



# CHAPTER 5

## Fitness and Mobility Capacity in School-aged Children with Cerebral Palsy: a Longitudinal Analysis

Astrid CJ Balemans, Leontien van Wely, Jules G Becher,  
Annet J Dallmeijer

Submitted

## ABSTRACT

**Introduction:** The purpose of this study was to determine the longitudinal relationship among fitness components and between fitness and mobility capacity in children with cerebral palsy (CP).

**Methods:** Forty-six children with a bilateral (N = 24) or a unilateral spastic CP (N = 22), aged 7-13 years, participated in aerobic and anaerobic fitness measurements on a cycle ergometer and isometric muscle strength tests. Mobility capacity was assessed with the gross motor function measure (GMFM) and a walking capacity test. Longitudinal relationships (3 or 4 measurements over one year) were determined using repeated measurements analyses.

**Results:** In children with bilateral CP, aerobic fitness was strongly related to anaerobic fitness ( $p < 0.001$ ), while aerobic fitness showed a weak relationship with muscle strength ( $p < 0.05$ ). Anaerobic fitness was not related to muscle strength. No relationships between fitness components were found in unilateral CP. Anaerobic fitness and muscle strength were significant determinants for GMFM and walking capacity in bilateral but not in unilateral CP.

**Conclusion:** The strong longitudinal relationship between aerobic and anaerobic fitness indicates that increasing either aerobic or anaerobic fitness corroborates with improvements in the other fitness component in children with bilateral CP only. While increasing anaerobic fitness might be beneficial for mobility capacity in children with bilateral CP, this is less likely for children with unilateral CP.

## INTRODUCTION

Emerging evidence shows that fitness is an important health indicator in all individuals.<sup>5;14</sup> Adequate fitness might be even more important for persons with a physical disability, who often have lower levels of fitness and higher risks of developing secondary health conditions including obesity, diabetes and cardiovascular disease.<sup>18</sup> It has also been suggested that adequate fitness in children with a physical disability associates with a better capacity to execute daily activities.<sup>3</sup> The most common physical disability in childhood is cerebral palsy (CP), a group of motor disorders characterized by activity limitations due to motor impairments.<sup>17;26</sup> As decreased fitness levels might increase the impact of CP on activities of daily living,<sup>7</sup> improving fitness might therefore be an important issue to consider when aiming to lower the impact of the disorder on activities of daily living, and to improve general health in persons with CP.

There is accumulating evidence that children with CP have decreased aerobic fitness, anaerobic fitness and muscle strength.<sup>2;36;39</sup> Decreases in muscle strength and anaerobic fitness might be expected in light of the impaired muscle activation in CP, whereas aerobic fitness seems to be more closely related to physical inactivity.<sup>2</sup> While muscle strength training has received increasing attention over the years,<sup>40</sup> interventions aimed at improving aerobic fitness have been undertaken less often.<sup>25</sup> Studies addressing aerobic fitness training focused on adolescents<sup>4</sup> or used field tests to estimate aerobic fitness, which also depend on walking ability such as time to complete a running test.<sup>42</sup> Objective assessments that measure aerobic fitness directly, such as peak oxygen uptake, the anaerobic threshold, oxygen pulse and ventilatory factors have been evaluated less frequently following interventions in children with CP.<sup>23;35</sup> More recently, it was suggested that more high intensity short duration (anaerobic) components should be included in fitness training programmes.<sup>20</sup> This type of training better suits the activity patterns of children and is promising in regard to improving aerobic and anaerobic fitness.<sup>1;20</sup> However, a recent multi-component intervention including both fitness training including strength and anaerobic exercises, combined with a lifestyle intervention, showed no effect on physical fitness.<sup>13</sup> In both groups a large inter-individual variation in changes in physical fitness was found, indicating that some subjects improved, while others deteriorated despite their group allocation. A secondary analysis on these data allows investigation of relationships between changes in physical fitness which might have been averaged out in the group mean values, and also allows subgroup differentiation in these relationships.<sup>19</sup>

Apparently, a better understanding of limited maximal exercise is required to provide guidance in setting up training programmes aimed at improving fitness in children with CP. A recent cross-sectional study suggested that aerobic and anaerobic fitness are limited by reduced muscle strength.<sup>11</sup> This was indicated by the stronger correlations between these fitness components in adults with CP than in controls.<sup>11</sup> Due to the differences in coordination and muscle function between unilateral and bilateral involved CP, a different relation among fitness components might also be expected. The strong cross-sectional correlations

in the previously mentioned study may be influenced by the large inter-individual variability of fitness levels in subjects with CP. This cross-sectional relationship does not preclude a relation between changes in fitness components. Therefore, longitudinal relationships might provide better insights into whether a change in one fitness component is related to a change in the other.<sup>34</sup>

Associations of muscle strength and anaerobic fitness with mobility capacity were shown in cross-sectional studies.<sup>24;41</sup> However, strength training studies showed that improvements in muscle strength did not necessarily lead to increased mobility capacity.<sup>29;30;32</sup> Moreover, evidence for a better mobility capacity as a result of improved anaerobic and aerobic fitness is even sparser.<sup>42</sup> The above-mentioned differing relationships in unilateral and bilateral CP may also apply to relations between fitness and mobility capacity. Since mobility capacity is highly related to the severity of the motor disorder, the anatomical involvement is likely to influence this relation.<sup>15</sup> Therefore, stronger relationships among fitness components and between fitness and mobility capacity are expected for children with a bilateral CP than for children with a unilateral CP, since children with a bilateral CP are more limited by muscle strength and coordination. The purpose of this study was to investigate the longitudinal relations between changes in the fitness components aerobic fitness, anaerobic fitness and muscle strength, and between fitness and mobility capacity in children with CP.

## **METHODS**

### **Participants**

This study is a secondary analysis of a 6-month single-blinded, randomized controlled trial, with a 6-month follow-up, to assess the effects of a physical activity stimulation program (*LEARN 2 MOVE 7-12* study).<sup>38</sup> Children with spastic CP were recruited in special schools for children with disabilities and pediatric physiotherapy practices in the Netherlands, between September 2009 and February 2012. The intervention program included 49 children with CP. A total of 46 children completed the study. Children were included when the following criteria were met: 1) 7-13 years old; 2) Gross Motor Function Classification System (GMFCS) level I, II or III; 3) no history of botulinum toxin injections and/or serial casting in the past 3 months or surgery in the past 6 months; 4) no contraindications for maximal exercise; and 5) less active than the international physical activity norm, no regular participation in sports or (physiotherapeutic) fitness program, and experience of problems related to daily life mobility or sports.<sup>37</sup> The institutional ethics committee of the VU University Medical Center approved this study and all participants above 12 years of age and all parents signed an informed consent.

### **Study design**

This study included measurements at baseline, 4 months, 6 months and 12 months, which were performed at approximately the same time during the day within each child.

A measurement session took place at the outpatient unit of a university medical centre and included determination of anthropometry, physical fitness and mobility capacity. The children were instructed not to eat or drink (except for water) 1.5 hours prior to the measurement session. All measurements, except for the walking test, were performed in a laboratory with a temperature between 19-22 °C and a relative humidity of 41-50%. Fitness testing consisted of a maximal aerobic exercise test, an anaerobic Wingate test, isometric strength testing of the knee extensors and the hip abductors, after which mobility capacity was determined by measuring gross motor function and walking capacity.

## Outcome measures and materials

### *Anthropometry*

Measures of body height [m] and weight [kg] were determined using an electronic scale (DGI 250D, KERN DE version 3.3 10/2004; Kern & Sohn GmbH Balingen-Frommern). Body mass index (BMI [ $\text{kg}/\text{m}^2$ ]) was calculated. Skinfold was measured at the supra-iliac and the sub scapular site using a Holtain skinfold caliper (ProCare BV, Groningen, the Netherlands) with an accuracy of 0.2 mm, providing a summed score of skinfold thickness [mm].<sup>31</sup>

### *Physical fitness*

Physical fitness was comprised of aerobic fitness, anaerobic fitness and isometric muscle strength. Aerobic fitness was determined during a maximal aerobic exercise test performed on a cycle ergometer (Corival V2; Lode B.V., Groningen, the Netherlands). The protocol was based on the McMaster protocol and consisted of a resting period enabling habituation to the mask used for gas analysis, 3 min of warming-up, 5 min of submaximal cycling and a maximal phase with the load increased each minute. The load increments were adapted to match the abilities of the child.<sup>2</sup> Respiratory gas was collected breath-by-breath with a gas analysis system. The corresponding software (Quark CPET, Cosmed, S.r.l., Rome, Italy) was calibrated with a known mixture prior to testing. Heart rate was measured with a heart rate monitor (Cosmed, S.r.l., Rome, Italy). Maximal aerobic parameters were calculated over the 30 s with the highest sustained load. Achievement of maximal aerobic exercise was checked using objective criteria: heart rate (HR) > 180 or respiratory exchange ratio (RER) > 1.00, and if subjective exhaustion was present.<sup>2</sup> Maximal aerobic parameters were  $\text{VO}_2$  peak, peak  $\text{O}_2$  pulse ( $\text{VO}_2/\text{HR}$  [ $\text{mL}\cdot\text{beat}^{-1}$ ]), peak ventilation ( $\text{VE}_{\text{peak}}$  [ $\text{L}\cdot\text{min}^{-1}$ ]). The anaerobic threshold (AT) was determined with the V-slope method by 2 independent raters. The test-retest reliability of this protocol for  $\text{VO}_2$  peak was shown to be excellent in children with CP, with an intraclass correlation coefficient (ICC) of 0.94 and a standard error of measurement (SEM) of  $2.06 \text{ ml}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$ .<sup>6</sup> Anaerobic fitness was measured using the 20 s Wingate anaerobic cycling test, which is a sprint test against a constant breaking torque. The test was performed for 20 s, providing the delivered mean power over 20 s ( $\text{P20}_{\text{mean}}$  [ $\text{W}\cdot\text{kg}^{-1}$ ]), which was used for analyses. Reliability assessment of  $\text{P20}_{\text{mean}}$  using this protocol showed high ICCs of 0.96-0.99 and SEMs of 0.148-0.270  $\text{W}\cdot\text{kg}^{-1}$ .<sup>8</sup> Isometric muscle strength of the knee extensors and hip abductors of the most-affected leg was determined via hand-held dynamometry.

The 'make test' was performed with the child pushing against the dynamometer (MicroFet; Biometrics, Almere, the Netherlands) for 3 s with maximal force, while the leg was fixed by the assessor according to standardized procedure.<sup>44</sup> Strength (N) was multiplied by the moment arm resulting in the torque [Nm], and the mean of 3 measurements was calculated. Intersession reliability was found to be good with ICC > 0.82 and SEMs of 0.55 N·kg<sup>-1</sup>, (knee extensors) 1.21 N·kg<sup>-1</sup> (hip abductors).<sup>44</sup> Measurements were performed by the same assessor in each measurement session.

#### *Mobility capacity*

Gross motor function was determined with the Gross Motor Function Measure item sets (GMFM-IS).<sup>28</sup> The Gross Motor Ability Estimator (GMAE) was used to calculate the GMAE score.

Walking ability was assessed using the 1 min walk test. Children were asked to walk as fast as possible, without running, on an oval walkway for 1 min. Covered distance [m] was registered. This test was reliable in children with CP with an ICC of 0.94 and limits of agreement of 13.1 m.<sup>21</sup>

#### **Statistics**

Longitudinal relationships among fitness components and between fitness components and mobility capacity were determined with a univariate random coefficient analysis. This analysis corrects for the dependency of observations within one individual by estimating a random intercept and slope (if this shows a better fitting model).<sup>34</sup> Effect modification was investigated to determine whether the relationships were different for children with unilateral involvement and bilateral involvement. To assess determinants of mobility capacity a multiple random coefficient regression model was applied, using a forward selection procedure. Fitness variables that were significantly associated with mobility capacity ( $p < 0.05$ ) in the univariate analysis were included as determinants in the multiple regression model. Variables with strongest association were added to the model first and included if level of significance was  $p < 0.05$ . Data on walking capacity was corrected for height if the regression coefficient changed > 10%. Analyses were carried out using IBM SPSS Statistics, version 20 (SPSS Inc, Chicago, Illinois, USA).

## **RESULTS**

### **Participants**

Characteristics of the participants are listed in Table 5.1. All children agreed to participate in all measurements, except for two children who were unable to perform measurements at 12 months for practical reasons. At all measurement sessions, there were missing data for a variety of reasons including refusal to wear the mask, lack of motivation or equipment problems.

**TABLE 5.1 Patient Characteristics**

	All children (N=46)	Bilateral CP (N=24)	Unilateral CP (N=22)
Boy/Girl	26/20	12/12	14/8
Age [year]	9y7mo (1y8mo)	9y6mo (1y4mo)	9y8mo (2y)
Height [cm]	136.8 (12.4)	133.3 (8.6)	140.7 (14.7)
Weight [kg]	34.8 (11.1)	31.7 (7.9)	38.2 (13.1)
BMI [ $\text{kg}\cdot\text{m}^{-2}$ ]	18.2 (3.3)	17.7 (3.3)	18.7 (3.1)
Skinfold [mm]	26.3 (10.3)	26.0 (10.9)	26.7 (9.7)
GMFCS (I/II/III)	26/12/8	8/8/8	18/4/0

Abbreviations: CP: Cerebral palsy; GMFCS: Gross Motor Function Classification System.

### Longitudinal relations

Table 5.2 shows the descriptives of fitness and mobility capacity. Significant interactions showed that relations among fitness components and of fitness components with mobility capacity were different for children with unilateral involvement than for those with a bilateral involvement. Therefore, descriptives and relations were presented for the whole group, and separately by anatomical involvement (Table 5.3, 5.4 and 5.5).

#### *Relations between fitness components*

Table 5.3 and 5.4 show the relations among the fitness components aerobic fitness, anaerobic fitness and muscle strength. In children with bilateral CP, aerobic fitness parameters ( $\text{VO}_2\text{peak}$  ( $p < 0.001$ ), AT and  $\text{O}_2\text{pulse}$  ( $p < 0.05$ )) were related to anaerobic fitness, while aerobic fitness ( $\text{VE}/\text{VO}_2$  only) showed a weak relationship with muscle strength (knee extensors and hip abductors) ( $p < 0.05$ ). Anaerobic fitness was not related to muscle strength. In children with unilateral CP, no relations among fitness components were found.

#### *Relations with mobility capacity*

Univariate analyses showed that in children with a bilateral CP,  $\text{VO}_2\text{peak}$  ( $p < 0.05$ ), anaerobic fitness ( $p < 0.001$ ) and knee extensor strength ( $p < 0.05$ ) were related to gross motor function, while  $\text{VO}_2\text{peak}$  ( $p < 0.05$ ), anaerobic fitness ( $p < 0.001$ ) and hip abduction strength ( $p < 0.001$ ) were related to walking capacity (Table 5.5). Fitness components and mobility capacity were not related in children with a unilateral CP (Table 5.5). The multivariate models showed that anaerobic fitness and muscle strength were the most important determinants of gross motor function and walking capacity in children with a bilateral CP (Table 5.6), whereas in children with a unilateral CP, no fitness components were determinants for mobility capacity.

## DISCUSSION

This was the first study to investigate the longitudinal relationships among the fitness components aerobic fitness, anaerobic fitness and muscle strength, and between fitness and mobility capacity in children with CP. The results showed that in children with bilateral CP, changes in aerobic fitness were strongly related to changes in anaerobic fitness, while

**TABLE 5.2 Descriptives of Mobility Capacity and Fitness**

	Descriptives (Mean (SE))							
	Baseline	N	4 months	N	6 months	N	12 months	N
<b>Mobility capacity</b>								
<b>GMAE score</b>								
<u>Overall</u>	78.8 (2.0)	46	na		79.2 (2.0)	46	80.0 (2.1)	44
Bilateral	72.5 (3.2)	24	na		73.6 (3.1)	24	73.8 (3.3)	23
Unilateral	85.6 (1.5)	22	na		85.2 (1.6)	22	86.7 (1.5)	21
<b>Walking capacity [m]</b>								
<u>Overall</u>	89.0 (3.0)	46	92.7 (3.2)	44	93.9 (3.2)	45	91.8 (3.4)	42
Bilateral	77.5 (3.9)	24	81.2 (3.8)	25	81.6 (4.1)	23	79.8 (4.2)	23
Unilateral	101.6 (2.6)	22	107.8 (2.9)	19	106.8 (3.0)	22	106.2 (3.2)	19
<b>Fitness</b>								
<b>Aerobic fitness</b>								
<b>VO<sub>2</sub>peak [ml·kg<sup>-1</sup>·min<sup>-1</sup>]</b>								
<u>Overall</u>	31.4 (1.0)	38	34.0 (1.2)	34	33.9 (1.1)	35	33.5 (1.2)	35
Bilateral	29.0 (1.5)	18	34.1 (2.1)	17	31.4 (1.8)	14	32.8 (2.2)	18
Unilateral	33.5 (1.2)	20	33.8 (1.1)	17	35.5 (1.3)	21	34.2 (1.0)	17
<b>Anaerobic threshold [ml·kg<sup>-1</sup>·min<sup>-1</sup>]</b>								
<u>Overall</u>	16.8 (0.7)	41	19.7 (0.7)	40	18.7 (0.8)	40	17.4 (0.7)	41
Bilateral	15.5 (1.2)	20	19.5 (1.0)	23	17.6 (1.2)	19	17.2 (1.0)	22
Unilateral	18.1 (0.9)	21	20.0 (0.9)	17	19.8 (1.1)	21	17.6 (0.9)	19
<b>O<sub>2</sub> pulse [ml·beat<sup>-1</sup>]</b>								
<u>Overall</u>	5.9 (0.4)	37	6.7 (0.4)	34	7.0 (0.4)	35	7.0 (0.4)	35
Bilateral	5.0 (0.3)	18	5.9 (0.3)	17	5.8 (0.3)	14	6.2 (0.4)	18
Unilateral	6.8 (0.6)	19	7.6 (0.6)	17	7.7 (0.6)	21	7.9 (0.7)	17
<b>VE/VO<sub>2</sub></b>								
<u>Overall</u>	42.2 (1.3)	38	38.8 (0.9)	34	38.6 (0.9)	35	37.7 (1.1)	35
Bilateral	44.4 (2.1)	18	39.1 (1.2)	17	39.4 (1.7)	14	38.4 (1.9)	18
Unilateral	40.3 (1.4)	20	38.5 (1.3)	17	38.0 (1.1)	21	36.9 (1.0)	17
<b>Anaerobic fitness</b>								
<b>P20mean [W·kg<sup>-1</sup>]</b>								
<u>Overall</u>	3.5 (0.2)	44	3.6 (0.2)	44	3.4 (0.2)	44	3.6 (0.2)	43
Bilateral	3.0 (0.3)	23	2.9 (0.3)	25	2.7 (0.3)	23	3.0 (0.2)	22
Unilateral	4.1 (0.3)	21	4.4 (0.3)	19	4.2 (0.2)	21	4.2 (0.3)	21
<b>Muscle strength</b>								
<b>Knee ext [Nm·kg<sup>-1</sup>]†</b>								
<u>Overall</u>	1.17 (0.05)	46	1.21 (0.05)	44	1.20 (0.05)	46	1.17 (0.05)	44
Bilateral	1.13 (0.07)	24	1.18 (0.08)	25	1.13 (0.08)	24	1.06 (0.08)	23
Unilateral	1.21 (0.06)	22	1.26 (0.06)	19	1.27 (0.07)	22	1.29 (0.06)	21
<b>Hip Abd [Nm·kg<sup>-1</sup>]†</b>								
<u>Overall</u>	0.84 (0.04)	46	0.83 (0.03)	44	0.89 (0.04)	46	0.80 (0.04)	44
Bilateral	0.76 (0.06)	24	0.81 (0.05)	25	0.82 (0.07)	24	0.73 (0.05)	23
Unilateral	0.92 (0.05)	22	0.86 (0.03)	19	0.95 (0.04)	22	0.89 (0.05)	21

Abbreviations: Knee ext: isometric knee extensor muscle strength; Hip Abd: isometric hip abductor muscle strength; na: not assessed; GMAE: Gross Motor Ability Estimator; P20mean: mean anaerobic power; †: of the non-dominant leg.

changes in aerobic fitness were weakly related to changes in muscle strength. In children with a unilateral CP, no longitudinal relationships between fitness components were found. Changes in anaerobic fitness, and to a lesser extent changes in muscle strength, determined mobility capacity in children with a bilateral CP, while none of the fitness components were related to mobility capacity in children with a unilateral CP.

Anaerobic fitness was positively and strongly related to aerobic fitness in children with a bilateral CP. It appears that low anaerobic capacity might limit aerobic fitness, while improving capacity to achieve short duration high intensity exercise (anaerobic capacity) might contribute to a higher peak oxygen uptake. This strong relationship was also found in a cross-sectional study undertaken in adults with CP, with a weaker relationship found in adults without CP.<sup>11</sup> The relationship between aerobic and anaerobic fitness is in agreement with the finding that aerobic fitness can be improved through anaerobic training in children with CP.<sup>42</sup> In our study, the longitudinal component of the regression coefficient is indicative for the actual relationship in changes between parameters. In more detail, when exercise is performed with a gradual increase of intensity, as in the maximal aerobic exercise test, the oxygen debt that occurs with every step increase requires an anaerobic contribution to achieve a new steady state.<sup>43</sup> Therefore, for children with physical activity patterns that are characterized by short, intermittent activities, anaerobic training programs might also have potential for improving aerobic fitness which might eventually be beneficial to general health.<sup>1,2</sup>

Muscle strength appears to be a less important limiting factor for aerobic and anaerobic fitness, as indicated by weak relationships. This corresponds to the weak relationships reported in adults with CP for isometric knee muscle strength of the more impaired leg and aerobic and anaerobic fitness<sup>11</sup>, although the relationships for muscle strength of the less impaired leg were stronger.<sup>11</sup> However, these conclusions were based on a combined sample of unilateral and bilateral involved adults with CP. As indicated in our study, results may differ for persons with CP with a different anatomical involvement. In addition, our study included prepubertal children in whom muscle characteristics differ from adults due to hormonal factors.<sup>27</sup> Our results show that  $VE/VO_2$  relates to muscle strength in children with bilateral CP. This might be indicative for a better ventilation efficiency due to changes in muscle volume, which could be beneficial in terms of muscle strength and oxidative capacity.<sup>45</sup>

The lack of relationships among fitness components in children with a unilateral CP in this study cannot be compared to previous studies as these relationships have not previously been evaluated for these subgroups separately. However, in the adult study, stronger correlations among fitness components were shown for adults with CP compared to those without CP.<sup>11</sup> This finding was attributed to the limiting effect of muscle strength in CP. The stronger correlations for the less impaired leg might indicate that muscle strength of the less impaired leg is more important for cycling performance. This might explain the lack of

TABLE 5.3 Relations between Aerobic Fitness with Anaerobic Fitness and Muscle Strength

Aerobic fitness	VO <sub>2</sub> peak [ml·kg <sup>-1</sup> ·min <sup>-1</sup> ]			Anaerobic threshold [ml·kg <sup>-1</sup> ·min <sup>-1</sup> ]			O <sub>2</sub> pulse [ml·beat <sup>-1</sup> ]			VE/VO <sub>2</sub>		
	B (SE)	95% CI	p	B (SE)	95% CI	p	B (SE)	95% CI	p	B (SE)	95% CI	p
<b>Anaerobic fitness</b>												
<b>P20mean [W·kg<sup>-1</sup>]</b>												
Overall	<b>1.79 (0.48)</b>	<b>0.85 to 2.73</b>	<b>0.000</b>	<b>0.93 (0.34)</b>	<b>0.26 to 1.60</b>	<b>0.008</b>	0.22 (0.14)	-0.05 to 0.49	0.112	-0.15 (0.56)	-1.27 to 0.97	0.791
Bilateral	<b>3.31 (0.67)</b>	<b>1.98 to 4.64</b>	<b>0.000</b>	<b>1.10 (0.47)</b>	<b>0.16 to 2.05</b>	<b>0.023</b>	<b>0.43 (0.21)</b>	<b>0.00 to 0.85</b>	<b>0.049</b>	0.25 (0.83)	-1.40 to 1.90	0.766
Unilateral	0.52 (0.65)	-0.77 to 1.81	0.429	0.49 (0.57)	-0.63 to 1.61	0.388	0.05 (0.18)	-0.30 to 0.41	0.760	-0.07 (0.84)	-0.17 to 1.59	0.933
<b>Muscle strength</b>												
<b>Knee ext [Nm·kg<sup>-1</sup>]<sup>†</sup></b>												
Overall	1.40 (1.52)	-1.60 to 4.41	0.357	<b>2.52 (1.26)</b>	<b>0.02 to 5.02</b>	<b>0.048</b>	-0.01 (0.36)	-0.72 to 0.71	0.987	<b>3.97 (1.84)</b>	<b>0.32 to 7.62</b>	<b>0.033</b>
Bilateral	3.00 (2.07)	-1.09 to 7.10	0.149	2.56 (1.67)	-0.74 to 5.86	0.127	-0.37 (0.50)	-1.36 to 0.62	0.456	<b>4.93 (2.46)</b>	<b>0.07 to 9.79</b>	<b>0.047</b>
Unilateral	-0.11 (2.13)	-4.34 to 4.11	0.958	1.93 (1.93)	-0.19 to 5.74	0.320	0.35 (0.49)	-0.62 to 1.32	0.478	3.28 (2.67)	-1.99 to 8.55	0.221
<b>Hip Abd [Nm·kg<sup>-1</sup>]<sup>††</sup></b>												
Overall	<b>3.96 (1.95)</b>	<b>0.11 to 7.81</b>	<b>0.044</b>	2.30 (1.68)	-1.02 to 5.62	0.173	0.15 (0.46)	-0.76 to 1.07	0.741	<b>5.63 (2.41)</b>	<b>0.87 to 10.39</b>	<b>0.021</b>
Bilateral	5.38 (3.08)	-0.71 to 11.48	0.083	3.03 (2.31)	-1.55 to 7.61	0.192	-0.19 (0.79)	-1.74 to 1.37	0.814	<b>7.97 (3.52)</b>	<b>1.00 to 14.75</b>	<b>0.025</b>
Unilateral	3.11 (2.56)	-1.95 to 8.17	0.226	0.71 (2.52)	-4.27 to 5.69	0.778	0.34 (0.58)	-0.80 to 1.48	0.557	4.34 (3.31)	-2.20 to 10.89	0.192

Abbreviations: B: unstandardised coefficient; Knee ext: isometric knee extensor muscle strength; Hip Abd: isometric hip abductor muscle strength; †: of the non-dominant leg. Significant results are marked in bold text.

**TABLE 5.4 Relations between Anaerobic Fitness and Muscle Strength**

Anaerobic fitness	P20mean [W·kg <sup>-1</sup> ]		
	B (SE)	95% CI	<i>p</i>
<b>Muscle strength</b>			
<b>Knee ext [Nm·kg<sup>-1</sup>]†</b>			
<b>Overall</b>	0.38 (0.19)	0.00 to 0.76	0.051
Bilateral	0.37 (0.26)	-0.14 to 0.88	0.150
Unilateral	0.44 (0.28)	-0.11 to 0.99	0.117
<b>Hip Abd [Nm·kg<sup>-1</sup>]†</b>			
<b>Overall</b>	0.19 (0.24)	-0.29 to 0.67	0.431
Bilateral	0.52 (0.38)	-0.24 to 1.28	0.179
Unilateral	0.07 (0.33)	-0.58 to 0.72	0.839

Abbreviations: B: unstandardised coefficient; Knee ext: isometric knee extensor muscle strength; Hip Abd: isometric hip abductor muscle strength; †: of the non-dominant leg. Significant results are marked in bold text.

relationships between muscle strength of the more impaired leg in this study of children with a unilateral CP, with the least affected leg determining aerobic and anaerobic performance on a bicycle.

The strong longitudinal relationship between anaerobic fitness and mobility capacity indicates that improving anaerobic fitness might lead to a better mobility capacity in children with a bilateral CP. This confirms the suggestion of Bar-or et al. that peak mechanical power (i.e. anaerobic fitness) is a more important determinant of the ability to execute activities than aerobic fitness.<sup>3</sup> This supports the positive correlations between running sprint power and gross motor function found in a cross-sectional study including both unilateral and bilateral involved children with CP.<sup>41</sup> In addition, gross motor function increased following combined aerobic and anaerobic fitness training.<sup>42</sup>

As regards muscle strength, knee extensors were associated with a better gross motor function, which confirms previous findings.<sup>9</sup> The stronger association between hip abductors and walking ability confirms earlier findings on the relationship between hip abductors and walking capacity.<sup>12</sup> However, it is remarkable, given that gross motor function is strongly associated with the severity of motor impairment,<sup>16</sup> that the higher innervated knee extensors show a stronger association with GMFM than the lower innervated hip abductors, which are more severely impaired as a consequence of the motor impairment.<sup>13</sup> In conclusion, improving gross motor function through fitness training focused on increasing anaerobic power, and to a lesser extent through increasing muscle strength, seems to have potential in children with a bilateral CP.

Increasing anaerobic power might not contribute to improvement of gross motor function and walking capacity in children with a unilateral CP, as indicated by the lack of a relationship in this group. The differing relationships in unilateral and bilateral involved CP might explain the fact that gross motor function and walking capacity did not improve with increased knee extension and hip abduction muscle strength, since no distinction was made between

**TABLE 5.5 Relations of Fitness with Mobility Capacity**

	GMAE			Walking capacity		
	B (SE)	95 %CI	p	B (SE)	95 %CI	p
<b>Aerobic fitness</b>						
<b>VO<sub>2</sub>peak [ml·kg<sup>-1</sup>·min<sup>-1</sup>]</b>						
Overall	0.063 (0.124)	-0.184 to 0.309	0.613	<b>0.32 (0.20)<sup>1</sup></b>	<b>-0.08 to 0.71</b>	<b>0.112</b>
Bilateral	<b>0.295 (0.143)</b>	<b>0.011 to 0.579</b>	<b>0.042</b>	<b>0.52(0.22)<sup>1</sup></b>	<b>0.09 to 0.96</b>	<b>0.019</b>
Unilateral	-0.208 (0.167)	-0.541 to 0.125	0.217	0.23 (0.27) <sup>1</sup>	-0.30 to 0.76	0.395
<b>Anaerobic threshold [ml·kg<sup>-1</sup>·min<sup>-1</sup>]</b>						
Overall	0.023 (0.109)	-0.193 to 0.239	0.832	0.19 (0.19)	-0.17 to 0.56	0.299
Bilateral	0.099 (0.143)	-0.186 to 0.384	0.491	0.34 (0.24)	-0.14 to 0.81	0.164
Unilateral	-0.106 (0.160)	-0.424 to 0.213	0.512	-0.04 (0.27)	-0.56 to 0.49	0.895
<b>Anaerobic fitness</b>						
<b>P20mean [W·kg<sup>-1</sup>]</b>						
Overall	<b>2.75 (0.67)</b>	<b>1.43 to 4.07</b>	<b>0.000</b>	<b>5.4 (1.1)<sup>1</sup></b>	<b>3.2 to 7.5</b>	<b>0.000</b>
Bilateral	<b>5.14 (0.90)</b>	<b>3.34 to 6.94</b>	<b>0.000</b>	<b>7.9 (1.3)<sup>1</sup></b>	<b>5.4 to 10.5</b>	<b>0.000</b>
Unilateral	1.54 (0.91)	-0.26 to 3.34	0.093	2.5 (1.5) <sup>1</sup>	-0.46 to 5.4	0.097
<b>Muscle strength</b>						
<b>Knee ext [Nm·kg<sup>-1</sup>]<sup>†</sup></b>						
Overall	3.25 (1.86)	-0.43 to 6.94	0.083	3.5 (3.3) <sup>1</sup>	-3.1 to 10.1	0.294
Bilateral	<b>7.88 (2.49)</b>	<b>2.93 to 12.82</b>	<b>0.002</b>	8.4 (4.3) <sup>1</sup>	-0.1 to 17.0	0.053
Unilateral	-0.97 (2.61)	-6.14 to 4.21	0.712	1.3 (4.9) <sup>1</sup>	-8.4 to 11.1	0.788
<b>Hip Abd [Nm·kg<sup>-1</sup>]<sup>†</sup></b>						
Overall	1.01 (2.47)	-3.89 to 5.92	0.683	<b>12.4 (4.2)<sup>1</sup></b>	<b>4.1 to 20.7</b>	<b>0.003</b>
Bilateral	7.65 (4.01)	-0.29 to 15.58	0.059	<b>27.7 (5.8)<sup>1</sup></b>	<b>16.1 to 39.2</b>	<b>0.000</b>
Unilateral	-0.77 (3.35)	-7.44 to 5.90	0.820	5.1 (5.9) <sup>1</sup>	-6.5 to 16.7	0.386

Abbreviations: B: unstandardised coefficient; Knee ext: isometric knee extensor muscle strength; Hip Abd: isometric hip abductor muscle strength; <sup>1</sup> corrected for height; <sup>†</sup>: of the non-dominant leg. Significant results are marked in bold text.

children with bilateral or unilateral CP in these studies.<sup>29;30;32</sup> A previous study found better gross motor function and walking capacity in children with a unilateral CP than in children with a bilateral CP within a given GMFCS level and suggested these components to be evaluated separately for unilateral and bilateral CP.<sup>10</sup> While improvements in mobility capacity might be expected following anaerobic fitness training in children with a bilateral CP, this is not expected for children with a unilateral CP. A different approach is required for improving mobility capacity in children with either a unilateral or a bilateral CP.

### Limitations

A limitation of this study was that isometric muscle strength of the dominant leg was not measured, while the least affected leg might determine aerobic and anaerobic performance. Another limitation was that only two muscle groups were included in the measurements.

**TABLE 5.6 Multiple Random Coefficient Regression Models for Mobility Capacity in Children with Bilateral Cerebral Palsy**

GMAE	B (SE)	95% CI	<i>p</i>	Walking capacity	B (SE)	95% CI	<i>p</i>
<b>Constant</b>	52.7 (4.6)	43.5 to 61.9	<0.001	<b>Constant</b>	5.5 (27.7)	50.0 to 61.0	0.844
P20mean	3.7 (1.1)	1.6 to 5.8	0.001	P20mean	5.9 (1.5)	2.9 to 8.8	<0.001
Knee ext <sup>†</sup>	9.1 (3.0)	3.1 to 15.2	0.004	Hip Abd <sup>†</sup>	15.2 (6.2)	2.9 to 27.4	0.016
				Height <sup>‡</sup>	0.3 (0.2)	0.1 to 0.8	0.124

Abbreviations: B: unstandardised coefficient; Knee ext: isometric knee extensor muscle strength; Hip Abd: isometric hip abductor muscle strength; <sup>‡</sup> as confounder; <sup>†</sup>: of the non-dominant leg.

## Conclusions

The strong longitudinal relationship between aerobic and anaerobic fitness indicates that increasing either aerobic or anaerobic fitness corroborates with improvements in the other fitness component in children with bilateral CP only. While increasing anaerobic fitness might be beneficial for mobility capacity in children with bilateral CP, this is less likely for children with unilateral CP. Different interventions are required for improving mobility capacity in children with either a unilateral or a bilateral CP.

## Perspective

There is increasing evidence that children with cerebral palsy (CP) have lower aerobic and anaerobic fitness. Similar declines have been detected in adults with CP.<sup>22</sup> As inactive children are more likely to become inactive adults,<sup>33</sup> interventions to improve fitness should start during childhood. Over the years, most training programs in children with CP have focused on increasing muscle strength. The results of the present study show that changes in anaerobic fitness strongly relate to aerobic fitness, as well as to mobility capacity in children with a bilateral CP. A focus on increasing anaerobic fitness in this group might contribute to better aerobic fitness, which is related to a better general health, in addition to an improved capacity for mobility in children with a bilateral CP. In the case of children with a unilateral CP, more research is required to reveal the limiting factors for exercise and mobility capacity that can subsequently be addressed in intervention programs.

## Acknowledgements

The authors would like to acknowledge Prof. dr. J. Twisk for his advice on with statistical analysis, Kim van Hutten for her help with data collection and Joyce van Tunen for her help with data analysis. This study was supported by a grant from The Netherlands Organisation for Health Research and Development (ZonMw) and the Phelps foundation for spastics.

## References

1. Bailey RC, Olson J, Pepper SL, Porszasz J, Barstow TJ, Cooper DM. The level and tempo of children's physical activities: an observational study. *Med Sci Sports Exerc.* 1995;27(7):1033-41.
2. Balemans ACJ, Van Wely L, De Heer SJA, Van Den Brink J, De Koning JJ, Becher JG, Dallmeijer AJ. Maximal aerobic and anaerobic exercise responses in children with cerebral palsy. *Med Sci Sports Exerc.* 2013;45(3):561-8.
3. Bar-Or O. Role of exercise in the assessment and management of neuromuscular disease in children. *Med Sci Sports Exerc.* 1996;28(4):421-7.
4. Bar-Or O, Inbar O, Spira R. Physiological effects of a sports rehabilitation program on cerebral palsied and post-poliomyelitic adolescents. *Med Sci Sports.* 1976;8(3):157-61.
5. Blair SN, Cheng Y, Holder JS. Is physical activity or physical fitness more important in defining health benefits? *Med Sci Sports Exerc.* 2001;33(6 Suppl):S379-S399.
6. Brehm MA, Balemans ACJ, Becher JG, Dallmeijer AJ. Reliability of a progressive maximal cycle ergometer test to assess peak oxygen uptake in children with mild to moderate cerebral palsy. *Phys Ther.* 2014;94(1):121-8.
7. Carlon SL, Taylor NF, Dodd KJ, Shields N. Differences in habitual physical activity levels of young people with cerebral palsy and their typically developing peers: a systematic review. *Disabil Rehabil.* 2013;35(8):647-55.
8. Dallmeijer A, Scholtes V, Brehm M, Becher J. Test-Retest Reliability of the 20-sec Wingate Test to Assess Anaerobic Power in Children with Cerebral Palsy. *Am J Phys Med Rehabil.* 2013;92(1).
9. Damiano DL, Martellotta TL, Sullivan DJ, Granata KP, Abel MF. Muscle force production and functional performance in spastic cerebral palsy: relationship of cocontraction. *Arch Phys Med Rehabil.* 2000;81(7):895-900.
10. Damiano D, Abel M, Romness M, Oeffinger D, Tylkowski C, Gorton G, Bagley A, Nicholson D, Barnes D, Calmes J, Kryscio R, Rogers S. Comparing functional profiles of children with hemiplegic and diplegic cerebral palsy in GMFCS Levels I and II: Are separate classifications needed? *Dev Med Child Neurol.* 2006;48(10):797-803.
11. De Groot S, Dallmeijer AJ, Bessems PJC, Lamberts ML, van der Woude LHV, Janssen TWJ. Comparison of muscle strength, sprint power and aerobic capacity in adults with and without cerebral palsy. *J Rehabil Med.* 2012;44(11):932-8.
12. Ferland C, Lepage C, Moffet H, Maltais DB. Relationships between lower limb muscle strength and locomotor capacity in children and adolescents with cerebral palsy who walk independently. *Phys Occup Ther Pediatr.* 2012;32(3):320-32.
13. Fowler EG, Staudt LA, Greenberg MB. Lower-extremity selective voluntary motor control in patients with spastic cerebral palsy: increased distal motor impairment. *Dev Med Child Neurol.* 2010;52(3):264-9.
14. Garber CE, Blissmer B, Deschenes MR, Franklin BA, Lamonte MJ, Lee IM, Nieman DC, Swain DP. American College of Sports Medicine position stand. Quantity and quality of exercise for developing and maintaining cardiorespiratory, musculoskeletal, and neuromotor fitness in apparently healthy adults: guidance for prescribing exercise. *Med Sci Sports Exerc.* 2011;43(7):1334-59.
15. Gorter JW, Rosenbaum PL, Hanna SE, Palisano RJ, Bartlett DJ, Russell DJ, Walter SD, Raina P, Galuppi BE, Wood E. Limb distribution, motor impairment, and functional classification of cerebral palsy. *Dev Med Child Neurol.* 2004;46(7):461-7.

16. Hanna SE, Bartlett DJ, Rivard LM, Russell DJ. Reference curves for the Gross Motor Function Measure: percentiles for clinical description and tracking over time among children with cerebral palsy. *Phys Ther.* 2008;88(5):596-607.
17. Himmelman K, Hagberg G, Uvebrant P. The changing panorama of cerebral palsy in Sweden. X. Prevalence and origin in the birth-year period 1999-2002. *Acta Paediatr.* 2010;99(9):1337-43.
18. Hombergen SP, Huisstede BM, Streur MF, Stam HJ, Slaman J, Bussmann JB, van den Berg-Emons R. Impact of cerebral palsy on health-related physical fitness in adults: systematic review. *Arch Phys Med Rehabil.* 2012;93(5):871-81.
19. Horn SD, DeJong G, Deutscher D. Practice-based evidence research in rehabilitation: an alternative to randomized controlled trials and traditional observational studies. *Arch Phys Med Rehabil.* 2012;93(8 Suppl):S127-S137.
20. Kessler HS, Sisson SB, Short KR. The potential for high-intensity interval training to reduce cardiometabolic disease risk. *Sports Med.* 2012;42(6):489-509.
21. McDowell BC, Kerr C, Parkes J, Cosgrove A. Validity of a 1 minute walk test for children with cerebral palsy. *Dev Med Child Neurol.* 2005;47(11):744-8.
22. Nieuwenhuijsen C, van der Slot WMA, Dallmeijer AJ, Janssens PJ, Stam HJ, Roebroek ME, Berg-Emons HJG. Physical fitness, everyday physical activity, and fatigue in ambulatory adults with bilateral spastic cerebral palsy. *Scand J Med Sci Sports.* 2011;21(4):535-42.
23. Nsenga AL, Shephard RJ, Ahmaid S. Aerobic training in children with cerebral palsy. *Int J Sports Med.* 2013;34(6):533-7.
24. Parker DF, Carriere L, Hebestreit H, Salsberg A, Bar-Or O. Muscle performance and gross motor function of children with spastic cerebral palsy. *Dev Med Child Neurol.* 1993;35(1):17-23.
25. Rogers A, Furler BL, Brinks S, Darrah J. A systematic review of the effectiveness of aerobic exercise interventions for children with cerebral palsy: an AACPD evidence report. *Dev Med Child Neurol.* 2008;50(11):808-14.
26. Rosenbaum P. A report: the definition and classification of cerebral palsy April 2006. *Dev Med Child Neurol.* 2007;49(6):480.
27. Round JM, Jones DA, Honour JW, Nevill AM. Hormonal factors in the development of differences in strength between boys and girls during adolescence: a longitudinal study. *Ann Hum Biol.* 1999;26(1):49-62.
28. Russell DJ, Avery LM, Walter SD, Hanna SE, Bartlett DJ, Rosenbaum PL, Palisano RJ, Gorter JW. Development and validation of item sets to improve efficiency of administration of the 66-item Gross Motor Function Measure in children with cerebral palsy. *Dev Med Child Neurol.* 2010;52(2):e48-e54.
29. Scholtes VA, Becher JG, Comuth A, Dekkers H, Van Dijk L, Dallmeijer AJ. Effectiveness of functional progressive resistance exercise strength training on muscle strength and mobility in children with cerebral palsy: a randomized controlled trial. *Dev Med Child Neurol.* 2010;52(6):e107-e113.
30. Scholtes VA, Becher JG, Janssen-Potten YJ, Dekkers H, Smallegenbroek L, Dallmeijer AJ. Effectiveness of functional progressive resistance exercise training on walking ability in children with cerebral palsy: a randomized controlled trial. *Res Dev Disabil.* 2012;33(1):181-8.
31. Tanner JM, Whitehouse RH. Revised standards for triceps and subscapular skinfolds in British children. *Arch Dis Child.* 1975;50(2):142-5.
32. Taylor NF, Dodd KJ, Baker RJ, Willoughby K, Thomason P, Graham HK. Progressive resistance training and mobility-related function in young people with cerebral palsy: a randomized controlled trial. *Dev Med Child Neurol.* 2013;55(9):806-12.

33. Telama R, Yang X, Viikari J, Valimaki I, Wanne O, Raitakari O. Physical activity from childhood to adulthood: a 21-year tracking study. *Am J Prev Med.* 2005;28(3):267-73.
34. Twisk JWR. Longitudinal data analysis. A comparison between generalized estimating equations and random coefficient analysis. *Eur J Epidemiol.* 2004;19(8):769-76.
35. Unnithan VB, Katsimanis G, Evangelinou C, Kosmas C, Kandrali I, Kellis E. Effect of strength and aerobic training in children with cerebral palsy. *Med Sci Sports Exerc.* 2007;39(11):1902-9.
36. Van Den Berg-Emons RJ, van Baak MA, de Barbanson DC, Speth L, Saris WH. Reliability of tests to determine peak aerobic power, anaerobic power and isokinetic muscle strength in children with spastic cerebral palsy. *Dev Med Child Neurol.* 1996;38(12):1117-25.
37. Van Wely L, Balemans ACJ, Becher JG, Dallmeijer AJ. Physical activity stimulation program for children with cerebral palsy did not improve physical activity: a randomised controlled trial. *J Physiother.* 2014;60(41).
38. Van Wely L, Becher JG, Reinders-Messelink HA, Lindeman E, Verschuren O, Verheijden J, Dallmeijer AJ. LEARN 2 MOVE 7-12 years: a randomized controlled trial on the effects of a physical activity stimulation program in children with cerebral palsy. *BMC Pediatr.* 2010;10:77.
39. Verschuren O, Takken T. Aerobic capacity in children and adolescents with cerebral palsy. *Res Dev Disabil.* 2010;31(6):1352-7.
40. Verschuren O, Ada L, Maltais DB, Gorter JW, Scianni A, Ketelaar M. Muscle strengthening in children and adolescents with spastic cerebral palsy: considerations for future resistance training protocols. *Phys Ther.* 2011;91(7):1130-9.
41. Verschuren O, Ketelaar M, Gorter JW, Helders PJM, Takken T. Relation between physical fitness and gross motor capacity in children and adolescents with cerebral palsy. *Dev Med Child Neurol.* 2009;51(11):866-71.
42. Verschuren O, Ketelaar M, Gorter JW, Helders PJM, Uiterwaal CSPM, Takken T. Exercise training program in children and adolescents with cerebral palsy: a randomized controlled trial. *Arch Pediatr Adolesc Med.* 2007;161(11):1075-81.
43. Wasserman K, Hansen JE, Sue DY, Stringer WW, Whipp BJ. Measurements During Integrative Cardiopulmonary Exercise Testing. *Principles of exercise testing and interpretation.* 4th ed. Lippincott Williams & Wilkins; 2005. p. 76-110.
44. Willemse L, Brehm MA, Scholtes VA, Jansen L, Woudenberg-Vos H, Dallmeijer AJ. Reliability of isometric lower-extremity muscle strength measurements in children with cerebral palsy: implications for measurement design. *Phys Ther.* 2013;93(7):935-41.
45. Wilmore JH, Costill DL. Respiratory regulation during exercise. *Physiology of Sport and Exercise.* Champaign, IL: Human Kinetics; 2004. p. 242-69.