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# **Research Report**

# Effects of the Fitkids Exercise Therapy Program on Health-Related Fitness, Walking Capacity, and Health-Related Quality of Life

Elles M.W. Kotte, Janke F. de Groot, Alexander M.F. Winkler, Barbara C.H. Huijgen, Tim Takken

**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 disabilities or chronic conditions.

**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 variations in health-related fitness, walking capacity, and HRQoL during the intervention period. The models were adjusted for sex, height, and weight.

AQ: 1

Bio

**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 the self-reported and parent-reported physical and emotion domains and for the parent-reported total score for 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.

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AQ: 2

AQ: 3

n the Netherlands, 14% to 20% of all children and adolescents (0-18 years of age) have long-term or chronic diseases.1 There is strong evidence that physical fitness is compromised in these young people.<sup>2-6</sup> 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 adiposity7 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 Therapist Practice (2nd ed) states that a part of physical therapist practice is to "provide prevention and promote health, wellness, and fitness."9(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 healthrelated fitness and health-related quality of life (HRQoL) in this population.<sup>2</sup> In keeping with the opinion of the American Physical Therapy Association, the Fitkids program is conducted under the 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 the 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.

### Method Participants

**Study sample.** Fifty-two children and adolescents who were referred to the Fitkids program by their treating physicians participated in this study (30 boys and 22 girls). Inclusion was based on willingness to participate. Participants were 6 to 17 years of age at the time of inclusion and were diagnosed with any type of chronic disease or disability. Children and adolescents who depended on a wheelchair or 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 the willingness of the therapists to participate and the ability of the therapists to administer the included exercise tests according to the test manuals. The inclusion of participants started after approval by 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.

#### Procedure

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 minutes) and a cool-down period (±10 minutes), which were based on simple competitive or strategic games, and a core component ( $\pm 40$ minutes), 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<sup>2</sup>) 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.<sup>10,11</sup>

#### **Outcome Measures**

Outcome measures, conducted according to *International Classification of Functioning, Disability and Healtb* guidelines,<sup>12</sup> 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).

AQ: 5

AO: 4

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 than the original protocol.13 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 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 refused to do more work despite verbal encouragement. Participants were not allowed to use the handrails. According to Cumming et al,14 the Bruce test is highly reproducible (r=.94)and valid (r=.88 for 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).<sup>15</sup> 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)15 and children with cerebral palsy.<sup>16,17</sup> 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 runs, the physical therapist counted backward from "10" to "1" and then gave the cue "go." The mean power, considered the most important parameter for

the MPST, was the average power output of the 6 sprints.<sup>15</sup> Power (W) was calculated as force  $\times$  velocity, where force (kg  $\times$  m/s<sup>2</sup>) = body mass (kg)  $\times$  acceleration (m/s<sup>2</sup>) and velocity (m/s) = distance (m)/time (seconds).

Muscle strength was assessed by use of the strength subscale of the Bruininks-Oseretsky Test of Motor Proficiency (2nd ed) (BOT-2).<sup>18,19</sup> 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.<sup>19</sup>

Activity. Walking capacity was assessed by use of the Six-Minute Walk Test (6MWT), conducted according to American Thoracic Society guidelines.<sup>20</sup> The test-retest reliability of the 6MWT has been reported to be high, with an intraclass correlation coefficient of .84 to .98.21-23 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.<sup>24,25</sup> In the present study, the 6MWT was performed in a 15-m straight corridor. Participants were instructed to cover the greatest possible distance in 6 minutes at a selfselected 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.

#### Fitkids Exercise Therapy Program

HRQoL. The 25-question version of the DUCATQOL (DUX-25)<sup>26</sup> 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 AQ:8 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 .94.28 Sample 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 the baseline characteristics of the study population, we compared initial test results with test results for people who were healthy. To inves-

AQ: 11

AO: 9

AO: 7

#### AQ: 22 Table 1.

Demographic Characteristics of Participants<sup>a</sup>

Characteristic	x	SD	Range	No. of Participants
Weight (kg)	50.1	17.7	20.2 to 105.0	
Height (cm)	149.0	14.7	112.0 to 175.0	
BMI (kg/m²)	22.0	4.9	13.9 to 35.8	
Weight for age (SD score)	1.2	1.4	-2.5 to 3.6	
Height for age (SD score)	0.2	1.2	-4.2 to 3.3	
BMI for age (SD score)	1.4	1.3	-2.3 to 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
Mental retardation				2
Mitochondrial disorder				1
Motor retardation				9
Neurofibromatosis type 1				2
Neurological diseases <sup>b</sup>				3
Orthopedic disorders <sup>c</sup>				7
Sickle cell anemia				1
Turner syndrome				1

AO: 23

AQ: 12

<sup>a</sup> For age (in years), the median was 10.6, and the interquartile range was 8.5 to 12.9. BMI=body mass index, ADHD=attention deficit hyperactivity disorder, DCD=developmental coordination disorder.

<sup>b</sup> Microcephaly, Prader-Willi syndrome, and Erb palsy.

<sup>c</sup> Osteochondritis dissecans, progressive scoliosis (n=3), multidigit camptodactyly, clubfoot, and Perthes disease.

tigate longitudinal changes in healthrelated fitness, walking capacity, and HRQoL over time, we applied multilevel modeling with the multilevel program MLwiN 2.23 (Centre for Multilevel Modelling, University of Bristol, Bristol, United Kingdom.<sup>27</sup> An advantage of multilevel modeling over the traditional repeatedmeasurement approach is that all of the available results can be incorporated within the analysis,27 even if the number of measurements varies

between the participants because of missing data, assuming that the missing data are random.<sup>29,38</sup> 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.30,31 These predictors were entered into the model to find the best model fit. The model fit was evaluated by comparing the deviance  $(\ln -2 \log \text{ likeli-}$ hood) 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 (ie, lacking predictors), and as predictors are added to the model, the deviance changes. A simpler model can be rejected with a decrease in the deviance and a P value of less than .05. An alpha level of .05 was adopted for all tests of significance.

#### Role of the Funding Source

The study was financed by the Johan Cruyff Foundation, the Rabobank Foundation, and SIA RAAK (2011-13-035P).

#### 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 the baseline characteristics are shown in Aq: 13 Table 2.

T1 Q: 13 T2, AO:24-27

AQ: 14

AO: 15

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%) (eg, 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 the missing data were 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 on the MPST, the total point score on the BOT-2, and the 6MWD are depicted in the Figure. 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.

#### **Multilevel Modeling**

**F1** 

T3

T4

**Body function.** The estimated models for the half-Bruce treadmill test and the MPST are shown in Table 3, and the estimated models for the strength subscale of the BOT-2 are 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 AQ: 16 height did not significantly influence outcome on the half-Bruce treadmill and, therefore, were not test included in the final model. After 3 and 6 months of training, estimated improvements in endurance time on the half-Bruce treadmill test were  $1.05 \text{ minutes} (P \le .001) \text{ and } 1.62 \text{ min-}$ utes ( $P \le .001$ ), respectively. Moreover, the predicted endurance time of 8.30 minutes (X=6.68, SD=1.62) at t2 was significantly higher than  $(6.68 \pm 1.05 \pm 7.73)$ that at t1  $(P \le .001)$ . The level 2 variance of 0.58 indicated the difference among participants, meaning that 58% of

#### Table 2.

Baseline Characteristics of Participants<sup>a</sup>

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

<sup>a</sup> ET=endurance time, SDS=standard deviation score: difference between the observed value and the predicted value divided by the standard deviation from reference values, MPST=Muscle Power Sprint Test, MP=mean power, 6MWD=6-minute walking distance, AE=age equivalent: typical age in a norm group with a similar score, BOT-2=Bruininks-Oseretsky Test of Motor Proficiency.

<sup>b</sup> Based on reference values from van der Cammen-van Zijp et al<sup>32</sup> ( $\leq$ 10 years) and Binkhorst et al<sup>33</sup> (>10 years).

Based on reference values from Douma-van Riet et al.<sup>15</sup>

 $^{\it d}$  Based on reference values from Geiger et al.  $^{\rm 30}$ 

<sup>e</sup> Based on unpublished data.

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, there-



#### Figure.

Bar plots of 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 of training (t2). \*P<.05.

fore, 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 ( $P \le .001$ ), respectively. Moreover, the predicted mean power at t2 was significantly higher than that at t1 (P = .02). Furthermore, 79% of the total variance in mean power was attributable to differences between participants. The fol-

#### Table 3.

Multilevel Model for the Half-Bruce Treadmill Test, MPST, and 6MWT<sup>a</sup>

Parameter	Half-Bruce	Treadmill Test		MPST MP		6MWT
			Fixed Effe	cts		
	β	95% CI	β	95% CI	β	95% CI
Intercept	6.68	5.93 to 7.42 <sup>b</sup>	-405.62	-610.24 to -201.01 <sup>b</sup>	294.39	87.73 to 501.05 <sup>b</sup>
t1	1.05	0.69 to 1.40 <sup>b</sup>	17.38	2.46 to 32.30 <sup>b</sup>	22.86	8.45 to 37.27 <sup>b</sup>
t2	1.62	1.27 to 1.98 <sup>b</sup>	35.36	20.67 to 50.05 <sup>b</sup>	35.67	21.25 to 50.08 <sup>b</sup>
Height			3.78	2.41 to 5.15	1.62	0.24 to 3.00
Weight						
Sex						
	•		Random Eff	fects		·
	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				
			Devianc	e		
Final model	520.9		1,544.3		1,644.4	
Empty model	641.2		1,689.6		1,763.9	

<sup>a</sup> 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. <sup>b</sup> P<.05.

lowing equations were derived from the final model:

mean power at t0 = -405.62

+ 3.78  $\times$  height (cm)

mean power at t1 = -405.62

$$+ 17.38 + 3.78 \times \text{height (cm)}$$

mean power at  $t^2 = -405.62$ 

 $+35.36 + 3.78 \times \text{height (cm)}$ 

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 ( $-405.62+17.38+3.78 \times 150$ ).

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 a 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 the SLJ were 7.90 cm ( $P \le .001$ ) and 8.52 cm ( $P \le .001$ ), 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*≤.001), 3.45 (*P*≤.001), 6.96 seconds (*P*≤.001), and 9.27 seconds ( $P \le .001$ ), respectively. After 6 months of training, estimated improvements were 4.54 ( $P \le .001$ ), 3.38 (*P*≤.001), 8.85 seconds  $(P \le .001),$ and 13.27 seconds  $(P \le .001)$ , respectively. No significant differences in muscle strength were found between t1 and t2. More-

 Table 4.

 Multilevel Model for the Bruininks-Oseretsky Test of Motor Proficienco<sup>a</sup>

	dn-V		95% CI	20.64 to 36.43 <sup>b</sup>	4.96 to 13.59 <sup>b</sup>	8.99 to 17.56 <sup>b</sup>					95% CI	0.04 to 0.48	-0.30 to 1.39	-0.70 to 1.09				
			β	28.54	9.27	13.27					Variance	0.26	0.55	0.19		1,281.2	1,386.0	
	/all Sit		95% CI	22.11 to 37.10 <sup>b</sup>	2.90 to 11.03 <sup>b</sup>	4.82 to 12.89 <sup>b</sup>					95% CI	0.04 to 0.43	-0.28 to 1.39	-0.72 to 1.14				
	N		β	29.11	6.96	8.85					Variance	0.23	0.56	0.21		1,269.1	1,376.7	
	it-ups		95% CI	-34.02 to 5.42 <sup>b</sup>	1.83 to 5.06 <sup>b</sup>	1.78 to $4.98^{b}$	0.07 to 0.33				95% CI	0.05 to 0.45	-0.24 to 1.16	-0.77 to 1.35				
	S	Fixed Effects	β	-14.30	3.45	3.38	0.20			andom Effect:	Variance	0.25	0.46	0.29	Deviance	978.9	1,107.0	
ourse of the second sec	: Push-ups		95% CI	6.15 to 13.08 <sup>b</sup>	2.77 to 6.40 <sup>b</sup>	2.74 to 6.35 <sup>b</sup>		)	/	R	95% CI	0.06 to 0.55	-0.24 to 0.96	-0.78 to 1.45				is of training.
	Knee		β	9.62	4.58	4.54	/				Variance	0.30	0.36	0.34		984.2	1,079.2	=after 6 month
	ng Long Jump		95% CI	-191.52 to -8.45 <sup>b</sup>	2.77 to 13.03 <sup>b</sup>	3.39 to 13.65 <sup>b</sup>	1.01 to 2.53	-1.86 to -0.60			95% CI	0.02 to 0.43	-0.42 to 1.89	-0.56 to 0.64				3 months of training, t2
	Standi		β	- 99.98	7.90	8.52	1.77	-1.23			Variance	0.23	0.74	0.03		1,339.0	1,489.7	erval, t1=after
	Parameter			Intercept	t1	t2	Height	Weight	Sex			For level 1	For level 2	For level 3		Final model	Empty model	<sup>a</sup> CI=confidence inte

over, 74% of the total variance in outcome on the SLJ was attributable to differences among participants. For the knee push-ups, sit-ups, wall sit, and v-up, the total variances in outcome attributable to differences among participants were 36%, 46%, 56%, and 55%, respectively. Furthermore, 3% of the total variance in outcome on the 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 the 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 the 6MWD were 22.9 m (P≤.001) and 35.7 m  $(P \le .001)$ , respectively. No significant differences in the 6MWD were found between t1 and t2. Moreover, 78% of the total variance in the 6MWD was attributable to differences among participants.

**HRQoL.** No significant intervention effects on self-reported and parent-reported family and social domains and on the self-reported total HRQoL score were found. Therefore, these domains were not included in the multilevel models. The estimated models for the selfreported 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 selfreported physical and emotional HRQoL were 4.64% ( $P \le .001$ ) and 4.67% (P=.01), respectively. Estimated improvements in parentreported physical and emotional HRQoL were 6.93% (P'.001) and 3.08% (P=.09), respectively, after 3 months of training and 7.25%  $(P \le .001)$  and 5.40%  $(P \le .00)$ , respectively, after 6 months of training. After 3 and 6 months of training, estimated improvements in the parent-reported total HROoL score were 3.13% (P=.02) and 4.56%  $(P \leq .00)$ , 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 HROoL 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 the parent-reported total HRQoL score was attributable to differences T5 among participants.

#### 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 the parent-reported total HRQoL score were found after 3 months of training. After 6 months of training, significant improvements in selfreported physical HRQoL and parentreported emotional HROoL 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 improvements during this period were found only for aerobic fitness and anaerobic fitness. A possible reason for the lack of significant improvements in muscle strength, walking capacity, and HRQoL during months 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.<sup>34</sup> Another possible reason is the law of "diminishing returns";

there is a decline in the effectiveness of a training program after a certain level of fitness has been achieved.<sup>34</sup>

The results of the present study are AQ: 17 in line with those found for pediatric patients who were diverse and homogeneous. Verschuren et al35 found improvements in aerobic fitness and anaerobic fitness in children with cerebral palsy after an 8-month training program with standardized exercises for aerobic fitness and anaerobic fitness (a 25% increase in watts of mean power measured with the MPST); the increase in the present study was 19% (154.1 W at t0; 184.6 W at t2) (Figure). Nsenga AQ: 18 Leunkeu et al<sup>36</sup> found significant improvements in the 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.37 In the study by Basaran et al,38 the 6MWD in children with asthma improved 4% after a moderately intensive basketball training program for 8 weeks. In the present study, a 7.5% increase in the 6MWD was found after 6 months of training (535.3 m at t0; 575.5 m at t2) (Figure).

> 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 an individual child.

The multilevel analysis used in the present study allowed us to identify

Se	ptem	ıber	201	4

<b>Table 5.</b> Multilevel Mode	l for the DU	X-25 Questionnair	<i>в</i> а.							
		Child	l Form				Par	ent Form		
Parameter	4	hysical	E	notional		Physical	E	notional		Total
	-		-		Fixed Effect	2				
	β	95% CI	β	95% CI	β	95% CI	β	95% CI	ß	95% CI
Intercept	61.65	54.07 to 69.23 <sup>b</sup>	72.71	67.98 to 77.43 <sup>b</sup>	63.42	58.67 to 68.18 <sup>b</sup>	70.61	66.14 to 75.07 <sup>b</sup>	72.51	68.91 to 76.10 <sup>b</sup>
t1	1.32	-2.31 to 4.95	3.90	0.08 to 7.72 <sup>b</sup>	6.93	3.00 to 10.87 <sup>b</sup>	3.08	-0.54 to 6.69	3.13	0.49 to 5.76 <sup>b</sup>
t2	4.64	1.18 to 8.10 <sup>b</sup>	4.67	0.99 to 8.35 <sup>b</sup>	7.25	3.50 to 10.99 <sup>b</sup>	5.40	1.99 to 8.81 <sup>b</sup>	4.56	2.08 to 7.05 <sup>b</sup>
Height										
Weight				/						
Sex	15.79	6.15 to 25.43								
					Random Effe	sets				
	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	-0.21 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	
<sup>a</sup> DUX-25=25-que: <sup>b</sup> P<.05.	tion version of i	the DUCATQOL, CI=(	confidence inter	rval, t1=after 3 mont	hs of training, t	2=after 6 months of t	raining.			

#### Fitkids Exercise Therapy Program

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.<sup>39-41</sup> The results of the present study showed height to be a significant predictor of the 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.42 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. The 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 are 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,<sup>43</sup> who suggested that with growth (ie, the increases in height and weight and other body changes that occur as a child matures), power output increases.

Previous research<sup>44</sup> indicated that jumping performance increased during growth in 11- to 16-year-old children. In our model, height and weight were found to be significant predictors 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<sup>45</sup> concluded that anthropometric variables that were deemed to influence the 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 to a small degree in the study of Veligekas.<sup>45</sup>

Participants' sex was revealed to be a significant predictor of the selfreported physical HRQoL. This result 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 the HRQoL.46-48 These data provide evidence that girls may experience 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.49

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 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 relatively high level 2 variance was found, indicating that a high level of the total variance in the test outcome was attributable to differences between participants. This result may have been due to the heterogeneous characteristics (eg, 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 differences between Fitkids centers did not contribute to the total variance in the test outcome. The Fitkids Foundation has organized several AQ: 19 training days to educate physical therapists at Fitkids centers in the administration of various aerobic exercise tests. It appears that these training days have been useful and that the 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 less standardized administration of the test and more variation in outcome.

A 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 incorporation of the Fitkids exercise therapy program into pediatric physical therapist practice, physical therapists are able to help children with a chronic condition or disability overcome barriers to fitness and sports and 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 longterm effects of the Fitkids exercise therapy program.

Ms Kotte, Dr Winkler, and Dr Takken provided concept/idea/research design and project management. Ms Kotte, Dr de Groot, Dr Huijgen, and Dr Takken provided writing and data analysis. Ms Kotte provided data collection. Dr Winkler provided fund procurement and administrative support. Dr Takken provided facilities/equipment and institutional liaisons. Dr de Groot, Dr Winkler, and Dr Takken provided consultation (including review of manuscript before submission).

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This study was approved by the Medical Ethics Committee of the University Medical Center Utrecht (number 11–325/C).

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# AUTHOR QUERIES

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- 1—AQ1: OK to replace the term "gender" with the term "sex" throughout? **PTJ generally prefers** to use the term "sex," which refers to the biological characteristics of males and females. The term "gender" includes more than sex and serves as a cultural indicator of someone's personal and social status.
- 2—AQ2: References 32 and 33 are cited only in the footnotes of Table 2. Can they be cited someplace in the text? Also, note that references in PTJ articles should be cited in numeric order (some are cited out of order); editorial staff will renumber references, as needed, after author review of the rapid proof. Some long paragraphs were divided into shorter paragraphs for ease of reading.
- 3—AQ3: "typical children" was changed to "children without chronic conditions or disabilities" in accordance with the PTJ preference for "people-first" language and because "typical" is nonspecific.
- 4-AQ4: Sense of sentence beginning "Each session" OK as edited?
- 5—AQ5: "healthy subjects" was changed throughout to "people who were healthy" in accordance with PTJ preferences for "people first" language and for "people" or "participants" rather than "subjects." Please check this change and similar changes throughout.
- 6-AQ6: Sense of sentence beginning "Outcome measures" OK as edited?
- 7—AQ7: In the sentence "Participants were instructed to cross the line," insertion of "finish" before "line" OK?
- 8—AQ8: Sense of sentence beginning "Power (W)" OK as edited? If not, please clarify.
- 9—AQ9: Please spell out "DUCATQOL" here and in footnote **a of Table 5**.
- 10—AQ10: Name and manufacturer of the statistical software program edited according to current manufacturer's information. According to the manufacturer, versions produced by SPSS Inc prior to the IBM acquisition (versions 18 and earlier) would be given an origin or publisher of SPSS Inc in Chicago, and versions released after the acquisition would be given an origin or publisher of IBM Corp in Armonk, NY.
- 11—AQ11: "normal population" was changed to "people who were healthy" in accordance with the PTJ preference for "people-first" language and with the recommendation of the AMA Manual of Style to avoid the use of "normal" to describe people's health status.
- 12-AQ12: Inserted name and address of the source of MLwiN 2.23 correct?.

1

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- 13—AQ13: Should refs 28 and 29 be cited instead of refs 29 and 38 after "assuming that the missing data are random"? If refs 29 and 38 are correct here, please note that both references are cited out of numeric sequence. Editorial staff will renumber references as needed after author review of the rapid proof.
- 14—AQ14: "−2 ln" was changed to "ln −2" in accordance with **AMA Manual of Style formatting** (10th ed., page 910).
- 15—AQ15: An acknowledgment of the funding source for this study has been added at the end of the Method section (with the specific paragraph heading "**Role of the Funding Source**") in accordance with PTJ policy. This information has been repeated at the end of the text for print and online downstreaming purposes.
- 16—AQ16: PTJ style is to allow no more than 3 levels of heads. Therefore, headings below the third level were changed to third-level headings. Please check organization of all headings throughout.
- 17—AQ17: Please check the paragraph beginning "The results of the present study are in line" carefully. OK to replace plus signs with "increase" throughout the paragraph?
- 18—AQ18: Please note that ref 35 repeats ref 4 in the reference list. Editorial staff will delete the repeated reference and renumber references as needed after author review of the rapid proof Also, in "the increase in the present study was 19%," should "19%" be "17%"? 154.1/184.6=83%, and 100%-83%=17%. Or is one of the values in parentheses incorrect?
- 19-AQ19: Sense of sentence beginning "This result indicated" OK as edited?
- 20—AQ20: Please specify year of publication in reference 34.
- 21—AQ21: Please note that ref 35 repeats ref 4 in the reference list. Editorial staff will delete the repeated reference and renumber references as needed after author review of the rapid proof.
- 22—AQ22: Tables 1 and 2 were reorganized in accordance with PTJ style. Please check both tables carefully.
- 23—AQ23: Data for age were moved to a footnote because they were the only data given as median and interquartile range.
- 24—AQ24: What is a "norm group"? Should "norm" be changed to "normative"?
- 25—AQ25: "14" was changed to "15" because reference 15 is Douma et al.

# AUTHOR QUERIES

## AUTHOR PLEASE ANSWER ALL QUERIES

- 26—AQ26: "26" was changed to "30" because reference 30 is Geiger et al.
- 27—AQ27: Whose unpublished data? Please provide initials and last names, professional degrees, and the date when the data were obtained (eg, "J. J. Smith, MD, unpublished data, January 2010").