cardiopulmonary exercise testing.[44] This test was also frequently used in Fitkids practices (i.e., physical therapy practices affiliated with the Fitkids Foundation). While this protocol has good validity and reproducibility and pediatric reference values have been published,[45-47] the use in local physical therapy practices can be difficult as the test requires a treadmill ergometer that can operate at an incline of 22%. Many physical therapy practices are embedded in health- and sports centers with only standard treadmill ergometers available that can operate at a maximum incline of 15%. This constraint has encouraged our research group to develop a new practical treadmill test (i.e., the Fitkids Treadmill Test (FTT)) that can be used when limited to a treadmill ergometer with a maximal incline of 15%. Before application of a new exercise test in clinical practice, insight in the clinimetric properties of the test is crucial. Therefore, we aimed to investigate the validity and reproducibility of the FTT in a group of healthy children and adolescents. This study is outlined in **chapter 4**. In addition, reference values (i.e., sets of sex- and age-related values obtained in typically developing participants) increase the usefulness of an exercise test as a physical therapist can interpret the measured exercise variable. **Chapter 5** outlines the sex- and age-related reference values for FTT performance in children and adolescents who were healthy, typically developing and 6-18 yr of age.

Figure 2. Children performing the Fitkids Treadmill Test with and without respiratory gas analysis.

Additionally, before a new test becomes well-embedded in clinical practice, its utility should be demonstrated in clinical populations. Clinical utility refers to the usefulness of a test for clinical practice.[48] Moreover, there are several factors affecting the use of a new test in physical therapy practice. Insight in these factors might facilitate improved uptake of a new test in clinical practice. In **chapter 6**, results of the clinical utility of the FTT in different diagnostic groups are presented and factors affecting the use of the FTT in clinical practice are outlined.

MEASURING PHYSICAL ACTIVITY

To determine whether the Fitkids intervention is successful in increasing participation in physical activity in children and adolescents with CMCs, feasible assessment tools to measure physical activity in clinical practice are needed.[49] In current practice, physical activity is often assessed in a subjective (or self-reported) manner, which include tools such as physical activity diaries or recall questionnaires. Subjective measures have practical and cost-saving advantages and are easy to use.[50] However, given the multi-dimensional character of physical activity, self-reports are easily biased as children might have difficulty recalling their past activity behavior.[51] Questionnaire assessment is also greatly complicated in young children by their propensity for repeated brief bouts of vigorous physical activity.[52] The most frequently used alternative for measuring physical activity is an accelerometer. Accelerometers have the ability to capture a variety of metrics such as minutes of activity, intensity of activity and bouts of activity by quantifying acceleration resulting from physical activity-associated bodily motion at a fixed point of the body.[53,54] However, the use of accelerometers in pediatric physical therapy practice is still a challenge and not implemented within regular care yet. Large-scale use of e-health applications like accelerometers strongly depends on whether healthcare users and healthcare providers experience enough added value from the e-health application in comparison to current practice.[55] Therefore, we aimed to explore the value and feasibility of using an accelerometer to monitor free-living physical activity in children with CMCs in pediatric physical therapy in comparison to subjective physical activity questionnaires. This study is outlined in **chapter 7**.

AIMS AND OUTLINE

The main objective of this thesis is to investigate if the Fitkids exercise therapy program can improve physical fitness, walking capacity, HRQoL and participation in physical activity in children and adolescents with CMCs. A secondary objective is to investigate the clinimetric properties of the FTT as well as to describe sex- and age-related reference values for FTT performance in healthy children and adolescents 6-18 yr. The final two chapters of this thesis cover studies evaluating the applicability of new measurement instruments in physical therapist practice. The specific aims of the studies outlined in this thesis are:

PART 1. Fitkids intervention

Chapter 2: (1) to describe the demographics and medical diagnoses of the children participating in Fitkids, (2) to specify the exercise tests used to determine the initial aerobic fitness of children participating in Fitkids and, (3) to determine the initial aerobic fitness of children participating in Fitkids.

Chapter 3: to determine the effect of the Fitkids exercise therapy program on health-related fitness, walking capacity and HRQoL in children and adolescents with chronic conditions or disabilities in a random sample of Fitkids centers throughout the country.

PART 2. Fitkids Treadmill Test

Chapter 4: to investigate the validity and the reproducibility of the new developed FTT in healthy children and adolescents between 6-18 yr.

Chapter 5: to provide normative values for FTT performance in healthy, typically developing, children and adolescents aged 6-18 yr.

PART 3. New measurement tools: applicability in clinical practice

Chapter 6: (1) to evaluate the utility of the FTT in different diagnostic groups and, (2) to explore potential factors affecting the use of the FTT in clinical practice.

Chapter 7: (1) to explore the added value of using an accelerometer to monitor free-living physical activity in children with CMCs in physical therapy practice in comparison to subjective physical activity questionnaires and, (2) to investigate the feasibility of using an accelerometer to monitor free-living physical activity in children with CMCs.

PART 1

Fitkids intervention

CHAPTER 2

Fitkids exercise therapy program in the Netherlands

ABSTRACT

Purpose: To describe the demographics, medical diagnoses, and initial aerobic fitness levels of children participating in Fitkids: an exercise therapy program for children with chronic conditions or disabilities in the Netherlands.

Methods: We reviewed data of children who were in the program on September 2010.

Results: In total, 2482 children from 105 Fitkids centers were included. Results showed the large heterogeneity of the population regarding demographic characteristics and medical diagnoses. Significantly reduced scores on 6-minute walk test and half Bruce treadmill test were observed.

Conclusion: The Fitkids population has great heterogeneity. In addition, a plethora of fitness tests were used, and registration of data in the Fitkids database was suboptimal. Moreover, this study showed the impaired aerobic fitness of children participating in Fitkids. Future research should investigate the effectiveness of the Fitkids program.

Published as

Kotte EM, Winkler AM, Takken T. Fitkids exercise therapy program in the Netherlands. *Pediatr Phys Ther.* 2013; 25(1): 7-13.

INTRODUCTION

Regular physical activity is important for healthy children as well as for children with chronic medical conditions. Physical activity is generally thought to be associated with benefits for psychosocial health, functional ability and health-related quality of life.[56] Moreover, physical activity has been proven to reduce the risk of coronary heart disease[57,58] and some cancers[59] in adults. Conditions associated with physical inactivity include obesity, type 2 diabetes, and hypertension.[60-63] Regular physical activity increases exercise capacity and physical fitness.[16] *Physical fitness* is defined as a set of attributes that people possess or achieve to perform physical activity.[16] Impaired physical fitness has been reported in children and adolescents with all kinds of chronic health conditions.[7,64-67]

Although children with a chronic health condition or disability are restricted in sports participation because of their impaired physical functioning, reduced physical fitness seems also related to hypoactivity in daily life.[64,68] Because of a sedentary lifestyle, which is defined as an unusual level of physical activity that is less than 60 minutes of moderate-intensity activity per day,[69] the majority of children with chronic health conditions or disabilities are not meeting public-health guidelines of healthy physical activity levels. The sedentary lifestyle could be a result from the tendency of parents to be overprotective, anxiety, or perceived restrictions in sports participation. Or alternatively, the sedentary lifestyle could be a result of the limited access to nearby facilities and programs for these children, whether for leisure, recreation, or competition.[70,71] Besides the potential negative health effects, children with a chronic condition or disability who are inactive do not benefit from the controlling or slowing effect of physical activity on the progression of the chronic disease. Moreover, these children are at risk to become socially isolated from peers who are healthy because of their poor physical fitness.[72]

On the basis of these experiences, the Dutch foundation 'Artsen voor Kinderen' (Physicians for Children) in 2003 developed an exercise therapy program (Fitkids) for children with a chronic condition or disability in the Netherlands. In 2006, Fitkids was housed in a distinct foundation, the Fitkids foundation, which has the responsibility to ensure quality of the Fitkids program and to expand the program in the Netherlands. Fitkids can be regarded as a facilitator for an active lifestyle. The program focuses on improving physical health and, as a result, on optimizing health-related quality of life. The main goal is to bring children to a higher level of durable activity, which allows them to join peers who are healthy in physical activity at a regular sports club. In Fitkids, children from the age of 6 to 18 years with all kinds of disabilities can participate, for example, children with asthma, diabetes, congenital heart defect, rheumatic disease, autism, cystic fibrosis, cerebral palsy, developmental coordination disorder, and Down syndrome. Because of this diversity in children with different chronic diseases and/or disabilities within the program, the program is under supervision of experienced pediatric physical therapists. All therapists receive an intervention protocol and personal instruction to guarantee uniformity of the Fitkids program between the therapists and Fitkids centers. Within the program, children train in small, heterogeneous groups based on age, gender and chronic condition or disability. Fitkids includes a warm-up and cool down based on simple competitive and/or strategic games. Furthermore, the program is set up to improve cardiovascular fitness. For this, treadmills, cross trainers, stationary exercise bikes, rowing machines and other indoor exercise equipment is used, which discriminates Fitkids from other pediatric exercise programs based on more functional exercises. Except for the warm-up and cool down, the training is performed individually. The physical therapy program consists of 2 to 4 phases each lasting 3 months in which the intensity of support is reduced. Since the start in 2003, 150 Fitkids centers have currently been opened, which makes Fitkids a nationwide program in the Netherlands, and which enables children to train in a pediatric physical therapy practice near their home. Studies describing the effectiveness of such a large nationwide program have not been performed yet. However, it is necessary to establish the effectiveness of the Fitkids program. The current study is the first step in this process toward improved evidence-based practice. In the current study, we want to describe the population of children currently participating in Fitkids. The specific aims of the current study were to: (1) describe the demographics and medical diagnoses of the children participating in Fitkids; (2) specify the exercise tests used to determine the initial aerobic fitness of children participating in Fitkids; and (3) determine the initial aerobic fitness of children participating in Fitkids.

METHODS

Study population

More than 6000 children had participated in the Fitkids program from May 2003 to September 2010. For this study, we reviewed data of children who were in the Fitkids program on September 2010. The outcomes that were analyzed for the study were obtained by pediatric physical therapists during the initial assessment of the participants. The data were entered and stored in a Web-based patient record, the Fitkids database, located at http://www.fitkids.nl. Outcomes included demographic characteristics of the participants (e.g., gender, age, height, weight, body mass index [BMI]), medical diagnoses, type of aerobic exercise test used to test the child's initial aerobic fitness, and initial test results. The analysis was performed on anonymous patient care data. Therefore, there was no medical ethics approval required for this study according to the Dutch law.

Demographic characteristics

Body mass and height of the children were determined with an electronic scale and a height-measuring device (wall-mounted measuring stick). Body mass index (kg/m^2) was derived from weight and height. Standard deviation scores (SDSs) were calculated from Dutch growth charts.[73,74] Overweight and obesity were defined as +1 or more and +2 or more SD BMI for age, respectively.

2

Medical diagnoses

Data concerning medical diagnosis of the children were based on the information provided by the treating physician in the medical referral letter.

Aerobic fitness

A variety of validated exercise tests were used to determine the initial aerobic fitness of the children participating in Fitkids. To some extent, the pediatric physical therapists were free to choose an aerobic exercise test which was most optimal to use in their center on the basis of the equipment available and patient characteristics. However, to guarantee uniformity between Fitkids centers, 3 aerobic exercise tests were recommended by the Fitkids foundation (e.g., the 6-minute walk test [SMWT], the half Bruce treadmill test, and the shuttle-run test [STR]). Test conditions, which are described later, were outlined in a Fitkids test manual, which was provided by the foundation. Training days were organized to educate the therapists in the administration of the recommended exercise tests. Furthermore, at set dates, centers were audited whether they followed the Fitkids test manual.

Six-Minute Walk Test

The SMWT was conducted according to the American Thoracic Society guidelines.[75] Subjects were instructed to walk from one end to the other of the walking path at their own pace, while attempting to cover the largest possible distance in 6 minutes. Physical therapists encourage subjects with the standardized statements 'You're doing well' or 'Keep up the good work'. The 6-minute walking distance (SMWD) was recorded as performance outcome measure. Percentage of predicted SMWD was calculated using an age- and height-based equation fitted to normative data by Geiger and colleagues.[76]

Equation boys: 196.72 + (39.81 x age [years]) - (1.36 x (age [years]^2) + (132.28 x height [m])
Equation girls: 188.61 + (51.5 x age [years]) - (1.86 x age [years]^2) + (86.1 x height [m])

Half Bruce Treadmill Test

The half Bruce treadmill test was used because it has smaller increments in workload in comparison with the original protocol.[77] The half Bruce treadmill test has eleven 1.5-minute stages. The first stage starts at a speed of 2.7 km/h and a gradient of 10%. Except for the last 2 stages, which have an increment of 0.4 km/h, each subsequent stage has an increment of 0.6 to 0.7 km/h. All subsequent stages have an increment of 1% in gradient. Children were urged to continue to the point of severe fatigue. The test was finished when a child refused to continue the test despite verbal encouragements. Maximal endurance time was recorded as the outcome measure. As reference values for the half Bruce Treadmill Test are lacking, those for the original Bruce protocol were used. According to Binkhorst et al,[45] there is no significant difference between maximal endurance times between the 2 test protocols. Reference values published by van der Cammen et al[47] were used for children aged up to 10 years. For the remaining children, the reference values of Binkhorst et al[45] were used.

Shuttle-Run Test (10- or 20 m)

The SRT requires children to walk or run between 2 markers delineating the respective course of 10 or 20m.[78,79] Speed of movement was incremented and determined by an audio signal, which was played through a standard CD player. The endpoint of the 10-m SRT was reached when, on 2 consecutive paced signals, the children were more than 1.5 m away from the marker. The end point of the 20-m SRT was reached when, on 2 consecutive paced signals, the children were more than 3-m away from the marker. Level and number of shuttles reached before a child was unable to keep up with the audio signal was recorded as the performance outcome measure. As reference values of the 10-m SRT for healthy children are lacking[80] and recent reference values of the 20-m SRT are only known for the 13- to18-year-old adolescents,[81] test outcomes of both the 10- and 20-m SRT were not compared with reference standards.

Data analysis

Statistical analyses were performed using the Statistical Package for the Social Sciences (SPSS-18.0, SPSS Inc, Chicago, IL). The distribution of the variables was checked with the Kolmogorov-Smirnov test. Because of the skewed nature of the age, weight, length and BMI distributions, medians and interquartile ranges were presented for the total population as well as for boys and girls separately. The nonparametric Mann-Whitney *U* test was used to test for differences in demographic characteristics between boys and girls. For initial test results, normality assumptions were only met for the 10- and 20-m SRT. The Wilcoxon signed rank test was used to test for significant differences between SMWD and distances predicted by the equations of Geiger et al.[76] Differences between maximal endurance times of the half Bruce treadmill test and reference values were analyzed using the SDS of the endurance time, that is, the difference between the observed and predicted value divided by the SD from the reference values. Statistical significance of the SDS endurance time was analyzed using a signed rank test. Significance level for all tests was set at $\alpha < .05$.

RESULTS

Study population

Demographic characteristics

A total of 2482 children were included in the analysis (1311 boys/1171 girls), from 105 Fitkids centers. The demographic characteristics and SDSs are presented in Table 1 for the total population as well as for boys and girls separately. Because of the skewed nature of the variables' distribution, medians and IQRs are given. Because of incomplete datasets, each variable is based on a different number of children. The median age of the children was 10.8 years (interquartile range 9.0-12.9). Median weight for height, height for age, weight for age, and BMI for age SDSs were respectively 2.1 (interquartile range 1.1-2.8), 0.1 (interquartile range -0.8-0.9), 1.7 (interquartile range 0.6-2.6) and 2.1 (interquartile range 1.0-2.6). Significant differences were found in age, weight, and BMI between boys and girls (Table 1).

Table 1. Demographic characteristics of participants.

	Boys (n=1311)			**Girls** (n=1171)				**Total**	
	n	Median	IQR	n	Median	IQR	*P*[a]	Median	IQR
Age (y)	968	10.7	8.9-12.5	882	11.0	9.2-13.2	.002	10.8	9.0-12.9
Weight (kg)	895	49.0	39.0-63.5	823	53.0	41.9-68.0	.000	51.0	40.0-65.2
Height (cm)	899	148.0	137.0-159.0	813	150.0	139.0-160.8	NS	149.0	138.0-160.0
BMI (kg/m^2)	887	23.0	18.9-26.8	810	24.3	20.7-28.2	.000	23.7	19.8-27.5
Weight for height (SDS)	884	2.2	1.0-2.9	809	2.1	1.3-2.7	NS	2.1	1.1-2.8
Height for age (SDS)	772	0.1	-0.8-1.0	700	0.1	-0.8-0.8	NS	0.1	-0.8-0.9
Weight for age (SDS)	768	1.7	0.4-2.7	703	1.7	0.7-2.5	NS	1.7	0.6-2.6
BMI for age (SDS)	763	2.1	0.9-2.7	696	2.1	1.2-2.6	NS	2.1	1.0-2.6

[a] Based on nonparametric Mann-Whitney *U* test.
Abbreviations: BMI, body mass index; IQR: interquartile range; NS: not significant; SDS, SD score.

Medical diagnoses

In total, the study sample showed 67 different primary diagnoses. The 20 most common primary diagnoses of the children participating in Fitkids are presented in Table 2. Twenty-eight percent of the children had a primary medical diagnosis of obesity, whereas 18% were diagnosed with motor retardation and 10% with autism or autism-related disorder. Each of the remaining disorders/diseases (e.g., asthma, orthopedic disorders, neurological disorders, intellectual disability, Down syndrome, cerebral palsy and attention deficit hyperactivity disorder) are reflected in less than 10% of the children.

Used exercise tests

As recommended in the Fitkids test manual, the SMWT (n=1233), the half Bruce treadmill test (n=306), the 10-m SRT (n=82) and the 20-m SRT (n=16) are often used in the initial assessment of Fitkids. Other exercise tests applied during the initial assessment, which were not outlined in the Fitkids test manual, include the Physical Working Capacity (PWC) 170 (n=62), the shuttle-walk test (SWT) (n=14), the Cooper test (n=11) or half-Cooper test (6 minutes; n=7) and a number of tests from the EUROFIT test manual[82]: 10 x 5 m sprint test (n=10), sit and reach test (n=12), standing long jump (n=14), sit- ups (n=5) and the handgrip strength test (n=2). Result of the five tests that were most commonly used are outlined later, that is, the SMWT, the half Bruce treadmill test, the 10- and 20-m SRT and the PWC170. No exercise test results were recorded for 681 children.

Table 2. Medical Diagnoses (primary).

	Boys (n= 1311)	**Girls** (n=1171)	**Total**
Obesity	303 (23.1)	403 (34.4)	706 (28.4)
Motor retardation	267 (20.4)	186 (15.9)	453 (18.3)
Autism spectrum disorders	192 (14.6)	53 (4.5)	245 (9.9)
Asthma	91 (6.9)	95 (8.1)	186 (7.5)
Orthopedic disorders	53 (4.0)	55 (4.7)	108 (4.4)
Neurological disorders	40 (3.1)	38 (3.2)	78 (3.1)
Intellectual disability	26 (2.0)	39 (3.3)	65 (2.6)
Down Syndrome	26 (2.0)	37 (3.2)	63 (2.5)
Cerebral Palsy	34 (2.6)	23 (2.0)	57 (2.3)
Attention Deficit Hyperactivity Disorder	36 (2.7)	10 (0.9)	46 (1.9)
Visual impairment	19 (1.4)	18 (1.5)	37 (1.5)
Diabetes	13 (1.0)	21 (1.8)	34 (1.4)
(Congenital) heart defects	20 (1.5)	11 (0.9)	31 (1.2)
Hypermobility syndrome	18 (1.4)	11 (0.9)	29 (1.2)
Developmental coordination disorder	20 (1.5)	8 (0.7)	28 (1.1)
Spina bifida	10 (0.8)	10 (0.9)	20 (0.8)
Neuromuscular diseases	18 (1.4)	4 (0.3)	20 (0.8)
Juvenile arthritis	5 (0.4)	13 (1.1)	18 (0.7)
Chronic fatigue syndrome	5 (0.4)	13 (1.1)	18 (0.7)
Cancer	10 (0.8)	7 (0.6)	17 (0.7)

Values given as number (% of total)

Aerobic fitness

Six-Minute Walk Test

Initial test results of the SMWT are presented in Table 3. In total, 654 boys and 579 girls performed the SMWT during the Fitkids initial assessment. However, because of incomplete data, the percentage of predicted SMWD could only be calculated for 527 boys and 451 girls and only these children were included in the analyses. Median walking distance was 510.0 meter for boys as well as for girls. Compared with reference values from Geiger et al., the walking distance was significantly reduced for both boys and girls. The median predicted distances for boys and girls were 78.7%; $P < .0001$ and 80.3%; $P < .0001$, respectively (Table 3).

Half Bruce Treadmill Test

Initial test results of the half Bruce treadmill test are presented in Table 4. In total, 161 boys and 145 girls performed the half Bruce treadmill test during the Fitkids initial assessment. However, because of incomplete data, the SDS of the endurance time could only be calculated for 154 boys and 140 girls and only these children were included in the analyses. Median maximal endurance time was 9.5 minutes (i.e., boys 10.0 minutes and girls 9.3 minutes). Both in boys and girls, the maximal endurance time was significantly below the norm (median SDS endurance time, -1.7; $P < .0001$ and -1.6; $P < .0001$, respectively).

Table 3. Initial test results of the 6-minute walk test.

6-Minute Walk Test	**Boys** (n=527)		**Girls** (n=451)		**Total**	
	Median	IQR	Median	IQR	Median	IQR
6-m walking distance (m)	510	451-580	510	441-576	510	449-578
Predicted distance (%)	78.7	69.4-88.4[a]	80.3	68.7-89.4[a]	79.3	69.0-88.7

[a] $P < .0001$ for differences between walking distances and norm values.

Table 4. Initial test results of the half Bruce treadmill test.

Half Bruce Treadmill Test	**Boys** (n=154)		**Girls** (n=140)		**Total**	
	Median	IQR	Median	IQR	Median	IQR
Maximal endurance time (min)	10.0	8.3-12.0	9.3	8.0-10.3	9.5	8.1-10.9
SD score maximal endurance time	-1.7	-2.9 to -0.8[a]	-1.6	-2.5 to -0.8[a]	-1.7	-2.7 to -0.8

[a] $P < .0001$ for differences between maximal endurance times and norm values.

Shuttle-Run Test (10 or 20-m)

Initial test results of the 10-m SRT are presented in Table 5. In total 40 boys and 42 girls performed the 10-m SRT during the Fitkids initial assessment. Mean level achieved was 8.7 (i.e., boys level, 9.3 and girls level, 8.2). Initial test results of the 20-m SRT are presented in Table 6. In total, 12 boys and 4 girls performed the 20-m SRT during the Fitkids initial assessment. Mean level achieved was 3.2 (i.e., boys level, 2.9 and girls level, 4.1).

Physical Working Capacity 170

Initial test results of the PWC170 are presented in Table 7. In total 27 boys and 35 girls performed the PWC170 during the initial assessment. Median PWC achieved at 170 beats per minute was 111 W (i.e., boys 125 W and girls 105 W). Because of lack of recent reference standard values for the PWC170, we did not compare the scores with reference standards.

Table 5. Initial test results of the 10-m SRT.

10-m Shuttle Run Test	**Boys** (n=40)		**Girls** (n=42)		**Total**	
	Mean ± SD	Range	Mean ± SD	Range	Mean ± SD	Range
Level	9.3 ± 3.2	1.0-15.5	8.2 ± 3.1	1.5-15.5	8.7 ± 3.2	1.0-15.5

Table 6. Initial test results of the 20-m SRT.

20-m Shuttle Run Test	**Boys** (n=12)		**Girls** (n=4)		**Total**	
	Mean ± SD	Range	Mean ± SD	Range	Mean ± SD	Range
Level	2.9 ± 2.2	1.0-7.5	4.1 ± 2.6	0.5-6.0	3.2 ± 2.3	0.5-7.5

Table 7. Initial test results of the Physical Working Capacity 170.

Physical Working Capacity 170	**Boys** (n=27)		**Girls** (n=35)		**Total**	
	Median	IQR	Median	IQR	Median	IQR
Watt	125	100-175	105	90-150	111	100-150

DISCUSSION

The purpose of this study was three-fold. First, to describe the demographics and medical diagnoses of children participating in Fitkids. Second, to determine the exercise tests used to determine the initial aerobic fitness of children participating in Fitkids. And, third, to determine the initial aerobic fitness of children participating in Fitkids. Results reveal a large heterogeneity of the Fitkids population with regard to primary medical diagnoses and demographic characteristics. The study sample included children reflecting 67 different primary conditions. The heterogeneity of the sample is also reflected in the relatively large range in weight, height and BMI or age of children participating in Fitkids.

Furthermore, a plethora of aerobic exercise tests were used to test aerobic fitness. Although physical therapists received a Fitkids manual in which the test procedures of the recommended tests were outlined, test procedures were sometimes slightly modified. For example, according to the Fitkids manual, rail holding is not allowed during the Bruce treadmill test. However, some therapists permitted children to hold the railing. Rail holding is known to increase endurance time and reduce physiological strain during sub-maximal exercise.[83] Moreover, according to the Fitkids manual, the recommended length of the walking course of the SMWT test was 15m. However, some Fitkids centers do not have the capacity to use 15 m, so shorter walking courses were used. A shorter walking course requires the performer to reverse directions more frequently, thus reducing the SMWD. Variability in test procedures within Fitkids centers makes it difficult to compare test results obtained in different centers. Better standardization and more training days are needed to educate the therapists in the administration of the recommended exercise tests. Besides, the need to follow test procedures as outlined in the Fitkids test manual should be further emphasized among therapists.

In addition, results of this study reveal that children participating in Fitkids have reduced aerobic fitness compared with peers who are healthy. The SMWD was significantly reduced for both boys and girls (±20% reduction). Test results of the half Bruce treadmill test indicate significantly reduced maximal endurance times for boys as well as for girls (median SDS endurance time, -1.7 and -1.6, respectively).

Because of the lack of recent reference standard values of the PWC170, and the 10-m/20-m SRT for the 6-to-12-year-old children, we were not able to compare initial test outcomes of these tests with a reference population. New reference values for the PWC170 and 10-m/20-m SRT should be established. Moreover, test results of the 20-m SRT show the relative short exercise times of children participating in Fitkids. Mean exercise time was 3.2 minutes, which is too short to reach a maximum cardiopulmonary effort. The optimal duration for a maximal exercise test is 6 to 10 minutes in children and 8 to 12 minutes in adolescents.[84] Therefore, the 20-m SRT is not recommended for use in the Fitkids population.

Results of this study point out a relatively high prevalence of children who are overweight and/or obese within the Fitkids program (median BMI for age SDS=2.1). Of the children with weight and height recorded in the database, 19% (284 children) were overweight (≥+1 SD BMI for age), whereas more than half of these children (784 children) were obese (≥+2 SD BMI for age). As these numbers are based on the children with weight and height recorded in the database rather than on the total number of children in this study, these numbers should be interpreted with some caution. It is likely that pediatric physical therapists did record BMI of children with weight problems more frequently and, therefore, these numbers could indicate an overestimation of the prevalence of overweight and obesity in the Fitkids population. However, Schönbeck and van Buuren[85] reported in 2011 that 13% to 15% of Dutch children in the general population suffered from overweight and obesity, so the overweight and obesity conditions are dramatically increased in the Fitkids population: in a best-case scenario, 32% of total population is obese, whereas more than 50% are obese in a worst-case scenario. The prevalence of overweight and obesity has been increasing in youth at an alarming rate. This is of concern because in this age group overweight has been associated with numerous health risks.[86] It should be questioned whether Fitkids is relevant for obese children because it is difficult to lose weight using exercise therapy.[87] However, the main goal for the children in Fitkids is to improve physical fitness and optimize, if applicable, psychosocial well-being. Moreover, children who are obese and demonstrate significant motor retardation are often excluded from obesity intervention programs. Probably, these children should first participate in Fitkids to improve gross motor function and fitness and, thereafter, they might participate in multidisciplinary obesity programs in which nutritional advice is combined with a more active lifestyle to gain a more healthy weight.

Limitations of this study

Limitations of this study include the problem of missing data. Although 2482 children were included in the database, only 1439 complete data sets including gender, date of birth, primary medical diagnosis, age, height, weight, type of initial test and initial test outcome were available. For 681 (27%) children the initial test outcome was missing, and for 785 children (32%) weight and/or height were missing. With regard to the process towards improved evidence-based practice, it is of high priority to optimize the process of recording data. Currently, pediatric physical therapist record demographic characteristics and test outcomes of patients participating in Fitkids twice: once in their own administration and recording systems and once in the Fitkids Web-based database. The Fitkids foundation could facilitate the recording of Fitkids data, with, for example, a data management system that supports Fitkids therapists with the data entry.

Clinical usefulness

Determining the physical fitness levels of children with chronic conditions is important. Patients included in studies are, in general, a small selected group of subjects with an existing interest in fitness or activity. The current database consists of a large unselected nation-wide group of patients who were participating in Fitkids as a part of their medical care.

CONCLUSIONS AND PERSPECTIVES

This study showed the heterogeneity of the Fitkids population regarding demographic characteristics and chronic condition or disability. In addition, there were a plethora of fitness tests used, and the recording of data in the Fitkids database was suboptimal. Moreover, this study showed the impaired aerobic fitness of children participating in Fitkids. Fitkids aims to improve the physical health of these children as well as health-related quality of life. However, the effectiveness of Fitkids has not been established yet. Future research should investigate the effectiveness of the exercise therapy program Fitkids, taking the population heterogeneity into account.

CHAPTER 3

Effects of the Fitkids exercise therapy program on health-related fitness, walking capacity, and health-related quality of life

ABSTRACT

Background: Children with disabilities have an increased risk for reduced fitness and reduced health-related quality of life (HRQoL). Fitkids, a nationwide exercise therapy program in the Netherlands, was developed to improve fitness and HRQoL in children with disabilities.

Objective: The study objective was to determine the effects of the Fitkids program on health-related fitness, walking capacity and HRQoL in children with chronic conditions or disabilities.

Design: This was a quasi-experimental single-group longitudinal study.

Methods: Fifty-two children and adolescents who were referred to the Fitkids program participated in this study. Participants received a graded exercise training program for 6 months, with frequencies of 1 hour 2 times per week in the first 3 months and 1 hour per week during months 4 to 6. Health-related fitness (aerobic fitness, anaerobic fitness, and muscle strength), walking capacity and HRQoL were evaluated at baseline and after 3 and 6 months of training. Multilevel modeling was used to quantify the contributions of repeated measures, participants, and Fitkids centers to the variations in health-related fitness, walking capacity and HRQoL during the intervention period. The models were adjusted for sex, height, and weight.

Results: After 6 months of training, significant intervention effects were found for aerobic fitness, anaerobic fitness and muscle strength. A significant effect also was found for walking capacity. On the HRQoL measure, significant improvements were found for self-reported and parent-reported physical and emotion domains and for the parent-reported total score of HRQoL.

Limitations: No control group was included in this study.

Conclusions: The Fitkids exercise therapy program has significantly improved health-related fitness, walking capacity and HRQoL in children and adolescents with chronic conditions or disabilities.

Published as

Kotte EM, de Groot JF, Winkler AM, Huijgen BC, Takken T. Effects of the Fitkids exercise therapy program on health-related fitness, walking capacity, and health-related quality of life. *Phys Ther.* 2014 Sep;94(9):1306-18.

INTRODUCTION

In the Netherlands, 14% to 20% of all children and adolescents (0-18 years of age) have long-term or chronic diseases.[88] There is strong evidence that physical fitness is compromised in these young people.[6,37,89-91] From these same studies, it is known that physical activity or exercise training enhances physical fitness in these young people and reduces the risk of secondary impairments. For example, a higher level of physical fitness is associated with a lower level of total adiposity[92] and is inversely associated with cardiovascular risk factors.[8] Therefore, the promotion of physical activity among people with disabilities is of major concern. However, despite many efforts to promote the participation of children with chronic conditions or disabilities in fitness and activity programs, many of these children still have a sedentary lifestyle and are less fit than children without chronic conditions or disabilities. The American Physical Therapy Association's *Guide to Physical Therapy Practice*, second edition, states that a part of physical therapist practice is to 'provide prevention and promote health, wellness, and fitness'.[93(p40)] In the Netherlands, the Fitkids exercise therapy program was developed to address the need for children and adolescents who are 6 to 18 years of age and have chronic conditions or disabilities to become more active and to promote health-related fitness and health-related quality of life (HRQoL) in this population.[89] In keeping with the opinion of the American Physical Therapy Association, the Fitkids program is conducted under supervision of pediatric physical therapists. In the Fitkids program, children and adolescents with different chronic conditions or disabilities train within a single group (heterogeneous grouping) to reduce any stigma associated with their disabilities. Moreover, because 150 Fitkids centers have been opened in the Netherlands, thus limiting the barrier of transport, children are able to train in their own communities. The costs associated with the Fitkids program are largely covered by health insurance, thus limiting the barrier of costs for becoming active. Information on effectiveness of the Fitkids exercise therapy program is currently lacking. The aim of this study was to determine the effects of the Fitkids exercise therapy program on health-related fitness, walking capacity and HRQoL in children and adolescents with chronic conditions or disabilities in a random sample of Fitkids centers throughout the Netherlands.

METHODS

Participants

Study sample

Fifty-two children and adolescents who were referred to the Fitkids program by their treating physician, participated in this study (30 boys and 22 girls). Inclusion was based on willingness to participate. Participants were 6 and 17 years of age at the time of inclusion and were diagnosed with any type of chronic disease or disability. Children and adolescents who depend on a wheelchair or who were not able to read or understand the Dutch language were excluded.

Recruitment

From each province (n=12) in the Netherlands, 1 Fitkids center was selected to participate in the study based on willingness of the therapists to participate and the ability to administer the included exercise tests according to the test manuals. The inclusion of participants started after approval of the Medical Ethics Committee of the University Medical Center Utrecht. The participants and their parents individually received written and verbal information about the study and an informed consent form. Written informed consent was obtained from the parents or legal guardian of each participant and separately from each participant who was 12 years of age or older.

Procedures

Exercise training program

Participants received a graded exercise program (regular Fitkids exercise therapy program) for 6 months. Participants were instructed to train 1 hour 2 times per week in the first 3 months and 1 hour per week during months 4 to 6. Each session consisted of a warm-up period (± 10 min) and cool down period (± 10 min), which were based on simple competitive or strategic games, and a core component (± 40 min), which was performed individually. Treadmills, cross-trainers, stationary exercise bikes, rowing machines, and other types of indoor exercise equipment were used. The exercise sessions were supervised by experienced pediatric physical therapists. More detailed information on the Fitkids program can be obtained from the Fitkids Foundation (e-mail: info@fitkids.nl).

Anthropometrics

Body weight and height were measured with an electronic scale and a wall-mounted centimeter scale, respectively. The body mass index (in kg/m^2) was derived from weight and height and compared with reference values for people who were healthy and matched for age and sex; *z* scores were calculated.[73,74]

Outcome measures

Outcome measures, conducted according to *International Classification of Functioning, Disability, and Health* guidelines,[31] were health-related fitness (defined as aerobic fitness, anaerobic fitness and muscle strength), walking capacity, and HRQoL measured at baseline (t0), after 3 months of training (t1), and after 6 months of training (t2).

Body function

Aerobic fitness was reflected by the endurance time achieved on the modified Bruce protocol (the 'half-Bruce' protocol). The modified protocol was used because it has smaller increments in workload in comparison with the original protocol.[77] The half-Bruce treadmill test has eleven 1.5-minute stages. The first stage starts at a speed of 2.7 km/h and a gradient of 10%. Except for the two latest stages, which have a speed increment of 0.4 km/h, each subsequent stage has an increment of 0.6 to 0.7 km/h. All stages have a gradient increment of 1%. Participants were urged to continue to the point of severe fatigue. The test was finished when a child refuses to do more work despite verbal encouragement. Participants were not allowed to use the handrails. According to Cumming et al.[94],

3

the Bruce test is highly reproducible (r=.94) and valid (r=.88 between maximal endurance time and maximal oxygen uptake).

Anaerobic fitness was measured by use of the mean power (measured in watts) derived from the Muscle Power Sprint Test (MPST).[95] The MPST is a reliable method for determining anaerobic performance in both children who are healthy (the intraclass correlation coefficient for mean power was .98)[95] and children with cerebral palsy.[96,97] Participants were instructed to complete six 15-m runs at a maximum pace. The 15-m distance was marked by 2 lines taped on the floor. Participants were instructed to cross the finish line. Between the runs, the participants were allowed a timed 10-second rest period before preparing for the next sprint. Participants were given the cues 'ready', '3', '2', '1', and 'go' for the first run. For the second through the sixth run the physical therapist counted backward from '10' to '1'and then gave the cue 'go'. The mean power, considered the most important parameter for MPST, was the average power output of the 6 sprints.[95] Power (W) was calculated as force x velocity, where force (kg x m/s^2) = body mass (kg) x acceleration (m/s^2) and velocity (m/s) = distance (m)/time (s).

Muscle strength was assessed by use of the strength subscale of the Bruininks-Oseretsky Test of Motor Proficiency (2nd ed) (BOT-2).[98,99] This subscale contains 5 items, which were administered in a fixed sequence, such as standing long jump (SLJ), knee push-ups, sit-ups, wall sit, and v-up. In addition to the item scores, an overall score on the strength subscale was recorded (total point score). The strength subscale of the BOT-2 seems to be valid and reliable.[99]

Activity

Walking capacity was assessed using the 6-minute walk test (6MWT), conducted according to the American Thoracic Society guidelines.[75] The test-retest reliability of the 6MWT has been reported to be high, with an intraclass correlation coefficient of .84 to .98.[100-102] The minimal important change of the 6MWT was investigated in recent clinimetric studies on adult patients with chronic conditions and was found to be 24 to 45 m.[103,104] In the present study, the 6MWT was performed on a 15-m straight corridor. Participants were instructed to cover the greatest possible distance in 6 minutes at a self-selected walking speed. Physical therapists encouraged participants with the following standardized statements: 'You're doing well' and 'Keep up the good work'. Participants were allowed to stop and rest during the test; however, they were instructed to resume walking as soon as they felt able to do so. The 6-minute walking distance (6MWD) was recorded as the primary outcome measure.

Health-related quality of life (HRQoL)

The 25-question version of the Dutch Children AZL-TNO Questionnaire Quality of Life (DUX-25)[105] was used to assess how the participants and their parents evaluated HRQoL in their daily functioning. The DUX-25 includes 4 functional domains: physical, emotional, family and social. In addition, a total HRQoL score with a high discriminative quality can be obtained, allowing its use in individual patients. The responses are rated on a visual analog scale with faces ranging from smiling to crying. Item scores are converted to a scale from 1 to 100, with higher scores representing a higher HRQoL. The DUX-25 was completed by both the child (Child Form) and a parent (Parent Form). The internal consistency (Cronbach-alpha) of the DUX-25 domains varies from .65 to .84, and that of the total score is 0.94.[106] Example questions from the emotional and physical domains, respectively, are as follows: 'How much do you (does your child) like school?' and 'How do you (does your child) rate his/her physical fitness?'.

The tests were administered on the same day and in a fixed order with sufficient resting time between the tests. The 6MWT was performed before the half-Bruce treadmill test, which was followed by the MPST and the strength subscale of the BOT-2. To ensure the uniformity of the test procedures, we gave the physical therapists written instructions on the application and scoring of the exercise tests before they administered the tests.

Data analysis

We used IBM SPSS Statistics for Windows, version 20.0 (IBM Corp, Armonk, New York) for descriptive statistics. The distribution of the variables was checked with the Kolmogorov-Smirnov test. To explore baseline characteristics of the study population, we compared initial test results with test results for people who were healthy. To investigate longitudinal changes in health-related fitness, walking capacity and HRQoL over time, we applied multilevel modelling with the multilevel program MLwiN 2.23 (Centre for Multilevel Modelling, University of Bristol, Bristol, United Kingdom).[107] An advantage of multilevel modelling over the traditional repeated measurement approach is that all of the available results can be incorporated within the analysis,[107] even if the number of measurements varies between the participants because of missing data, assuming that the missing data are random.[108,109] By means of the multilevel analyses, we were able to determine whether the changes in health-related fitness, walking capacity, and HRQoL differed significantly across participants and Fitkids centers. A 3-level multilevel structure was used; level 1 represented repeated measures within participants (t0, t1, and t2), level 2 represented differences among participants, and level 3 represented differences among Fitkids centers.

Possible predictors for the multilevel model were sex, height, and weight because these variables were considered to influence health-related fitness and walking capacity.[76,110] These predictors were entered into the model to find the best model fit. The model fit was evaluated by comparing the deviance (Ln -2 log likelihood) of the empty model from the final model. Deviance is a measure of how well the model fits the data or how much the actual data deviate from the predictions of the model. The larger the deviance, the poorer the fit to the data. The empty model is the null model (i.e., lacking predictors), and as predictors are added to the model, the deviance changes. A simpler model can be rejected with a decrease in deviance and a

3

P value of less than .05. An alpha level of .05 was adopted for all tests of significance.

RESULTS

Fifty-one participants completed the exercise training program. One child dropped-out after 3 months of training because of an injury. The average adherence was 81.7% of offered training sessions. The demographic characteristics of the study population are shown in Table 1, and baseline characteristics Table 2.

Table 1. Demographic characteristics of participants.[a]

Characteristic	Mean	SD	Range	No of participants
Weight (kg)	50.1	17.7	20.2 - 105.0	
Height (cm)	149.0	14.7	112.0 - 175.0	
BMI (kg/m2)	22.0	4.9	13.9 - 35.8	
Weight for age (SD score)	1.2	1.4	-2.5 - 3.6	
Height for age (SD score)	0.2	1.2	-4.2 - 3.3	
BMI for age (SD score)	1.4	1.3	-2.3 - 3.3	
Primary medical diagnosis				
ADHD				2
Asthma				10
Auditory disorder				1
Autism spectrum disorders				4
Chronic fatigue syndrome				1
Cerebral palsy				1
DCD				1
Diabetes Mellitus type 2				1
Gastrointestinal disorders				1
Ehlers-Danlos syndrome				1
Hypermobility syndrome				3
Intellectual disability				2
Mitochondrial disorder				1
Motor retardation				9
Neurofibromatosis type 1				2
Neurological diseases[b]				3
Orthopedic disorders[c]				7
Sickle cell anemia				1
Turner syndrome				1

[a] For age (in years), the median was 10.6, and the interquartile range was 8.5 to 12.9. BMI = body mass index, SD = Standard Deviation, ADHD = Attention Deficit Hyperactivity Disorder; DCD = Developmental Coordination Disorder.
[b] Microcephaly, Prader-Willi syndrome, Erb palsy.
[c] Osteochondritis dissecans, progressive scoliosis (n=3), multidigit camptodactyly, club foot, Perthes disease.

Table 2. Baseline characteristic of the participants.[a]

Characteristics	Measure	Mean	SD	Range		
Fitness level						
Aerobic capacity	Half-Bruce ET (min)	6.8	1.8	3.7	to	12.0
	SDS[b]	-3.9	1.3	-7.1	to	-0.8
Anaerobic capacity	MPST-MP (Watt)	154.1	93.0	22.5	to	428.7
	SDS[c]	-1.5	1.0	-3.2	to	1.5
Walking capacity	6MWD (meters)	535.2	78.6	345.0	to	690.0
	Pred. distance (%)[d]	83.2	11.9	57.0	to	115.0
Muscle Strength	AE BOT-2 (years)	8.1	3.0	4.0	to	17.2
Health-related quality of life						
Child form						
Physical domain	Mean (%)	72.1	18.7	12.5	to	100.0
	SDS[e]	-0.2	1.1	-3.6	to	1.4
Emotional domain	Mean (%)	73.6	18.5	25.0	to	100.0
	SDS[e]	0.0	1.2	-3.1	to	1.7
Family domain	Mean (%)	86.8	10.5	60.0	to	100.0
	SDS[e]	0.2	0.7	-1.6	to	1.1
Social domain	Mean (%)	78.1	13.5	17.9	to	100.0
	SDS[e]	0.1	1.0	-4.5	to	1.7
Total score	Mean (%)	77.3	13.3	34.0	to	99.0
	SDS[e]	-0.0	1.0	-3.4	to	1.7
Parent form						
Physical domain	Mean (%)	63.8	17.9	33.3	to	100.0
	SDS[e]	-0.5	1.0	-2.2	to	1.5
Emotional domain	Mean (%)	70.8	17.7	28.6	to	100.0
	SDS[e]	-0.2	1.1	-2.9	to	1.7
Family domain	Mean (%)	80.6	13.7	50.0	to	100.0
	SDS[e]	0.1	0.9	-1.9	to	1.3
Social domain	Mean (%)	75.9	14.6	14.3	to	100.0
	SDS[e]	0.2	1.1	-4.5	to	2.0
Total score	Mean (%)	72.5	13.8	36.0	to	100.0
	SDS[e]	-0.2	1.0	-2.9	to	1.8

[a] ET = endurance time, SDS = standard deviation score: difference between the observed value and the predicted value divided by the standard deviation from the reference values, MPST = Mean Power Muscle Power Sprint Test, MP = mean power, 6MWD = 6-minute walking distance, AE = age equivalent: typical age in a normative group with a similar score, BOT-2 = Bruininks-Oseretsky Test of Motor Proficiency.
[b] Based on reference values from van der Cammen-van Zijp et al[47] (≤ 10 years) and Binkhorst et al[45] (> 10 years).
[c] Based on reference values from Douma-van Riet et al.[95]
[d] Based on reference vales from Geiger et al.[76]
[e] Based on unpublished data (HM Koopman, PHD; 2007).

On average, 8% of all measurements were missing in the present study. The reasons for missing data were incomplete or missing test forms (41.9%), injuries (36.4%), vacation (10.6%) and incorrect measurements (11.1%) (i.e., the time needed to complete runs on the MPST was recorded to the tenth of a second rather than to the hundredth of a second). We assumed that these missing data were

3

random and, therefore, would not have an effect on the results.

The scores for endurance time on the half-Bruce treadmill test, the mean power of the MPST, the total point score of the BOT-2, and the 6MWD are depicted in the Figure 1. Comparisons of the measurements at baseline, 3 months of training, and 6 months of training indicated that participants showed improvements in their aerobic fitness, anaerobic fitness, muscle strength, and walking capacity over time.

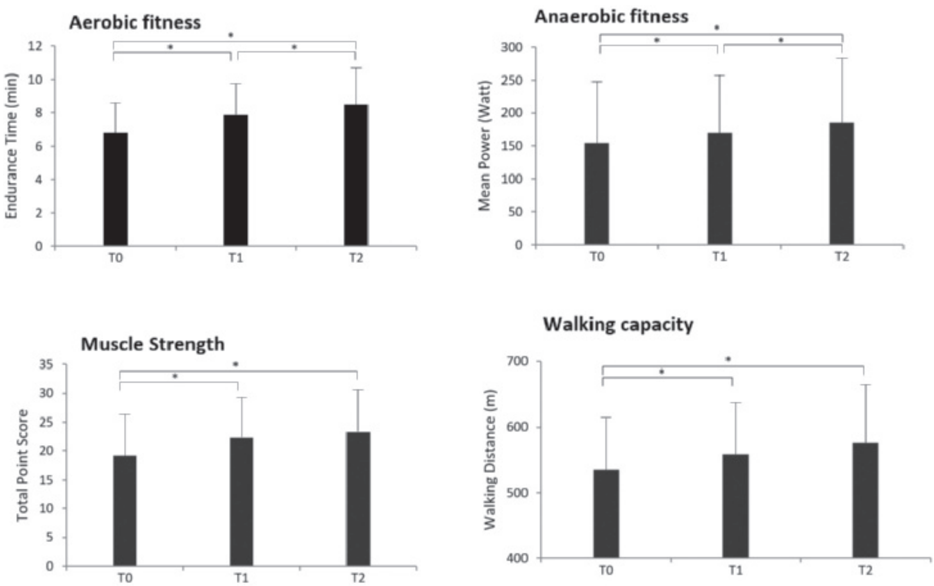

Figure 1. Bar plots on the changes in baseline health-related fitness and walking capacity after 3 and 6 months of training. The bars show the mean test outcome and standard deviation at baseline (t0) and after 3 months of training (t1) and 6 months (t2) of training. **P*<.05.

Multilevel modeling

Body function

The estimated models for the half-Bruce treadmill test and the MPST are shown in Table 3, and the estimated model for the strength subscale of the BOT-2 is shown in Table 4. Because differences among Fitkids centers (level 3) did not significantly contribute to the variation in mean power, this level was excluded from the final model. Thus, multilevel modeling for the MPST resulted in a 2-level model, with timing of the measurement defined as level 1 and participants defined as level 2.

Aerobic fitness

Sex, weight, and height did not significantly influence outcome on the half-Bruce treadmill test and, therefore, were not included in the final model. After 3 and 6 months of training, estimated improvements in endurance time on the half-Bruce treadmill test were 1.05 minutes ($P \leq .001$) and 1.62 minutes ($P \leq .001$), respectively.

Moreover, the predicted endurance time of 8.30 minutes (6.68 minutes + 1.62 minutes) for t2 was significantly higher than for t1 (6.68 minutes + 1.05 minutes = 7.73 minutes) ($p \leq .001$). The level 2 variance of 0.58 indicates the difference among participants, meaning that 58% of the total variance in endurance time was attributable to differences among participants. Furthermore, 21% of the total variance was attributable to differences among Fitkids centers.

Anaerobic fitness

Height significantly influenced mean power on the MPST and, therefore, was included in the final model. Sex and weight did not significantly predict mean power on the MPST and, therefore, were not included in the final model. After 3 and 6 months of training, estimated improvements in mean power were 17.38 W (P=.02) and 35.36 W ($P \leq .001$), respectively. Moreover, the predicted mean power at t2 was significantly higher than at t1 (P=.02). Furthermore, 79% of the total variance in mean power was attributable to differences between participants. The following equations are derived from the final model:

Mean power at t0 = -405.62 + 3.78 x height

Mean Power at t1 = -405.62 + 17.38 + 3.78 x height

Mean Power at t2 = -405.62 + 35.36 + 3.78 x height

Thus, the mean power at baseline (t0) and after 3 months (t1) and 6 months (t2) of training could be predicted with the multilevel model. For example, after 3 months of training in the Fitkids program, the predicted mean power on the MPST for a child with a height of 150 cm would be 178.8 W (-405.62 + 17.38 + 3.78 x 150).

Table 3. Multilevel Model for the Half-Bruce Treadmill Test, MPST and 6MWT.[a]

	Half-Bruce Treadmill Test		**MPST MP**		**6MWT**	
	Fixed effects					
	β	**95% CI**	**β**	**95% CI**	**β**	**95% CI**
Intercept	6.68	5.93 to 7.42[b]	-405.62	-610.24 to -201.01[b]	294.39	87.73 to 501.05[b]
t1	1.05	0.69 to 1.40[b]	17.38	2.46 to 32.30[b]	22.86	8.45 to 37.27[b]
t2	1.62	1.27 to 1.98[b]	35.36	20.67 to 50.05[b]	35.67	21.25 to 50.08[b]
Height	-	-	3.78	2.41 to 5.15	1.62	0.24 to 3.00
Weight	-	-	-	-	-	-
Sex	-	-	-	-	-	-
	Random effects					
	Variance	**95% CI**	**Variance**	**95% CI**	**Variance**	**95% CI**
For level 1	0.21	0.03 to 0.38	0.21	-0.09 to 0.51	0.21	-0.09 to 0.51
For level 2	0.58	-0.27 to 1.43	0.79	-0.87 to 2.45	0.78	-0.87 to 2.45
For level 3	0.21	-0.73 to 1.15	-	-	-	-
	Deviance					
Final model	520.9		1,544.3		1,644.4	
Empty model	641.2		1,689.6		1,763.9	

[a] MPST = Muscle Power Sprint Test, 6MWT = Six-Minute walk Test, MP = mean power, CI = confidence interval, t1 = after 3 months of training, t2 = after 6 months of training.

[b] $P < .05$.

Muscle Strength

For the SLJ, height and weight were included in the final model as independent factors because these factors significantly influenced the model. A positive effect was found for height, whereas as negative effect was found for weight. Moreover, for the sit-ups, the final model included height because height significantly influenced the number of sit ups. After 3 and 6 months of training, estimated improvements in SLJ were 7.90 cm ($p \leq .001$) and 8.52 cm ($p \leq .001$), respectively. After 3 months of training, estimated improvements in the number of knee push-ups, the number of sit-ups, the duration of the wall sit, and the duration of the v-up were 4.58 ($P \leq .001$), 3.45 ($P \leq .001$), 6.96 seconds ($P \leq .001$) and 9.27 seconds ($P \leq .001$), respectively. After 6 months of training, estimated improvements were 4.54 ($P \leq .001$), 3.38 ($P \leq .001$), 8.85 seconds ($P \leq .001$) and 13.27 seconds ($P \leq .001$). No significant differences in muscle strength were found between t1 and t2. Moreover, 74% of the total variance in outcome on SLJ is attributable to differences among participants. For the knee push-ups, sit-ups, wall sit and v-up, the total variance in outcome attributable to differences among participants were 36%, 46%, 56%, 55%, respectively. Furthermore, 3% of the total variance in outcome on SLJ was attributable to differences among Fitkids centers. For the knee push-ups, sit-ups, wall sit and v-up, the total variances in outcome attributable to differences among Fitkids centers were 34%, 29%, 21%, and 19%, respectively.

Activity

The estimated model for the 6MWT is shown in Table 3. Because differences among Fitkids centers (level 3) did not significantly contribute to the variation in 6MWD, this level was excluded from the final model. Thus, multilevel modeling for the 6MWT resulted in a 2-level model, with timing of the measurement defined as level 1 and participants defined as level 2. Height significantly influenced the model and, therefore, was included in the final model. Weight also significantly influenced the model, but the average score became insignificant after weight was added to the model; therefore, weight was not included in the final model. Sex did not significantly predict outcome on the 6MWT, and, therefore, was not included in the final model. After 3 and 6 months of training, estimated improvements in 6MWD were 22.9 m ($P \leq .001$) and 35.7 meters ($P \leq .001$). No significant differences in 6MWD were found between t1 and t2. Moreover, 78% of the total variance in 6MWD was attributable to differences among participants.

HRQoL

No significant intervention effects on self-reported and parent-reported family and social domains and on self-reported total score of HRQoL were found. Therefore, these domains were not included in the multilevel models. The estimated models for the self-reported and parent-reported physical and emotional domains and for the parent-reported total HRQoL score are shown in Table 5. Because differences among Fitkids centers (level 3) did not significantly contribute to the variation in outcome on the functional domains of quality of life, this level was excluded from the final model. Thus, multilevel modeling for the DUX-25 resulted in a 2-level model, with timing of the measurement defined as level 1 and participants defined as level 2. Participants' sex significantly influenced outcome on the self-reported physical domain and, therefore, was incorporated into the final model for this domain (boys=1; girls=0). After 3 months of training, estimated improvements in self-reported physical and emotional HRQoL were 1.32% (P=.48) and 3.90% (P=.05), respectively. After 6 months of training, estimated improvements in self-reported physical and emotional HRQoL were 4.64% (P≤.001) and 4.67% (P=.01), respectively. Estimated improvements in parent-reported physical and emotional HRQoL were 6.93% (P≤.001) and 3.08% (P=.09), respectively, after 3 months of training and 7.25% (P≤.001) and 5.40% (P≤.00), respectively, after 6 months of training. After 3 and 6 months of training, estimated improvements in the parent-reported total HRQoL score were 3.13% (P=.02) and 4.56% (P≤.00), respectively. No significant differences in self-reported and parent-reported HRQoL were found between t1 and t2. Moreover, 80% of the total variance in self-reported physical HRQoL and 72% of the total variance in self-reported emotional HRQoL were attributable to differences among participants. With regard to the Parent Form, 72% of the total variance in reported physical HRQoL and 73% of the total variance in reported emotional HRQoL were attributable to differences among participants. In addition, 79% of the total variance in parent-reported total HRQoL score was attributable to differences among participants.

Table 4. Multilevel Model for the Bruininks-Oseretsky Test of Motor Proficiency.[a]

	Standing Long Jump		Knee Push Ups		Sit Ups		Wall Sit		V-Up	
Fixed effects										
	β	**95% CI**	**β**	**95% CI**	**β**	**95% CI**	**β**	**95% CI**	**β**	**95% CI**
Intercept	-99.98	-191.52 to -8.45[b]	9.62	6.15 to 13.08[b]	-14.30	-34.02 to 5.42[b]	29.11	21.11 to 37.10[b]	28.54	20.64 to 36.43[b]
t1	7.90	2.77 to 13.03[b]	4.58	2.77 to 6.40[b]	3.45	1.83 to 5.06[b]	6.96	2.90 to 11.03[b]	9.27	4.96 to 13.59[b]
t2	8.52	3.39 to 13.65[b]	4.54	2.74 to 6.35[b]	3.38	1.78 to 4.98[b]	8.85	4.82 to 12.89[b]	13.27	8.99 to 17.56[b]
Height	1.77	1.01 to 2.53	-	-	0.20	0.07 to 0.33	-	-	-	-
Weight	-1.23	-1.86 to -0.60	-	-	-	-	-	-	-	-
Sex	-	-	-	-	-	-	-	-	-	-
Random effects										
	Variance	**95% CI**	**Variance**	**95% CI**	**Variance**	**95% CI**	**Variance**	**95% CI**	**Variance**	**95% CI**
For level 1	0.23	0.02 to 0.43	0.30	0.06 to 0.55	0.25	0.05 to 0.45	0.23	0.04 to 0.43	0.26	0.04 to 0.48
For level 2	0.74	-0.42 to 1.89	0.36	-0.24 to 0.96	0.46	-0.24 to 1.16	0.56	-0.28 to 1.39	0.55	-0.30 to 1.39
For level 3	0.03	-0.56 to 0.64	0.34	-0.78 to 1.45	0.29	-0.77 to 1.35	0.21	-0.72 to 1.14	0.19	-0.70 to 1.09
Deviance										
Final model	1,339.0		984.2		978.9		1,269.1		1,281.2	
Empty model	1,489.7		1,079.2		1,107.0		1,376.7		1,386.0	

[a] CI = confidence interval, t1 = after 3 months of training, t2 = after 6 months of training, [b] $P < .05$.

Table 5. Multilevel Model for the DUX-25Questionnaire.[a]

	Child form				Parent form					
	Physical		Emotional		Physical		Emotional		Total	
	Fixed Effects									
	β	95% CI	β	95% CI	β	95% CI	β	95% CI	β	95% CI
Intercept	61.65	54.07 to 69.23[b]	72.71	67.98 to 77.43[b]	63.42	58.67 to 68.18[b]	70.61	66.14 to 75.07[b]	72.51	68.91 to 76.10[b]
t1	1.32	-2.31 to 4.95	3.90	0.08 to 7.72[b]	6.93	3.00 to 10.87[b]	3.08	-0.54 to 6.69	3.13	0.49 to 5.76[b]
t2	4.64	1.18 to 8.10[b]	4.67	0.99 to 8.35[b]	7.25	3.50 to 10.99[b]	5.40	1.99 to 8.81[b]	4.56	2.08 to 7.05[b]
Height	-	-	-	-	-	-	-	-	-	-
Weight	-	-	-	-	-	-	-	-	-	-
Sex	15.79	6.15 to 25.43	-	-	-	-	-	-	-	-
	Random effects									
	Variance	95% CI	Variance	95% CI	Variance	95% CI	Variance	95% CI	Variance	95% CI
For level 1	0.20	-0.09 to 0.50	0.28	-0.12 to 0.68	0.28	-0.13 to 0.69	0.27	-0.12 to 0.65	0.21	-0.10 to 0.53
For level 2	0.80	-0.87 to 2.46	0.72	-0.84 to 2.28	0.72	-0.83 to 2.27	0.73	-0.84 to 2.31	0.79	-0.86 to 2.43
For level 3	-	-	-	-	-	-	-	-	-	-
	Deviance									
Final model	1,078.2		1,073.9		1,091.6		1,062.2		989.0	
Empty model	1,094.6		1,080.5		1,107.9		1,071.5		1,001.7	

[a] DUX-25 = 25-question version of the Dutch Children AZL-TNO Questionnaire Quality of Life, CI = confidence interval, t1 = after 3 months of training, t2 = after 6 months of training, [b] *P*<.05.

3

DISCUSSION

The present study investigated the effects of the Fitkids exercise therapy program on health-related fitness, walking capacity, and HRQoL in children and adolescents with chronic conditions or disabilities in a random sample of Fitkids centers throughout the Netherlands. Significant improvements in aerobic fitness, anaerobic fitness, muscle strength, and walking capacity were found after 3 months of training. In addition, significant improvements in self-reported emotional HRQoL, parent-reported physical HRQoL, and parent-reported total HRQoL score were found after 3 months of training. After 6 months of training, significant improvements in self-reported physical HRQoL and parent-reported emotional HRQoL also were found.

Although there was a trend toward significant further improvements in walking capacity and muscle strength between 3 and 6 months of training, except for the knee push-ups and the sit-ups, significant further improvement during this period were only found for aerobic fitness and anaerobic fitness. A possible reason for the lack of significant improvements in muscle strength, walking capacity, and HRQoL during month 3 to 6 of the intervention period is the training frequency. Earlier research indicated that training for 1 hour per week is sufficient for maintaining the effects of training but is insufficient for producing further improvements in health outcomes.[111] Another possible reason is the law of the 'diminishing returns'; there is a decline in effectiveness of a training program after a certain level of fitness has been achieved.[111]

The results of the present study are in line with those found in diverse homogenous pediatric patient groups. Verschuren at al.[37] found improvements in aerobic and anaerobic fitness in children with cerebral palsy after an 8-month training program with standardized exercises for aerobic and anaerobic fitness (a 25% increase in watts of mean power measured with the MPST); the increase in the present study was 22% (157,6 W at t0; 192,96 W at t2). Nsenga Leunkeu et al.[112] found significant improvements in 6MWD in children with cerebral palsy after 8 weeks of moderate walking exercise 3 times per week. Moreover, significant training effects on the 6MWD (an increase of 4.4%) after a supervised exercise training program in children with cystic fibrosis were found by Gruber et al.[113] In the study by Basaran et al,[114] the 6MWD in children with asthma improved 4% after a moderately intensive basketball training program for 8 weeks. In the present study, a 6.7% increase in the 6MWD was found after 6 months of training (535.7 m at t0; 571.4 m at t2).

Although we found significant improvements in health-related fitness, walking capacity, physical HRQoL, and emotional HRQoL, the population of children and adolescents in the present study was heterogeneous, and studies referring to the measurement properties of the exercise tests used for many pediatric chronic conditions are scarce. Therefore, it is difficult to conclude whether the estimated improvements were clinically meaningful for the individual child.

The multilevel analysis used in the present study allowed us to identify the significant determinants that influence outcome. We chose to adjust the models for sex, weight, and height. From the literature, it is known that the main predictor variables for the 6MWD in adults who are healthy are sex, age and height.[115-117] The results of the present study showed height to be a significant predictor of 6MWD. This relationship can be attributed to the longer lengths of steps in taller people. The length of the step is one of the main determinants in gait speed.[118] Adding age along with height to our model did not result in a more optimal model fit, probably because of the strong association of height and age in our population. A reason for preferring height over age in our study was based on the assumption that children of the same chronological age vary in height on the basis of their maturity status; children who are more mature are taller than children who less mature.

The multilevel model for the MPST showed that height was a significant independent predictor of mean power. This finding is consistent with the findings of Armstrong et al,[119] who suggested that with growth (i.e., the increases in height and weight and other body changes that occur as a child matures), power output increases.

Previous research[120] indicated that jumping performance increased during growth in 11-16-year-old children. In our model height and weight were found to be a significant predictor of SLJ performance. It is likely that body dimensions significantly affect SLJ performance; for instance, taller people may jump farther than shorter people with the same leg muscle power. However, Veligekas[121] concluded that anthropometric variables that were deemed to influence SLJ, such as leg length, weight and body mass index, did not contribute to SLJ performance in 9- to 12-year-old children. Body height contributed to SLJ performance only at a small degree in the study of Veligekas et al.[121]

Participants' sex was revealed to be a significant predictor of the self-reported physical HRQoL. This indicated that the physical HRQoL at baseline and after 3 and 6 months of training differed significantly between boys and girls, with girls reporting a lower physical HRQoL than boys. This finding is in line with cross-sectional studies reporting that the HRQoL of girls in their early teenage years is significantly lower than that of boys, being mainly evident in the physical and psychological aspects of HRQoL.[122-124] These data provide evidence that girls may experiences greater impairments in physical and psychological quality of life than boys. The higher reported physical HRQoL of boys may be due to their physical activities with peers, such as collective games and other leisure activities.[125]

Body height did not influence outcome on the half-Bruce treadmill test in our population. This result was somewhat surprising. We expected that children with short legs would have shorter

3

endurance times than those with longer legs, so we expected to find a negative relationship between body height and endurance time on the half-Bruce treadmill test. However, the heterogeneity of our population may have affected this relationship.

For all test outcomes, a relative high level 2 variance was found, indicating that a high level of the total variance in test outcome was attributable to difference among participants. This result may have been due to the heterogeneous characteristics (e.g., health status, disability, and age) of the included participants. Monitoring of the intervention effects in a larger sample of children and adolescents referred to the Fitkids program is warranted to identify subgroups for whom the exercise therapy program is more or less effective.

Differences among Fitkids centers (level 3) did not contribute to the variation in outcome on the 6MWT and the MPST, indicating that the difference between Fitkids centers did not contribute to the total variance in test outcome. The Fitkids Foundation has organized several training days to educate physical therapists in Fitkids practices (i.e., physical therapist practices affiliated with the Fitkids Foundation) in the administration of various aerobic exercise tests. It appears that these training days have been useful and that therapists are well educated in providing Fitkids training sessions as well as the administration of the exercise tests. The Fitkids Foundation will continue to supervise the centers with regard to whether they are adhering to the prescribed, standardized test manuals and will continue to organize supplementary training days. In addition, new therapists or Fitkids centers should receive thorough instructions about the outcome measures. Differences among Fitkids centers (level 3) contributed to the variation in outcome on the various items of the strength subscale of the BOT-2. A possible explanation is that the physical therapists were less familiar with the administration of this test, leading to a less standardized administration of the test and more variation in outcome.

The strength of the present study was that we included an unbiased clinical sample of children and adolescents who were referred to the Fitkids exercise therapy program. This sample was a reflection of the children and adolescents participating in the Fitkids program. It is known from the literature that, in general, the effects of interventions are overestimated in randomized clinical trials. An additional strength of the present study was the use of statistical multilevel methods, in which various measurements were allowed per participant. The absence of a control (waiting list) group may be considered a limitation of the present study because it is unknown what happens to children's health if no exercise training is given. In addition, the absence of a follow-up period may be considered a limitation of the present study. We are interested in gaining insight into the long-term effects of the Fitkids program on health-related fitness, walking capacity, and HRQoL as well as on activity and exercise participation

Practical implications

Through incorporating of the Fitkids exercise therapy program into pediatric physical therapy practice, physical therapists are able to help children with a chronic condition or disability to overcome barriers to fitness and sports and to improve their physical and mental health. These practices reflect the important role, recognized by the American Physical Therapy Association, that pediatric physical therapists should play in the promotion of health, wellness, and fitness among children.

In conclusion, the Fitkids exercise therapy program significantly improved health-related fitness, walking capacity, and HRQoL in children and adolescents with chronic conditions or disabilities. Future studies should investigate the long-term effects of the Fitkids exercise therapy program.

ACKNOWLEDGEMENTS

This research was financed by the Johan Cruyff Foundation, the Rabobank Foundation, and the Dutch Foundation Innovation Alliance SIA RAAK, project number 2011-13-035P.

PART 2

Fitkids Treadmill Test

CHAPTER 4

Validity and reproducibility of a new treadmill protocol: the Fitkids Treadmill Test

ABSTRACT

Purpose: This study aimed to investigate the validity and reproducibility of a new treadmill protocol in healthy children and adolescents: the Fitkids Treadmill Test (FTT).

Methods: Sixty-eight healthy children and adolescents (6-18 yr) were randomly divided into a validity group (14 boys and 20 girls; mean ± SD age, 12.9 ± 3.6 yr) that performed the FTT and Bruce protocol, both with respiratory gas analysis within 2 wk, and a reproducibility group (19 boys and 15 girls; mean ± SD age, 13.5 ± 3.5 yr) that performed the FTT twice within 2 wk. A subgroup of 21 participants within the reproducibility group performed both FTT with respiratory gas analysis. Time to exhaustion (TTE) was the main outcome of the FTT.

Results: VO_{2peak} measured during the FTT showed excellent correlation with VO_{2peak} measured during the Bruce protocol (r=0.90; P<.01). Backward multiple regression analysis provided the following prediction equations for VO_{2peak} ($L \cdot min^{-1}$) for boys and girls, respectively: VO_{2peak} FTT = -0.748 + (0.117 × TTE FTT) + (0.032 × body mass) + 0.263, and VO_{2peak} FTT = -0.748 + (0.117 × TTE FTT) + (0.032 × body mass) [R^2=0.935, SEE=0.256 $L \cdot min^{-1}$]. Cross-validation of the regression model showed an R^2 value of 0.76. Reliability statistics for the FTT showed an intraclass coefficient of 0.985 (95% confidence interval, 0.971-0.993; P<.001) for TTE. Bland-Altman analysis showed a mean bias of -0.07 min, with limits of agreement between +1.30 and -1.43 minutes.

Conclusion: Results suggest that the FTT is a useful treadmill protocol with good validity and reproducibility in healthy children and adolescents. Exercise performance on the FTT and body mass can be used to adequately predict VO_{2peak} when respiratory gas analysis is not available.

Published as

Kotte EM, de Groot JF, Bongers BC, Winkler AM, Takken T. Validity and Reproducibility of a New Treadmill Protocol: the Fitkids Treadmill Test. *Med Sci Sports Exerc.* 2015 Oct;47(10):2241-7.

INTRODUCTION

Standardized exercise testing remains an important tool that provides valuable diagnostic and prognostic information in daily clinical practice. Exercise testing allows individualized assessment of exercise tolerance and can be used to monitor exercise training programs. For use in daily clinical practice, non-sophisticated, inexpensive, reliable, and valid exercise tests are of increasing interest, as they might help to increase the use of exercise testing.[43]

The Bruce treadmill protocol is the most frequently used protocol in children and adolescents using a treadmill for cardiopulmonary exercise testing (CPET),[44] and pediatric reference values have been published.[45-47] Although the Bruce treadmill protocol has good validity and reproducibility, the use in outpatient physical therapy practices can be difficult because the test requires a treadmill ergometer that can operate at an incline of 22%. Many of these practices are embedded in health and sports centers with only standard treadmills available that can operate at a maximum incline of 15%. Therefore, the angle of inclination of the Bruce treadmill protocol is a major concern in outpatient physical therapy practices.

Although there are several other established maximal treadmill protocols, including the Balke protocol,[126] the Cornell protocol[127] and the German Society of Pediatric Cardiology protocol,[128] the maximum incline of these treadmill protocols exceeds 15% as well. In addition to this practical issue, many of the established maximal treadmill protocols are too demanding for children with a disability or chronic disease because of the high incline in the first stage of the protocol, leading to a premature exhaustion of the muscles of the lower limbs before achieving cardiac or respiratory limits.[129]

In summary, there is a need for a maximal treadmill protocol that can be used in outpatient physical therapy practices when limited to a treadmill ergometer with maximal incline of 15%. In this article, a new maximal treadmill protocol, the so-called Fitkids Treadmill Test (FTT), is introduced. The protocol starts with 0% incline, making this protocol useful in children and adolescents with a disability or chronic disease. Before application in clinical practice, insight in the clinimetric properties of the FTT is crucial. The current study is the first step in the identification of the clinimetric properties of the FTT and aimed to investigate the validity and the reproducibility of the new developed FTT in healthy children and adolescents between 6 and 18 yr of age.

METHODS

Participants

Healthy children and adolescents (6-18 yr of age) were recruited from primary and secondary schools as well as from different sports clubs located in the Netherlands (convenience sample). The inclusion of participants started after approval of the Central Committee on Research Involving Human Subjects in the Netherlands. The participants and their parents individually received written information about the study and informed consent forms. Signed informed consent forms were obtained from the parents or legal guardian of each participant and separately from each participant who was 12 yr or older. The modified physical activity readiness questionnaire (PAR-Q) was used to evaluate the health status of willing participants and to assess safety for performing maximal exercise. Exclusion criteria were as follows: use of medication affecting exercise capacity, cardiovascular, or respiratory disease, impaired motor development, morbid obesity (body mass index (BMI) >2.5 SDS), or positive responses to one or more of the modified PAR-Q questions.

Study design

Participants were randomly divided into a validity or a reproducibility group. To assess the validity of the FTT, the validity group performed the FTT and the Bruce treadmill test with respiratory gas analysis, in a counterbalanced order within 2 wk. To assess the reproducibility of the FTT, the reproducibility group performed the FTT twice within 2 wk. A subgroup of 21 participants within the reproducibility group performed both FTT with respiratory gas analysis.

Anthropometry

Body mass and height were determined to the nearest 0.5 kg and 0.1 cm, respectively, using an analog scale (Medisana PSD; Medisana Benelux NV; Kerkrade, the Netherlands), and a stadiometer (Seca 213; Seca, Hamburg, Germany), respectively. For both measurements, participants wore light clothes and no shoes. BMI was derived from body mass and height. The BMI for age SD scores was calculated using Dutch reference values.[130] Subcutaneous fat of the biceps, triceps, subscapular, and suprailiac regions was measured using a Harpenden skinfold caliper. The average of 3 measures of each area was used. Body density was estimated using the equations proposed by Deurenberg et al[131] and used to estimate percent body fat on the basis of the Siri equation.[132] Body surface area (BSA) was calculated using the equation of Haycock et al,[133] which has been validated in infants, children and adults.

Exercise testing

Exercise tests were performed on a motor-driven treadmill ergometer (Lode Valiant; Lode BV, Groningen, the Netherlands) using the Lode Ergometry Manager software (Lode BV, Groningen, the Netherlands). During all treadmill tests, heart rate (HR) was monitored using a soft strap with a heart rate sensor (Polar H1 transmitter; Polar, Kempele, Finland). The participants of the validity group and 21 participants of the reproducibility subgroup breathed through a face mask (Hans Rudolph Inc., Kansas City, MO) during both maximal treadmill tests, which was connected to a mobile gas analysis system (Cortex Metamax B^3; Cortex Medical GmbH, Leipzig, Germany) with an in-built gas analyzer. The mobile respiratory gas analysis system was calibrated for respiratory gas analysis measurements (ambient air and a gas mixture of 17% oxygen and 5% carbon dioxide) and volume measurements (3-L syringe). Values for oxygen uptake (VO_2), carbon dioxide production (VCO_2), minute ventilation (VE), and respiratory exchange ratio (RER) were collected at 10-s intervals. The Cortex Metamax B^3 is a valid and reliable system for measuring ventilatory parameters during exercise.[134-136]

Peak VO_2 (VO_{2peak}), peak VE (VE_{peak}), and peak RER (RER_{peak}), measured as the average value over the last 30 s before peak exercise, and peak HR (HR_{peak}), defined as the highest value achieved during the last 30 s before test termination, were used for analysis. The test was deemed maximal when at least one of the following criteria was met: an HR_{peak} >180 beats·min^{-1} or an RER_{peak} >1.0.[137] Before and directly after the exercise test, participants were asked to rate their level of perceived exhaustion on an OMNI scale for perceived exertion (0-10). Perceived exertion of the FTT and the Bruce treadmill test were determined by subtracting the pretest OMNI score from the posttest OMNI score (ΔOMNI; posttest OMNI score minus pretest OMNI score).

Fitkids Treadmill Test

The FTT protocol consists of a 90-s warm-up period (3.5 km·h^{-1}, 0% grade) followed by the initiation of the test at 3.5 km·h^{-1} and a 1% gradient for 90 s followed by incremental increases in both speed (0.5 km·h^{-1}) and incline (2%) every 90 s until an incline of 15% was attained (see Table 1). After this last step, the incline is held at 15% and incremental increases of speed (0.5 km·h^{-1}) are performed every 90 s until volitional exhaustion. After the test, participants are monitored for 2 min to ensure normal recovery of HR (2.0 km·h^{-1} with a flat treadmill). The main outcome measure of the FTT is time to exhaustion (TTE) and is defined as the point at which the participant chooses to stop despite strong verbal encouragement. TTE is calculated as the total duration of the test minus the duration of the warm-up phase.

Table 1. Protocol FTT.

Stage	Stage duration (s)	Speed (km·h^{-1})	Speed (mph)	Gradient (%)
Warm-up	90	3.5	2.175	0
1	90	3.5	2.175	1
2	90	4.0	2.485	3
3	90	4.5	2.796	5
4	90	5.0	3.106	7
5	90	5.5	3.417	9
6	90	6.0	3.728	11
7	90	6.5	4.039	13
8	90	7.0	4.350	15
9	90	7.5	4.661	15
…				

Speed is not limited until voluntary exhaustion.
Maximum gradient is limited to 15%.

Bruce test

The Bruce treadmill protocol consisted of a 90-s warm-up period (2.74 km·h^{-1} and a flat treadmill) followed by the initiation of the test at 2.74 km·h^{-1} and a 10% gradient for 3 min followed by incremental increases in speed and incline every 3 min until volitional exhaustion, as described elsewhere.[77] After the test, participants were monitored for 2 min to ensure a normal recovery of HR (2.0 km·h^{-1} with a flat treadmill). The main outcome measure of the Bruce protocol is TTE and is defined as the point at which the participant chooses to stop despite strong verbal encouragement. TTE is calculated as the total duration of the test minus the duration of the warm-up phase.

During both tests, participants are not allowed to use the handrails, except for touching the handrail with one or two fingers to maintain body position near the center of the moving belt.

Statistical analysis

The IBM SPSS Statistics for Windows version 20.0 (IBM Corp, Armonk, NY) was used for data analyses. The distribution of the variables was assessed using visual inspection (histogram, boxplot and normal Q-Q plot) and the Shapiro-Wilk test for normality. Except for ΔOMNI scores in both groups and VE_{peak} in the validity group, all variables were normally distributed, with skewness and kurtosis *z*-scores between -1.96 and +1.96.

For both the validity group and the reproducibility group, data were checked on significant differences for TTE, cardiopulmonary variables, and perceived exertion between both tests. Paired-samples *t*-tests were completed for normally distributed data, whereas Wilcoxon signed-rank tests were completed for nonnormally distributed data.

Validity

To examine the validity of the FTT, correlation coefficients were calculated between TTE reached at the FTT and TTE reached at the Bruce test as well as between the cardiopulmonary variables reached at both tests. Backward multiple linear regression analysis was used to generate an equation to predict VO_{2peak} achieved at the FTT. Pearson product correlations were calculated between anthropometric variables (age, height, body mass, percent body fat, fat free mass (FFM), BMI, and BSA) and VO_{2peak} attained at the FTT. The best predictor candidate for VO_{2peak} attained at the FTT was entered into the model along with sex (0=girls, 1=boys) and TTE of the FTT. The goodness of fit and precision of the regression equation were evaluated using multiple coefficient of determination (R^2) and the absolute standard error of estimate (SEE). The model was tested for random errors, homoscedasticity and multicollinearity. The regression model was cross-validated using the data from the respiratory gas analysis obtained in the reproducibility subgroup (*n*=21). Before the cross-validation analysis, the validity group and the reproducibility group were checked on significant differences in participant characteristics. Independent sample *t*-tests were completed for normally distributed data, and Mann-Whitney Tests were performed for nonnormally distributed data.

Reproducibility

For test-retest reliability analysis, the two-way mixed (type, absolute agreement) intraclass correlation coefficient (ICC) was calculated for TTE achieved at both FTT and for cardiopulmonary variables achieved at both FTTs. ICC values higher than 0.75 were considered acceptable.[138] To determine whether improvements are meaningful for a single patient, limits of agreement (LOA) were calculated for the main outcome measure of the FTT, TTE, according to the procedure described by Bland and Altman.[139] TTE was defined as the main outcome measure in the reproducibility analysis because many clinicians might not be able to collect expired gasses during exercise. A *P*-value <.05 was considered statistically significant.

RESULTS

Of the 74 participants approached for this study, none were excluded because of a positive response in the PAR-Q. The study population was randomly divided into a validity group and a reproducibility group. From both the validity group and the reproducibility group, 3 participants were excluded because of the following reasons: invalid respiratory gas analysis (*n*=1), illness during the second day of testing (*n*=2), dyspnea during the test (*n*=1), scheduling issues (*n*=1) and a painful leg during testing (*n*=1). Eventually, the validity group and reproducibility group both consisted of 34 participants. Participant descriptive statistics are found in Table 2.

Table 2. Participant characteristics.

	Validity group (n=34)		**Reproducibility group** (n=34)		***P* Value**
Sex (boys/girls)	14/20		19/15		
Age (yr)	12.9 ± 3.6	[6.5-18.6]	13.5 ± 3.5	[6.6-18.2]	.501
Body mass (kg)	48.9 ± 16.8	[20.0-80.8]	49.2 ± 15.9	[21.0-80.5]	.928
Body height (m)	1.57 ± 0.18	[1.15-1.85]	1.61 ± 0.19	[1.20-1.89]	.374
BMI ($kg{\cdot}m^{-2}$)	19.1 ± 3.2	[13.6-25.3]	18.3 ± 2.3	[12.9-22.5]	.258
BSA (m^2)	1.44 ± 0.34	[0.80-2.04]	1.47 ± 0.33	[0.83-2.05]	.775
FFM (kg)	39.1 ± 13.0	[17.2-64.0]	40.1 ± 13.2	[16.7-61.9]	.750
Body fat (%)	19.4 ± 4.5	[12.8-27.2]	18.6 ± 4.2	[10.8-26.1]	.486
BMI for age (SD score)	-0.02	(-0.45-0.37)	0.30	(-0.20-1.24)	.048[a*]

Date are presented as mean ± SD [range].
Abbreviations: BMI=body mass index, BSA=body surface area; FFM=fat free mass; SD=standard deviation.
[a] Nonparametric Mann-Whitney Test, data presented as median (interquartile range).
[*] *P* <.05.

Validity

Participants from the validity group performed both the FTT and the Bruce test without any adverse effects. They all met the subjective criteria of maximal effort (sweating, unsteady walking, facial flushing, and clear unwillingness to continue despite strong verbal encouragement) and the objective criteria of maximal effort (HR_{peak} >180 beats·min^{-1} and/ or an RER_{peak} >1.0) during both the FTT and the Bruce treadmill protocol. Seventeen of the 34 participants (50%) reached the maximal incline of 15%. The mean ± SD between-visit time is 6.2 ± 1.4 d.

Results of the FTT and the Bruce protocol completed by the validity group are presented in Table 3. Significant higher values of TTE were found for the FTT. The cardiopulmonary variables and rate of perceived exertion were not significantly different between the FTT and the Bruce protocol. Pearson correlation coefficients for TTE, HR_{peak}, VO_{2peak} (L·min^{-1}) and RER_{peak} achieved at the Bruce protocol and the FTT were 0.98, 0.67, 0.90 and 0.64 (all *P*<.01), respectively, which are categorized as moderate to strong correlations according to Dancey and Reidy's categorization.[140] For VE_{peak} a Spearman correlation coefficient of 0.96 was found (*P*<.01).

Table 3. FTT and Bruce test results of the validity group.

	FTT (n=34)		**Bruce test** (n=34)		***P* Value**
TTE (min)	13.5 ± 3.7	[9.2-22.2]	12.1 ± 3.0	[7.9-18.0]	<.001*
HR_{peak} (beats·min^{-1})	194.9 ± 6.9	[181-208]	194.1 ± 6.3	[183-207]	.360
VO_{2peak} (L·min^{-1})	2.5 ± 1.0	[1.1-4.4]	2.5 ± 1.0	[1.0-4.5]	.779
VO_{2peak} (mL·kg^{-1}·min^{-1})	51.1 ± 7.6	[28.0-71.0]	51.8 ± 8.2	[34.0-72.0]	.366
RER_{peak}	1.1 ± 0.1	[1.0-1.3]	1.1 ± 0.1	[1.0-1.3]	.947
VE_{peak} (L·min^{-1})	75.3	(56.3-96.6)	71.09	(55.0-103.3)	.918[a]
ΔOMNI	10	(9.0-10.0)	9.5	(8.75-10)	.491[a]

Date are presented as mean ± SD, [range].
[a] Nonparametric Wilcoxon singed-ranks test; data are presented as median (interquartile range).
*P≤.001.
ΔOMNI, OMNI difference addressing the participants' level of fatigue (posttest OMNI score minus pretest OMNI score).

To construct an equation to predict VO_{2peak} attained at the FTT, Pearson correlation coefficients were calculated between anthropometric variables and VO_{2peak} attained at the FTT. FFM seemed to be the best predictor candidate to include into the regression model (*r*=0.934, *P*=.01). However, FFM is impractical for daily use and was therefore not included into the final model. Body mass, which was the second-best predictor and easily available in daily practice, was included into the final model along with sex and TTE on the FTT. The model that incorporated FFM can be found in the Supplemental Digital Content (see Document, Supplemental Digital Content 1, Equations to predict VO_{2peak} achieved during the FTT from the attained TTE at the FTT and the FFM, http://links.lww.com/MSS/A530).

4

The following equations for boys and girls were generated to predict VO_{2peak} achieved at the FTT from the attained TTE at the FTT and body mass:

Boys: VO_{2peak} FTT = -0.748 + (0.117 × TTE FTT) + (0.032 × body mass) + 0.263

Girls: VO_{2peak} FTT = -0.748 + (0.117 × TTE FTT) + (0.032 × body mass)

In the prediction equations, 'VO_{2peak} FTT' represents the predicted VO_{2peak} in liters per minutes, 'TTE FFT' is the total duration of the FTT in minutes minus the duration of the warm up phase in minutes, and 'body mass' is expressed in kilograms (R^2 = 0.935, SEE= 0.256).

For cross-validation purposes, the VO_{2peak} estimated by the equations was plotted against the measured VO_{2peak} during the first FTT in the reproducibility subgroup who performed the FTT with respiratory gas analysis (Fig 1). Estimated VO_{2peak} (3.19 ± 0.51 L·min^{-1}) did not differ significantly from the observed VO_{2peak} (3.31 ± 0.88 L·min^{-1}; P=.291; R^2 = 0.76).

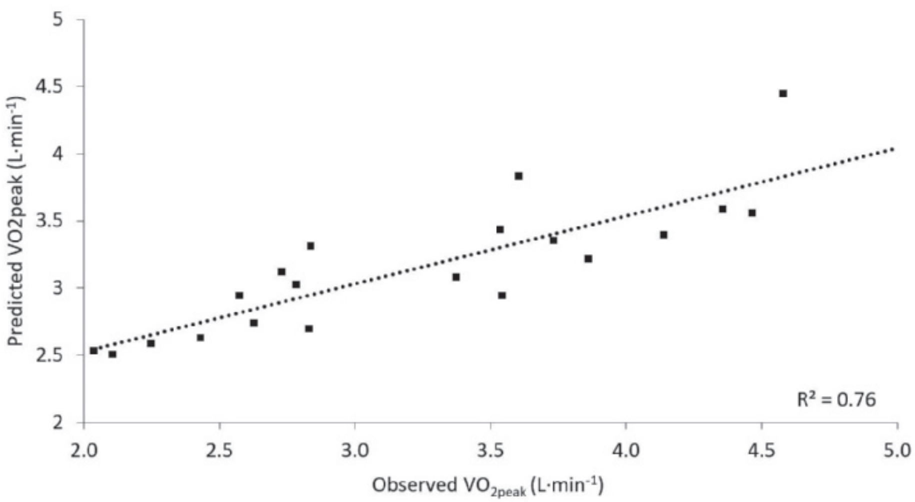

Figure 1: Cross-validation plot: linear relation between the predicted VO_{2peak} values based on the prediction equation of the regression model and the VO_{2peak} measured during the first FTT in the reproducibility subgroup.

Reproducibility

Participants from the reproducibility group performed the FTT twice without any adverse effects. They all met the subjective criteria of maximal effort (sweating, unsteady walking, facial flushing and clear unwillingness to continue despite strong verbal encouragement) and the objective criteria of maximal effort (HR_{peak}>180 beats·min^{-1} and/or an RER_{peak}>1.0) during both FTT. One participant showed an HR_{peak} of 174 and 169 beats·min^{-1}, respectively, on the first and second FTT; however, RER_{peak} measured during these tests were 1.28 and 1.25, respectively. Twenty-two of the 34 participants (65%) reached the maximal incline of 15%. The mean ± SD between-visit time was 8.9 ± 3.8 d.

The results of both FTT performed by the reproducibility group are shown in Table 4. There were no significant differences in TTE and HR_{peak} between the two FTT. In addition, for perceived exertion (ΔOMNI), no significant difference was found between both FTT. For the cardiopulmonary variables, a significant higher RER_{peak} was found during the second FTT. To quantify the relation between TTE achieved at both tests and between the cardiopulmonary variables achieved at both tests, ICC values were calculated. The ICC for TTE, which is the main outcome for the test-retest reliability statistics, was 0.985 (95% confidence interval (CI), 0.971-0.993; *P*<.001). The ICC values for HR_{peak}, VO_{2peak}, RER_{peak} and VE_{peak} were 0.767 (95% CI, 0.584-0.876; *P*<.001), 0.963 (95% CI, 0.912-0.985; *P*<.001), 0.631 (95% CI, 0.269-0.834; *P*<.001) and 0.948 (95% CI, 0.877-0.978; *P*<.001), respectively. To analyze agreement between the two FTT, a Bland Altman plot for TTE is depicted in Figure 2. The mean bias between the two FTTs was -0.07 minutes. The LOA for TTE were +1.30 minutes and -1.43 minutes.

Table 4. FTT results of the reproducibility group.

		First FFT		**Second FFT**		**P Value**
TTE (min)	n=34	14.3 ± 4.0	[7.7-24.5]	14.3 ± 4.1	[8.0-24.3]	.584
HR_{peak} (beats· min^{-1})	n=34	197 ± 8	[174-215]	196 ± 10	[169-222]	.157
VO_{2peak} (L·min^{-1})	n=21	3.3 ± 0.9	[2.0-5.1]	3.3 ± 0.8	[2.2-5.0]	.645
VO_{2peak} (mL·kg^{-1}·min^{-1})	n=21	54.5 ± 12.7	[26.0-80.0]	55.2 ± 10.1	[40.0-77.0]	.644
RER_{peak}	n=21	1.1 ± 0.1	[1.0-1.31]	1.2 ± 0.1	[1.0-1.4]	.029*
VE_{peak} (L·min^{-1})	n=21	105.8 ± 25.8	[64.4-152.2]	107.3 ± 27.0	[65.7-151.2]	.407
Δ OMNI	n=34	10	(8.0-10.0)	10	(8.38-10.0)	.241[a]

Date are presented as mean ± SD, [range].
[a] Nonparametric Wilcoxon singed-ranks test; data are presented as median (interquartile range).
*$P \leq .001$.
ΔOMNI, OMNI difference addressing the participants' level of fatigue (posttest OMNI score minus pretest OMNI score).

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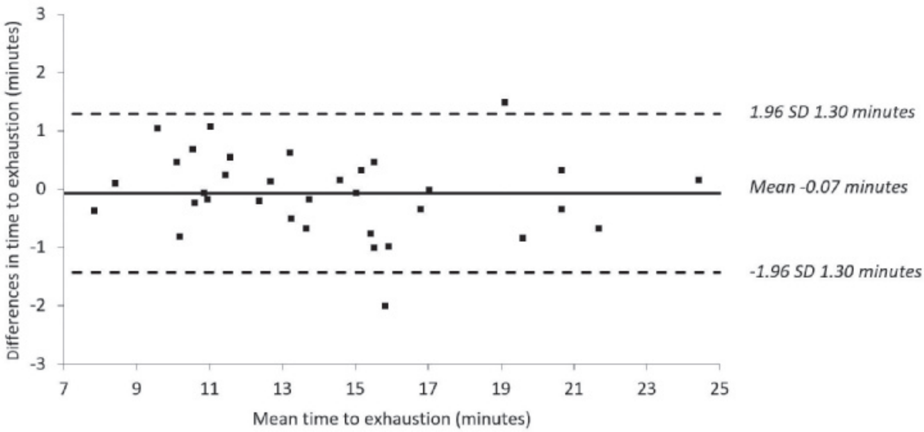

Figure 2: Bland-Altman plot of TTE as attained at the first FTT versus the second FTT.

DISCUSSION

The aim of the current study was to investigate the validity and reproducibility of a new maximal treadmill protocol for children and adolescents between 6 and 18 yr of age: the FTT. The results of the present study suggest that the FTT is a useful treadmill protocol with good validity and reproducibility. Moderate-to-strong significant correlations between TTE and cardiopulmonary variables attained at the FTT and the Bruce protocol were found, indicating that the FTT is a valid test for the assessment of aerobic exercise capacity in healthy children and adolescents. At the same time, there were no significant differences between the VO_{2peak} measured during the Bruce protocol and the VO_{2peak} measured during the FTT. These results suggest that the tests produce quite similar estimates and that the tests may be interchangeable because of these similar results.

The current study showed that VO_{2peak} can be adequately predicted from TTE and body mass in boys and girls, explaining 94% of the total variance in VO_{2peak}. When applying the multiple prediction equations developed in the validity group to the reproducibility subgroup, no significant difference was found between the observed VO_{2peak} (3.31 ± 0.88 L·min^{-1}) and the VO_{2peak} estimated from the prediction equation (3.19 ± 0.51 L·min-1; P =.291). The multiple correlation of the cross-validation regression equation was good (R^2= 0.76). This means that, for clinicians who do not have the resources to directly measure VO_{2peak}, the TTE during the FTT is an adequate alternative that gives insight in the aerobic fitness of healthy children and adolescents. Nevertheless, measuring VO_{2peak} using respiratory gas analysis during incremental exercise is still considered to be the gold standard for aerobic fitness by the World Health Organization.[41]

Several other studies have predicted aerobic fitness from functional performance during exercise testing, both in adults and the pediatric population. Bruce et al[77] developed the first predictive equations, which were population specific for active and sedentary men with and without cardiac conditions as well as for healthy adults (Pearson correlation coefficients between predicted VO_{2peak} and measured VO_{2peak} ranged from r= 0.87 to r= 0.92). Foster et al[141] later developed a more generalized equation, dependent only on Bruce treadmill test performance, to predict VO_{2peak} ($L \cdot min^{-1}$) (R^2=0.98, SEE= 3.35 $mL \cdot kg^{-1} \cdot min^{-1}$ or 8.5%). This generalized equation was developed for use in cardiac patients and in healthy sedentary and active individuals. Buono et al[142] predicted VO_{2peak} ($L \cdot min^{-1}$) during a timed distance run on an oval dirt track in a healthy children and adolescents. The equation in their study (based on mile run time, sex, skinfold thickness and body mass) explained 84% of the total variance in VO_{2peak} (R^2= 0.84 and SEE 4.3 $mL \cdot kg^{-1} \cdot min^{-1}$ or 9%). Bongers et al[143,144] concluded that VO_{2peak} ($L \cdot min^{-1}$) could be validly predicted from the attained peak work rate at the steep ramp test in healthy children and adolescents (R^2=0.917, SEE=0.24 $L \cdot min^{-1}$ or 9%) and Dencker et al[145] found that VO_{2peak} ($L \cdot min^{-1}$) could be predicted from the peak work rate reached during CPET in healthy children (R^2=0.83, SEE=0.11 $L \cdot min^{-1}$ or 8.4%). In the study of Dencker et al,[145] CPET was performed on an electronically braked cycle ergometer using a protocol with an initial workload of 30 W and an increase of 15 $W \cdot min^{-1}$ (1 W every 4 s). The equation developed in the current study is in agreement with previous studies reporting that the addition of body mass[142,146] and sex[142] improves the prediction of VO_{2peak} from TTE in children.

Test-retest reproducibility encompasses both reliability and agreement. Whereas the first refers to the consistency or stability of a test after repeated trials, the latter analyzes the variation within the individual scores during a test-retest situation[147,148] and is used to determine the clinical value of a measurement. The test-retest reliability of the FTT can be considered excellent with an ICC for TTE of 0.985 (95% CI, 0.971-0.993; P<.001). This means that the measurement error is small compared to the variability between the participants and that the discrimination of persons is hardly affected by measurement error. These results are comparable with those reported by Cumming et al.[94] They investigated the test-retest reliability of the Bruce protocol in 20 schoolchildren age 7-13 yr and reported a correlation coefficient of 0.94 between TTE achieved on trial 1 and TTE achieved on trial 2. The mean TTE values in the study of Cumming et al[94] were 13.9 ± 2.1 min for trial 1 and 13.7 ± 1.9 min for trial 2. The mean TTE of the FTT observed in the current study were 14.2 ± 4.0 min for trial 1 and 14.3 ± 4.1 min for trial 2. The results of the current study are also comparable with those of Johnston et al[149] who determined test-retest reproducibility of cardiopulmonary variables during CPET in children using a treadmill protocol. The reported ICC for VO_{2peak}, HR_{peak} and VE_{peak} were 0.96, 0.87 and 0.91, respectively, against 0.96, 0.77 and 0.95 for VO_{2peak}, HR_{peak} and VE_{peak}, respectively, found in the current study.

The agreement analysis revealed narrow LOA (+1.30 to -1.43 minutes), indicating that the agreement of the FTT is good. The average

4

difference of TTE attained at the two FFT was roughly 4 s. There was no evidence for a significant learning effect as reflected by a symmetrically distribution of the differences around the zero-difference line. A change score in TTE between two concessive measurements within an individual can only be considered to represent a real change if it is outside the LOA.

A limitation of this study is that only healthy participants were tested, so the equations currently developed are appropriate only for healthy children and adolescents. As the FTT is developed for use in outpatient physical therapy practices, predicting models for the clinical population should be developed and evaluated. An additional limitation of the current study is the small sample size used in the cross-validation analysis. The predictive accuracy of the developed regression equation seems proportionally biased, in which true values less than 2.5 $L{\cdot}min^{-1}$ are systematically overestimated and true values more than 4.0 $L{\cdot}min^{-1}$ are systematically underestimated. The predictive accuracy of the developed regression equation should be evaluated using a larger sample in future studies. Future studies should also look at reference values for different populations.

In conclusion, the results of the current study suggest that the FTT is a useful treadmill protocol with good validity and reproducibility in healthy children and adolescents age 6-18 yr. The use of the FTT can be favored instead of the Bruce protocol when limited to a standard treadmill ergometer with maximum incline of 15%. We have shown that exercise performance on the FTT and body mass can be used to simple predict VO_{2peak} in healthy children and adolescents in situations where it is not possible to measure VO_{2peak} with respiratory gas analysis. Further testing of the FTT in clinical populations is warranted.

ACKNOWLEDGEMENTS

This research was financed by the Johan Cruyff Foundation, the Rabobank Foundation, and the Dutch Foundation Innovation Alliance SIA RAAK PRO-4-03.

The authors are very grateful to Lode BV, Groningen, the Netherlands, and ProCare BV, Groningen, the Netherlands, for the technical support during this study. The authors thank the participating schools: Basisschool de Wiekslag, Tubbergen, the Netherlands and Bonhoeffer college, Castricum, the Netherlands. They also want to thank the Child Development and Exercise Center, University Medical Center Utrecht, Utrecht, the Netherlands. Finally, the authors are especially grateful to all of the participants.

CHAPTER 5

Fitkids Treadmill Test: age- and sex-related normative values in Dutch children and adolescents

ABSTRACT

Background: Recent research has shown that the Fitkids Treadmill Test (FTT) is a valid and reproducible exercise test for the assessment of aerobic exercise capacity in children and adolescents who are healthy.

Objective: The study objective was to provide sex- and age-related normative values for FTT performance in children and adolescents who were healthy, developing typically, and 6 to 18 years of age.

Design: This was a cross-sectional, observational study.

Methods: Three hundred fifty-six children and adolescents who were healthy (174 boys and 182 girls; mean age = 12.9, SD = 3.7) performed the FTT to their maximal effort to assess time to exhaustion (TTE). The least-mean-square method was used to generate sex- and age-related centile charts (P3, P10, P25, P50, P75, P90, and P97) for TTE on the FTT.

Results: In boys, the reference curve (P50) showed an almost linear increase in TTE with age, from 8.8 minutes at 6 years of age to 16.1 minutes at 18 years of age. In girls, the P50 values for TTE increased from 8.8 minutes at 6 years of age to 12.5 minutes at 18 years of age, with a plateau in TTE starting at approximately 10 years of age.

Limitations: Youth who were not white were underrepresented in this study.

Conclusion: The current study describes sex- and age-related normative values for FTT performance in children and adolescents who were healthy, developing typically, and 6 to 18 years of age. These age-and sex-related normative values will increase the usefulness of the FTT in clinical practice.

Published as

Kotte EM, de Groot JF, Bongers BC, Winkler AM, Takken T. Fitkids Treadmill Test: age- and sex-related normative values in Dutch children and adolescents. *Phys Ther.* 2016 Nov; 96(11):1764-1772.

INTRODUCTION

Exercise testing is being used with increasing frequency by pediatric physical therapists to assess the physical fitness of children and adolescents or to implement training programs.[150] Studies have shown that physical fitness is a powerful maker of health in youth.[151,152] With the use of cardiopulmonary exercise testing, therapists and exercise physiologists are able to determine peak oxygen uptake, which is the measure most commonly used for assessing aerobic fitness.[24,41] However, direct measurement of peak oxygen uptake requires sophisticated respiratory gas exchange equipment and specific training. Therefore, interest in methods in which aerobic fitness is estimated by use of predictive equations from functional outcomes during exercise tests is growing.[43]

Several valid and reliable strategies for estimating aerobic fitness in daily clinical practice by use of a cycle ergometer or treadmill are available.[77,143] Recently, our research group developed a new practical treadmill protocol for assessing aerobic fitness in children and adolescents: the Fitkids Treadmill Test (FTT). This development was based on a practical request articulated by physical therapists working with the Fitkids program. The FTT has 2 practical advantages over other established treadmill protocols. First, the protocol starts with a 0% incline, making it useful in children and adolescents with limited motor performance or those using an ankle-foot orthosis. Second, the maximal incline of the protocol is restricted to the maximal incline of standard treadmills, which is 15%, as these treadmills are most often available in outpatient physical therapy practices. A treadmill was chosen instead of a cycle ergometer because almost all children in the Fitkids program can be appropriately tested on a treadmill; even younger children (younger than 8 years), who have relatively underdeveloped knee extensor strength and do not fit on a standard cycle ergometer because of their short leg length.

The main outcome measure of the FTT is time to exhaustion (TTE), which is defined as the point at which a participant can no longer exercise against the speed and incline of the treadmill, despite strong verbal encouragement. In a recent study[153], good validity and reproducibility of the FTT in healthy children and adolescents were reported. Aerobic fitness can be accurately predicted from FTT performance (TTE) and body mass in boys and girls who are healthy (R^2=0.935).[153] At this point, sex- and age-related normative values for the FTT are lacking. Normative values will increase the usefulness of the FTT in clinical practice, as a physical therapist or exercise physiologists can determine whether a child's aerobic fitness is likely to be above average, average, or below average on the basis of FTT performance. The aim of the present study was to provide normative values for TTE on the FTT in children and adolescents, who were healthy, developing typically, and 6 to 18 years of age.

METHODS

Participants

Children and adolescents who were healthy and 6 to 18 years old were eligible to participate in this cross-sectional, observational study. The majority of the children and adolescents who were healthy were recruited from a primary school and several secondary schools, whereas a minority of the adolescents were recruited from local recreational sport clubs. At the schools, the selection procedure was based on class lists; only name and age were available.

Randomly selected participants were provided with an information package. The inclusion of participants started after approval of the Central Committee on Research Involving Human Subjects in the Netherlands. In total, 441 information packages were distributed to both the children and their parents. The modified Physical Activity Readiness Questionnaire was used to evaluate the health status of the children and adolescents who were willing to participate as well as to assess safety for performing maximal exercise. Exclusion criteria were a positive response to one or more questions of the modified Physical Activity Readiness Questionnaire, the use of medication affecting exercise capacity, cardiovascular or respiratory disease, musculoskeletal disease, metabolic disease, impaired motor development or morbid obesity (body mass index [BMI] standard deviation score (SDS) > 2.5).

To construct sex- and age-related normative values, we used the least mean squares method. It is not possible to perform a power calculation for setting up normative values with the least mean squares method. However, a minimum of 10 boys and 10 girls for each age seemed to be a feasible and sufficient number of participants for collecting and constructing generalizable and robust normative values. For the lowest and highest ages and within the age range of 12 to 14 years, we aimed to include 15 boys and 15 girls for an optimal fit of the data at both ends of the reference curve and because we expected a major development in exercise capacity due to puberty. Informed consent was signed by both parents as well as by children 12 years and older. Assent was attained from children younger than 12 years of age.

Anthropometry

Before exercise testing, body mass, body height, and sitting height were determined to the nearest 0.5 kg and 0.1 cm with an analog scale (Medisana PSD; Medisana Benelux NV, Kerkrade, the Netherlands) and a stadiometer (Seca 213, Seca, Hamburg, Germany). For these measurements, participants were wearing light clothes and no shoes. The BMI was derived from body mass and body height, whereas leg length was calculated by subtracting sitting height from body height. Standard deviation scores were calculated for BMI for age with Dutch reference values.[130] Subcutaneous fat of the biceps, triceps, subscapular, and suprailiac skinfolds was measured with a Harpenden skinfold caliper (Baty International, West Sussex, United Kingdom). The sum of the average of 3 measures for each measurement site was used to estimate body density with the equations proposed by Deurenberg et al.[131] To estimate percent body fat and subsequently to calculate fat-free mass, we used the Siri equation.[132] Body surface area was calculated with the equation of Haycock et al,[133] which has been validated in infants, children and adults.

Physical activity levels and sedentary time

Physical activity levels and sedentary time were assessed with the Dutch Standard Physical Activity Questionnaire for youth (Indicators for Monitoring Youth Health).[154] For children younger than 12 years of age, parents were asked how many days, in a typically week, their child walks or bikes to school, plays sports at school, plays sports at a sports club, and plays outside (outside school hours). In addition, the average duration of these activities on a typically day was assessed. Sedentary screen-based behavior was assessed in a similar manner, by asking parents about their child's television watching (including video's and DVDs, and YouTube) and computer playing. Children 12 years of age and older completed the questionnaire themselves. Participants were categorized as 'inactive' (<180 minutes of physical activity a week), 'semi-inactive' (180 - 299 minutes of physical activity a week), 'semi-active' (300 - 419 minutes of physical activity a week), or 'normally active' (>420 minutes of moderate-to-vigorous intensity physical activity a week) according to the Dutch public health guidelines for recommended levels of physical activity for children and adolescents.[155]

Fitkids Treadmill Test

Participants recruited from the primary and secondary schools were tested in a quiet room at their school, and the FTT was performed on a motor driven treadmill ergometer (Lode Valiant, Lode BV, Groningen, the Netherlands). Adolescents recruited from sports clubs were tested at a local fitness center, and a calibrated treadmill ergometer of the fitness center was used. To ensure that the setup at the sports clubs was similar to that at the schools, we tested the participants mainly outside hours when the fitness center was open. When it was not possible to test the participants during these hours, we chose to position the treadmill ergometer out of sight of other athletes during testing. During testing, heart rate was monitored with a heart rate belt (Polar H1 transmitter, Polar, Kempele, Finland).

The FTT protocol consists of a 90-second warm-up period (3.5 $km{\cdot}h^{-1}$; 0% incline) followed by the initiation of the test at 3.5 $km{\cdot}h^{-1}$ and 1% incline for 90 seconds. After this initial period, speed is increased by 0.5 $km{\cdot}h^{-1}$, and incline is increased by 2% every 90 seconds. The maximal incline is limited to 15%, but speed is increased with no limitation. The incremental increases in both speed and incline are continued until volitional exhaustion is reached, as described elsewhere.[153] The test is terminated when the participant can no longer keep up with the speed of the treadmill, despite strong verbal encouragement (standardized) (Appendix). At this point, a recovery phase of 90 seconds is initiated at a speed of 2.0 km·h^{-1} on a flat treadmill to ensure normal heart rate recovery.

The 90-second interval of the FTT is based on the interval used in the modified Bruce and Dubowy treadmill protocols. Smaller increments will facilitate better responsiveness in the protocol after an intervention such as an exercise training intervention. The same protocol was used for adolescents and younger children. Participants were instructed not to hold the handrails, except for touching the handrail with 1 or 2 fingers to regain balance during changes of speed and angle of inclination.

The TTE (in minutes, 1 decimal) was defined at peak exercise. The TTE was calculated as the total duration of the test minus the duration of the warm-up phase. The peak heart rate (HR_{peak}) was defined as the highest value achieved during the last 30 seconds before test termination. The test was deemed maximal when HR_{peak} was greater than 180 beats·min^{-1} and when subjective indicators of maximal effort occurred (e.g., sweating, unsteady walking, facial flushing and clear unwillingness to continue despite strong verbal encouragement).[156] Before and directly after the exercise test, participants were asked to rate their level of perceived exhaustion on an OMNI scale for perceived exertion (0-10). The scale starts with 0, indicating that the participant is not tired at all, and ends with 10, meaning that the participant is very, very tired. The level of perceived exertion was determined by subtracting the pretest OMNI score from the posttest OMNI score (i.e., change in OMNI score).[157]

Data analysis

Version 20.0 of IBM SPSS Statistics for Windows (IBM Corp, Armonk, New York) was used for data analysis. The distribution of the variables was assessed with visual inspection (histogram, boxplot, and normal quartile-quartile plot) and the Shapiro-Wilk test for normality. Differences between boys and girls in anthropometric variables and exercise variables were examined with Mann-Whitney *U* tests for nonnormally distributed data and the independent sample *t* test for normally distributed data. Determinants of exercise capacity were identified with Spearman correlation coefficients between TTE on the FTT and anthropometric variables. We used the least mean squares method to generate sex- and age-related centile charts (P3, P10, P25, P50, P75, P90, and P97) for TTE (LMS Chart maker Pro, Medical Research Council, London, United Kingdom). A *P* value of less than .05 was considered statistically significant.

RESULTS

Participants

Of the 441 children and adolescents who received an information package on the study, 373 children and adolescents (85%) were willing to participate and 361 were tested (82%). Twelve children (3%) were not tested for the following reasons: 6 had one or more positive answers on the modified Physical Activity Readiness Questionnaire, one 10-year-old girl fainted during skinfold thickness measurements, and 5 children were excluded because of morbid obesity (BMI SDS >2.5). The remaining 361 children performed the FTT, after which 5 children (1%) were excluded from the analysis for of the following reasons: hyperventilation during the FTT (n=1), painful Achilles tendon (n=1), painful leg (n=1), software problems (n=1), and dizziness during the FTT (n=1). Eventually, the data from 356 children and adolescents (81%), 174 boys and 182 girls, with a mean age of 12.9 years (SD = 3.7), were used for analysis (convenience sample). A flowchart of the inclusion procedure is shown in Figure 1. Participant characteristics are shown in Table 1.

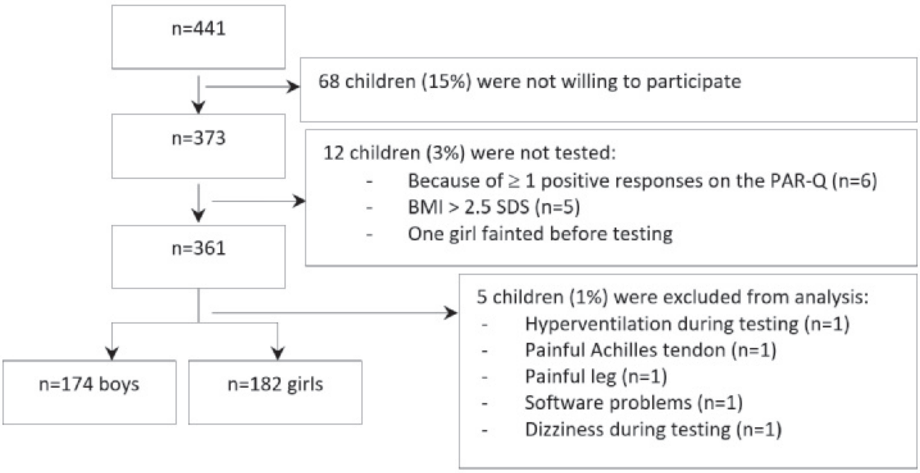

Figure 1. Flowchart of the inclusion procedure. BMI = body mass index, FTT = Fitkids Treadmill Test, PAR-Q = Physical Activity Readiness Questionnaire, SDS = standard deviation score.

Table 1. Participant characteristics.[a]

	Boys				Girls				
	n	Mean	SD	Range	n	Mean	SD	Range	*P*
Age (y)									
Total	174	13.0	3.7	6.3 to 18.8	182	12.8	3.7	6.1 to 18.9	.700
6-12 y	81	9.6	2.2	6.3 to 12.9	88	9.6	2.2	6.1 to 13.0	.900
13-18 y	93	15.9	1.7	13.1 to 18.8	94	15.8	1.6	13.3 to 18.9	.726
Body mass (kg)									
Total	174	48.5	17.8	18.5 to 92.0	182	46.6	16.3	18.0 to 83.0	.347
6-12 y	81	33.4	9.4	18.5 to 58.5	88	33.8	11.4	18.0 to 66.0	.742
13-18	93	61.7	11.8	32.5 to 92.0	94	58.7	9.6	40.0 to 83.0	.042[b]
Body height (m)									
Total	174	1.60	0.21	1.13 to 1.97	182	1.56	0.18	1.17 to 1.86	.016[b]
6-12 y	81	1.42	0.14	1.13 to 1.71	88	1.41	0.14	1.17 to 1.70	.769
13-18 y	93	1.76	0.10	1.44 to 1.97	94	1.69	0.07	1.53 to 1.86	<.001
BMI (kg/m^2)									
Total	174	18.2	2.8	12.6 to 25.8	182	18.5	3.4	10.7 to 28.5	.426
6-12 y	81	16.3	2.1	12.6 to 22.9	88	16.4	2.6	10.7 to 25.7	.995
13-18 y	93	19.8	2.3	15.1 to 25.8	94	20.5	2.7	16.2 to 28.5	.138
BSA (m^2)									
Total	174	1.45	0.36	0.77 to 2.23	182	1.40	0.33	0.76 to 2.06	.218
6-12 y	81	1.13	0.21	0.77 to 1.66	88	1.14	0.25	0.76 to 1.73	.753
13-18 y	93	1.72	0.21	1.13 to 2.23	94	1.65	0.16	1.31 to 2.06	.007[b]
FFM (kg)									
Total	174	40.3	14.3	16.1 to 74.0	180	36.3	11.6	15.1 to 59.5	.011[b]
6-12 y	81	27.9	7.1	16.1 to 46.6	87	26.8	8.0	15.1 to 48.2	.188
13-18 y	93	51.2	9.2	27.7 to 74.0	93	45.2	6.1	32.1 to 59.5	<.001

5

Table 1 (continued). Participant characteristics.[a]

	Boys				Girls				
	n	**Mean**	**SD**	**Range**	**n**	**Mean**	**SD**	**Range**	***P***
Body fat (%)									
Total	174	16.4	3.5	7.4 to 27.2	180	20.7	4.0	11.9 to 30.4	<.001
6-12 y	81	15.8	3.7	7.4 to 27.2	87	18.9	3.8	11.9 to 30.4	<.001
13-18 y	93	16.9	3.2	10.8 to 26.1	93	22.4	3.5	15.5 to 30.4	<.001
BMI for age (SDS)[c]									
Total	174	0.0	1.0	-3.2 to 2.3	182	0.0	1.1	-5.1 to 2.5	.936
6-12 y	81	-0.1	1.2	-3.2 to 2.3	88	-0.2	1.2	-5.1 to 2.5	.478
13-18 y	93	0.1	0.9	-2.0 to 2.2	94	0.2	0.9	-1.7 to 2.5	.362
Characteristic	**Total No. of Participants**	**n**	**%**		**Total No. of Participants**	**n**	**%**		
Inactive[d]									
Total	156	0	0		170	2	1		
6-12 y	80	0	0		86	1	1		
13-18 y	76	0	0		84	1	1		
Semi-inactive[d]									
Total	156	5	3		170	12	7		
6-12 y	80	3	4		86	4	5		
13-18 y	76	2	3		84	8	10		
Semi-active[d]									
Total	156	16	10		170	25	15		
6-12 y	80	7	9		86	14	16		
13-18 y	76	9	12		84	11	13		
Normally active[d]									
Total	156	135	87		170	131	77		
6-12 y	80	70	88		86	67	78		
13-18 y	76	65	86		84	64	76		

Table 1 (continued). Participant characteristics.[a]

	Boys				Girls				
	n	**Mean**	**SD**	**Range**	**n**	**Mean**	**SD**	**Range**	***P***
Sedentary time of > 2h/d									
Total	166	73	44		177	64	36		
6-12 y	81	31	38		86	27	31		
13-18 y	85	42	49		91	37	41		

[a] BMI = body mass index, BSA=body surface area; FFM=fat free mass.
[b] Significant at P<.05.
[c] Standard deviation score (SDS).
[d] Based on Dutch public health guidelines for recommended levels of physical activity for children and adolescents (5-18 years of age).[187]

5

Test Performance

All participants included in the analysis performed the FTT without any adverse effects, and they all met the subjective criterion of maximal effort. All participants also met the objective criterion of maximal effort during the FTT (HR_{peak} of > 180 beats·min^{-1}), except for one girl, who reached an HR_{peak} of 174 beats·min^{-1}. However, on the basis of the subjective indicators of maximal effort for this girl, we did include her data in the analysis. Figure 2 shows a scatter plot of the HR_{peak} reached during the FTT in relation to age for the total population.

The FTT results are shown in Table 2. Compared with girls, boys had a prolonged TTE (*P*<.001) and a slightly lower HR_{peak} (*P* =.011) on the FTT. The main TTEs on the FTT were 13.6 minutes (SD = 3.1) for boys and 11.6 minutes (SD = 1.9) for girls. The difference in mean HR_{peak} between boys and girls (197 versus 198 beats·min^{-1}) was not clinically relevant. No statistically significant differences in perceived exhaustion (change in OMNI score) between boys and girls were obtained.

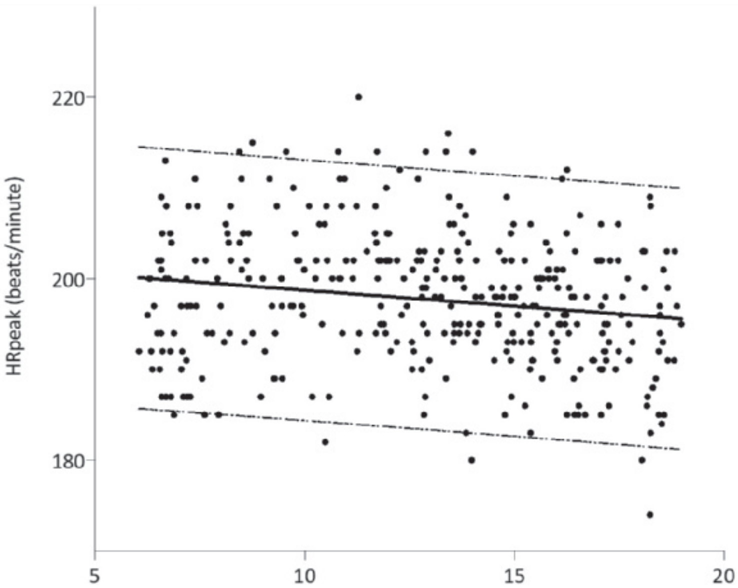

Figure 2. Age in relation to peak heart rate (HR_{peak}) reached on the Fitkids Treadmill Test for the total study population.

Table 2. Fitkids Treadmill Test results.[a]

	Boys (n=174)			**Girls** (n=182)			
	Mean	**SD**	**Range**	**Mean**	**SD**	**Range**	***P***
TTE (min)	13.6	3.1	7.5-24.5	11.6	1.9	7.5-18.3	<.001
HR_{peak} (beats·min^{-1})	197	7	180-214	198	7	174-220	.011[b]
ΔOMNI	9.2	1.2	3.0-10.0	8.9	1.4	2.0-10.0	.061

[a] TTE = time to exhaustion, HR_{peak} = peak heart rate, ΔOMNI = change in OMNI score (based on 173 boys).
[b] Significant at *P*<.05.

As expected, strong positive correlations were found between TTE on the FTT and age, body mass, body height, body surface area, fat-free mass, and leg length in boys (*r* values ranging from .679 to .799, with *P*<.001 for all coefficients) (Table 3). A moderate positive correlation was found between TTE on the FTT and BMI in boys (*r*=.501; *P*<.001). No correlation was found between TTE on the FTT and body fat in boys. In girls, moderate positive correlations were found between TTE on the FTT and age, body mass, body height, body surface area, fat-free mass, and leg length (*r* values ranging from .433 to .582, with *P*<.001 for all coefficients). A weak positive correlation was found between TTE on the FTT and BMI in girls (*r*=.325, *P*<.001). In accordance with the results for boys, no correlation was found between TTE on the FTT and body fat in girls.

Figure 3 shows age-related normative centile charts for TTE on the FTT for boys and girls. For practical considerations, we chose to use age instead of body height in the normative centile charts. Age and body height are highly correlated in children and had similar correlations with endurance times in our study population (correlation between age and TTE on the FTT: *r*=.649 [*P*<.001]; correlation between height and TTE on the FTT: *r*=.648 [*P*<.001]). In boys, the normative curves (P50) showed an almost linear increase in TTE with age, from 8.8 minutes at 6 years of age to 16.1 minutes at 18 years of age. In girls, the P50 values for TTE increased from 8.8 minutes at 6 years of age to 12.5 minutes at 18 years of age, with a plateau in TTE starting at approximately 10 years of age.

Table 3. Spearman correlation between time to exhaustion on the Fitkids Treadmill Test and anthropometric variables.[a]

	Boys (n=174)		**Girls** (n=182)	
Variable	**r**	***P***	**r**	***P***
Age (y)	.779	<.001	.582	<.001
Body mass (kg)	.720	<.001	.515	<.001
Body height (m)	.679	<.001	.433	<.001
BMI	.501	<.001	.325	<.001
BSA	.693	<.001	.446	<.001
FFM (kg)[b]	.720	<.001	.494	<.001
Body fat (%)[b]	-.046	NS	.111	NS
Leg length	.688	<.001	.522	<.001

[a] BMI = body mass index, BSA=body surface area; FFM=fat free mass, NS = not significant.

[b] FFM and body fat were not determined in 2 girls, so FFM and body fat values were based on 180 girls.

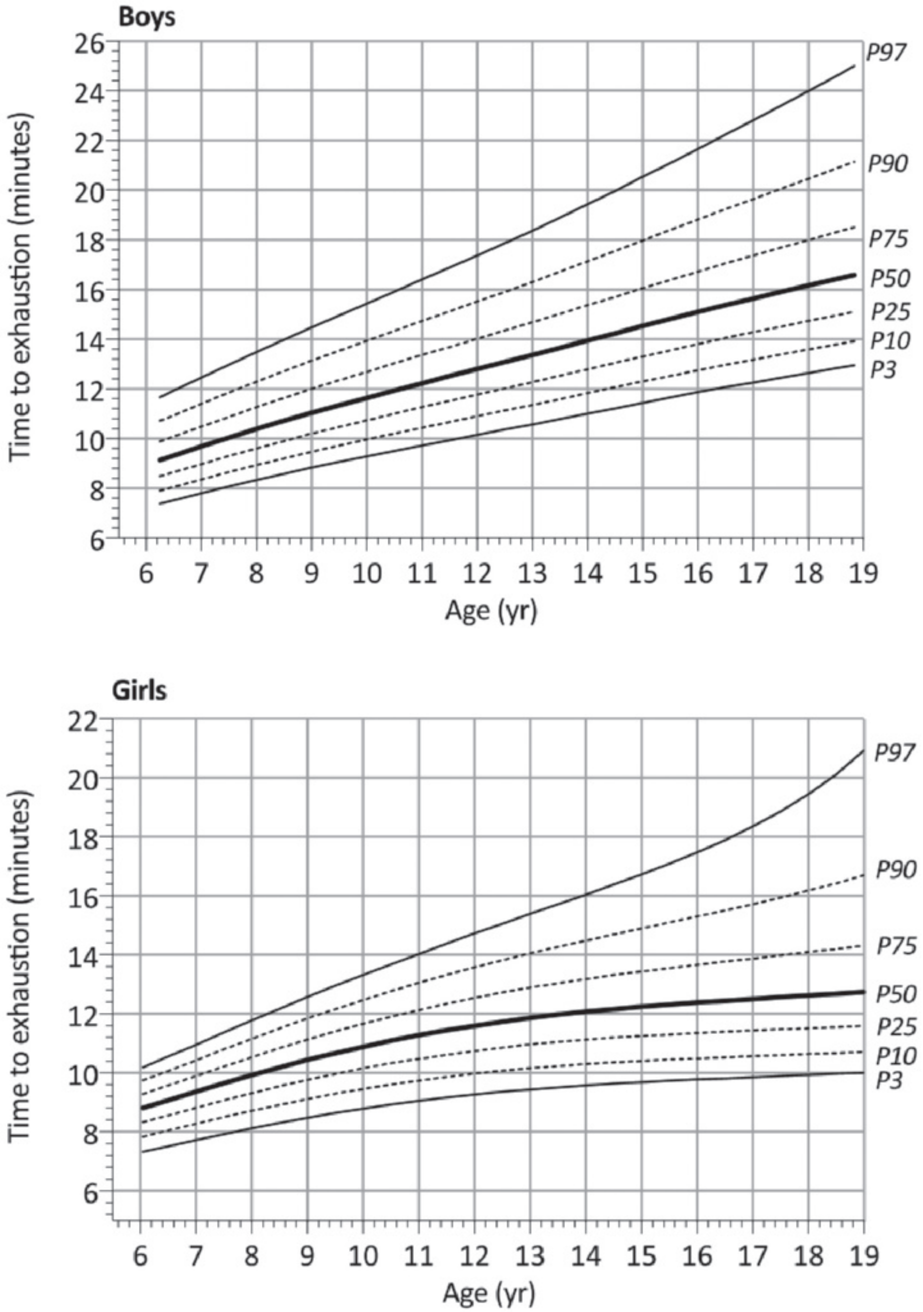

Figure 3. Age-related centile charts for time to exhaustion (TTE) on the Fitkids Treadmill Test (FTT) for boys and girls separately. The following equations can be used to predict the 50th centile (P50) for TTE on the FTT (minutes) from age (years): for boys, P50 TTE = (0.5870 x age) + 5.688 (R^2=.99); for girls, P50 TTE = (0.8817 x age) + (-0.02359 x age squared) + 4.384 (R^2=0.99).

DISCUSSION

The present study provides sex- and age-related normative values for FTT performance (TTE) in children and adolescents who were healthy, developing typically, and 6 to 18 years old. Because the FTT starts with a flat treadmill, has small incremental steps, and has a lower maximal incline than most established maximal-effort treadmill protocols, it is useful in children and adolescents with limited motor performance or balance problems or those using an ankle-foot orthosis as well.

Over the past 3 decades, normative values have been reported for standard maximal-effort treadmill exercise protocols, such as the Bruce protocol or the Balke protocol, as well as for several stepwise protocols with increments in speed, incline, or both.[158] Various shortcomings of these studies hinder implementation of the protocols in clinical practice. Many studies[159-168] assessed outcome measures that require sophisticated respiratory gas-exchange equipment. Others studies[45,47,61,128,163,166,167,169] used a treadmill protocol that requires an advanced treadmill with a high slope. Additionally, several studies included limited samples of participants in terms of sample size,[161,164,166] age range,[159,162] environmental conditions (altitude),[168] or ethnic background (nonwhite).[161,165] Some studies assessed outcome measures with individualized treadmill protocols. For instance, in the protocol used by Al-Hazzaa et al,[165] the speed of the treadmill depended on a child's age and ability to run comfortably on a treadmill. Studies assessing outcome measures with individually tailored treadmill protocols cannot be compared with other studies. A recent extensive overview of existing pediatric norms is available elsewhere.[158]

To our knowledge, no published studies have addressed most of these shortcomings, and no pediatric normative values have been published for exercise parameters that do not require respiratory gas analysis or that use a treadmill protocol that can be applied to a standard treadmill with a maximal incline of 15%. Although Dubowy et al,[128] van der Cammen-van Zijp and colleagues,[47,169] and Binkhorst et al[45] used protocols with a high incline (>15%), these studies are of interest for our setting in the Netherlands because they established pediatric normative values for exercise parameters (maximal endurance times) that do not require respiratory gas analysis in a white study population. Dubowy et al[128] used a stepwise protocol with incremental speed and incline every 90 seconds. They included 1,195 participants who were 3.0 to 75.0 years old.[128] Van der Cammen-van Zijp and colleagues[47,169] and Binkhorst et a.[45] used the Bruce protocol. Binkhorst et al[45] included 279 Dutch children who were healthy (6 - 18 years of age) and van der Cammen-van Zijp at al[47] included 267 Dutch children who were healthy (6 - 13 years old). In a separate study, van der Cammen-van Zijp et al[169] also described normative values for maximal endurance times in the Bruce treadmill protocol for eighty 4- and 5-year-old children who were healthy. The present study included 356 children and adolescents who were 6 to 18 years of age. Although Dubowy et al[128] included a large sample, the exact number of children and adolescents included were not mentioned. With respect to the studies by van der Cammen-van Zijp and colleagues,[47,169] normative values were established for a slightly broader pediatric age range in the present study (6-18 years in the present study versus 4-13 years in the studies of van der Cammen-van Zijp and colleagues).

In a comparison of the normative curves established for TTE in the present study with those provided by Dubowy et al,[128] similar patterns were obtained. In boys, the normative curve for TTE on the FTT showed an almost linear increase with age. In girls, the increase in TTE on FTT started to level off at approximately 10 years of age. The endurance time achieved by male participants in the study of Dubowy et al[128] increased until the age of 19 years, whereas in female participants it decreased continuously from puberty. Van der Cammen-van Zijp et al[47] and Binkhorst et al[45] also obtained similar patterns.

LIMITATION

A limitation of the present study is that youth who were not white were underrepresented.

FUTURE RESEARCH

Further study of the FTT is warranted and should include investigation of the clinimetric properties and responsiveness in clinical populations, such as children with cardiovascular disease, pulmonary disease, limited motor performance, or balance problems.

CONCLUSION

In conclusion, the present study provides sex- and age-related normative values for FTT performance in children and adolescents who were healthy, developing typically, and 6 to 18 years of age. In boys, the normative curves showed an almost linear increase in TTE with age. In girls, values started to level off at approximately 10 years of age.

APPENDIX

Encouragement

Because the duration of the load phase of the Fitkids Treadmill Test (FTT) differed among the participants, it was difficult to provide standardized encouragement throughout the test for each participant. During the first part of the FTT, encouragements such as 'You are doing great, come on' and 'Keep on going, great work' were used. When it became clear that a participant was struggling during the test, encouragements such as 'OK, keep running; the speed and incline are increasing, and you should try to keep up with the speed of the treadmill' were used. When a participant became exhausted, encouragements such as 'Come on, this is the most important part of the test; try to perform one last sprint, give everything you have got' were used.

ACKNOWLEDGEMENTS

This study was financed by the Johan Cruyff Foundation, the Rabobank Foundation, and the Dutch Foundation Innovation Alliance SIA RAAK PRO-4-03.

The authors are very grateful to Lode BV, Groningen, the Netherlands, and ProCare BV, Groningen, The Netherlands, for the technical support during this study. The authors thank the participating schools: Basisschool de Wiekslag, Tubbergen, the Netherlands, and R.K. Scholengemeenschap Canisius, Almelo and Bonhoeffer college, Castricum, the Netherlands. They also thank the Child Development and Exercise Center, University Medical Center Utrecht, Utrecht, the Netherlands, and the participating sports clubs. The authors thank the students for their assistance during data collection. Finally, the authors are especially grateful to all of the participants.

PART 3

NEW MEASUREMENT TOOLS: APPLICABILITY IN CLINICAL PRACTICE

CHAPTER 6

Fitkids Treadmill Test: clinical utility and factors associated with its use among physical therapists

ABSTRACT

Background: Although the Fitkids Treadmill Test (FTT) has been validated and normative values are available for 6-to-18-year-old children who are healthy, these facts do not automatically imply an uptake of the test in day-to-day practice of physical therapists.

Objective: The objectives of this study were to evaluate the utility of the FTT in different diagnostic groups and to explore potential factors affecting the use of the FTT in clinical practice.

Design: Mixed methods with both quantitative and qualitative data were used in this study.

Methods: Outcome parameters from the FTT were retrieved from the Fitkids database. For evaluation of the utility of the FTT, 2 indicators, exercise duration and maximal effort, were used. An online survey was sent to physical therapists in Fitkids practices Fitkids to identify factors affecting the use of the FTT in clinical practice.

Results: The percentage of children in each of the diagnostic groups who reached the minimal duration of a maximal exercise test ranged from 94% to 100%. The percentage of children who reached a HR_{peak} of >180 beats·min^{-1} ranged from 41% for children with cognitive, psychological or sensory disorders to 92% for children with metabolic diseases. The most important facilitator for the use of the FTT was the fact that most therapists were convinced of the additional value of the FTT. Main barriers were physical therapists' attitudes (resistance to change/ lack of experience) and, on the environmental level, the absence of a treadmill ergometer in physical therapist practice.

Limitations: Structured interviews would have provided a larger amount of information on potential factors affecting the use of the FTT in clinical practice

Conclusion: This study has shown the clinical utility of the FTT in different diagnostic groups in pediatric physical therapy practice. Responding to the factors identified in this study should enable improved uptake the FTT in clinical practice.

Published as

Kotte EMW, de Groot JF, Winkler AMF, Veenhof C, Takken T. Fitkids Treadmill Test: clinical utility and factors associated with its use among physical therapists. *Phys Ther.* 2019 Apr 1;99(4):428-439.

INTRODUCTION

Comprehensive assessment of aerobic fitness in children, especially those with a medical condition or disability, is an important part of pediatric physical therapy.[150] Various valid and reliable treadmill protocols for estimating aerobic fitness are available, but there are questions concerning the utility of most of these protocols in clinical practice by physical therapists in Fitkids practices (i.e., physical therapist practices affiliated with the Fitkids Foundation). To overcome these concerns, the Fitkids Treadmill Test (FTT) has been developed.[153] The advantages of the FTT over other existing and well-established maximal treadmill protocols[47,126-128] are 2-fold. First, the maximal incline of the protocol is restricted to the maximal incline of standard treadmills, which is 15%, as these treadmills are most often available in community physical therapy practices. Second, the protocol starts with 0% incline making this protocol useful in children and adolescents with a disability or chronic disease. A high incline in the first stage of the protocol often leads to premature exhaustion of the muscles of the lower limbs before achieving cardiac or respiratory limits in this population. Earlier research has shown the FTT to be valid and reproducible for the assessment of aerobic fitness in children and adolescents who are healthy.[153] Also, reference values for 6-to-18-year-old children who are healthy have been published.[170] Although the FFT has been evaluated to be valid and reproducible in children and adolescents who are healthy, these facts do not automatically imply an uptake of the test in the routine day-to-day practice of physical therapists. Clinical utility is clearly of key concern. Literally, clinical utility refers to 'the usefulness of an intervention for, or in, clinical practice'.[48] An uptake of the FTT in routine practice implies that therapists should change their established behavior in exercise testing. A widely used approach for understanding the difficulties in changing established behavior in clinical practice is based on a framework of Cabana et al.[171] The determinant framework of Cabana et al specifies the barriers and facilitators to the implementation of clinical practice guidelines. In these guidelines, the use of standardized measurement instruments is recommended in order to support clinical decision-making. However, the use of these standardized outcome measures is inconsistent in physical therapy practice.[172-175] Regarding maximal treadmill protocols, the Bruce treadmill protocol is the most commonly used protocol in clinical practice. In a study regarding the daily practice of exercise testing among pediatric cardiology and pulmonary centers in the United States, it was stated that the (modified) Bruce protocol was used by 79%.[44] The modified protocol has smaller increments in workload in comparison with the original Bruce protocol. This value is broadly in line with those in other studies.[176] This widespread popularity of the (modified) Bruce protocol is perhaps somewhat surprisingly, because of the frequently reported disadvantages of the (modified) Bruce protocol, especially in very young children and those with chronic illness.[177,178] Before the introduction the FTT, the (modified) Bruce protocol was also widely used in Fitkids practices.[89] However, because of the limited inclination of the available treadmill ergometer in most Fitkids practices, the protocol was often adjusted.

There is only limited knowledge about effective strategies to increase use of outcome measures.[175] However, several studies have identified barriers and facilitators to routine use of outcome measures among physical therapists in clinical practice of which many are similar to those specified by Cabana et al.[171,172,174,175,179,180] The barriers identified by Cabana et al are grouped based on whether they affect physician knowledge (lack of awareness and lack of familiarity), attitude, (lack of agreement with (specific) guidelines, skills, motivation, outcome expectancy, self-efficacy and habits/routines) or behavior. Three external factors that are considered to be related to behavior are the guideline itself, the patient, and social/environmental factors.[171]

Identification of facilitators and barriers influencing the use of the FTT among physical therapists in Fitkids practices seems useful for improving uptake of the FTT in routine day-to-day practice of physical therapists. As the recommendation by the Fitkids Foundation to use the FTT in clinical practice can be interpreted as being a new guideline to monitor aerobic fitness it seems legitimate using the theoretical framework of Cabana et al[171] in this identification. Summarized, for the FTT to become well embedded in clinical practice, its utility should be demonstrated in clinical populations and a tailored implementation strategy should be launched based on prospectively identified barriers and facilitators to the use of the FTT in clinical practice. Therefore, the 2 goals of the present study were to evaluate the utility of the FTT in different diagnostic groups (part 1 of the study) and to explore potential factors affecting the use of the FTT in clinical practice (part 2 of the study).

METHODS

Fitkids Treadmill Test

The FTT is an incremental treadmill test consisting of 90-second stages with increments in both speed and grade as described previously.[153,170] The test starts after a warming-up period (3.5 km/h, 0% grade) at 3.5 km/h and a 1% gradient, followed by incremental increases in both speed (0.5 km/h) and incline (2%). The maximal incline is 15%. Main outcome measures of the FTT are time to exhaustion (TTE) and peak heart rate (HR_{peak}). The TTE is calculated as the total duration of the test minus the duration of the warm-up phase.

6

Knowledge transfer activities

The FTT was developed in close cooperation with physical therapists and pediatric physical therapists in Fitkids practices. Since 2014, the FTT is adopted as one of the core exercise tests outlined in the Fitkids test manual. Concurrently, the validity and the reproducibility of the FTT were investigated and in 2015 the article concerning the validity and reproducibility was published.[153] In 2016 reference values of the FTT in 356 Dutch children who were healthy were published.[170] The Fitkids foundation used the intranet located at our website, social media and newsletters to inform the therapists about the FTT. The protocol, standard operation procedures and reference values could also be obtained from the intranet and were distributed on several meetings. Different training days and workshops were organized over the past few years to ensure that therapists got familiar with the protocol. The knowledge transfer activities were part of the 'Fit for the Future' consortium[181] and were based on the policy of the Royal Dutch Society for Physical Therapy[180], that information about measurement instruments should be disseminated through publication in professional journals, newsletters, and guidelines.

Study design

The clinical quantitative data that were used to describe the study population and to evaluate the utility of the FTT in clinical practice were extracted from the Fitkids database. The Fitkids database is a web-based electronic patient record of children participating in the program. Data that were entered between March 2014 and May 2017 were used for this study. Fitkids is a nationwide exercise therapy program in the Netherlands. Fitkids aims to increase health-related fitness, health-related quality of life, and participation in physical activity and/ or sports in children and adolescents who are 6 to 18 years old and have a chronic condition or disability.

The data in the database were obtained by physical therapists and pediatric physical therapists during the initial assessment of Fitkids and concerned medical diagnosis, height, weight, HR_{peak}, and maximal TTE. The medical diagnosis of the children was based on the information provided by the treating physician in the medical referral letter. Body mass index (in kg/m^2) was calculated from weight and height and standard deviation scores (SDS) were calculated using Dutch growth charts.[182] Reference values published in our earlier research[170] were used to compare the maximal TTE of children in the present study with that of children who were healthy.

Part 1: Evaluation the utility of the FTT

To assess the utility of the FTT in clinical practice, 2 indicators were used. The first was exercise duration (determined as the total duration of the FTT, including the duration of the warm-up phase), in terms of percentage of children and adolescents who reached the minimal duration for a maximal exercise test that was set at 6 minutes for children who were 6 to 12 years old and 8 minutes for those who were 12 years and older[84]. The second was maximal effort, in terms of percentage of children and adolescents who reached a HR_{peak} of >180 beats·min^{-1} when performing the FTT. The indicators were considered sufficient when reached by 75% of the included children in each of the diagnostic groups.

Part 2: Exploring potential factors affecting the use of the FTT in clinical practice

To identify barriers and facilitators to the use of the FTT in clinical practice, an online questionnaire was sent to physical therapists working in Fitkids practices. The survey was sent with the use of Surveymonkey (www.surveymonkey.com). For a solid basis of the survey questionnaire, we used existing questionnaires to formulate questions and create response options.[183,184] These questions were supplemented with useful subjects for collecting specific information about the factors affecting the use of the FTT in clinical practice (i.e., questions on test characteristics of the FTT). Different types of response categories were used: open field questions, multiple-choice bullets, visual analog scale (ranging from 0 to 10), and binary questions. The survey started with some questions to gather demographic information of the physical therapist. To ensure clarity and assess face validity of the survey items and response options, a draft of the questionnaire was created by E.M.W.K. using the framework of Cabana et al[171] to reflect to underlying domains of barriers and facilitators affecting the use of the FTT in daily clinical practice. These domains are knowledge, attitude and external factors. The questionnaire was reviewed and discussed in our research group. Some questions of the survey were answered by all respondents (1-7,17-20,23,24), some were answered by only those who were using the FTT (defined as users) (8-15,21,22) and question 16 was answered only by those who were not using the FTT in clinical practice (defined as non-users). The appendix provides detailed information on the exact content of the online survey. For the purpose of this article, all questions are translated from Dutch to English.

Ethical issues

The analysis of the quantitative data was performed on preidentified anonymous patient care data. The data was collected in the course of standard care to evaluate the effects of the Fitkids exercise training program. According to the Dutch Medical Research Involving Human Subjects Act, this study did not require ethics approval. Parents and children gave their written permission to use the data for research purposes.

Data analysis

The IBM SPSS Statistics for Windows, version 24.0 (IBM Corp, Armonk, New York) was used for data analysis. For analysis of the quantitative data, the distribution of the data was determined and checked for normal distribution. The medical conditions of the included children were classified according to the diagnostic groups proposed by the American College of Sports and Medicine[185], to which we have added 1 category, being the category of motor developmental delays. Differences between maximal TTE of the FTT and reference values were analyzed using the SDS of TTE, that is, the difference between the observed and predicted value divided by the SD of the reference values. The analysis concerning the evaluation of the utility of the FTT focused on determining whether the indicators were sufficient in each diagnostic group. The analysis of the online survey focused on identifying facilitators and barriers, based on the Cabana framework.[171]

RESULTS

Study population

Data were based on a database export that was done on April 24, 2017. The export file consisted of test results of 415 children (260 boys and 155 girls) from 49 Fitkids practices. The median age of the children was 10.7 years (interquartile range: 9.1-12.3), the median body mass index (kg/m^2) was 22.4 (interquartile range: 18.3-25.9), and the median body mass index for age score 2.2 (interquartile range: 0.9-3.1). Because of incomplete datasets, the body mass index scores are based on 275 children. The medical conditions of the included children are outlined in Table 1.

FTT

Initial test results of the included children are presented in Table 2. The median maximal TTE ranged from 9.0 minutes in children with neuromuscular disorders to 11.5 minutes in children with cardiovascular diseases. Compared to the reference values, test results of the FTT showed significantly reduced maximal TTE for all medical diagnosis groups (median SDS TTE between -2.1 [in children with metabolic disorders] and -0.8 [in children with cardiovascular diseases]). The FTT was deemed maximal when HR_{peak} was >180 beats·min^{-1}. Median HR_{peak} ranged from 179 beats·min^{-1} in children with cardiovascular diseases and children with cognitive, psychological or sensory disorders to 197 beats·min^{-1} in children with metabolic diseases.

Table 1. Classification of primary medical diagnoses according to the American College of Sports Medicine.[a]

Classification	Examples of diagnosis	Total No. (%) participants
Cardiovascular diseases	congenital heart defects	2 (0.5)
Pulmonary diseases	asthma, cystic fibrosis	36 (9)
Metabolic diseases	diabetes	13 (3)
Immunological and hematological disorders	chronic fatigue syndrome, cancer, abdominal organ transplant, juvenile arthritis	18 (4)
Musculoskeletal/ orthopedic disabilities	hypermobility syndrome, toeing-in/ toeing-out, fractures	64 (15)
Neuromuscular disorders	cerebral palsy, spina bifida, Erb's palsy, epilepsy, ataxia	31 (8)
Cognitive, psychological, and sensory disorders	autism spectrum disorders, ADHD, intellectual disability, headache, hyperventilation, sensory processing disorder	94 (23)
Motor developmental delays	DCD	147 (35)
Unexplained condition		10 (2)

[a] ADHD = attention deficit hyperactivity disorder; DCD = developmental coordination disorder.

Table 2. Initial test results for the FTT.[a]

Classification	Maximal TTE (min)		SD of maximal TTE		HR_{peak} (beats·min^{-1})		% of participants who reached minimal exercise duration[b]	% of participants who reached maximal effort[c]
	Median	IQR	Median	IQR	Median	IQR		
Cardiovascular diseases	11.5[d]		-0.8		179		100	50
Pulmonary diseases	9.7	9.0 to 10.5	-1.3	-2.3 to -0.5	190	178 to 197[e]	97	71[e]
Metabolic diseases	9.5	7.7 to 10.8	-2.1	-3.9 to -0.6	197	188 to 200	100	92
Immunological and hematological disorders	10.5	9.0 to 11.5	-1.3	-3.5 to 0.2	187	176 to 194	100	61
Musculoskeletal/ orthopedic disabilities	9.2	8.2 to 11.0	-1.7	-2.7 to -0.2	180	168 to 193[f]	100	48[f]
Neuromuscular disorders	9.0	7.5 to 10.3	-1.9	-4.1 to -0.8	187	180 to 194[g]	97	76[g]
Cognitive, psychological, and sensory disorders	9.3	8.3 to 10.5	-1.8	-3.2 to -1.0	179	164 to 191[h]	94	41[h]
Motor developmental delays	9.7	8.5 to 11.0	-1.5	-2.9 to 0.0	188	178 to 196[i]	99	64[i]

[a] FTT = Fitkids Treadmill Test, HR_{peak} = peak heart rate, TTE = time to exhaustion.
[b] The minimal duration for a maximal exercise test was set at 6 minutes for children who were 6-12 years old and 8 minutes for those who were 12 years old and older.
[c] Defined as an HR_{peak} >180 beats·min^{-1}.
[d] Based on n=2.
[e] Based on n=35.
[f] Based on n=62.
[g] Based on n=29.
[h] Based on n=87.
[i] Based on n=135.

Part 1: Evaluate the utility of the FTT

Overall, 98% (405/415) of the children reached the minimal duration for a maximal exercise test which was set at 6 minutes for children who were 6 to 12 years old and 8 minutes for those who were 12 years old and older. With respect to maximal effort, 58% (228/391) of the participants with an HR_{peak} registration in the database met the objective criteria of a maximal effort during the FTT (HR_{peak} >180 beats·min^{-1}). Table 2 shows the percentages in each of the diagnostic groups.

Part 2: Exploring potential factors affecting the use of the FTT in clinical practice

Of the 166 Fitkids practices in the Netherlands at the time of the online survey, 155 were active. In total, 142 therapists who were involved in exercise testing in children participating in Fitkids completed the online survey from 141 Fitkids centers (response rate 91% [141/155]).

Survey respondents

The characteristics of the 142 survey respondents are presented in Table 3. Sixty-three percent of the therapists reported using the FTT in clinical practice.

Table 3. Survey respondents (n=142).

Characteristic	No. (%) Users (n=90)	Nonusers (n=52)
Sex		
M	11 (12)	7 (14)
F	79 (88)	45 (87)
Age, y		
21-25	6 (7)	3 (6)
26-35	47 (52)	24 (46)
36-45	21 (23)	7 (14)
46-55	9 (10)	12 (23)
>55	7 (8)	6 (12)
Working experience		
0-2	4 (4)	3 (6)
3-4	6 (7)	3 (6)
5-6	8 (9)	3 (6)
7-8	14 (16)	7 (13)
9-10	20 (22)	7 (13)
>10	38 (42)	29 (56)

Table 3 (continued). Survey respondents (n=142).

Characteristic	No. (%) Users (n=90)	Nonusers (n=52)
Profession		
Pediatric physical therapist	67 (74)	45 (87)
Pediatric physical therapist in training	11 (12)	1 (2)
General physical therapist	12 (13)	6 (12)
Experience in using measurement instruments, y		
0-2	28 (31)	11 (21)
3-4	17 (19)	11 (21)
5-6	17 (19)	10 (19)
>6	28 (31)	20 (39)

Knowledge related factors

In general, therapists were aware of the existence of the FTT. Only 1 respondent explicitly reported not to be aware of the FTT (0.7%). Users of the FTT rated a mean knowledge score of 7.4 on a scale from 1 to 10. Fifty-four percent (49/90) of the users reported to have a lot of knowledge of the FTT (score 8, 9 or 10). Nonusers were not asked to rate their knowledge on a scale from 0-10 but were asked for specific knowledge of the FTT by answering three questions: What is the main outcome measure of the FTT? Is rail holding permitted? And, are reference values of the FTT available? The users also answered these questions. Ninety-seven percent (87/90) of the users were aware of the main outcome measure of the FTT; the value for nonusers was 56% (27/48). Ninety-two percent (83/90) of the users reported to know that rail holding is not permitted; the value for nonusers was 73% (36/49). Ninety-four percent (85/90) of the users reported that they were aware of the existence of the reference values; the value for nonusers was 78% (38/49).

Attitude related factors

Ninety-six percent (85/89) of the users were convinced of the additional value of the FTT; the value for nonusers was 62% (31/50). The reasons why the test was found to be of additional value are summarized in Table 4 for both the users as the nonusers. In total, 112 reasons were reported by 79 users and 26 reasons were reported by 22 nonusers. All reasons reported were classified in 5 main categories. With respect to motivation to use the FTT, users rated a mean motivation score of 8.5 on a scale from 1 to 10, whereas the nonusers rated a score of 5.1. With respect to skills in using the FTT, users rated a mean score of 7.5 on a scale from 1 to 10. Fifty-six percent (50/90) of the users reported having good skills in using the FTT (score 8, 9 or 10), whereas 10% (9/90) reported having only a little skill in using the FTT (score of ≤ 5). Nonusers did not answer this question but were assumed to have no skills in using the FTT.

Table 4. Reasons why therapists found the FTT of added value.[a]

Reason	No. (%) of reasons reported by:	
	Users (n=79)	**Nonusers** (n=22)
Maximal incline of the protocol of 15%	27 (24)	7 (27)
Because the FTT is more appropriate when testing children with a chronic disease or disability than the (modified) Bruce treadmill test	24 (21)	6 (23)
Recently developed reference values	23 (21)	5 (19)
The FTT provides valuable insight into (improvement of) health-related fitness (evaluates the effect of the Fitkids program)	21 (19)	7 (27)
The FTT is easy to use in general and easy to conduct	17 (15)	1 (4)

[a] FTT = Fitkids Treadmill Test.

Users were asked whether they found themselves well educated in using the FTT and what their experience is in using the FTT. Seventy-seven percent (69/90) (highly) agreed that they were sufficiently trained in using the FTT in clinical practice. Fifty-seven percent (51/90) reported having a lot of experience in using the FTT. The mean number of FTTs conducted by 27 of these therapists was 41. Both the users as the nonusers were asked whether they were interested in additional training in using the FTT. Of the users, 27% (24/90) reported that they were interested; the value for nonusers was 28% (14/50).

External barriers

Twenty-seven percent (14/52) of the nonusers reported not meeting the practical requirement of having a treadmill ergometer available (either in their practice or at the time of exercise testing). Also, factors related to the measurement instrument itself may hamper the implementation of the FTT. Therefore, we asked whether the FTT was found to be time-consuming to conduct. Sixteen percent (14/90) of the users agreed with the statement 'conducting the FTT is time consuming'. As therapists who found the FTT highly demanding for children with a medical condition are less likely to use the FTT, we stated that 'the FTT is a highly demanding physical test for children who participate in the Fitkids program'. Of the users, 7% (6/90) (highly) agreed with this statement. Physical therapists are also less likely to use the FTT in clinical practice if they assume that children who are visiting the practice are not able to reach their maximal effort on the FTT. Of the users, 32% (29/90) disagreed with the statement that 'children often reach their maximal effort when performing the FTT'. The users who disagreed with the statement reported the following reasons for limiting maximal effort. In total, 61 reasons were reported by 29 therapists: the FTT is difficult because holding onto the front handrail during the test is not permitted (38%; 23/61), painful legs (31%; 19/61), walking on the treadmill with increments in speed and inclination is too difficult (18%; 11/61), motivational problems/lack of perseverance (10%; 6/61), and the physiological response to (maximal) aerobic exercise is unfamiliar or unknown to children (3%; 2/61).

Reasons for not using the FTT in clinical practice are summarized in Table 5. In total 44 nonusers reported a reason and 50% (22/44) of the reasons reported could be classified in the category attitude related, of which 39% (17/44) were related to resistance to change (habits) and 11% (5/44) to lack of experience. The other 50% of the reasons reported were related to external barriers, which could be classified into 3 main categories: patient (9%; 9/44), organizational (39%; 17/44) and measurement related (2%; 1/44). There were no reasons reported that could be classified in the category knowledge related.

Table 5. Factors for not using the FTT in clinical practice reported by the non-users.[a]

Classification	**No. (%) of nonusers** (n=44)
Attitude related	
Resistance to change (habits)	17 (39)
Lack of experience	5 (11)
External barrier	
Patient	
Different preferences	1 (2)
Child cannot be tested because the test is too demanding	1 (2)
Contraindications to maximal exercise (cardiovascular problems, exercise intolerance)	2 (5)
Organization	
No treadmill ergometer available	14 (32)
No time for individual testing	3 (7)
Measurement instrument	
Test is time-consuming	1 (2)

[a] FTT = Fitkids Treadmill Test.

6

Table 6 provides an overview of factors that seemed to have influenced the implementation of the FTT in clinical practice.

Table 6. Overview of important factors in the implementation of the FTT.[a]

Factor	Users (n=90)	Non-users (n=52)
Knowledge related		
Awareness of FTT	100%	98%
Mean knowledge score	7.4	
Percentage of respondents that correctly answered 3 questions related to the FTT	84%	44%[b]
Attitude related		
Convinced of added value	96%[c]	*62%*[d]
Mean motivation score	8.5	5.1
Good skills in using the FTT	56%	Assumed to have no skills
Sufficiently trained in using the FTT	77%	
Interested in additional training in using the FTT	27%	28%[d]
Resistance to change		39%[e]
Lack of experience		11%[e]
External barriers		
The FTT is time consuming	16%	
FTT is highly demanding	7%	
Children are able to reach for their maximal effort	32% *disagreed*	
Availability of a treadmill ergometer	100%	73%

[a] FTT = Fitkids Treadmill Test.
[b] Based on n=48.
[c] Based on n=89.
[d] Based on n=50.
[e] Based on n=44.

DISCUSSION

The objectives of the study were to evaluate the utility of the FTT in different diagnostic groups and to explore potential factors affecting the use of the FTT in clinical practice. The test results of the FTT showed significantly reduced median maximal TTE for all diagnostic groups (SDS ranging from -2.1 to -0.8), indicating sufficient discriminatory ability of the FTT. The findings of reduced aerobic fitness levels in children participating in Fitkids are consistent with the findings in our previous studies.[89,186]

Part 1: Evaluation of the utility of the FTT

The first indicator to evaluate the utility of the FTT was successful as the minimal exercise duration of 6 minutes in children (6-12 years old) and 8 minutes in adolescents (12 years old and older) was reached in more than 75% of the included children in each diagnostic group. It is important to well inform the therapists on the minimal duration of a maximal cardiorespiratory exercise test. Maximal cardiopulmonary exercise tests with extremely short durations should be interpreted with caution and used evaluative as the oxygen uptake may not have been reached its maximum. The second indicator, to evaluate utility, was successful if 75% of the children in each diagnostic group reached a HR_{peak} of >180 beats·min^{-1}. This indicator was, however, only reached in children with metabolic diseases (92%) and neuromuscular diseases (76%). In fact, 42% of the participants with a HR_{peak} registration in the database had a $HR_{peak} \leq$ 180 beats·min^{-1}. From the online survey, we learned that 68% of the therapists who are using the FTT in clinical practice found the test feasible on HR_{peak} as they agreed with the statement that 'children often reach their maximal effort when performing the FTT'. It is known from literature that, for instance, children with intellectual disabilities have lower predicted HR_{peak}.[187] The recommended lower limit of normal for HR_{peak} of 180 beats·min^{-1} to identify maximal effort seems therefore not applicable in this population. Moreover, in our study on reference values for the FTT, 356 Dutch children who were healthy were included and a lower limit of normal of 174 beats·min^{-1} was found in girls (the mean HR_{peak} ± SD values in boys and girls were 197 ± 7 and 198 ± 7 beats·min^{-1}, respectively).[170] Finally, some children might have problems with movement coordination or have, for instance, an equinus position of the foot. A maximal treadmill test is likely to be symptom-limited in these children resulting in a symptom-limited HR_{peak}. There are other validated field tests that better suits these children.[79,100,188] We advise therapists to choose measurement instruments that are tailored to the individual child and to use subjective symptoms for maximal effort, such as sweating, facial blushing and a clear unwillingness to continue despite encouragement. Moreover, as it is questioned whether therapists push and encourage children enough to reach their maximal effort during a maximal exercise test, we aim at training therapists in conducting maximal exercise tests.

From the online survey, it became clear that there are 3 main reasons for limiting maximal effort during the FTT: the test is difficult because holding onto the front handrail during the test is not permitted, painful legs, and walking on a treadmill machine with increments in speed and inclination is too difficult. These reasons are likely to be related to being not familiar with treadmill walking. To ensure the opportunity for a successful evaluation, children must be familiarized with the testing procedure.[177] With respect to the FTT, it is essential that children are familiar with walking and running on an inclined treadmill ergometer before the child performs the FTT. Based on the current findings, we advise therapist to conduct 2 FTTs on 2 different sessions, as is also recommended for other field walking tests.[189] Ideally, we recommend a practice session before the actual test. In the context of Fitkids, we recommend performing a practice session during the Fitkids lesson in the week before the actual test session is planned. We are aware, however, that this might not always be feasible in clinical practice.

Part 2: Exploring potential factors affecting the use of the FTT in clinical practice

A substantial part of the therapists working in the Fitkids centers (63%) have adapted their established behavior in exercise testing. The percentage of Fitkids therapists using the FTT in clinical practice approaches the percentage of those using the (modified) Bruce treadmill test in clinical practice of pediatric cardiology and pulmonary centers in the United States (79%).[44] However, we are aware of the fact that therapists in Fitkids practices are a demarcated population of therapists in the Netherlands and that the use of the FTT in general is much lower. The online survey yielded information on factors we can respond to improve uptake of the FTT in routine day-to-day practice of physical therapists.

Discussion of factors influencing the use of the FTT on the basis of factors proposed by Cabana et al[171]

The online survey revealed a high level of awareness of the FTT among the physical therapists in the Fitkids practices and revealed that the knowledge of the FTT among the users was high. It can be concluded that the strategy to improve knowledge as used by the Fitkids Foundation was quite successful. However, the nonusers revealed to have far less detailed knowledge on the FTT and this might have contributed to the fact that these therapists do not use the FTT yet. Important reasons that were reported for not using the FTT although being convinced of the additional value were, attitude related, resistance to change or habits and at the organizational level the absence of a treadmill ergometer. It seems likely that therapists who cannot use the test because of the absence of a treadmill ergometer are less interested in the provided information on the FTT and therefore have less detailed knowledge.

From literature it is known that habits cause resistance to change at the individual level.[190] Miller at al[191] investigated the influence of providing information about a change on resistance to change and concluded that detailed 'quality' information about a change minimizes resistance at the individual level. Keeping this in mind, still a lot can be achieved by distributing detailed information on the FTT in the population of therapists that are not using the FTT in clinical practice yet. Currently, we are working on an integrated digital knowledge transfer system with information, detailed protocols and information videos to more widely distribute information on the FTT.

Logically, the nonusers were less motivated to use the FTT in clinical practice. Next to habits, a lower level of motivation is also known to cause resistance to change at the individual level.[190] Therefore, it is of interest which internal or external factors may help to motivate therapists who are nonusers to use the FTT. The fact that nonusers lack skills in using the FTT may result in a low motivation, because of lack of self-efficacy**.** Self-efficacy refers to the belief of being capable of carrying out a specific task (i.e., conducting the FTT). An individual's sense of capability is strongly related to his perception, motivation, and performance as people rarely attempt to perform a task when they expect to be unsuccessful.[192] Appropriate educational workshops or training days would raise practical skills and maybe motivate therapist in using the FTT among the nonusers and this might facilitate further use of the FTT in clinical practice. Training days will also facilitate further use as it will help those therapists who reported not to use the FTT because of lack of experience.

The most dominant, attitude related, facilitator for use of the FTT in clinical practice seems the fact that most therapists (83%; 116/139) indicated being convinced of the added value of the FTT to the quality of exercise testing in clinical practice. This is related to the fact that the FTT was developed in close cooperation with the therapists. More than half of the nonusers group (62%) indicated being convinced of the additional value of the FTT. Both in the users as the nonusers group, the maximal incline of 15% was mentioned as the most important reason for the FTT being of added value. Part of the therapists may have reported this reason out of practical considerations because the test can be conducted on a standard treadmill ergometer, whereas others may have reported this reason because they found a maximal gradient of 15% sufficient for the target group. Migdley et al.[193] recommend that treadmill tests should not surpass 15% degree in inclination. Although the reason for this recommendation remains more or less unclear, the recommendation may come from the fact that intolerable treadmill grades may limit the ability to elicit a maximal cardiopulmonary effort.

With respect to external factors, it can be concluded that the test characteristics of the FTT might have facilitated the use of the FTT in clinical practice, because only a small minority found the test time-consuming (16%) and highly demanding for children with a medical condition or disability (7%). The FTT is a valid and reliable test, with reference values available, which can be executed on a standard treadmill ergometer, starting with a flat treadmill. As there is no such test available like the FTT it may be concluded that the superiority of the FTT to other maximal treadmill protocols also have facilitated the use of the test in clinical practice. Moreover, it is likely to assume that the commitment of the therapists to the Fitkids Foundation has contributed to the fact that the FTT is already being used by 63%. The Fitkids foundation has developed the FTT and because of the commitment between the Foundation and the Fitkids practices, physical therapists in Fitkids practices are more likely to use the FTT in clinical practice. Moreover, the Fitkids Foundation and the nearly 170 affiliated Fitkids practices can be interpreted as being a social network. There is a significant relation between individuals' attitudes during a recently implemented change and the attitudes of others in their network.[194] Increase use of the FTT outside Fitkids practices would therefore be a bit more complicated because of lack of this commitment or lack of a social network. Finally, the fact that usage of the FTT does not entail additional costs is likely to have facilitated the use of the test in clinical practice.

LIMITATIONS

An online survey was used to identify factors influencing the use of the FTT in clinical practice. Using structured interviews would have allowed us to continue asking about underlying thoughts and would have provided us with a larger amount of information. Moreover, clinical utility is a much broader multi-dimensional judgement including usefulness, benefits and drawbacks of a new intervention. This study mainly focused on the clinical usefulness of the FTT. Furthermore, the narrow geographical location of the Netherlands in which the study data were collected might also be a limitation of this study. Finally, the results on the utility of the FTT in children with cardiovascular diseases should be interpreted with caution as these results are based on only 2 children.

CONCLUSIONS

The clinical utility of the FTT has been demonstrated in children with neuromuscular and metabolic diseases. In general, the FTT seems also useful in the remaining diagnostic groups of this study. However, prior to testing, it should always be questioned whether the individual child is able to reach his/her maximal effort during the FTT. Therapists should be trained in conducting maximal exercise tests and children should be familiar with the testing procedure to enable the opportunity of successful evaluations. Only successful evaluations should be interpreted in clinical practice. The online survey revealed important factors to the use of the FTT in clinical practice. Responding to the factors identified in this study enables an improved uptake the FTT in clinical practice.

ACKNOWLEDGEMENTS

This study was financed by the Johan Cruyff Foundation and the Dutch Foundation Innovation Alliance (SIA RAAK), project number SIA RAAK PRO-4-03. This funding source had no influence on the study's design, conduct, and reporting.

APPENDIX

Online survey

1. What is your age?
   - ☐ 21-25 years
   - ☐ 26-35 years
   - ☐ 36-45 years
   - ☐ 46-55 years
   - ☐ >55 years

2. How many years of working experience do you have as a physical therapist?
   - ☐ 0-2 years
   - ☐ 3-4 years
   - ☐ 5-6 years
   - ☐ 7-8 years
   - ☐ 9-10 years
   - ☐ > 10 years

3. What is your main profession?
   - ☐ General physical therapist
   - ☐ Pediatric physical therapist
   - ☐ Sport physical therapist
   - ☐ Other, namely

4. Who many years of experience in using measurement instruments do you have?
   - ☐ 0-2 years
   - ☐ 3-4 years
   - ☐ 5-6 years
   - ☐ > 6 years

5. Do you have the availability of a treadmill ergometer?
   - ☐ Yes
   - ☐ No

6. How motivated are you to use the FTT on a scale from 0 to 10?
   (0= not motivated; 10= highly motivated)
   0 – 1 – 2 – 3 – 4- 5 – 6 – 7 – 8 – 9 – 10

7. I am using the FTT in clinical practice
   - ☐ Yes
   - ☐ No

8. What is your experience in using the FTT?
   - ☐ No experience
   - ☐ Little experience
   - ☐ A lot of experience

9. How many FTT have you already conducted?
   - ☐ <5
   - ☐ Between 5 and 10
   - ☐ Between 10 and 20
   - ☐ Between 20 and 30
   - ☐ Between 30 and 40
   - ☐ Between 40 and 50
   - ☐ More than 50

10. Which maximal treadmill test did you use before using the FTT?
    (multiple answers possible)
    - ☐ Half-Bruce treadmill protocol
    - ☐ Bruce treadmill protocol
    - ☐ Balke protocol
    - ☐ Other, namely

11. On a scale of 0 to 10, what is your level of knowledge of the FTT?
    (0= no knowledge; 10= lots of knowledge)
    0 – 1 – 2 – 3 – 4- 5 – 6 – 7 – 8 – 9 – 10

12. On a scale of 0 to 10, what is your skill level in using the FTT?
    (0= not skilled; 10=incredibly skilled)
    0 – 1 – 2 – 3 – 4- 5 – 6 – 7 – 8 – 9 – 10

13. Please indicate to what extent you agree or disagree with the following statement:
    'The FTT is a highly demanding physical test for children who participate in the Fitkids program'.
    - ☐ Highly disagree
    - ☐ Disagree
    - ☐ Agree nor disagree
    - ☐ Agree
    - ☐ Highly agree

14. Please indicate to what extent you agree or disagree with the following statement:
    'Conducting the FTT is time consuming'.
    - ☐ Highly disagree
    - ☐ Disagree
    - ☐ Agree nor disagree
    - ☐ Agree
    - ☐ Highly agree

15. Please indicate to what extent you agree or disagree with the following statement:
    'I am sufficiently trained in using the FTT'
    - ☐ Highly disagree
    - ☐ Disagree
    - ☐ Agree nor disagree
    - ☐ Agree
    - ☐ Highly agree

16. Please report reasons for not using the FTT in clinical practice

17. I am interested in additional training in using the FTT
    - ☐ Yes
    - ☐ No

18. Is the FTT of additional value in exercise testing in children with a chronic disease or disability?
    - ☐ Yes, the FTT is of additional value
    - ☐ No, the FTT is not of additional value

19. What is, to your knowledge, the additional value of the FTT?

20. Children are not permitted to hold onto the front handrail during the test.
    Are you aware of that?
    - ☐ Yes
    - ☐ No

21. Please indicate whether you agree with the following statement: ‘children often reach their maximal effort when performing the FTT (HR_{peak} >180 beats·min^{-1})’.
    - ☐ Yes, I agree
    - ☐ No, I don’t agree

22. A reason for terminating the FTT before maximal exercise is:
    (multiple answers possible)
    - ☐ Painful legs
    - ☐ Walking on the treadmill with increments in speed and inclination is too difficult
    - ☐ The FTT is difficult because holding onto the front handrail during the test is not permitted
    - ☐ Other, namely

23. Reference values of the FTT in healthy Dutch children were published.
    Are you aware of that?
    - ☐ No, I am not aware of the reference values of the FTT
    - ☐ Yes, I am aware of the reference value of the FTT

24. The main test outcome of the FTT is time to exhaustion, which is the total duration of the test minus the warm-up phase.
    Are you aware of that?
    - ☐ Yes, I am aware
    - ☐ No, I am not aware

CHAPTER 7

Monitoring physical activity in primary pediatric physical therapy practice: difficulties, opportunities and a case study illustration

ABSTRACT

Background: To use physical activity (PA) as an outcome measure for physical therapy interventions, use of feasible assessment tools to objectively measure PA in clinical practice are essential.

Purpose: To explore the 'added value' and feasibility of objectively measured PA in pediatric physical therapy practice in comparison to currently used subjective questionnaires.

Methods: Twelve children wore an Activ8 accelerometer continuously for 7 days and filled out the Dutch Standard PA Questionnaire for youth at baseline, and after 3 and 6 months of training. The added value was determined by differences in outcomes with regard to amount, type, context and bouts of PA. An in-depth data analysis on one case was presented to illustrate these differences in more detail. Feasibility of the Activ8 was defined by wear time, reasons for non-compliance and measurement issues, children's acceptability, and therapist's experiences.

Results: Subjectively measured PA overestimated objective measured PA. The in-depth analysis showed that the value of using objective data in pediatric physical therapy lies in more detailed description of PA per day and differences between weekdays and weekend days. Feasibility was limited due to difficulties applying an accelerometer, reluctance of children and their parents to wear an accelerometer for 7 days and technical difficulties in reading and charging the accelerometer.

Limitations: This study was limited by the small and clinically heterogeneous sample of patients included and the small and (self) selected group of therapists.

Conclusion: Objectively measured PA has additional value in clinical decision making as it gives therapists more detailed information on PA. At the same time, steps have to be taken in optimizing accelerometers for use in physical therapy practice.

Submitted as

Kotte EMW, Veenhof C, Winkler AMF, Horemans HLD, Takken T, de Groot JF. Monitoring physical activity in primary pediatric physical therapy practice: difficulties, opportunities and a case study illustration.

INTRODUCTION

Over the past decade, countless studies reported on the health benefits of sufficient physical activity (PA) and there is indisputable evidence of the effectiveness of regular PA in the prevention of several chronic diseases and premature death.[195-197] PA is also being increasingly recognized as an important strategy for disease management.[198] From this perspective, the American College of Sports Medicine (ACSM) launched '*Exercise is Medicine*' as a global health initiative. Priority of this initiative is to encourage health care professionals to include PA when designing treatment plans and to refer patients to evidence-based exercise programs.

To include PA as an outcome measure for pediatric physical therapy interventions, feasible assessment tools to measure PA in physical therapy are needed.[49] In current practice, PA is often assessed in a subjective, self-reported, manner. Subjective measures have practical and cost-saving advantages. However, given the multi-dimensional character of PA, self-reports are easily biased as children might have difficulty recalling their past activity behavior.[51] Questionnaire assessment is also greatly complicated in young children by their propensity for repeated brief bouts of vigorous PA.[52] Additionally, questionnaires are entirely dependent on a person's subjective assessment, which depends to a large extent on memory, time, personal characteristics and current social norms and values.[199]

Objective PA data might serve as input for more optimal counselling of parents and children to adopt more active lifestyles. However, objective measures have distinct strengths and weaknesses as well. There will always be a trade-off between accuracy and available resources when choosing the best approach for measuring PA amongst youth.[200] In recent years, a growing body of literature on clinimetric quality of commercially available accelerometers has become available for both use in healthy participants[201-203] and the assessment of PA in youth.[204-206] So far, these accelerometers have mostly been developed and used in research and not in clinical practice.

Using accelerometers in pediatric physical therapy practice might be of added value to overcome the limitations of subjective questionnaires. Accelerometry could provide more precise and detailed information on PA, which can be useful in clinical decision making and PA counselling. Use of telemonitoring applications or monitoring techniques like accelerometers are expected to keep healthcare accessible and affordable for persons with chronic medical conditions (CMCs) without a trade of in quality of care.[207,208] At the same time, large-scale use of e-health applications strongly depends on whether healthcare users and healthcare providers experience enough added value from the e-health application in comparison to current practice.[55] Therefore, this study aims to (1) explore the added value of using an accelerometer to monitor free-living PA in children with CMCs in pediatric physical therapy practice in comparison to subjective PA questionnaires (part 1 of the study), (2) investigate the feasibility of using an accelerometer to monitor free-living PA in children with CMCs (part 2 of the study). This pilot study is an essential first step towards a larger intervention trial on the effect of the Fitkids intervention (an exercise therapy program in the Netherlands for children with CMCs) on PA levels of children with CMCs.

METHODS

Training of participating therapists

In September 2015 and June 2017, we initiated two training sessions for Fitkids physical therapists who voluntarily agreed in participating in this practice-based study. Both training sessions were followed by a period of 7 months in which children referred to the Fitkids exercise therapy program were asked to participate in the study. Main goal of the training sessions for therapists was to become familiar with the research protocol, the accelerometer, the accelerometer wear requirements (placement and fixation) and the accelerometer software. During the training we provided therapists with a web lecture and interactive workshop on the barriers and facilitating factors of PA in children and adolescents with physical disabilities. Data were collected between September 2015 and September 2017.

Study sample

Children and their parents were invited to participate in this study when entering the Fitkids program. According to the Dutch Medical Research Involving Human Subjects Act, our study did not require ethics approval. The children and their parents individually received written and verbal information about the study and an informed consent form. Written informed consent was obtained from the parents or legal guardian of each child, and also separately from each participant aged 12 years and older.

Fitkids intervention

The Fitkids intervention consisted of exercise training and PA counseling. Participating children received a graded exercise program for 6 months as previously described.[186] Children participated in the exercise training twice a week for 1 hour in the first 3 months and 1 hour per week during month 4 to 6 under supervision of a pediatric physical therapy practice. The main components of PA counseling were: (1) increasing awareness of parents and children on the benefits of regular PA for children with CMCs, (2) providing information on the possibilities of increasing PA in daily life and (3) finding a suitable sport to participate in after the Fitkids program as sport participation is one of the domains, besides playing outside and active transport, that contributes to daily PA.[17,20] For more detailed information on the Fitkids program please contact the Fitkids foundation (info@fitkids.nl) or visit www.fitkids.nl.

Measures and procedure

At baseline, data regarding age (yr), sex (male/female), medical condition, body weight, and body height were collected. Body mass index (BMI in kg/m^2) was derived from weight and height and compared with reference values for healthy subjects matched for age and sex. Z-scores were calculated using the growth TNO calculator at https://groeiweb.pgdata.nl/calculator.asp. Data on PA was collected at three points: at baseline and after 3 and 6 months of training. Data were collected as part of standard care and de-identified before being sent to the existing Fitkids database.

Objectively measured PA

Activ8 Professional Accelerometers (2M Engineering, Valkenswaard, The Netherlands) were used to collect objectively measured PA. The Activ8 is a commercially available small device (30x32x10 mm) that measures acceleration in three planes. The Activ8 aims to detect the actual PA in terms of body postures and movements (i.e., sitting, standing, walking, cycling, and running). The algorithm used has recently been validated for use in typically developing youth and ambulatory youth with a motor disability.[49] The wear requirements (i.e., thigh placement and skin taped fixation using Tegaderm (3M)) used to validate the algorithm were also used in the current study. The Activ8 was wrapped in household film guarantee the device being waterproof. The children wore the device continuously for 7 consecutive days at the three data collection points. Children and their parents were blind to all activity data collected while wearing the device.

For data analyses, first wake time and sleep onset were determined by visual inspection of the raw data. Wake time was defined as the detection of standing or walking in the morning time continuously for a couple of minutes. Sleep onset was determined by the absence of upright activities, continuously for a couple of minutes around bedtime. To ensure representativeness of PA behavior, the criteria for analysis were: (1) > 10 hours awake time per day,[209-212] (2) at least one measured weekend day and at least two weekdays.[209] Subsequently, Activ8 output was analyzed in Excel to quantify minutes walking, cycling and running. Custom-written Matlab scripts were used to generate graphs. With respect to objective measured PA, two outcome measures were defined. The number of minutes a child spent in walking, cycling and running activities during the day (i.e. time between wake time and sleep onset) was defined as an indication of total PA. Subsequently, the number of minutes a child spent in cycling and running activities was defined as an indication of moderate-to-vigorous PA (MVPA). MVPA is a category of activity intensity that is strongly associated with increased health benefits. MVPA is defined as any activity over 3 METs. A MET is a ratio of a person's working metabolic rate compared to their resting metabolic rate. At this moment, the intensity and cut-off points of the Activ8 have not been validated, and therefore we choose to use the youth compendium of physical activities[213] to estimate the energy costs of physical activities. According to Butte et al[213], slow walking up to 3.2 km/ hour does not exceed 3.0 MET in children under 12 years of age, whereas walking at higher speed does as well as riding a bike and running at various speed.

Subjectively measured PA

The Dutch Standard Physical Activity Questionnaire for youth was used for collecting subjective measured MVPA.[214] This questionnaire is currently used in Fitkids centers and designed to monitor population-based trends in general MVPA levels and recreational sedentary activity in youth. The PA questions have been validated.[214] To assess weekly MVPA, the frequency and average duration of four specific types of PA during a typical week were added together, using the statistic syntax of the authors. The types of PA included: (1) walking or cycling to school, (2) sports at school, (3) sports at a club, and (4) sports outside a club (children ≥12 years of age) or playing outside (children younger than 12 years of age). Parents filled out the questionnaire for children younger than 12 years of age. Children ≥12 years of age filled out the questionnaires themselves.

Study design

Part 1: The 'added value' of using objective measures in the assessment of PA

Added value was determined by differences in available information from both objectively and subjective measured PA with regard to amount, type, context and bouts of PA. An in-depth data analysis on one case was presented to illustrate these differences in more detail and to show how this information could be used in clinical decision making in pediatric physical therapy practice.

Part 2: The feasibility of the Activ8

Different aspects of feasibility were assessed in this study. First, to capture accelerometer adherence, the number of valid days of PA monitoring (i.e., ≥10h) was determined as well as average daily wear time in hours. Second, reasons for missing data were outlined (e.g., non-compliance and measurement issues). Third, children's perception of the acceptability of wearing the accelerometer were assessed together with the therapist's experiences with applying the Activ8 in clinical practice to examine subjective feasibility. Children were asked to report their experience with wearing the accelerometer by asking them: 'What is your experience with wearing the accelerometer?' Data from therapist's experiences were collected by semi-structured telephone-based interviews during the intervention period and additional e-mail contact.

RESULTS

In total, 16 physical therapists participated in the present study (13 pediatric physical therapists, 2 pediatric physical therapist students and 3 general physical therapists). The mean age of the participating physical therapists was 39.0 yr and their mean working experience was 16 yr.

Study population

In total, 12 children with a variety of diagnoses were included. A flowchart of the inclusion procedure is shown in Figure 1. Demographic characteristics of the included cases are depicted in Table 1. One child (case 7) dropped-out after 3 months of training because of progressive knee problems.

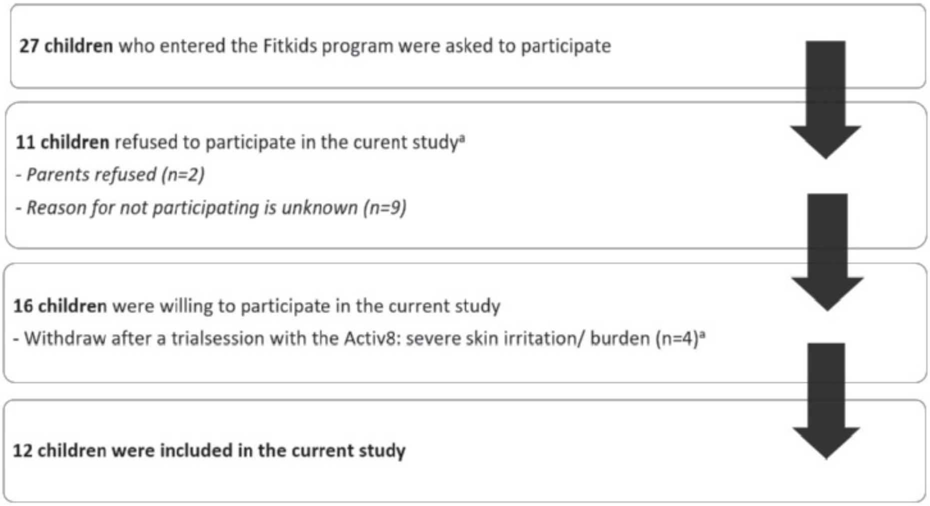

Figure 1. Flowchart of the inclusion procedure.
[a] These children did not participate in the study but continued the Fitkids program.

Table 1. Demographic characteristics of participants.[a]

	M/F	Age (yr)	Height (m)	Weight (kg)	Weight for age (SDS)	Height for age (SDS)	BMI for age (SDS)	Medical condition
Case 1	F	12	1.48	51.3	1.0	-1.28	1.98	DCD
Case 2	F	13	1.64	63.6	1.38	0.03	1.72	Exercise-induced asthma
Case 3	F	9	1.41	39.8	1.63	0.27	1.73	Hypothyroidism
Case 4	F	9	1.38	46	2.5	0.05	2.86	Hypermobility syndrome
Case 5	M	9	1.50	38	1.23	1.15	0.51	Autism-spectrum disorder
Case 6	M	11	1.45	57	2.41	-0.86	3.26	Orthopedic disability
Case 7	F	11	1.46	45.3	0.86	-0.98	1.56	Hypermobility syndrome
Case 8	F	9	1.34	43.3	2.22	-0.59	2.85	Intellectual disability
Case 9	M	14	1.64	75.8	2.0	-1.18	2.82	PDD-NOS
Case 10	M	10	1.50	70.7	3.70	0.52	4.14	Motor developmental delay
Case 11	M	10	1.39	28.6	-1.01	-0.78	-0.94	PDD-Nos & Orthopedic disability
Case 12	M	9	1.50	47.1	2.37	1.08	2.23	Orthopedic disability

[a] SDS: Standard Deviation Score; DCD: Developmental Coordination Disorder; PDD-Nos: Pervasive Developmental Disorder.

Part 1: The value of using objective measures in the assessment of PA

In Table 2 objective and subjective measured PA at the three data collection points are presented. Based on objective measures, the median time spent in walking, cycling and running activities was on average 163 minutes/day, 185 minutes/day and 165 minutes/day on T0, T1, and T2, respectively. The median time engaged in MVPA, based on cycling and running activities, was 35 minutes/day, 43 minutes/ day, and 33 minutes/day on T0, T1 and T2, respectively. Based on self-reported measures, the time spent in MVPA was on average 41 minutes/day, 75 minutes/day, and 53 minutes/day on T0, T1 and T2, respectively. Results show a significant difference between subjective reporting's and objective measured *MVPA* (*P*=.005), with self-reporting's overestimating objective measures. The observed overestimation was on average 211% (224 minutes per week).

Table 2. PA of children at the three data collection points.

		Activ8				**Dutch Standard PA Questionnaire for youth**		**Diff *MVPA*[a]**
		Measurement days	*Walking, cycling & running*	*MVPA (cycling & running)*	*Total*	MVPA		
		n	*min/day*[b]	*min/day*[b]	*min/week*[c]	*min/week*	*min/day*[d]	
Case 1	**T0**	7	127	105	758	405	58	353
	T1	7	204	83	563	495	71	68
	T2	7	191	47	420	420	60	0
Case 2	**T0**	6	71	18	160	240	34	-80
	T1[e]	-	-	-	-	255	36	
	T2	7	105	31	192	150	21	42
Case 3	**T0**	-	-	-	-	225	32	
	T1	6	142	28	194	270	39	-76
	T2	7	162	15	110	-[f]	-	
Case 4	**T0**	6	178	18	139	-[f]	-	
	T1	7	160	24	188	-[f]	-	
	T2	6	175	113	761	-[f]	-	
Case 5	**T0**	6	209	44	284	-[f]	-	
	T1	6	188	23	160	-[f]	-	
	T2	5	154	29	251	-[f]	-	
Case 6	**T0**	7	165	37	298	-[f]	-	
	T1	6	211	97	665	-[f]	-	
	T2	-	-	-	-	-[f]	-	
Case 7	**T0**	6	140	28	211	260	37	-49
	T1	7	124	28	186	350	50	-164
	T2	-	-	-	-	-	-	
Case 8	**T0**	7	239	6	77	225	36	-148
	T1	6	266	31	204	225	36	-21
	T2	6	227	19	152	225	36	-73
Case 9	**T0**	-	-	-	-	-	-	
	T1	7	113	4	34	45	6	-11
	T2	6	99	4	48	45	6	3

7

Table 2 (continued). PA of children at the three data collection points.

		Activ8				Dutch Standard PA Questionnaire for youth		Diff *MVPA*[a]
		Measurement days	*Walking, cycling & running*	*MVPA (cycling & running)*	*Total*	MVPA		
		n	*min/day*[b]	*min/day*[b]	*min/week*[c]	*min/week*	*min/day*[d]	
Case 10	**T0**	3	130	8	63	180	26	-117
	T1	6	200	22	207	450	64	-243
	T2	6	188	42	240	400	57	-160
Case 11	**T0**	6	170	48	296	395	56	-99
	T1	3	244	71	497	1630	232	-1133
	T2	6	157	25	184	450	64	-266
Case 12	**T0**	6	199	29	212	395	56	-183
	T1[e]	-	-	-	-	1025	146	
	T2	5	195	18	175	935	134	-760
Average	***T0***	6	*163*	*35*	*250*	*291*	*41*	
	T1	6	*185*	*43*	*290*	*527*	*75*	
	T2	6	*165*	*33*	*253*	*375*	*53*	

[a] Difference in MVPA: total weekly MVPA obtained using the accelerometer minus total reported MVPA obtained using the questionnaire.
[b] Data presented as median values.
[c] If accelerometer data was not based on 7 days, we choose to adjust accelerometer data to 7 days by dividing total MVPA by number of valid measurement days and multiplying outcome with 7.
[d] Daily MVPA measured by use of the questionnaire is calculated by dividing weekly MVPA by 7.
[e] The T1 measurements of case 2 and case 12 were excluded because these measurements did not include a valid weekend day.
[f] Questionnaires were lost by the treating physical therapist.

In-depth data analysis on one case

Case description

Case 10 was a ten-year-old boy with a sustained motor developmental delay and learning difficulties. He lived with his older brother and parents and had attended special education since first grade of primary school. He received additional physical therapy to adjust for his reduced ability to learn and automate motor skills. Performing dual tasks remained difficult and he was unable to ride a bike on his own at baseline. He had no intrinsic motivation to exercise and did not experience any pleasure doing it. His parents requested for help to increase his physical fitness, discover the fun of exercise and increase participation in PA. A plan of care to address activity limitations and participation restrictions was developed by the treating pediatric physical therapist.

Objective versus subjective measures

Based on accelerometer measures, the total time this boy spent in walking, cycling and running activities can be presented and the variation becomes clear (Fig 2A), for example within one week: between weekdays, and between weekdays and weekend days. In this specific case, figure 2B shows that at T0, the boy spent less than 20 minutes/day in MVPA, without much variation between the days. His objectively assessed MVPA increased to 240 minutes/week at T2 (207 minutes at T1), while also showing more variation between the days. Subjectively measured MVPA increased to 450 minutes/week at T1 (360 minutes from playing outside and 90 minutes from PA classes at school) and 400 minutes/week at T2 (175 minutes of PA from active transport, 135 minutes from playing outside and 90 minutes from PA classes at school), an overestimation of approximately 100%. The difference between perception and reality in general could be a good starting point for increasing awareness of PA.

At the same time, differences between days in time spent in the various activities (walking, cycling or running) may give the therapist relevant feedback for PA counseling. Also, insight in time spent in sedentary activities (lying and sitting) and static activity (standing) during wake hours might contribute to an improved physical behavior. Figure 3 present the different activities on the 25th of February, which is a measurement day at T0 (Fig 3A), and on the September the 11th, which is a measurement day at T2 (Fig 3B). From these figures, the variation between days becomes clear immediately: on the 11th of September, this boy shows a slight increase in cycling activity whereas he was less active in walking compared to the 25th of February.

Figure 4 provides insight in the specific time ranges over the day in which a child accumulates time spent in specific postures (lying, sitting or standing) and activities (walking, cycling or running). From these plots, it can be concluded that this boy accumulates more walking activities in the morning time than the rest of the day and that the time spent sitting increases during the day. For therapy purposes, this plot can be used to gain insight into the specific time frames with considerable potential for improving physical behavior. Comparing the weekend day (Figure 4A) with both weekdays (Figure B and C) it can be seen that, for this boy, the distribution of PA over the day differs between week days and weekend days in a way that he mainly accumulates his activity between 8:30 AM and 2:00 PM during weekdays and between 11:30 AM and 4:00 PM during weekend days. Moreover, Figure 4 clearly points out that, during weekdays, this boy accumulates a level of higher activity during school time and there seems potential for improving physical behavior in the time period when school is finished (see plot B and C in Figure 4).

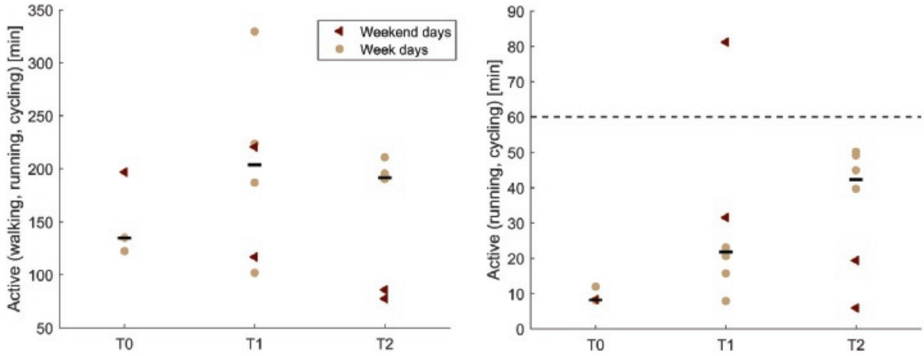

Figure 2. Data present the number of minutes the boy was active during weekdays and weekend days. Data presented for T0, T1, T2. The flat dash presents the median number of minutes the boy was active. In figure 2A, the number of minutes spent in walking, cycling and running activities are outlined, whereas in figure 2B, minutes running, and cycling are outlined as an indication of MVPA. The dotted line in figure 2B represents the recommended norm of 60 minutes of MVPA per day.

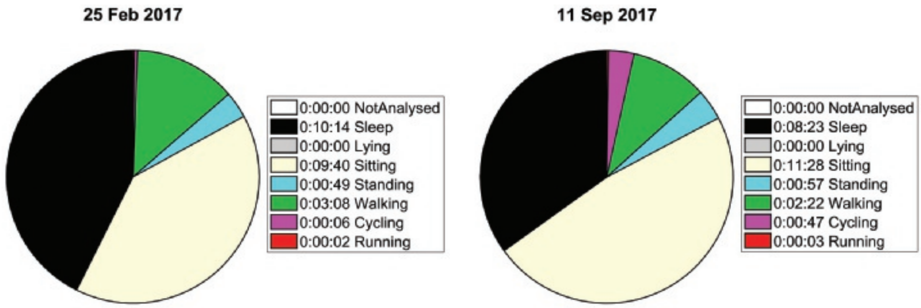

Figure 3. Activity pie presenting the time spent in different categories of physical and sedentary activities. Figure A: 25th of February 2017 and figure B: 11th of September 2017.

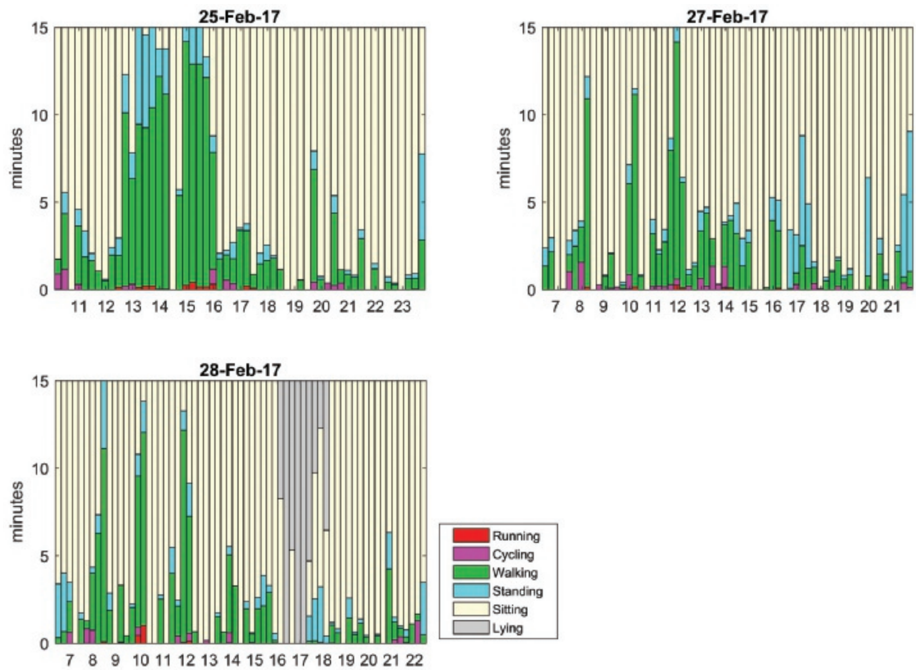

Figure 4. Distribution of the different categories of physical and sedentary activity over the day. A bar represents 15 minutes and data is based on awake time. 25th of February 2017: weekend day; 27th of February 2017: weekday; 28th of February 2017: weekday.

Part 2: The feasibility of the Activ8

Accelerometer adherence

Table 3 presents an overview of number of valid days per case and mean daily wear time. In total, 19 out of 252 days were excluded from analysis because less than 10 hours of accelerometer data were recorded (8%). Other reasons for non-compliance or measurement issues were: (1) the Activ8 did not record any activity during the week, possibly because of an error during the initialization of the Activ8, (2) the Activ8 was removed one day earlier because of a planned holiday, (3) missing measurement due to difficulties in saving data file by the physical therapist, (4) the child removed the Activ8, (5) low battery of the Activ8. In total, 37 out of 252 measurement days were excluded based on these reasons (15%). Based on the valid measurement days, the average daily wear time was 14.0 hours at T0 and 14.4 hours both at T1 and T2.

7

Table 3. Description of valid measurement days, mean daily wear time and reasons for missing data.

	T0			T1			T2		
	Measurement days (n)		**Daily wear time (hours)**[a]	**Measurement days (n)**		**Daily wear time (hours)**[a]	**Measurement days (n)**		**Daily wear time (hours)**[a]
	Week	Weekend		Week	Weekend		Week	Weekend	
Case 1	5	2	14.5 ± 1.5	5	2	14.4 ± 2.1	5	2	14.8 ± 1.8
Case 2	4[b]	2	12.0 ± 1.1	5	0[b]	14.2 ± 1.6	5	2	13.7 ± 1.6
Case 3	-[c]	-	-	4[b]	2	15.1 ± 1.2	5	2	14.7 ± 1.9
Case 4	4[b]	2	14.0 ± 1.6	5	2	13.5 ± 1.8	4[b]	2	15.1 ± 1.2
Case 5	4[b]	2	14.7 ± 0.9	4[b]	2	14.6 ± 0.5	3[b,d]	2	14.4 ± 0.9
Case 6	5	2	13.0 ± 2.2	4[b]	2	15.4 ± 1.5	-[e]	-	-
Case 7	4[b]	2	13.3 ± 1.4	5	2	12.8 ± 1.3	-[f]	-	-
Case 8	5	2	14.1 ± 0.7	4[b]	2	14.0 ± 0.5	4[b]	2	14.6 ± 0.7
Case 9	-[c]	-	-	5	2	13.9 ± 2.5	4[b]	2	14.7 ± 0.6
Case 10	2[g]	1[g]	15.2 ± 1.3	4[b]	2	13.9 ± 1.1	4[b]	2	13.9 ± 2.6
Case 11	4[b]	2	14.5 ± 0.6	2[h]	1[h]	15.3 ± 1.2	4[b]	2	14.6 ± 0.5
Case 12	4[b]	2	14.6 ± 1.6	2[h]	0[h]	15.4 ± 1.7	3[g]	2	13.7 ± 1.4

[a] Data presented as mean ± SD.
[b] Exclusion of measurement day because of less than 600 minutes awake.
[c] Activ8 did not record any activity during the week, possibly because of an error during the initialization of the Activ8.
[d] Activ8 was removed one day earlier because of a planned holiday of the child.
[e] Missing measurement.
[f] Drop-out because of progressive knee problems.
[g] Child removed the Activ8.
[h] Low battery.

Acceptability by children in wearing the Activ8

Two children indicated that they did not experience any problems wearing the accelerometer (case 11 and 12), cases 1 and 9 found it just a bit annoying whereas 7 children indicated skin irritation (redness and/ or itching; case 2-6, 8, 10). Case 7 did not rate her experience with the accelerometer because of early drop-out. At the same time, children and parents complained about the fact that the skin tape did not stick to a sweaty skin.

Experience of therapists with applying the Activ8

From the semi-structured telephone-based interviews and additional e-mail contact it became clear that using the Activ8 was difficult in several groups of children. The adolescents often refused to participate as they were afraid of reactions of others who probably could see something being attached to their upper leg (embarrassment of accelerometer visibility though clothing). Moreover, using the device in children with autism or related problems was challenging because of too many stimuli. In addition, the use of a device in children with cognitive problems appeared to be challenging and some children and parents refused to participate as they did not like the fact that other persons had insight in their (or their child's) activity levels. At the same time, some parents saw the added value of having precise information on activity levels, but their children did not. Some therapists reported that part of the population of children participating in Fitkids is too complicated for using this technology.

DISCUSSION

With this study we aimed to (1) explore the 'added value' of using an accelerometer to monitor free-living PA in children with CMCs in pediatric physical therapy practice in comparison to subjective PA questionnaires, (2) investigate the feasibility of using an accelerometer to monitor free-living PA in children with CMCs.

Part 1: The value of using objective measures in the assessment of PA

As PA is traditionally assessed by anamnesis or questionnaire in physical therapy practice, it was of interest how objective data relate to self-reported data. A discrepancy was found between both methods, with self-reported MVPA overestimating objectively measured MVPA. This observation is consistent with findings of a systematic review reporting an average difference between accelerometer and indirect measures of 147%.[215] In a study of Leblanc and Jansen, self-reported MVPA was over-estimated by an average of 183%[216], compared to 211% found in our study. In a more recent study, it was reported that the estimation of MVPA by questionnaires was higher by 117.6 minutes per week,[217] compared to 224 minutes per week found in our study. The added value of using objective measures in the assessment of PA is reflected in the discrepancy found. While self-reported measures are useful for capturing the context of the activity, the value of using objective data in the evaluation of PA in pediatric physical therapy practice lies in more detailed information (e.g., a detailed description of PA per day, differences between weekdays and weekend days and the possibility for real time feedback). This detailed information is precisely where a window of opportunity exists for creating individualized advises on integrating (more) PA in daily life of inactive or semi-inactive children and adolescents with CMCs. Moreover, a comparison of both objective and subjective PA measures might probable be most promising for use in clinical practice as it might increase awareness on misperceptions of PA levels. This awareness seems essential for realizing a behavior change towards a more active lifestyle.[218]

Using an accelerometer fits into the current changes in healthcare in which themes like self-management and efficiency of care become more evident.[219] There is a shift towards more technology supportive care throughout healthcare and the use of technology might also open doors to innovation in pediatric physical therapy practice. However, the technology should be useful within the context, fitting the needs of both children and physical therapists. With respect to using accelerometry in pediatric physical therapy, the device should be capable of capturing short bouts of PA changes which are typically for the pediatric population. Lankhorst et al[49] recently validated the algorithm which is used in the present study to classify separate postures and movements based on the orientation of the sensor with respect to gravity combined with movement intensity. They concluded that the algorithm had difficulties in detecting separate postures and movements during complex activities, similar to those in daily life, both in the typically as the non-typically developing youth. Misdetection was, among others, related to the (relative) inability of the algorithm to detect short-duration activities. Meanwhile, based on the results of Lankhorst et al[49] and others, the developers of the Activ8 adapted the algorithm to resolve the inaccurate detection. For broad-scale implementation of accelerometry in pediatric physical therapy practice it seems, however, recommendable evaluating the updated algorithm in a non-controlled free-living setting in children with CMC because the complex activities (i.e., a combination of postures and movements of short duration) included in the study of Lankhorst et al were executed in a controlled setting.[49]

Part 2: The feasibility of the Activ8 accelerometer

The feasibility of using the Activ8 accelerometer in pediatric physical therapy practice was limited. The difficulties in using the Activ8 in pediatric physical therapy practice are reflected in the low number of children participating in the present study. Only 27 children and adolescents were asked to participate, partly because the population of children participating in Fitkids was too complicated for being instructed to wear an accelerometer. This number may, however, also reflect the difficulty of implementing new technology in pediatric physical therapy practice in general. Moreover, uncertainty or unfamiliarity of therapists with accelerometry might have interfered parents' decision to participate in the present study. In addition, from the feasibility study it became clear that many children and adolescents did not like something being attached to their body. Placement and fixation of the Activ8 was of major concern and has interfered children's' decision to participate. Some therapists initiated a trial session in wearing the Activ8 and part of the children experienced itching and discomfort, resulting in children who did not want to participate. Based on this and the fact that 58% children of the included children reported (severe) skin irritation it can be concluded that skin taped fixation of the accelerometer is not most optimal for usual care in pediatric physical therapy practice, like is has been used in research.[49,220] The challenges of wearing an accelerometer for 7 days in pediatric physical therapy practice as outlined in this study count for the Activ8 in specific but are likely to be of concern when using other accelerometers as well.

Huygens et al emphasized that in developing e-health tools that are tailored to the needs of the users, it is important to include future users in the entire development process.[221] The Activ8 was primary developed for use in the adult population and for research purposes. This has resulted in adult-specific preferences and highly standardized (fixed) placement. Involving end-users (physical therapists, children and adolescents, and parents) from the onset of the development process would have probably led to better accelerometer wear requirements, design and usability. At this point, the involvement of physical therapists, children and adolescents, and parents is still recommended to modify Activ8 wear requirements with respect to attachment and placement which are useful in a free-living setting in children with CMCs.

Strengths and limitations

A strength of this study is that it was performed in physical therapy practice, rather than in a controlled, research setting. At the same time, this study was limited because of the small and clinically heterogeneous sample of patients included and the small and (self) selected group of physical therapists. Other limitations include: (1) the feasibility of the implementation of accelerometery in pediatric physical therapy practice (e.g., complexity of the accelerometer, required skills, required time for use and interpretation of the data) have not been investigated; (2) the Activ8 is not widely used in research and practice, (3) maybe the provided training for the therapists was not sufficiently enough.

Lessons learned

This study highlights the gap between what is useful in controlled research settings and in clinical practice in which measurements take places in non-controlled free-living settings. Moreover, self-reported measures seem to overestimate MVPA and therefore, therapists are advised to handle questionnaire-based MVPA data with care. Finally, developers of accelerometers are advised to include end-users in the development process.

Future research

Future research should focus on modifying the sensor wear requirements with respect to attachment and placement which is useful in a free-living setting in children with CMCs. Future research should also investigate the validity of the Activ8 in children with CMCs in an uncontrolled free-living setting using the modified sensor wear requirements. Additionally, cut-points for children with CMCs should be developed for accelerometry to estimate energy expenditure. To benefit from the advantages of objectively measured PA in clinical practice, the graphs and tables as outlined in this article should be implemented in the professional dashboard of the Activ8 and in the electronic medical record of patients. Finally, studies how to effectively implement objectively measured PA in pediatric physical therapy practice should be investigated and stakeholders and end-users should be involved in this process.

CONCLUSIONS

This practice-based study has shown the additional value of using an accelerometer to monitor free-living PA in children with CMCs in pediatric physical therapy practice. At the same time, feasibility was limited due to difficulties in applying an accelerometer, reluctance of children and their parents to wear an accelerometer and technical difficulties in reading and charging an accelerometer. Therefore, optimizing accelerometers for use in pediatric physical therapy practice is recommended parallel with increasing experiences with use and implementation of this kind of innovation or technology in pediatric physical therapy practice.

ACKNOWLEDGMENTS

The study was financed by the Johan Cruyff Foundation and the Dutch Foundation Innovation Alliance (SIA-RAAK), project number SIA RAAK PRO-4-03.

CHAPTER 8

General discussion, clinical implications and directions for future research

The main objective of this thesis was to investigate if the Fitkids exercise therapy program can improve physical fitness, walking capacity, HRQoL and participation in physical activity in children and adolescents with CMCs. A secondary objective was to investigate the clinimetric properties of the FTT as well as to describe sex- and age-related reference values for FTT performance in healthy children and adolescents 6-18 yr. The final two chapters of this thesis covered studies evaluating the applicability of new measurement instruments in physical therapist practice. Here, the findings of this thesis are placed in broader perspective and clinical implications as well as directions for future research are provided.

PART 1. FITKIDS INTERVENTION

Results of **chapter 2** showed the great heterogeneity of the population of children and adolescents participating in Fitkids, both in demographic characteristics and medical diagnoses. Results of this study also showed that children and adolescents participating in the Fitkids have reduced aerobic fitness levels compared to peers who are typically developing. Fitkids is an exercise therapy program that is implemented in local physical therapy practices and includes an unbiased clinical sample of youth with chronic medical conditions (CMCs) visiting these practices. Real-life setting interventions are often made available to a much broader audience than the restricted target group in most controlled trials.[222] Because of the heterogeneous target population as well as the flexibility of real-life interventions to fit into the disparate settings of clinical practice, it is not certain that findings from controlled studies can be directly translated into a real-life setting.[223,224] The results outlined in **chapter 3** add useful information to the body of evidence on effects of real-life interventions. As outlined in this chapter, the Fitkids intervention significantly improved health-related fitness, walking capacity and HRQoL in children and adolescents with CMCs.

Effects on health-related fitness and walking capacity

The results in our study, by using multilevel analysis, showed an average improvement of 24% in aerobic fitness after 6 months of training ($P \leq .001$). In comparison, Tomassoni and colleagues[225] found an improvement of 16% in endurance time on the modified Bruce treadmill test following a 12-week hospital based aerobic exercise program (2 times a week for 60 minutes) in children with a complex congenital heart defect after repair (4.5-15 years of age).[225] The length of the training program might explain the discrepancy found.

Furthermore, following the Fitkids intervention, an average improvement of 22% ($P \leq .001$) was found in anaerobic fitness measured by use of the mean power (W) derived from the Muscle Power Sprint Test (MPST). In contrast, Zwinkels and colleagues[28] recently found an improvement of 9% in mean power measured on the MPST following an 8 weeks high-intensity interval training (2 times a week for 20-24 minutes) in children with chronic conditions or physical disabilities (6-19 years of age). Both the length of the training program and the difference in anaerobic fitness at baseline (168 W vs 154 W in our study) might explain the discrepancy found. In addition, van Vulpen and colleagues[226] found an improvement of 83% in mean power on the MPST following a 14-week functional power training intervention (3 times a week for 60 minutes) in children with CP (4-10 years of age). This study clearly points out the value of training specificity (i.e., training the activity that needs improvement). The specificity of the strength training exercises in the study of van Vulpen ensured that the power generated by the plantar muscles was directly related to walking capacity. As compared with the restricted target group of children with CP included in the study of van Vulpen,[226] the Fitkids intervention is available for a much broader audience which hampers the ability of offering such a specific training in Fitkids.

Moreover, significant intervention effects of the Fitkids intervention were found on muscle strength. The descriptive statistics showed an improvement of 16% (4 points) in the total score of the BOT-2 subscale strength following the Fitkids intervention. The individual items of the BOT-2 subscale, analyzed by means of multilevel analysis, showed average improvements of 8% ($P \leq .001$) on distance from standing long jump and 47% ($P \leq .001$) on number of knee push-ups in 30s after 6 months of training. The average improvements found on the number of sit ups in 30s, the duration of the wall sit, and the duration of the v-up, after 6 months of training, were 24% ($P \leq .001$), 32% ($P \leq .001$) and 22% ($P \leq .001$), respectively. No significant differences in muscle strength were found between 3 and 6 months of training. Our results on distance from standing long jump are comparable with those found by Elbasan and colleageus.[227] They found a mean improvement of 10% on distance from standing long jump after a 6-week chest therapy and aerobic exercise training on a treadmill (3 times a week for 30 minutes) in young children (5-13 years) with cystic fibrosis.[227] Another training intervention comprised of both aerobic exercises and functional strength exercises also found similar effects.[228] Habers and colleagues[283] found an improvement of 4 points in total score on the BOT-2 subscale strength in the training group as compared to the waiting list control group in patients (8-18 years) with juvenile dermatomyositis following a 12-week home-based exercise intervention with additional functional strength exercises.

Furthermore, the children and adolescents included in **chapter 3** also improved their walking capacity following the Fitkids intervention. After 6 months of training, an average improvement of 36 m (6.7%) ($P \leq .001$) was found. Gruber and colleagues found and increase of 4% after a 4-6 weeks supervised exercise training program consisting of several sports activities (5 times a week for 45 minutes) in children with cystic fibrosis (6-18 years of age).[113] Both the training volume (15-22.5 hours vs 36 in our study) and the difference in baseline values (676 m versus 535 m in our study) might explain the discrepancy in results found. The improvement of 36 m observed in our study seems like a clinically meaningful improvement as a change of approximately 32 m in 6MWD is required to be considered clinically meaningful in individuals with chronic heart failure.[229] Additionally, clinimetric studies in adult patients with chronic conditions resulted in a minimal important change of 24 to 45 m.[103,104]

Effects on health-related quality of life

The Fitkids intervention improved child-reported and parent-reported physical and emotional functioning and also parent-reported total score of HRQoL (**chapter 3**). The results in our study, by using multilevel analysis showed average improvements ranging from 4.6% ($P \leq .001$) in child-reported physical functioning to 7.3%

($P \leq .001$) in parent-reported physical functioning. In contrast, the Fitkids intervention did not improve social functioning, such as playing with peers, and home functioning. These results are in line with those found in a systematic review on the association between exercise capacity, physical activity, and psychosocial functioning in children with congenital heart disease; a positive association was found between exercise capacity and physical domains of HRQoL, whereas a scare association was found between exercise capacity and psychosocial domains of HRQoL.[230] The absence of intervention effects on home functioning might also be explained by the fact that a ceiling effect was obtained for child-reported home functioning at baseline; 22% of the children reporting highest possible scores.[231] Moreover, the questions of the DUX-25 related to home functioning are quite delicate questions. An example question related to this domain is: 'When I think about my mother I feel',[105,232] this might have caused socially desirable answers. The fact that our population did not differ much from the reference group on self-reported ratings at baseline (SDS values between -0.2 and 0.2 on the different domains of HRQoL) might also have interfered results because participating in a standardized exercise therapy program is particularly beneficial for those with low baseline HRQoL scores.[233]

Effects on participation in physical activity

Although based on a small number of patients, preliminary data outlined in **chapter 7** reveal that there is no distinct relationship between participation in the Fitkids intervention and an increase in daily physical activity during the intervention period. However, unpublished data of that study pointed out that half of the children found an appropriate sport to participate in after the intervention period was competed. In this way, the Fitkids intervention might boost participation in physical activity post intervention.[18] A systematic review of Bloemen et al[234] clearly points out that there is evidence for no effect of physical training alone on objectively measured physical activity in children with physical disabilities. Although Fitkids has incorporated physical activity counseling additional to the exercise training this seems insufficient to elicit clear positive effects on participation in physical activity. The physical activity counseling incorporated in Fitkids focuses on: (1) increasing awareness of parents and children on the benefits of regular physical activity for children with CMCs, (2) providing information on the possibilities of increasing physical activity in daily life and (3) finding a suitable sport to participate in. A reason for the absence of clear intervention effects on participation in physical activity could be that the physical activity counseling components are insufficiently embedded in the intervention. This could, in turn, be explained by the fact that physical therapists mainly focus on bodily functioning rather than on participation as defined by the ICF.[31,234] This focus might result in an inadequate assessment of the individual determinants associated with participation in physical activities.[31,234] Determinants of physical activity are complex and multifactorial and include either personal factors (e.g., 'intention', 'attitude' and 'health condition', 'motivation') and environmental factors (e.g., 'lack of support from family', 'parental behavior', 'not being accepted by peers' or 'inadequate facilities').[235-240] Another reason for the absence of clear preliminary intervention effects on participation in physical activity might be that pediatric physical therapists find it difficult to support children and their parents to overcome the identified personal barriers to participation in physical activity.[241] An improved focus on changing behavior, for example by the inclusion of specific tools developed to support people in changing their behavior in the desired direction might help physical therapist in achieving treatment goals aimed at increasing participation in physical activity.[234,242,243]

In addition, to circumvent the barrier of 'limited possibilities in adapted sports', physical therapists should probably also increase focus on other pathways, than sport participation, to increase participation in physical activity in children with CMCs, like an increase in time spent in playing outside or an increase in active transport to school. In fact, given the results of recent research among parents commissioned by '*Jantje Beton*' (an organization dedicated to youth facilities in the Netherlands) increasing time spent in playing outside might be highly effective as results point out that 15% of the Dutch children never play outside.[244] The research, involving 12,000 parents, showed that children from the highest social class are least likely to play outdoors, in total 34% never did. By contrast, just 6% of children with the most disadvantaged backgrounds only played indoors. When aiming for an increase in time spent in playing outside, initiatives like the playground gang [In Dutch: Speeltuinbende] are highly valuable.[245] The 'playground gang' aims for accessible playgrounds for both the typically developing children as children with a disability so they can play together (www.speeltuinbendewijzer.nl). Additionally, from a recent national survey it became clear that active transport to school provides an excellent opportunity to increase leisure time physical activity in adolescents, especially when cycling.[18] In a study from New Zealand, it was concluded that a distance of approximately 2 km was associated with the best physical activity outcomes related to active transport (9% to 15% increase on weekdays).[246]

Furthermore, the involvement of parents in the determination of treatment goals aimed at participation in physical activity in physical therapist practice is also highly important since parental support is a significant determinant of children's participation in physical activity.[235,236,238] Research confirm the notion that parents are important because of an active role model,[247] because of parental support such as transportation to a sport club,[248] or because parents are of importance in developing sport identity (i.e., a concept that encompasses both the influence of movement skills, explicit sport knowledge (rules, tactics) and the social and cultural constraints on sport participation).[249,250] Sport identity is known to be an important determinant in long-term sport engagement.[251] The involvement of parents is also essential because they are often not aware of the importance of sufficient physical activity for their children.[252] Parents in turn report that their unfamiliarity with the local sport possibilities is an important barrier of sport participation of their children.[253] Therefore, pediatric physical therapists have an important role in the guidance of children with CMCs towards sport clubs.[252] An improved collaboration of pediatric physical therapists with the local physical activity sector (i.e., Care Sport Connector [in Dutch: *Buurtsportcoaches*][254] and sport professionals (i.e., trainers and coaches) might increase Fitkids intervention effects on participation in physical activity or sports of children with CMCs. An essential requirement for this is, however, that there are sufficient possibilities in adapted sports.[235-240] Fortunately, with new initiatives arising, the number of adapted sport facilities is still growing in the Netherlands. Examples of fairly new initiatives in adapted sports are 'race running', an initiative of the VuMC and the NSGK and, 'frame football', an initiative of the Cerebral Palsy Sport.[255,256] Acceptance of these initiatives within regular sports clubs may ensure inclusive sports.[257]

PART 2. FITKIDS TREATMILL TEST

When we initially started with the evaluation of the Fitkids intervention we faced the problem of the use of many local protocol modifications of the half-Bruce treadmill test. To overcome these concerns and the concerns regarding the utility of other established treadmill protocols to estimate aerobic fitness in children with CMCs in physical therapy practice,[47,126-128] we decided to develop a new protocol. The advantages of the FTT are three-fold: (1) the maximal incline of the protocol is restricted to 15% given the similarity with the maximal incline of standard treadmills ergometers available in local physical therapy practices, (2) the protocol starts with 0% incline making the protocol useful in children and adolescents with a disability or chronic disease and, (3) robust reference values for the typically developing youth between 6 and 18 yr are available.

From our study outlined in **chapter 4** it can be concluded that the FTT is a useful screening tool with good validity and reproducibility. According to the guidelines for correlations[140], VO_{2peak} measured during the FTT showed excellent correlation with VO_{2peak} measured during the Bruce protocol (r=0.90; P<.01). Moderate to strong correlations were found between cardiopulmonary variables (HR_{peak}, VO_{2peak} ($L{\cdot}min^{-1}$) and RER_{peak}) of both protocols. The prediction equations established **in chapter 4** can be used to predict VO_{2peak} reached on the FTT in children and adolescents who are healthy and between 6 and 18 yr. Furthermore, the reproducibility of the FTT can be considered as excellent with an intraclass correlation coefficient for time to exhaustion (TTE) of 0.985 (95% CI: 0.971 to 0.993; p<.001), meaning that the measurement error is small compared to the variability between the participants and that the discrimination of persons is hardly affected by measurement error. These results are comparable with those reported by Cumming et al[94] regarding the test-retest reliability of the Bruce protocol in school children between 7 and 13 years of age (correlation coefficient 0.94). The agreement analysis revealed narrow LOA (+1.30 to -1.43 minutes), indicating that the agreement of the FTT is good. A change score in TTE between 2 measurements within an individual can only be considered to represent a real change if it is outside the LOA. The reference values provided in **chapter 5** can be used by physical therapists to adequately interpret FTT performance. It is recommended to use the third percentile as cut-off point in daily clinical practice to indicate below normal FTT performance, as is also recommended in other studies.[143]

Although the FTT is a useful screening tool with good validity and reproducibility, there are a number of important issues to consider in order to enable successful evaluations of aerobic exercise capacity. A maximal exercise test, like the FTT, requires a maximal effort for adequate interpretation. From our study in **chapter 6** it became clear that there is a large group of children with CMCs who did not reach for a HR_{peak} of >180 beats·min^{-1}, an objective criterion of a maximal effort in children. From other research it is known that some children with CMCs have lower predicted HR_{peak}.[187] However, based on the results of **chapter 6** it became clear that there are a number of other reasons for a limited maximal effort, which are often linked to being unfamiliar with walking and running on an inclined treadmill ergometer. Therefore, we advise therapist to conduct a practice session before the actual test, as is also recommended for other field walking tests.[189] Our advice to conduct a practice session is justified by the fact that, in line with other studies,[94,149] no significant learning effect was observed in our study outlined in **chapter 4**. In addition, it is also the question whether physical therapists are too cautious in pushing children to reach for their maximal effort. There should be an enhanced attention on maximal exercise testing in the professional curriculum of pediatric physical therapists. Additionally, from clinical experience it is known that standardized assessment procedures of exercise test are frequently modified in clinical practice. There should be an increased awareness that, for instance, rail holding during the FTT limits the use of the reference values of that test.

PART 3. NEW MEASUREMENT TOOLS: APPLICABILITY IN CLINICAL PRACTICE

Pediatric physical therapists have expertise in key areas that are essential in the implementation and evaluation of health promotion and fitness programs, like Fitkids, for youth with CMCs.[150] In order to provide safe training recommendations and to evaluate whether interventions are successful, valid measurement instruments that are useful in physical therapy practice are indispensable. There is often a large gap between the development of measurement instruments in research and their use in clinical practice.[258] From research it is know that the implementation of a

8

new measurement instrument in physical therapy practice depends on several factors, including the measurement instrument itself.[180]

In **chapter 6** and **chapter 7** we have evaluated the applicability of two fairly new measurement instruments in physical therapist practice (in **chapter 6** the FTT and in **chapter 7** the Activ8). As previously described in this chapter, the FTT is a maximal treadmill test for the assessment of aerobic fitness, whereas the Activ8 is a commercially available accelerometer that measures physical activity in terms of body postures and movements. The development of the FTT was based on a practical request articulated by physical therapists in Fitkids practices (i.e., physical therapy practices affiliated with the Fitkids Foundation) and tailored to the needs of both children with CMCs and physical therapists. It can therefore be assumed that the test itself facilitates its use. In addition, the clinical utility of the FTT has been demonstrated in **chapter 6** which further promotes use of the FTT in physical therapy practice. In contrast, the feasibility of the Activ8 in physical therapy practice was limited as outlined in **chapter 7**, meaning that the Actc8 itself hampers uptake in physical therapy practice. The Activ8 was primary developed for use in the adult population and for research purposes. This has resulted in adult-specific preferences and a highly standardized placement of the accelerometer (i.e., thigh placement and skinned taped fixation), which in turn has led to moderately acceptability of the Activ8 by children and adolescents with CMCs because of (severe) skin irritation. As these wearing requirement has also interfered children's decision to participate in our study, skin taped fixation of an accelerometer is not advised for usual care in physical therapy practice. Optimizing sensor wear requirement with respect to attachment and sensor placement which is useful in a free-living setting in children with CMCs is warranted.

Except from the measurement instrument itself, there are other factors related to the uptake of a measurement instrument in physical therapy practice: (1) factors related to the physical therapist' competence (e.g., knowledge) and attitude (e.g., being convinced of the added value), (2) factors related to the organization (e.g., availability or time investment), and (3) factors related to patients (e.g., patient are not familiar with measurement instruments).[172,174,180] Results of the online survey outlined in **chapter 6** points out that the most dominant attitude related facilitator for use of the FTT in physical therapy practice was the fact that 83% of the therapists indicated to be convinced of the added value of the FTT to the quality of exercise testing in clinical practice. Based on the results outlined in **chapter 7**, there is no doubt that using an accelerometer to monitor free-living physical activity in children and adolescents with CMCs is of added value in comparison with subjective questionnaires. The added value is reflected in the significant overestimation of subjective reporting's, the more detailed information, the difference between days and the possibility for real time feedback. However, physical therapists might evaluate the added value far less positive because they experienced major difficulties in applying the Activ8 in several groups of children with CMCs (**chapter 7**). The difficulties in using the Activ8 were also reflected in the low number of children participating in the study outlined in **chapter 7**. As discussed in that chapter, unfamiliarity of therapists with accelerometry in clinical practice might have interfered parents' decision to participate. Although therapists were trained in using the Activ8, the training might not have been sufficient, particularly given the fact that physical therapists are less familiar with measurement instruments used to evaluate effects on participation rather than on bodily functioning and the difficulty of integrating new technology in daily physical therapy practice. In contrast, our study on the use of the FTT in clinical practice (**chapter 6**) was preceded by a long period in which information on the FTT was disseminated and training days were organized to ensure that physical therapists got familiar with the protocol. Users of the FTT rated a mean knowledge score of 7.4 (1-10), indicating that the knowledge transfer activities were effective (**chapter 6**).

Based on our findings, we have formulated recommendations for future development of measurement instruments for use in physical therapy practice: (1) the development should be based on practical requests articulated by physical therapists, (2) it is important that the measurement instrument is developed for a specific context (research vs clinical practice) and for a specific population (adults vs children), (3) as the measurement instrument should fit the needs of the end-users (i.e., physical therapists, children and adolescents and parents), it is highly important to include end-users in the entire development process.[221]

CLINICAL IMPLICATIONS

- As indicated by our findings, the Fitkids intervention improved health-related fitness, walking capacity and some HRQoL domains in children and adolescents with CMCs. By incorporating the Fitkids program in clinical practice, therapists are able to help children and adolescents with CMCs to improve their physical and mental health. Preliminary results reveal that there is no distinct relationship between participation in the Fitkids intervention and an increase in daily physical activity during the intervention period. This thesis, however, includes several recommendations to improve future intervention effects on participation in physical activity in children and adolescents with CMCs.

- When assessing physical fitness in children and adolescents with CMCs, several maximal exercise tests can be used. If one is limited to a treadmill ergometer with maximal incline of 15% and when someone does not have the availability of a mobile gas analysis system, the FTT can be used to estimate aerobic fitness from FTT performance. The FTT is a feasible, valid and

8

reproducible exercise test to estimate aerobic fitness in children and adolescents who are healthy. FTT performance and body mass can be used to predict VO_{2peak} in healthy children and adolescents between 6-18 yr. A change score in TTE between two concessive measurements within an individual can be considered to represent a real improvement if TTE is improved by 1.30 min or more. Finally, our results indicate that the FTT is useful in different diagnostic groups.

- Our results indicate that objectively measured physical activity is of 'added value' in comparison with subjective questionnaires as it offers detailed information on physical activity which can serve as input for more optimal counselling of children and adolescents (and their parents) to increase participation physical activity.

DIRECTIONS FOR FUTURE RESEARCH

Identification of subgroups for whom the Fitkids intervention is more or less effective

Our studies have shown that the Fitkids population is highly heterogenous. Monitoring intervention effects in a larger sample of children and adolescents referred to the Fitkids program might ensure the identification of subgroups for whom the Fitkids intervention is more or less effective.

Evaluation of intervention effects

The current thesis research objectively illustrated the short-term effects of the Fitkids intervention on health-related fitness, walking capacity and HRQoL in children and adolescents with CMCs. Because intervention effects tend to disappear without continued support,[6,91] future studies are required to evaluate effects of the Fitkids intervention on the long-term. In addition, future work is needed to investigate intervention effects on (long-term) daily physical activity and sports participation.

Behavioral components additional to exercise training

Future studies should investigate the feasibility of including specific behavioral change strategies or tools developed to support people in changing their behavior in a desired direction (i.e., increased physical behavior).[242] Future studies should also evaluate the effectiveness of these tools additional to exercise training on participation in physical activity in children and adolescents with CMCs. Including such specific strategies or tools in physical therapy practice will highlight the importance of changing behavior in order to achieve treatment goals aimed at participation in physical activity.

Collaboration with sport care connectors

The current physical activity policy in the Netherlands is aimed at connecting the sport, and physical activity sectors with other sectors, for example the primary care sector, to stimulate physical activity of individuals within the community. As part of this policy, Sport Care Connectors (in Dutch: *Buurtsportcoaches*) were introduced by the Dutch Ministry of Health, Welfare, and Sport in 2012.[254] Part of these Sport Care Connectors especially focus on promoting physical activity in children and adolescents with CMCs. Future research should focus on the collaboration of physical therapists with these Sport Care Connectors and other sport professionals (i.e., trainers and coaches). An improved collaboration has the potential to increase participation in sports and physical activity of children and adolescents with CMCs.

Clinimetric properties of the FTT in clinical populations

The FTT is developed for use in physical therapy practice. Future studies should therefore investigate the clinimetric properties of the FTT in specific clinical populations and should develop and evaluate prediction models for different diagnostic groups, such as children and adolescents with cardiovascular disease, pulmonary disease and those with a limited motor performance.[259]

Reference values for FTT performance in clinical populations

Future studies should focus on providing sex- and age-related reference values for FTT performance in different clinical populations. In general, comparison with persons who are typically developing is less well meaningful because the children of many diagnostic groups are performing significantly worse than the mean value for the general population. Disease-specific reference values contributes to a more optimal classification of the FTT performance of children and adolescents with CMCs.

Sensor wear requirements

Smaller and more user-friendly accelerometers are needed to increase the feasibility of device-based activity monitoring in a clinical setting. In addition, future research should focus on optimizing sensor wear requirements (i.e., attachment and sensor placement) which are useful in pediatric physical therapy practice to monitor free-living physical activity in children with CMCs.

Validation of accelerometers for detecting postures and movements in children with CMCs

Future studies should focus on validating accelerometers for detecting separate postures and movements in children with CMCs in an uncontrolled free-living setting using sensor wear requirements that are useful in pediatric physical therapy practice. Accordingly, there is need to explore new accelerometer data processing methods, like machine learning techniques, that have the potential for more accurate detection of separate postures and movements in youth with CMCs.[260]

Implementation of accelerometers in physical therapy practice

Use of telemonitoring or monitoring techniques like accelerometers fits into the current changes in healthcare in which themes like self-management and efficiency of care become more evident.[219] Future studies should focus on the implementation of feasible monitoring devices in pediatric physical therapy practice.

SUMMARY

PART 1. FITKIDS INTERVENTION

In **chapter 2**, the demographics, medical diagnoses, and initial aerobic fitness levels of children and adolescents participating in the exercise therapy program Fitkids were described. Results showed a great heterogeneity of the Fitkids population regarding demographic characteristics and medical diagnoses. Results of this study also showed that children participating in Fitkids have reduced aerobic fitness levels compared to peers who are typically developing. The results of this study have been used to further refine the Fitkids intervention and to prepare the intervention for effectiveness testing. In **chapter 3**, the effects of the Fitkids intervention were investigated in a cross-sectional observational study. Children and adolescents with CMCs received a graded exercise program for 6 months. Outcome measures included aerobic fitness, anaerobic fitness, muscle strength, walking capacity and HRQoL. These outcome measures were evaluated at baseline (t0), after 3 months of training (t1) and after 6 months of training (t2). After 3 months of training, significant improvements in health-related fitness, walking capacity, child-reported emotional functioning, parent-reported physical functioning, and parent-reported total HRQoL were found. After 6 months of training, significant improvements in child-reported physical functioning and parent-reported emotional functioning also were found. The Fitkids intervention did not improve social functioning, such as playing with peers, and home functioning. The absence of intervention effects on home functioning might be explained by the fact that a ceiling effect was obtained for child-reported home functioning at baseline. Although based on a small number of patients, preliminary data outlined in **chapter 7** reveal that there is no distinct relationship between participation in the Fitkids intervention and an increase in daily physical activity during the intervention period. Unpublished data of that study, however, pointed out that half of the included children were able to find an appropriate sport to participate in after the Fitkids intervention was completed.

PART 2. FITKIDS TREADMILL TEST

In **chapter 4** the validity and reproducibility of the Fitkids Treadmill Test (FTT) was evaluated in a group of children and adolescents who are healthy, typically developing, and between 6 -18 yr. The VO_{2peak} reached during the FTT showed excellent correlation with VO_{2peak} reached during the Bruce protocol (r = .90; P<.01). Backward multiple regression analysis provided the following prediction equations that can be used to estimate VO_{2peak} ($L{\cdot}min^{-1}$) from the time to exhaustion (TTE) reached on the FTT: for boys, VO_{2peak} FTT = -0.748 + (0.117 × TTE FTT) + (0.032 × body mass) + 0.263; for girls, VO_{2peak} FTT = -0.748 + (0.117 × TTE FTT) + (0.032 × body mass) (R^2=0.935, SEE=0.256 $L{\cdot}min^{-1}$). Cross-validation of the regression model showed an R^2 of 0.76. Compared to the TTE achieved at Bruce protocol, significant higher values of TTE achieved at the FTT were found in the validity group indicating that the FTT is less demanding than the Bruce protocol. Reliability statistics for the FTT showed an ICC of .985 (95% confidence interval: .971 to .993; P<.001) for TTE, which was the main outcome for the test-retest reliability statistics. Bland-Altman analysis showed a mean bias of -0.07 minutes, with limits of agreement between +1.30 and -1.43 minutes. In conclusion, it was reported that the FTT is a useful treadmill protocol with good validity and reproducibility in children and adolescents who are healthy. Exercise performance on the FTT and body mass can be used to adequately predict VO_{2peak} when respiratory gas analysis is not available. In **chapter 5** we described sex- and age-related normative values for FTT performance in children and adolescents who were healthy, typically developing, and between 6-18 yr. Strong positive correlations were found between TTE on the FTT and age, body mass, body height, body surface area, fat-free mass, and leg length in boys (r values ranging from .679 to .779 with P<.001 for all coefficients). In girls, moderate positive correlations were found between TTE on the FTT and age, body mass, body height, body surface area, fat-free mass, and leg length (r values ranging from .433 to .582, with P<.001 for all coefficients). Overall, age and body height had similar correlations with endurance times in our population (correlation between age and TTE on the FTT: r=.649 [P<.001]; correlation between body height and TTE on the FTT: r =.648 [P<.001]. For practical considerations we chose to use age in the normative centile charts. In boys, the reference curves (P50) showed an almost linear increase in TTE with age, from 8.8 minutes at 6 years of age to 16.1 minutes at 18 years of age. In girls, the P50 values for TTE increased from 8.8 minutes at 6 years of age to 12.5 minutes at 18 years of age, with a plateau in TTE starting at approximately 10 years of age. The reference values presented in this chapter can be used by physical therapists to adequately interpret FTT performance.

PART 3. NEW MEASUREMENT TOOLS: APPLICABILITY IN CLINICAL PRACTICE

In order to provide safe training recommendations and to evaluate whether interventions are successful, valid measurement instruments that are useful in physical therapy practice are indispensable. There is often a large gap between the development of measurement instruments in research and their use in clinical practice. In **chapter 6**, the clinical utility of the FTT was investigated on the basis of quantitative data obtained from the Fitkids database. In addition, this study explored potential factors affecting the use of the FTT in clinical practice. For the latter, an online survey was sent to physical therapists working in Fitkids practices (i.e., physical therapy practices affiliated with the Fitkids Foundation). To assess the utility of the FTT, we used 2 indicators. The first indicator was exercise duration (determined as the total duration of the FTT, including the duration of the warm-up phase), in terms of percentage of children and adolescents who reached the minimal duration for a maximal exercise test. The second was maximal effort, in terms of percentage of children and adolescents who reached a HR_{peak} of >180 beats·min^{-1} when performing the FTT. The indicators were considered sufficient when reached by 75% of the included children in each of the diagnostic groups. The first indicator to evaluate the clinical utility of the FTT was successful as the minimal exercise duration was reached in more than 75% of the included children in each diagnostic group. The second indicator to evaluate clinical utility was only reached in children with metabolic diseases (92%) and neuromuscular diseases (76%). The most important facilitator to the use of the FTT was the fact that the great majority was convinced of the additional value of the FTT. Main barriers were physical therapists' attitude (resistance to change/ lack of experience) and, on the environmental level, the absence of a treadmill ergometer within the physical therapist's practice. In **chapter 7**, the added value of using an accelerometer to monitor physical activity in children with CMCs in physical therapy practice was explored in comparison to subjective physical activity questionnaires. In addition, this study focused on the feasibility of using an accelerometer to monitor free-living physical activity in children with CMCs. The added value of objective measures was determined by differences in outcomes with regard to amount, type, context and bouts of PA. An in-depth data analysis on one case was presented to illustrate these differences in more detail. Feasibility of the accelerometer was defined by wear time, reasons for non-compliance and measurement issues, children's and parent's acceptability, and therapist's experiences. The results showed a significant difference between subjective measures and objective measures of moderate to vigorous

physical activity (P =.005) The in-depth analysis showed that the value of using objective data lies in more detailed description of physical activity per day and differences between weekdays and weekend days. Feasibility of the accelerometer was limited due to difficulties in applying the accelerometer, reluctance of children and their parents to wear the accelerometer for 7 days and technical difficulties in reading and charging. It was concluded that objectively measured physical activity has additional value in comparison with subjective measured physical activity as it gives therapists more detailed information. This detailed information can serve as input for more optimal counselling of children and adolescents (and their parents) to increase participation in physical activity. At the same time, steps have to be taken in optimizing accelerometers and accelerometer wear requirements for use in pediatric physical therapy practice.

SAMENVATTING IN HET NEDERLANDS

(Summary in Dutch)

ACHTERGROND

In Nederland groeien 1.3 miljoen kinderen en adolescenten op met een chronische ziekte of beperking. Het gaat om aandoeningen zoals diabetes, reuma, taaislijmziekte, aangeboren hartafwijkingen, ADHD en autisme spectrum stoornissen. Kinderen en adolescenten met een chronische ziekte of beperking zijn minder actief in het dagelijks leven en sporten minder vaak vergeleken met leeftijdsgenootjes zonder chronische ziekte of beperking. Mede door deze lagere mate van fysieke activiteit zijn deze kinderen minder fit. Een verminderde fitheid heeft negatieve consequenties voor de gezondheid. Het is daarom belangrijk dat kinderen met een chronische ziekte of beperking voldoende bewegen en sporten, zeker ook omdat sport een belangrijke sociale activiteit is. Het is voor kinderen en adolescenten met een chronische ziekte of beperking echter vaak lastiger om te sporten bij een sportvereniging. Niet voldoende adequate sportfaciliteiten is een vaak gehoorde belemmerende factor.

Fitness- en oefenprogramma Fitkids

Fitkids is een fitness- en oefenprogramma dat speciaal ontwikkeld is voor kinderen en adolescenten met een chronische ziekte, beperking of langdurige aandoening in de leeftijd van 6-18 jaar. De kinderen trainen in een groep onder professionele begeleiding van een (kinder)fysiotherapeut. Het programma is geïmplementeerd in ongeveer 165 (kinder) fysiotherapiepraktijken in Nederland. Iedere Fitkids training start met een gezamenlijke warming-up waarin o.a. verschillende (bal)spellen of circuittraining worden gedaan. Vervolgens gaan de kinderen aan de slag met een individueel trainingsschema waarbij gebruik wordt gemaakt van fitnessapparatuur. De training wordt afgesloten met een gezamenlijke cooling-down. Het fitness- en oefenprogramma Fitkids heeft als doel het verbeteren van de fysieke en mentale conditie van kinderen met een chronische ziekte of beperking. Het uiteindelijke doel van Fitkids is dat deze kinderen meer gaan bewegen in het dagelijks leven, waarbij de focus ligt op doorstroom naar een reguliere of aangepaste sportvereniging.

WAT IS ER GEDAAN EN HOE ZIJN DE ONDERZOEKEN UITGEVOERD?

Het hoofddoel van dit proefschrift was om te onderzoeken wat de effecten zijn van het fitness- en oefenprogramma Fitkids op fysieke fitheid, kwaliteit van leven en dagelijkse fysieke activiteit van kinderen en adolescenten met een chronische ziekte of beperking. Een tweede doelstelling was het onderzoeken van de klinimetrische eigenschappen van de Fitkids Treadmill Test (FTT) en het beschrijven van geslachts- en leeftijdsgerelateerde referentiewaarden voor deze test voor normaal ontwikkelde kinderen en adolescenten. In de laatste 2 hoofdstukken van dit proefschrift onderzochten wij de toepasbaarheid van nieuwe meetinstrumenten in de fysiotherapeutische praktijk.

Het doel van de studie beschreven in **hoofdstuk 2** was het beschrijven van de demografische kenmerken en medische diagnoses van kinderen en adolescenten die deelnemen aan het fitness- en oefenprogramma Fitkids (Fitkids populatie). Tevens werd in deze studie in kaart gebracht hoe fit deze kinderen zijn vergeleken met leeftijdsgenootjes zonder chronische ziekte of beperking. Voor dit onderzoek werd gebruik gemaakt van data uit de Fitkids database. De Fitkids database is een online patiëntendossier op www.fitkids.nl. Fysiotherapeuten werkzaam in Fitkids praktijken (fysiotherapie praktijken die aangesloten zijn bij Stichting Fitkids) kunnen hierop inloggen en met toestemming van ouders gegevens invoeren. Het betreft gegevens zoals lengte, gewicht, geslacht en medische diagnose. Deze gegevens worden geanonimiseerd ingevoerd. Tevens worden er testuitslagen van inspanningstesten ingevoerd die o.a. bij de start van het programma worden afgenomen. Door die testuitslagen te vergelijken met de referentiewaarden van de verschillende testen voor kinderen en adolescenten zonder chronische ziekte of beperking is bepaald hoe fit de kinderen zijn die bij Fitkids trainen in vergelijking met leeftijdsgenootjes zonder chronische ziekte of beperking.

In **hoofdstuk 3** zijn de effecten van het fitness- en oefenprogramma Fitkids onderzocht op duurconditie (aerobe fitheid), sprintconditie (anaerobe fitheid), functioneel inspanningsvermogen, functionele kracht en gezondheidsgerelateerde kwaliteit van leven (G-KvL). Kinderen en adolescenten met een chronische ziekte of beperking trainden gedurende 6 maanden bij Fitkids. Om de effecten van Fitkids te kunnen evalueren zijn er op 3 meetmomenten verschillende inspanningstesten afgenomen: voor de start van Fitkids (t0), na drie maanden trainen (t1) en na zes maanden trainen (t2). Naast een maximale inspanningstest voor de aerobe fitheid (halve Bruce protocol), werd de Muscle Power Sprint Test (MPST) afgenomen voor de anaerobe fitheid, de 6-minuten wandeltest voor het functioneel inspanningsvermogen, en verschillende functionele krachttesten van de Bruininks-Oseretsky Test (BOT-2) voor het evalueren van functionele kracht. Voor het evalueren van de effecten op G-KvL werden zowel de ouderversie als de kindversie van de DUX-25 afgenomen.

Het halve Bruce-loopbandprotocol is een maximale inspanningstest op een loopband die gebruikt wordt om de duurconditie van kinderen in kaart te brengen. Het is een test waarbij de hellingshoek en de snelheid van de loopband iedere 1.5 minuten volgens een vast protocol wordt verhoogd. De initiële snelheid is 2.7 km/uur en de initiële hellingshoek is 10%. De MPST is een test waarbij kinderen 6 keer over een afstand van 15 meter moeten sprinten met tussendoor 10 seconden rust. De 6-minutenwandeltest is submaximale inspanningstest waarbij kinderen gedurende 6 minuten op een zelfgekozen snelheid lopen waarbij het de bedoeling is dat er een zo groot mogelijke afstand wordt afgelegd. Een voorbeeld van een functionele krachttest uit de BOT-2 is de knie push up, waarbij kinderen in 30 seconde zoveel mogelijk knie push ups uitvoeren. De DUX-25 is een kwaliteit van leven vragenlijst bestaande uit 25 vragen waarbij de antwoorden zijn vormgegeven met behulp van smileys op een vijfpunts Likert Scale. De vragen kunnen worden onderverdeeld in vier domeinen: het domein fysiek, emotioneel, sociaal functioneren en thuissituatie. In totaal hebben 52 kinderen en adolescenten deelgenomen aan deze studie. De eerste drie maanden trainden de kinderen twee keer per week één uur en de laatste drie maanden één uur per week.

In **hoofdstuk 4** en **hoofdstuk 5** van dit proefschrift komen onderzoeken aan bod die betrekking hebben op de Fitkids Treadmill Test (FTT). De FTT is een maximale inspanningstest die uitgevoerd wordt op een loopband om de duurconditie (aerobe fitheid) van kinderen in kaart te brengen. De FTT is ontwikkeld als onderdeel van dit proefschrift. De aanleiding voor het ontwikkelen van deze test is dat de maximale hellingshoek van reeds bestaande loopbandprotocollen groter is dan de maximale helling van een standaard loopband (15%). Veel (kinder)fysiotherapeuten maken gebruik van zo'n standaard loopband. Het (halve) Bruce loopbandprotocol is de meest gebruikte inspanningstest op een loopband. Vanwege de hoge maximale hellingshoek van het (halve) Bruce protocol (22%) is het praktisch niet mogelijk deze test op een standaard loopband uit te voeren. Daar komt bij dat het Bruce protocol start met een hellingshoek van 10% wat het protocol minder geschikt maakt voor kinderen en adolescenten met een chronische ziekte of beperking. De FTT start, na de warming-up, met een helling van 1% en een snelheid van 3.5 km/uur. Iedere 1.5 minuten wordt de snelheid en hellingshoek van de loopband volgens een vast protocol verhoogd. De maximale helling van het FTT-protocol is 15%.

Alvorens een nieuwe test in te zetten in de klinische praktijk is inzicht in de klinimetrische eigenschappen van de test cruciaal. In **hoofdstuk 4** zijn de validiteit en de reproduceerbaarheid van de Fitkids Treadmill test onderzocht bij gezonde kinderen en adolescenten. Onder validiteit wordt verstaan dat de test meet wat het beoogt te meten. Reproduceerbaarheid wil zeggen dat bij herhaling van een meting dezelfde uitkomst wordt verkregen. Hierbij wordt onderscheid gemaakt tussen betrouwbaarheid en overeenkomst. Betrouwbaarheid is het vermogen van een test om personen van elkaar te onderscheiden ondanks de meetfout. Overeenkomst is van belang wanneer het doel is om veranderingen in testuitslag in de tijd aan te tonen. Vierenzeventig kinderen werden gerandomiseerd ingedeeld in een validiteitsgroep (n=34, 14 jongens, 20 meisjes, gemiddelde ± SD leeftijd: 12.9 ± 3.6 jaar) en een reproduceerbaarheidsgroep (n=34, 19 jongens, 15 meisjes, gemiddelde ± SD leeftijd: 13.5 ± 3.5 jaar). De kinderen werden gerekruteerd op verschillende scholen en sportclubs in Nederland. Deelnemers uit beide groepen hebben de inspanningstesten uitgevoerd met ademgasanalyse. De deelnemers in de validiteitsgroep hebben zowel de FTT als het Bruce protocol uitgevoerd binnen 2 weken. Het Bruce protocol fungeerde als de 'gouden standaard' en er is gekeken hoe verschillende inspanningsparameters behaald op beide testprotocollen met elkaar correleerden. De deelnemers in de reproduceerbaarheidsgroep hebben de FTT twee keer uitgevoerd binnen 2 weken. De 'intraclass' correlatiecoëfficiënt tussen de volhoudtijd (= duur van de test minus de duur van de warming-up) op de eerste FTT en de volhoudtijd op de tweede FTT was de belangrijkste uitkomstmaat t.a.v. betrouwbaarheid. De 'limits of agreement' (LOA) methode werd gebruikt als maat voor overeenkomst.

Referentiewaarden maken het mogelijk om het behaalde resultaat op een inspanningstest te interpreteren. Het doel van **hoofdstuk 5** was het opstellen van geslacht- en leeftijdsgerelateerde referentiewaarden voor kinderen en adolescenten in de leeftijd van 6 tot 18 jaar voor de geleverde prestatie op de FTT. Driehonderdzesenvijftig kinderen en adolescenten (174 jongens en 182 meisjes; gemiddelde ± SD leeftijd: 12.9 ± 3.7), hebben de FTT uitgevoerd waarbij de behaalde volhoudtijd de primaire uitkomstmaat was. De testen zijn hoofdzakelijk uitgevoerd op 2 scholen in het oosten van Nederland.

De **hoofdstukken 6 en 7** hebben betrekking op de toepasbaarheid van nieuwe meetinstrumenten in de fysiotherapeutische praktijk. Om een veilig trainingsschema te kunnen opstellen en om te kunnen evalueren of trainingsinterventies zoals Fitkids succesvol zijn, zijn valide meetinstrumenten onmisbaar. Meetinstrumenten die voor onderzoeksdoeleinden zijn ontwikkeld zijn lang niet altijd toepasbaar in de klinische praktijk. In **hoofdstuk 6** werd de klinische bruikbaarheid van de FTT onderzocht op basis van kwantitatieve gegevens verkregen uit de Fitkids database. Tevens werden in deze studie de potentiële factoren in kaart gebracht die het gebruik van de FTT in de klinische praktijk beïnvloeden. Voor dat laatste werd een online enquête verstuurd naar fysiotherapeuten werkzaam in Fitkids praktijken. Om de klinische bruikbaarheid van de FTT te beoordelen, werden er 2 indicatoren opgesteld. De eerste indicator was de duur van de inspanningstest (bepaald als de totale duur van de FTT, inclusief de duur van de warming-up), in termen van het percentage kinderen en adolescenten dat de minimale duur voor een maximale inspanningstest had bereikt. De tweede indicator was maximale inspanning, uitgedrukt in percentage kinderen en adolescenten die een HR_{peak} van >180 slagen·min^{-1} bereikten tijdens het uitvoeren van de FTT. De indicatoren werden voldoende geacht wanneer ze werden bereikt door 75% van de kinderen in elk van de diagnostische groepen.

Om te bepalen of het fitness- en oefenprogramma Fitkids erin slaagt om kinderen in het dagelijks leven meer te laten bewegen zijn er valide meetinstrumenten nodig om fysieke activiteit in de klinische praktijk te meten. Momenteel wordt de mate van fysieke activiteit vaak subjectief beoordeeld door het gebruik van beweegdagboekjes of vragenlijsten. Subjectieve meetinstrumenten zijn makkelijk in gebruik en zijn kostenbesparend. Echter, gezien het multidimensionale karakter van fysieke activiteit en het feit dat kinderen moeite hebben hun eerdere beweeggedrag (de uitvoering van lichaamshoudingen en bewegingen in het dagelijks leven) te herinneren, zijn subjectieve meetinstrumenten niet heel nauwkeurig. Naast subjectieve meetinstrumenten zijn er ook objectieve meetinstrumenten om het beweeggedrag van kinderen in kaart te brengen, zoals bijvoorbeeld een stappenteller of een activiteitenmonitor. In **hoofdstuk 7** werd de toegevoegde waarde van het gebruik van een activiteitenmonitor voor het monitoren van het beweeggedrag van kinderen met een chronische ziekte of beperking onderzocht in vergelijking met een vragenlijst. Bovendien concentreerde deze studie zich op de feasibility (haalbaarheid) van het gebruik van een activiteitenmonitor in Fitkids praktijken. In totaal hebben 12 kinderen deelgenomen aan dit onderzoek. Deze kinderen droegen op 3 meetmomenten gedurende 7 aaneengesloten dagen een activiteitenmonitor: voor aanvang van Fitkids (t0), na 3 maanden trainen (t1) en na 6 maanden trainen (t2). Overeenkomstig met de manier waarop het gebruikte algoritme gevalideerd is werd de sensor gedragen op het bovenbeen en vastgeplakt met huidvriendelijke tape. Tevens werd op t0, t1, en t2 middels een vragenlijst navraag gedaan naar het beweeggedrag van de kinderen in een normale week. De toegevoegde waarde van de activiteitenmonitor werd bepaald door verschillen in uitkomsten tussen de vragenlijst en de activiteitenmonitor met betrekking tot hoeveelheid fysieke activiteit, type activiteit, context informatie en 'bouts' (aaneengesloten periode van dezelfde activiteit). Om de verschillen tussen de vragenlijst en de activiteitenmonitor in meer detail te kunnen illustreren werd de data van 1 casus in meer detail beschreven. De feasibility van de activiteitenmonitor werd bepaald door de draagtijd, redenen voor niet-dragen, meetproblemen, de acceptatie van kinderen en de ervaringen van de (kinder)fysiotherapeut.

WAT HEBBEN WE GEVONDEN?

Hoofdstuk 2 toonde de grote heterogeniteit t.a.v. demografische kenmerken en medische diagnoses van kinderen en adolescenten die bij Fitkids trainen. In totaal toonde de data 67 verschillende diagnoses. De resultaten van dit onderzoek toonden tevens de verminderde duurconditie (aerobe fitheid) van deze kinderen in vergelijking met leeftijdsgenootjes zonder chronische ziekte of beperking.

De resultaten van **hoofdstuk 3** toonden aan dat het fitness- en oefenprogramma Fitkids positieve effecten heeft op de fysieke fitheid, het functioneel inspanningsvermogen en op de G-KvL van kinderen met een chronische ziekte of beperking. Na drie maanden trainen lieten de deelnemers een verbeterende fitheid zien, zowel de sprintconditie als de duurconditie verbeterde en tevens lieten de deelnemers een verbeterde spierkracht zien. Ook konden de deelnemers in zes minuten een grotere afstand lopen, wat duidt op een vooruitgang in het functioneel inspanningsvermogen. Na zes maanden trainen werden de effecten van de training op de sprintconditie en de duurconditie groter, echter de gevonden effecten op spierkracht en het functioneel inspanningsvermogen werden niet groter. De reden is waarschijnlijk dat één keer per week trainen voldoende is om trainingseffecten te behouden, maar onvoldoende om verdere verbeteringen te bewerkstellingen. In de studie zijn tevens verschillende scores voor kwaliteit van leven bepaald: een totaalscore en een score voor de domeinen fysiek, emotioneel, sociaal en thuis gerelateerd. Na drie maanden trainen vonden de kinderen dat ze het in emotioneel opzicht beter waren gaan doen. De ouders vonden op dat moment dat de kinderen er in fysiek opzicht op vooruit waren gegaan. Ook rapporteerden de ouders een hogere totaalscore in vergelijking met voor de training. Na zes maanden vonden de kinderen tevens dat ze het in fysiek opzicht beter waren gaan doen en vonden de ouders dat de kinderen vooruitgang hadden geboekt in emotioneel opzicht. Er werden geen effecten van de training gevonden op sociaal functioneren en ook ervaarden de kinderen de thuissituatie niet anders door de Fitkids training, zowel volgens zichzelf als wanneer dit beoordeeld werd door de ouders.

De resultaten van **hoofdstuk 4** toonden aan dat de FTT een valide en reproduceerbare inspanningstest is bij gezonde kinderen en adolescenten. In de validiteitsgroep werd een uitstekende correlatie gevonden tussen de VO_{2piek} behaald tijdens de FTT en de VO_{2piek} behaald tijdens het Bruce protocol (r = .90; P <.01). Achterwaartse multiple regressieanalyse leverde de volgende predictievergelijkingen op: voor jongens, VO_{2peak} FTT = -0.748 + (0.117 × volhoudtijd FTT) + (0.032 × lichaamsgewicht) + 0.263; voor meisjes, VO_{2peak} FTT = -0.748 + (0.117 × volhoudtijd FTT) + (0.032 × lichaamsgewicht) (R^2 = 0.935, SEE = 0.256 L · min-1). Aan de hand van deze predictievergelijkingen kan de VO_{2piek} (L · min-1) adequaat voorspeld worden aan de hand van de geleverde prestatie op de FTT en lichaamsgewicht. In de reproduceerbaarheidsgroep werd een 'intraclass' correlatiecoëfficiënt van 0.985 (P<.001) gevonden voor de volhoudtijd op de twee FTTs, wat betekent dat de meetfout klein is vergeleken met de variabiliteit tussen de deelnemers en dat de discriminatie van personen nauwelijks wordt beïnvloed door meetfouten. Het gemiddelde verschil tussen de twee FTTs bedroeg -0.07 minuten, met LOA tussen +1.30 en -1.43 minuten. Deze LOA geeft aan dat de overeenkomst van de FTT goed is. Een verandering in testuitslag in de tijd binnen een individu kan alleen worden beschouwd als een echte verandering als deze buiten de LOA ligt.

De data verzameld in **hoofdstuk 5** werden gebruikt om referentiewaarden op te stellen in de vorm van centielen. Bij jongens correleerde de behaalde volhoudtijd sterk met leeftijd, lichaamsgewicht, lichaamslengte, lichaamsoppervlakte en vetvrije massa en beenlengte (r waarden variërend van .679 tot .779; met P<.001). Bij meisjes werden er matige correlaties gevonden tussen behaalde volhoudtijd en leeftijd, lichaamsgewicht, lichaamslengte, lichaamsoppervlakte, vetvrije massa en beenlengte (r waarden variërend van .433 tot .582; met P<.001). Leeftijd en lengte hadden overeenkomstige correlaties met volhoudtijd in onze populatie. Om praktische redenen hebben we ervoor gekozen om leeftijd te gebruiken in de referentiegrafieken. De referentiewaarden voor jongens lieten een lineaire toename met leeftijd zien voor de behaalde volhoudtijd. De referentiewaarden voor meisjes lieten een lineaire toename met leeftijd zien tot ongeveer 10 jaar, waarna de waarden afvlakten.

Op basis van de resultaten in **hoofdstuk 6** werd geconcludeerd dat de FTT bruikbaar is in diverse patiëntengroepen. De eerste indicator om de klinische bruikbaarheid van de FTT te evalueren was succesvol omdat de minimale duur voor een maximale inspanningstest werd bereikt door meer dan 75% van de kinderen in elke diagnostische groep. De tweede indicator werd slechts behaald in de groep kinderen met metabole ziekten en de groep kinderen met neuromusculaire ziekten. Echter, wanneer een aantal zaken in ogenschouw genomen worden, lijkt de FTT ook bruikbaar in de overige patiëntengroepen. Zo dient een kind bekend te zijn met het lopen en rennen op een loopband met hellingshoek. Tevens moeten therapeuten zich vertrouwd voelen om kinderen maximaal te laten inspannen en moeten ze kinderen ook dusdanig aanmoedigen dat kinderen hun maximale inspanning durven te bereiken. Dit vergt ervaring. Meer aandacht voor het afnemen van maximale inspanningstesten in het professionele curriculum van kinderfysiotherapeuten zou hier helpend in zijn.

Tevens werden in deze studie de potentiële factoren in kaart gebracht die het gebruik van de FTT in de klinische praktijk beïnvloeden. De belangrijkste facilitator was het feit dat de grote meerderheid de toegevoegde waarde van de FTT zag. De belangrijkste factoren die het gebruik van de FTT in de klinische praktijk belemmerden waren de houding van fysiotherapeuten (weerstand tegen verandering/ gebrek aan ervaring) en de afwezigheid van een loopband in de fysiotherapiepraktijk. Deze informatie t.a.v. de factoren die het gebruik van de FTT beïnvloeden kan gebruikt worden om de FTT nog beter in te bedden in de klinische praktijk.

De resultaten in **hoofdstuk 7** toonden een significant verschil tussen de totale hoeveelheid subjectief gemeten fysieke activiteit en de totale hoeveelheid objectief gemeten fysieke activiteit, waarbij er subjectief veelal hogere waarden gerapporteerd werden. De gemiddelde overschatting was 211% (224 minuten/week). De gedetailleerde analyse van data van 1 casus toonde aan dat de waarde van het gebruik van een objectieve data ligt in een meer gedetailleerde beschrijving van de fysieke activiteiten per dag en verschillen tussen weekdagen en weekenddagen. De feasibility van de activiteitenmonitor was beperkt vanwege de terughoudendheid van kinderen en hun ouders om de activiteitenmonitor gedurende 7 dagen te dragen en technische problemen bij het uitlezen en opladen van de activiteitenmonitor. De plek van dragen (bovenbeen) in combinatie met de manier van bevestigen (huidvriendelijke tape) waren de belangrijkste redenen waarom ouders en kinderen terughoudend waren om deel te nemen aan de studie. Tevens rapporteerde meer dan de helft van de kinderen (ernstige) huiduitslag. Er werd geconcludeerd dat objectief gemeten fysieke activiteit een toegevoegde waarde heeft t.o.v. subjectief gemeten fysieke activiteit omdat het meer gedetailleerde informatie geeft over het beweeggedrag van kinderen en adolescenten met een chronische ziekte of beperking. Deze gedetailleerde informatie kan o.a. als input dienen voor een meer optimale begeleiding van kinderen en adolescenten (en hun ouders) om het beweeggedrag van deze kinderen te vergroten. Tegelijkertijd moeten stappen worden genomen om de activiteitenmonitors, de plek van dragen en de manier van bevestigen te optimaliseren voor gebruik in de kinderfysiotherapie.

Voorlopige resultaten van deze studie tonen tevens aan dat er op dit moment geen duidelijk verband bestaat tussen deelname aan het fitness- en oefenprogramma Fitkids en een toename in de dagelijkse fysieke activiteit tijdens de interventieperiode. De helft van de kinderen die deelnamen aan de studie vond echter wel een geschikte sport om aan deel te nemen na het afronden van het fitness- en oefenprogramma Fitkids. Hoewel niet onderzocht mag aangenomen worden dat deze sportparticipatie de dagelijkse fysieke activiteit van deze kinderen alsnog vergroot.

WAAR ZOU TOEKOMSTIG ONDERZOEK ZICH OP MOETEN RICHTEN?

In **hoofdstuk 8** beschreven we onze belangrijkste bevindingen en werden verschillende aanbevelingen gedaan voor toekomstig onderzoek. Zo is er bijvoorbeeld meer onderzoek nodig naar de (lange termijn) effecten van het fitness- en oefenprogramma Fitkids op fysieke activiteit en sportparticipatie.

Ook verdient het aanbeveling om onderzoek te doen naar de klinimetrische eigenschappen van de FTT in klinische populaties en het ontwikkelen en evalueren van predictievergelijkingen voor verschillende diagnostische groepen. Tevens zou toekomstig onderzoek zich moeten richten op het ontwikkelen van geslachts- en leeftijdsgerelateerde referentiewaarden voor FTT-prestaties voor verschillende diagnostische groepen. Dit gezien het feit dat, over het algemeen, kinderen en adolescenten met een chronische ziekte of beperking aanzienlijk lager presteren dan hun leeftijdsgenootjes zonder chronische ziekte of beperking. Referentiewaarden voor specifieke diagnostische groepen dragen daarom bij aan betere classificatie van FTT-prestaties van kinderen en adolescenten met een chronische ziekte of beperking.

Daarnaast zou toekomstig onderzoek zich moeten richten op de ontwikkeling van kleinere en meer gebruiksvriendelijke activiteitenmonitors die beter toepasbaarheid zijn binnen de kinderfysiotherapie. Tevens wordt geadviseerd om meer onderzoek te doen naar het valideren van activiteitenmonitors voor het detecteren van afzonderlijke houdingen en bewegingen bij kinderen met een chronische ziekte of beperking in een ongecontroleerde setting.

Dit proefschrift bevat verschillende aanbevelingen om toekomstige interventie-effecten op dagelijkse fysieke activiteit van kinderen en adolescenten met een chronische ziekte of beperking te verbeteren. Zo verdient het aanbeveling om toekomstig onderzoek te richten op het implementeren van gedragsmatige interventies, aanvullend op het huidige trainingsprogramma. Dit omdat gedragsmatige interventies, die zich richten op het kind en gezinssysteem, van meerwaarde lijken te zijn bij het verbeteren van het beweeggedrag van kinderen met een chronische ziekte of beperking. Toekomstige studies zouden ook de effectiviteit van deze gedragsmatige interventies moeten evalueren.

Daarnaast wordt geadviseerd om onderzoek te doen naar de samenwerking van (kinder) fysiotherapeuten met buurtsportcoaches en andere sportprofessionals (trainers en coaches) in de sport- en beweegsector. Het huidige sport- en beweegbeleid in Nederland is gericht op het verbinden van de sport- en beweegsector met andere sectoren om het beweeggedrag van individuen te stimuleren. Als onderdeel van dit beleid zijn buurtsportcoaches in 2012 geïntroduceerd door het Nederlandse ministerie van Volksgezondheid, Welzijn en Sport. Een deel van deze buurtsportcoaches zijn aangesteld om

het beweeggedrag van kinderen en adolescenten
met chronische ziekte of beperking verbeteren.

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LIST OF PUBLICATIONS

Kotte EMW, Veenhof C, Winkler AMF, Horemans HLD, Takken T, de Groot JF. Monitoring physical activity in primary pediatric physical therapy practice: difficulties, opportunities and a case study illustration. *Submitted for publication, revisions pending.*

Kotte EMW, de Groot JF, Winkler AMF, Veenhof C, Takken T. Fitkids treadmill test: clinical utility and factors associated with its use among physical therapists. *Phys Ther.* 2019 Apr 1; 99(4):428-439.

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CURRICULUM VITAE

Elles Kotte was born on March 18, 1978 in Almelo, the Netherlands. After graduating from secondary school (Canisius, Almelo) in 1995 she started as a radiographer student at the department of radiology of the Gelre Medical Center in Apeldoorn. This in-service training was combined with theoretical education at the Saxion University of Applied Sciences. After graduating (BSc), Elles continued working at the department of radiology of the Gelre Medical Center for one year. In 2000 she started the bachelor program Biomedical Health Sciences at the Radboud University Nijmegen and at the same time she worked as a radiographer at the department of radiology of the Radboud University Medical Center Nijmegen. During her master (Human Movement Sciences) she completed two internships at the department of pediatric physical therapy of the Radboud University Medical Center Nijmegen, under supervision of Prof. dr. Nijhuis- van der Sanden. During these internships, she became enthusiastic for research in the field of pediatric physical therapy. She received her Master of Science degree in 2005. Immediately after graduating she started as a junior researcher at Roessingh Research and Development and worked at a project aimed to investigate, develop, and demonstrate core architectures and frameworks for future Ambient Systems. After working there for 3 years, she started to work for the Fitkids Foundation. This thesis is the result of this assignment. She currently lives with her boyfriend, Jasper Hulshof, and their two children, Sam (6 years of age) and Daantje (3 years of age) in Oldenzaal. She continues working for the Fitkids foundation.

DANKWOORD (Acknowledgements)

'At the end of the day we can endure much more than we think we can'
Frida Kahlo

Promoveren is voor mij nooit een doel op zich geweest en omdat er thuis de afgelopen jaren grotere uitdagingen waren dan het afronden van een promotietraject, heb ik lange tijd gedacht dat het er ook niet meer van zou komen. Dat ik het toch heb kunnen afronden voelt wel ontzettend goed. Ik zou het nooit hebben gered zonder heel veel lieve mensen om mij heen. Ik wil dit dankwoord starten met de belangrijkste personen in mijn leven die het verdienen als eerste genoemd te worden.

Lieve Jasper, je wilde helemaal niet genoemd worden in mijn dankwoord, maar je bent de eerste ;-). Dank voor wie je bent, je onvoorwaardelijke steun, liefde en vertrouwen in mij. Zonder jou komt het allemaal niet goed. Vier jaar geleden belandden we, na de geboorte van Daantje, in een totaal nieuwe wereld. Een wereld van slopende onzekerheid, veel zorg en weinig slaap. Maar ondanks de rollercoaster waarin we zaten zijn we elkaar nooit kwijtgeraakt. Die credits zijn voor jou lieve Jasper. Je rust en tomeloze geduld zijn bewonderingswaardig en zo belangrijk voor mij. Ik hou van je!

Lieve Sam, mijn grote lieve vriendje! Wat ben je geweldig en wat is het leuk dat je erbij bent op mijn feestje. Zonder jou zou het feestje ook helemaal niet compleet zijn. Ondanks alle spanningen van de afgelopen jaren blijf jij onze stralende 'steady factor'. Je enthousiasme is aanstekelijk en je bent de allerliefste grote broer die Daantje zich kan wensen. Je zit altijd vol goede ideeën en ik had daarom gelijk naar je moeten luisteren toen je zei dat jij en Daantje op de voorkant van mijn boekje moesten komen te staan: jullie staan er prachtig op! Je zei laatst gekscherend: buut voor mama apenstaartje Fitkids toen we verstoppertje speelden. Je hebt helemaal gelijk lieve vent, ik ben ook veel te veel aan het werk geweest. Nu het boekje klaar is ga ik nog meer lol maken met jou!

Daantje, ons kleine meisje waar al zoveel over gezegd en geschreven is. Lieve Daan, jij leert mij dat er zoveel meer is dan gesproken taal. Je leert mij te vertragen, meegaan in jouw tempo. Bij jou in de buurt is iedereen de beste versie van zichzelf. Dat is misschien wel het mooiste cadeau dat jij mij hebt gegeven. Jouw onschuld is betoverend. *'Big things have small beginnings',* dat geldt zeker voor jou lieve Daan.

Lieve pap en mam! Niets dat ik hier schrijf doet recht aan hoe belangrijk jullie voor mij zijn. Mam, jouw energie en oog voor wat anderen nodig hebben zijn bewonderingswaardig. Je denkt altijd twee stappen

vooruit. De ontelbare nachtjes dat Daantje bij jullie slaapt, de oppasdagen inclusief boodschappen en weggestreken wasjes. Je doet het allemaal mam. Het maakt dat ik mijn werk weer kon oppakken en dit traject tot een goed einde heb kunnen brengen. Daar ben ik je ontzettend dankbaar voor. En pap, hoe fijn zijn de uitstapjes met Daan op maandag, de kopjes koffie met een krantje erbij. Ik bewonder je nuchterheid en humor. Zeker nu je geheugen je in de steek laat zijn dit hele waardevolle eigenschappen. Ons 'tweede thuis' op de Tutenberg heeft ons allemaal goed gedaan en ik hoop dat het ons gegund is daar nog vele jaren samen te mogen genieten.

Mijn schoonouders, lieve Henk en Hermien, ook jullie steun is van onschatbare waarde. De dinsdagavond, als Daantje bij jullie slaapt is nog altijd een ontzettend welkom rustpunt in de week. Even tijd voor Sam, al kwam het er de laatste maanden vaak op neer dat ik op de dinsdagavonden ongestoord kon werken. Op woensdagavond worden er steevast twee hele vrolijke kindjes thuisgebracht inclusief een pannetje met heerlijk eten. Wat een voorrecht. Op die avonden werd er door Henk altijd even naar de vorderingen omtrent mijn proefschrift gevraagd. Onder het genot van een… nee, ik hoef echt geen… glaasje wijn.

Mijn lieve zussen, Hayke en Hellen. Ik ben ontzettend blij met jullie! We zijn zo anders en toch ook zo hetzelfde. Ons werk kent vele raakvlakken. Hel, via je werk als kinderfysiotherapeut ben ik ooit bij Stichting Fitkids terechtgekomen. Ik ben je er nog dankbaar voor. En, Hayk, via jouw werk als fysiotherapeut participeer je in vele wetenschappelijke studies. Wellicht dat we in de toekomst onze krachten eens moeten gaan bundelen. Dank lieve zussen voor jullie interesse in mijn onderzoeken maar bovenal voor jullie steun de afgelopen jaren!

Lieve zwagers en schoonzussen, Thomas, Tim, Ieke en Alice. Dank voor de tripjes vanuit Groningen om even te horen hoe het met ons gaat (Ieke en Alice), de lunches bij Blond (Thomas), de biertjes onder de veranda (Tim) en gewoon voor jullie gezelligheid waarbij het ook vaak niet over werk ging.

Uiteraard zou dit proefschrift er ook niet zijn geweest zonder fijne collega's om mij heen. Allereerst mijn collega's van Stichting Fitkids en Stichting Artsen voor Kinderen. Lex Winkler. Beste Lex, jou wil ik ontzettend bedanken voor je vertrouwen vanaf onze allereerste ontmoeting bij 'De Twee Wezen' in Hengelo. Dank dat je mij de kans en ruimte hebt gegeven mij te kunnen ontwikkelen in een vakgebied die bij mij past en me erg aan het hart ligt. Jikke Wigmans, lieve Jikke, we hebben meer dan acht jaar samengewerkt. Een samenwerking waar ik met heel veel plezier op terugkijk. Dank voor alle lunches en koffietjes aan de Kuiperbergweg (tjee dat is lang geleden ;-), later aan de Beethovenstraat, de Gabriël Metsustraat en de laatste jaren aan Noorderakerweg. Ik denk met heel veel plezier terug aan alle scholingsdagen die we samen hebben georganiseerd en alle jaren dat we met ‘onze’ stormbaan op de open dag van de Cruijff Foundation in het Olympisch stadion stonden. Hester Noll, lieve Hester, 2 jaar geleden nam jij het stokje over van Jikke. Ik bewonder je inzet en gedrevenheid om Fitkids als merk sterker neer te zetten. Onze nieuwe huisstijl is zeker jouw verdienste. Het is prachtig geworden. Ik kom gauw een koffietje doen bij je op ons nieuwe kantoor. Roesja en Marloes, beiden werkzaam voor ‘Artsen voor Kinderen’. Al zagen we elkaar niet vaak en hebben jullie geen concrete bijdrage geleverd aan dit proefschrift toch wil ik jullie bedanken. Gewoon omdat jullie bijzonder mooie mensen zijn!

Daarnaast alle lof voor mijn copromotoren Tim Takken en Janke de Groot. Beste Tim, in het bijzonder heb ik veel geleerd van jouw inzicht en vermogen om feilloos en snel tot de kern van de zaak te komen. Ik bewonder je snelle schakelen, je nuchtere kijk en uiteraard je onuitputtelijke kennis op het gebied van klinische inspanningsfysiologie. Ontelbare keren ben je spreker geweest op één van onze scholingsdagen waarbij er de laatste jaren vaak een foto van je sportieve meiden in je presentatie voorbij kwam. Mooi! Goed voorbeeld doet goed volgen. Beste Janke, dank voor de fijne samenwerking en voor alles wat ik van je heb geleerd, zowel op wetenschappelijk als op persoonlijk vlak. Dank voor alle kritische, maar altijd zeer relevante commentaren op mijn artikelen. Vaak vroeg je even hoe het thuis ging. Ik heb dat als ontzettend fijn ervaren. Als ik naar Utrecht kwam voor een overleg over één van mij artikelen liep mijn hoofd vaak over van alle dingen die er thuis speelden. Het is dan fijn te merken dat daar ook even ruimte voor is. Voor jullie beiden geldt, ik hoop dat onze samenwerking niet stopt bij het afronden van dit boekje.

Uiteraard ook heel veel dank aan mijn promotor. Beste Cindy, je bent in een wat later stadium betrokken geraakt bij mijn promotietraject. Ik kan me voorstellen dat het voor jou niet makkelijk was om in te stappen in een traject dat al zo uitgestippeld was. Ik wil je ontzettend bedanken dat je de stap toch hebt genomen. Ik heb onze samenwerking als bijzonder fijn ervaren. Dank voor je vertrouwen in mij, het laagdrempelige contact en je kritische blik!

Mijn paranimfen, Manon en Eline. Jullie wisten vaak precies aan te voelen wanneer ik een steuntje in de rug kon gebruiken. Jullie bos bloemen was meer dan perfect getimed. Misschien komt het door onze Twentse roots? Manon, ik bewonder je gedrevenheid en positieve energie. Je bent een echter powervrouw en ondertussen ook nog eens een super lieve mama voor Faas en Lilly. Eline, ik weet nog dat ik een presentatie kwam geven op de VU. Sommige mensen blijven je gewoon bij en ik was dan ook blij verrast toen ik hoorde dat je op de HU kwam werken. Ik bewonder je zachte karakter en ben onder de indruk van je mooie dijkhuisje en uiteraard je prachtige kleine man Fosse. Onze samenwerking houdt zeker niet op bij het afronden van dit boekje.

De leden van de promotiecommissie, prof. dr. F.J.G. Backx, prof. dr. J.W. Gorter, prof. dr. M.J. Jongmans, prof. dr. E.E.S Nieuwenhuis en prof. dr. R. Nijhuis wil ik bedanken voor de tijd en aandacht die zij aan de beoordeling van mijn proefschrift hebben besteed. Prof. dr. R. Nijhuis wil ik extra danken. Lieve Ria, wij gaan wat verder terug. Ik weet nog dat ik voor het eerst op de afdeling kinderfysiotherapie van het Radboud UMC kwam. Toen nog op de oude locatie. Je enthousiasme voor toegepast onderzoek en daarbij altijd het kind centraal stellend heeft er zeker aan bijgedragen dat ik sta waar ik nu sta. Buiten dat ben je ook gewoon een heel mooi mens en heb ik ontzettend veel van je geleerd. Fijn dat onze wegen destijds kruisten en dat we contact zijn blijven houden.

Dank ook aan alle kinderen die vol enthousiasme hebben meegedaan aan één van mijn onderzoeken. Wat heb ik veel van jullie geleerd! Door jullie weet ik dat het fitness- en oefenprogramma Fitkids veel meer is dan een programma dat bijdraagt aan een betere conditie. Het geeft zelfvertrouwen, plezier, en draagt bij aan een gevoel van ergens bij horen. Ik heb heel veel blije koppies gezien, ontzettend veel dank daarvoor. Ook heel veel dank aan alle kinderen en jongeren die hebben meegedaan aan één van de onderzoeken t.a.v. de Fitkids Treadmill test. Jullie gingen tot het uiterste op de loopband. Dank daarvoor! Ik ben ook veel dank verschuldigd aan de schoolleiding en de leerkrachten van: basisschool 'De Wiekslag' en scholengemeenschap 'Canisius'. Jullie maakten het mogelijk dat alle inspanningstesten onder schooltijd konden worden uitvoeren. Ook verdient Procare het om hier genoemd te worden. Dank voor het leveren van de loopband om de metingen op de scholen te kunnen uitvoeren. Aafko Roggema, toen de metingen op de basisschool klaar waren kwam je hoogstpersoonlijk om samen met mijn vader de loopband naar de nieuwe locatie te verplaatsen. Topper! Dank daarvoor.

Uiteraard wil ik ook alle kinderfysiotherapeuten en fysiotherapeuten werkzaam bij de Fitkidscentra bedanken. Zonder jullie zou het nooit gelukt zijn om mijn onderzoeken tot een goed einde te brengen. Daarbij zijn jullie het gezicht van Fitkids. Uit ervaring weet ik hoe moeilijk het soms is om die ene geschikte plek te vinden waar een kind lekker kan sporten en bewegen. Ik weet dat jullie daarin vaak tot het uiterste gaan. Laten we er met z'n allen voor blijven zorgen dat we de kinderen blijven zien en blijven horen op een manier waar ze blij van worden.

Ook de co-auteurs van mijn artikelen wil ik bedanken voor de fijne samenwerking. Zonder jullie was het niet mogelijk geweest alle artikelen te schrijven. Barbara Huijgen, lieve Bar, wij waren elkaars kamergenoot bij Roessingh Research and Development. Toen jij naar Groningen vertrok en ik bij Stichting Fitkids ging werken bleven we contact houden. Heel erg bedankt voor je hulp bij de multilevel analyses. Het artikel moest geschreven worden op het moment dat je zelf ging promoveren, maar je had altijd wel even tijd om mij bij te staan met raad en daad. Gelukkig! Het was anders niet goed gekomen.

Bart Bongers, beste Bart, dank voor je input, feedback en voor de metingen die je hebt gedaan. Met een busje op weg naar Castricum om daar de laatste inspanningstesten te doen voor de normering van de Fitkids Treadmill test. Dank! Herwin Horemans, wat fijn dat je bent aangehaakt bij het laatste artikel. Dank dat ik daardoor gebruik kon maken van je expertise op het gebied van activiteiten monitoring. Je input was meer dan waardevol.

Ook ben ik veel dank verschuldigd aan Green Orange in Oldenzaal. Dank dat ik op mijn 'thuiswerkdagen' kon werken in dit prachtige bedrijf. Dank voor de gezelligheid en heerlijke lunches. Het is niet uit te leggen hoe belangrijk dit voor mij is geweest, zeker het eerste jaar na de geboorte van Daantje.

Edwin van Asseldonk. Beste Edwin, alias 'Matlab hulplijn'! Het begon al tijdens mijn afstuderen. Ondanks een cursus Matlab is het tussen Matlab en mij nooit wat geworden. Maar gelukkig bestaan er vrienden die Matlab leuk vinden en er dan ook nog eens heel goed in zijn. Het beste van twee werelden zou ik zeggen. Edwin dank voor je tijd en je interesse in mijn onderzoeken en je vermogen om vakkundig mijn wensen te verwerken in een Matlab script. Lieve Bianc, bedankt dat ik je mannetje af en toe van je mocht lenen, maar bovenal dank voor je oprechte interesse in mij en mijn gezinnetje en dat je zo leuk bent!

Ook veel dank aan Annelies, Elsje, Lisette en Sabine. Tijdens jullie afstudeerprojecten hebben jullie vele inspanningstesten afgenomen en zo bijgedragen aan de normering van de Fitkids Treadmill Test. Jullie inzet heeft me enorm geholpen. Dank! En Elsje, leuk dat je nu werkzaam bent binnen één van de Fitkidscentra.

Peter van Limbeek! Beste Peter, jij mag ook zeker niet ontbreken in dit dankwoord. Al was het gele vlakje wel even 'een dingetje'. De omslag is meer dan prachtig geworden. Dank!

Lieve allemaal. Ontzettend bedankt.

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ISBN 978-94-028-1695-2