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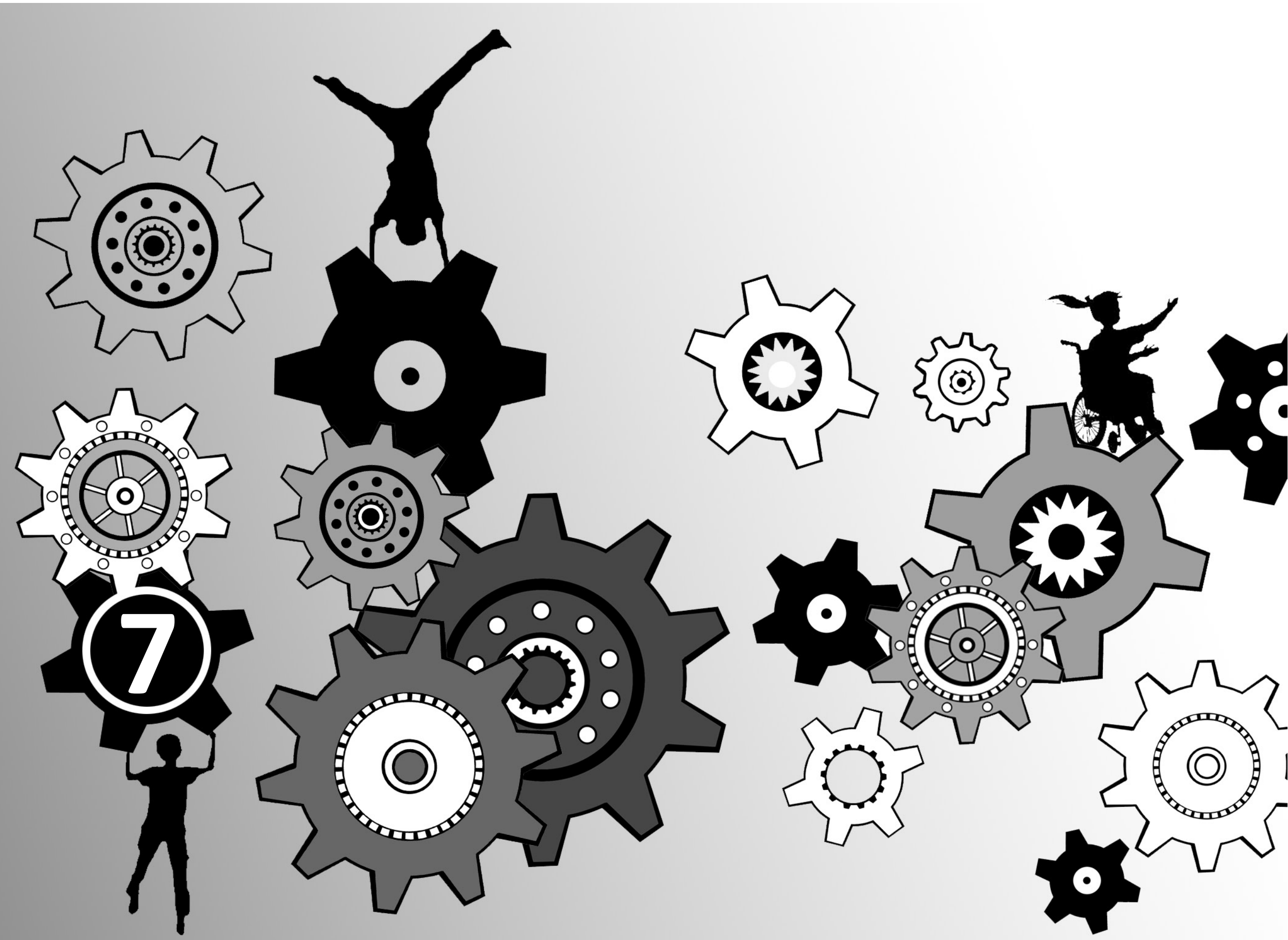

CHAPTER 7

Longitudinal Relationship between Physical Fitness, Walking-related Physical Activity and Fatigue in Children with Cerebral Palsy

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

Submitted

ABSTRACT

Objective: A vicious cycle of decreased physical fitness, early fatigue and low physical activity levels (PAL) is thought to affect children with cerebral palsy (CP). However, the relationship of changes in physical fitness to changes in PAL and fatigue is unclear. The objective of this study was to investigate the relation between changes in physical fitness, walking-related PAL and fatigue in children with CP.

Design: A secondary analysis of a randomized controlled trial with measurements at baseline, 6 months (after the intervention period) and 12 months.

Setting: Community.

Participants: 24 children with bilateral and 22 with unilateral spastic CP, aged 7-13, all walking, participated in this study.

Interventions: None.

Main Outcome Measures: Physical fitness was measured by aerobic capacity (VO_2peak), anaerobic threshold, anaerobic capacity and, isometric and functional muscle strength. Walking-related PAL was measured using an ankle-worn StepWatch™ activity monitor for 1 week. Fatigue was determined with the PedsQL multidimensional fatigue scale. Longitudinal relationships were analyzed by random coefficient analysis ($p < 0.05$).

Results: In children with bilateral CP, all fitness parameters showed a positive, significant relationship to walking-related PAL, whereas no relationship between physical fitness and walking-related PAL was seen in children with unilateral CP. No clinically relevant relationship between physical fitness and fatigue was found.

Conclusions: Children with bilateral spastic CP might benefit from an improved physical fitness to increase their PAL or vice versa, while this is not the case in children with unilateral CP. There seems no relationship between physical fitness and self-reported fatigue in all children with CP. Interventions aimed at improving PAL may be differently targeted in children with either bilateral or unilateral CP.

INTRODUCTION

It has been suggested that children with a physical disability may be trapped in a vicious circle of low physical fitness, early fatigue in daily activities and inactivity, resulting in deconditioning and a further decrease in physical activity.[19] The physical activity level (PAL) tends to deteriorate during the transition to adulthood, especially in persons with a physical disability.[18] From this perspective, establishing a healthy and active lifestyle during childhood is even more important for individuals with a disability, who are at higher risk for developing secondary conditions such as cardiovascular disease, diabetes and obesity.[19]

Cerebral palsy (CP) is the most common cause of physical disability in childhood[21], and is associated with low physical fitness[4], decreased PAL[8], and general fatigue[40]. The health-related components of physical fitness are defined by the American College of Sports Medicine (ACSM) as cardiorespiratory (aerobic) fitness, body composition, muscular strength and muscular endurance, and flexibility.[1] Anaerobic fitness is not a separate fitness component according this definition. However, anaerobic fitness is, next to aerobic fitness and muscular strength, an important determinant for physical activity and exercise in children who have short, intermittent activity patterns.[2;20] CP is defined as "a group of disorders of the development of movement and posture causing activity limitations that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain".[5;12] Due to motor impairments children with CP have higher energy requirements during certain activities such as walking.[10] Also, a lower anaerobic threshold in CP[4] implies that children with CP walk close to or above their AT[15], inducing early fatigue. The high energy requirements and the lower AT might cause a lower PAL as a compensatory mechanism to prevent fatigue, aggravating the above mentioned vicious circle.[25]

Recently, the focus of intervention programs to break this vicious circle of low physical fitness, early fatigue in daily activities and inactivity, resulting in deconditioning and a further decrease in physical activity has been on improving physical fitness and increasing PAL by fitness training and stimulating an active lifestyle.[36;39] It has been reported that physical fitness can be improved in children with CP[36;43], but it is not yet clear whether increased physical fitness levels lead to higher activity levels.[36] A recent multi-component physical activity intervention including both fitness training and a lifestyle intervention, showed no effect on physical fitness and walking-related PAL.[37] A large inter-individual variation in changes in both physical fitness and walking-related PAL was found in both groups. These findings indicate that some subjects improved, while others deteriorated despite their group allocation. Apparently, other factors than the intervention affected physical fitness and walking-related PAL. A secondary analysis on these data provide insight in the relationship between changes in physical fitness and walking-related physical activity, indicating whether improved physical fitness is related to higher PAL levels.

Understanding the longitudinal relationship between changes in physical fitness and PAL may contribute to the development of effective programs for improving both physical fitness and PAL in children with CP. While cross-sectional correlations may be influenced by the large inter-individual variability of physical fitness and physical activity levels in subjects with CP, longitudinal analyses provide information on intra-individual changes over time. This longitudinal relationship provides insight into whether a change in physical fitness is related to a change in walking-related PAL, which is in contrast to cross-sectional relationships which do not preclude a relation between changes in components.[35] Previous studies investigating the association between physical fitness and PAL using a cross-sectional design, found no associations between PAL and peak oxygen uptake (VO_2peak) in children[25] and adults with CP[28;33]. However, physical strain, defined as the oxygen consumption during walking expressed as a percentage of VO_2peak, did show an association with PAL.[25;33] Oxygen consumption during walking is elevated in CP and is closely related to the severity of the motor disorder.[17] Orthoses, orthopedic surgery or spasticity treatment have the potential to reduce the oxygen consumption during walking.[24;26;32] In addition, increasing the VO_2peak while the oxygen consumption during walking remains constant will reduce physical strain and may concomitantly improve walking-related PAL. Consequently, it is anticipated that changes in VO_2peak are related to changes in walking-related PAL. Our hypothesis is that if physical fitness changes in children with CP, this will be related to a change in PAL and to a change in fatigue. To investigate this, we performed a secondary analysis of the above mentioned intervention study.[37] Using a longitudinal design, the objective of this study was to investigate the relation between changes (either improvements or decreases, irrespective of group allocation) in physical fitness, walking-related PAL and fatigue in children with CP.

METHODS

Participants

This study included 24 children with bilateral CP and 22 children with unilateral CP who were recruited in specials schools or through physical therapy practices from August 2009 to February 2011. Measurements were performed at baseline, at 6 months (31 ± 2.4 weeks) and at 12 months (52 ± 3.4 weeks), and were part of a randomized controlled trial evaluating the effects of a physical activity stimulation program.[39] Inclusion criteria were: i) children with a spastic CP; ii) 7-13 years old; and iii) Gross Motor Function Classification System (GMFCS) level I, II (walking without aids) or III (walking with aids). Exclusion criteria were: i) contra indications for maximal exercise, ii) history of botulinum toxin injections and/or serial casting (<3 months), and iii) surgery (<6 months). This study was approved by the institutional ethics committee and all participants aged above 12 years and all participants' parents signed an informed consent form.

Procedure

Participants visited the out-patient unit. Measurements taken in the morning or in the afternoon were taken again at the same time 6 months later, and consisted of body composition assessments, physical fitness measurement and calibration of the StepWatch™ activity monitor. After this session children wore the StepWatch™ for one week during waking hours, excluding bathing time and swimming. Children were asked to fill in a questionnaire on experienced fatigue prior to each measurement session.

Measurements

Body composition

Body height and weight were measured on an electronic scale (DGI 250D, KERN DE version 3.3 10/2004; Kern&Sohn GmbH, Balingen-Frommern), which enabled calculation of body mass index (BMI). Consecutively, skin fold measurements were performed at the subscapular site and supra-iliac site of the non-dominant arm (bilateral) or non-affected side (unilateral) using a Holtain skinfold caliper (accuracy 0.2 mm; ProCare BV, Groningen, the Netherlands), providing a summed score of skin fold thickness.[34]

Physical fitness

Children performed a maximal aerobic exercise test on a cycle ergometer (Corival V2; Lode B.V., Groningen, the Netherlands) until exhaustion. The exercise test started with a 3 min warm up phase, followed by a 4-5 min steady-state phase at sub maximal exercise, 1 min rest, and finally a maximal phase starting with the load of the submaximal phase being incremented every minute based on height and GMFCS level.[11] Heart rate was measured using a heart rate monitor and gas was analyzed with a gas analysis system (Quark CPET, Cosmed, S.r.l., Rome, Italy) with the corresponding software (PFT CPET Suite, version 9.1b). Prior to testing, the flow sensor was calibrated with a 3 L syringe, and the O_2 and CO_2 concentration sensors were calibrated with ambient air and a reference gas of a known mixture. Data were included if the following criteria for achieving maximal exercise were met: either heart rate > 180 beats/min and/or if the respiratory exchange ratio > 1.00, and if subjective exhaustion was present.[3] VO_2peak was defined as the highest VO_2 over 30 s. Anaerobic threshold (AT) was determined by the V-slope method by two independent raters.[6] Reliability of VO_2peak assessments in CP children using this protocol was excellent (Intraclass Correlation Coefficient (ICC) of 0.94 and Standard Error of Measurement (SEM) of 2.06 $ml \cdot kg^{-1} \cdot min^{-1}$).[11]

A 20 s Wingate test on the cycle ergometer was performed; a full out sprint test against a constant workload, with the mean power output over 20 s (P20mean) representing anaerobic capacity. Wingate software (Wingate Software V1, Lode B.V., Groningen, the Netherlands) was used to apply the workload and to measure P20mean. Reliability of the 20 s Wingate test showed a high ICC of 0.99 and a SEM of 0.219 $W \cdot kg^{-1}$ for P20mean in children with CP.[16]

Isometric muscle strength of the knee extensors and the hip abductors was measured by use of the 'make test' with hand-held dynamometry (MicroFet; Biometrics, Almere, the Netherlands) at the non-dominant leg by taking the average over three measurements, preceded by a practice trial.[46] The child's limb was fixed by the assessor, and the child pushed for 3 s with maximal force against the dynamometer. Peak force [N] and the moment arm

[m] were measured.[46] This procedure's feasibility and good intersession reliability was shown in children with CP, with ICCs > 0.82 and a SEM of 11.3% (knee flexors) and 16.6% (hip abductors).[46]

Functional strength was measured with the lateral step up test (with both the dominant and the non-dominant leg) and the sit to stand test, where the number of repetitions over 30 s was determined.[42] The number of repetitions was summed over the three tests, resulting in a total score for functional muscle strength.[42]

Walking-related physical activity

Walking-related PAL was determined by measuring walking activity with the biaxial StepWatch™ Activity Monitor 3.0 (Cyma Corporation, Seattle WA, USA). This device was worn at the ankle of the dominant leg and measures accelerations of the leg in the frontal–sagittal plane per time interval.[14] The psychometric properties of this device have been shown to be good in typically developing children[9] and by adjusting the sensitivity settings, the Stepwatch can accurately record strides in children with CP.[38] Calibration was carried out with the subject walking on an oval 50 m track, while strides were counted manually and concurrently registered by the StepWatch™ device and sensitivity settings were adjusted until StepWatch™ recordings and manual counting agreed > 95%. StepWatch calibration resulted in an accuracy of 99.8 ± 3.4%. Mean values per minute were stored, providing average strides·min^{-1}. Walking-related PAL was expressed as total strides·day^{-1} and minutes at high stride rates (> 30 strides·min^{-1}) as used in previous studies[27;38], on an average weekday scaled by 4/5 school day and 1/5 weekend day. At least 3 school days and 1 weekend day were required, to provide reliable data.[22] A minimum registration duration of 10 hours on school days and 8 hours on weekend days was required for days to be included, as recommended in literature.[31] Days were excluded if more than 3 hours of data were missing within the awake time interval. The time interval awake was registered by the parent and/or child in a diary.

Fatigue

Fatigue experienced was assessed with the PedsQL Multidimensional Fatigue Scale that was completed by the child. This questionnaire encompasses questions on 'general fatigue', each including 6 items with a 5-point response where '0 = never a problem' and '4 = almost always a problem'. The items are reversely scored and linearly transformed to a 0-100 scale, with higher scores indicating less experienced fatigue.[40;41] Reliability was found to be good in CP children (α = 0.79) and their parents (α = 0.91).[40]

Statistical analysis

Patient characteristics were compared between bilateral and unilateral CP with a student *t*-test or a chi^2 test. In longitudinal analysis, repeated measurements over time are performed within the same participants. These repeated measurements for each subject are dependent of each other. The statistical method should take into account this within-subject correlation.

The random coefficient analysis adjusts for the within-subject correlation in longitudinal data.[35] Therefore, we performed a random coefficient analysis with a random intercept to determine the longitudinal relationship of the fitness parameters with walking-related PAL and fatigue.[35] The fitness parameters (VO_2peak, AT, P20mean, isometric and functional muscle strength) were included as the independent variables in separate models with either walking-related PAL or fatigue as the dependent variable. Each relation (fitness parameter) with walking-related PAL or fatigue was investigated separately. Analyses were corrected for age and height, by including age and/or height in the model, when regression coefficients of the fitness parameter changed > 10%. The sample size enabled inclusion of four independent variables. Effect modification by localization (unilateral or bilateral CP), gender and GMFCS level was also investigated. Statistical analyses were performed using IBM SPSS Statistics, version 20 (SPSS Inc, Chicago, Illinois, USA). The level of significance was set at $p < 0.05$.

RESULTS

Participants

Patient characteristics are shown in Table 7.1. 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 incomplete registrations (walking-related PAL), questionnaires that were not completed (fatigue), refusal to wear the mask, lack of motivation or equipment problems (physical fitness).

TABLE 7.1 Patient Characteristics

	Overall (N=46)	Bilateral CP (N=24)	Unilateral CP (N=22)	t/Chi^2*	p
Boy/Girl	26/20	12/12	14/8	.869*	0.35
Age [year]	9.6 (1.7)	9.5 (1.3)	9.7 (2.0)	-0.486	0.63
Height [cm]	136.8 (12.4)	133.3 (8.6)	140.7 (14.7)	-2.055	0.05
Weight [kg]	34.8 (11.1)	31.7 (7.9)	38.2 (13.1)	-2.037	0.05
BMI [kg·m^{-2}]	18.2 (3.3)	17.7(3.3)	18.7 (3.1)	-1.102	0.28
Skinfold [mm]	26.3 (10.3)	26.0 (10.9)	26.7 (9.7)	-0.209	0.84
GMFCS (I/II/III)	26 / 12 / 8	8 / 8 / 8	18 /4 / 0	13.117*	≤0.001

Values are presented as mean (SD).

Relations

Analysis revealed a significant interaction effect of bilateral or unilateral CP (indicating different relationships for bilateral than for unilateral CP) for all analyzed relationships between physical fitness and walking-related PAL, except for AT, isometric knee extension strength and hip abduction strength. There were no interaction effects of localization for the relationships between the fitness parameters and fatigue and no significant interaction effects with any of the investigated relationships for gender and GMFCS. Therefore, descriptives and associations are presented for the whole group, and separately for children with bilateral and unilateral CP. Descriptives of physical fitness, walking-related PAL and fatigue are presented in Table 7.2. Table 7.3 shows the regression coefficients of the longitudinal relationships. For children with bilateral CP significant positive relations with walking-related PAL (both parameters) were found for all fitness parameters. No relationship of fitness parameters to walking-related PAL was found for unilateral involved CP children. Functional muscle strength was significantly and positively related to fatigue in unilateral involved children, while all other fitness parameters were unrelated to fatigue in all children.

DISCUSSION

This was the first study to investigate the longitudinal relationship of physical fitness to walking-related PAL and experienced fatigue in children with CP. Our results showed a significant positive relationship of all fitness parameters to walking-related PAL in children with bilateral CP. For children with unilateral CP, no relation was found between physical fitness and walking-related PAL. Functional muscle strength was significantly related to fatigue.

These results support the hypothesis that if physical fitness changes in children with CP, this is related to a change in walking-related PAL. However, this relationship was only found in children with bilateral CP and could not be confirmed in children with unilateral CP. The fitness parameter showing the strongest relation to walking-related PAL was VO_2peak; a change of 1 $ml \cdot kg^{-1} \cdot min^{-1}$ VO_2peak translated to a change of 98 strides per day (Table 7.3). A recent study showed that children with CP were able to increase their VO_2peak by 7 $ml \cdot kg^{-1} \cdot min^{-1}$ compared to a control group[30], which would correspond to a clinically relevant increase of 686 strides per day (16% of our group mean) (Table 7.2). However, determining the relation between changes in physical fitness and walking-related PAL parameters does not necessarily ascertain causality, as the relation might also be the reverse. The longitudinal relation between these measures indicates that improving VO_2peak has the potential to increase walking-related PAL, but also that a higher walking-related PAL might lead to an improved VO_2peak in children with CP.

The results of our longitudinal study showed that changes in physical fitness are related to changes in walking-related PAL in children with bilateral CP. This is in contrast with previous cross-sectional designed studies, where no association between VO_2peak and PAL was found in adults and a relatively small sample of children with CP.[25;28;33] In addition, an intervention study showed an improvement in physical fitness with only a positive trend towards increased PAL, although VO_2peak for aerobic capacity was not included.[36] Though there might also be a cross-sectional component in the interpretation of the regression coefficient, our present longitudinal results strongly suggest that a change in physical fitness is related to a change in walking-related PAL in children with bilateral CP.

TABLE 7.2 Descriptives (Mean(SD)) of Physical Activity, Fatigue and Physical Fitness

	Baseline	*N*	6 mo	*N*	12 mo	*N*
Dependent variables						
Strides per day						
Overall	5242 (1685)	*45*	5733 (2014)	*40*	4904 (1651)	*39*
Bilateral	4373 (1234)	*23*	4910 (1912)	*21*	4318 (1711)	*22*
Unilateral	6151 (1632)	*22*	6642 (1750)	*19*	5662 (1244)	*17*
SR>30 [Min]						
Overall	49.9 (22.1)	*45*	60.5 (31.8)	*40*	49.1 (24.4)	*39*
Bilateral	38.8 (15.1)	*22*	47.0 (28.7)	*21*	40.7 (25.8)	*22*
Unilateral	61.5 (22.7)	*23*	75.4 (28.8)	*19*	59.9 (17.8)	*17*
Fatigue						
Overall	73.2 (16.6)	*46*	77.2 (15.1)	*43*	75.6 (15.9)	*42*
Bilateral	74.0 (13.9)	*24*	75.5 (14.4)	*23*	73.2 (16.9)	*23*
Unilateral	72.3 (19.5)	*22*	79.2 (16.1)	*20*	78.5 (14.5)	*19*
Independent variables						
VO_2peak [$ml \cdot kg^{-1} \cdot min^{-1}$]						
Overall	31.4 (6.2)	*38*	33.9 (6.5)	*35*	33.5 (7.1)	*35*
Bilateral	29.0 (6.3)	*18*	31.4 (6.6)	*14*	32.8 (9.2)	*18*
Unilateral	33.5 (5.4)	*20*	35.5 (6.0)	*21*	34.2 (4.0)	*17*
Anaerobic threshold [$ml \cdot kg^{-1} \cdot min^{-1}$]						
Overall	16.8 (4.7)	*41*	18.7 (5.2)	*40*	17.4 (4.2)	*41*
Bilateral	15.5 (5.2)	*20*	17.6 (5.1)	*19*	17.2 (4.5)	*22*
Unilateral	18.1 (3.9)	*21*	19.8 (5.2)	*21*	17.6 (4.0)	*19*
P20mean [$W \cdot kg^{-1}$]						
Overall	3.5 (1.5)	*44*	3.4 (1.4)	*45*	3.6 (1.4)	*43*
Bilateral	3.0 (1.6)	*23*	2.7 (1.4)	*24*	3.0 (1.3)	*22*
Unilateral	4.1 (1.3)	*21*	4.2 (1.0)	*21*	4.2 (1.2)	*21*
Knee ext [$Nm \cdot kg^{-1}$]*						
Overall	1.17 (0.31)	*46*	1.20 (0.35)	*46*	1.17 (0.35)	*44*
Bilateral	1.13 (0.34)	*24*	1.13 (0.37)	*24*	1.06 (0.37)	*23*
Unilateral	1.21 (0.28)	*22*	1.27 (0.32)	*22*	1.29 (0.30)	*21*
Hip abd [$Nm \cdot kg^{-1}$]*						
Overall	0.84 (0.27)	*46*	0.89 (0.29)	*46*	0.80 (0.24)	*44*
Bilateral	0.76 (0.31)	*24*	0.82 (0.33)	*24*	0.73 (0.24)	*23*
Unilateral	0.92 (0.21)	*22*	0.95 (0.21)	*22*	0.89 (0.21)	*21*
Functional strength [rep]						
Overall	42.8 (16.9)	*46*	52.0 (19.0)	*42*	54.5 (19.7)	*42*
Bilateral	34.0 (14.3)	*24*	43.1 (16.4)	*22*	43.7 (18.7)	*21*
Unilateral	52.5 (14.1)	*22*	61.8 (14.2)	*20*	65.2 (14.2)	*21*

The values in this table present the group means at baseline, at 6 months and at 12 months for the whole group, and separated for unilateral and bilateral CP. Descriptives of walking-related physical activity (strides per day and stride rate > 30 strides·min^{-1}) and fatigue (the dependent variables) and physical fitness (the dependent variables) are presented. Abbreviations: 6mo: 6 months; 12 mo: 12 months; SR >30: Stride rate > 30 strides·min^{-1} (high stride rate); Knee ext: isometric knee extensor muscle strength; Hip Abd: isometric hip abductor muscle strength; rep: repetitions; ***:** *of the non-dominant leg only.*

Chapter 7

Our results show that the relation between physical fitness and walking-related PAL for children with bilateral CP differs to that in unilateral CP. The present results showed that physical fitness and walking-related PAL were more severely reduced in bilateral than in unilateral CP, which is in agreement with previous studies.[23;29] The reduced physical fitness might be caused by decreased muscle strength and muscle volume at the affected side of the body, which influences the maximal capacity that can be achieved.[4;45] An explanation for the higher walking-related PAL compared to children with bilateral CP may be the lower oxygen cost of walking in children with unilateral CP, resulting in a lower physical strain when compared with children with bilateral CP.[17] Therefore, walking-related PAL in children with unilateral CP might be less influenced by lower physical fitness since the physical strain is lower. Also, other factors, like cognition, behavioral and environmental factors might have a greater impact on PAL in children with unilateral CP.[44] These results indicate that, for children with unilateral CP, a change in physical fitness does not necessarily lead to a change in walking-related PAL.

The relationship of changes in anaerobic capacity and muscle strength to a change in PAL in children with bilateral CP confirms that anaerobic capacity and muscle strength also contribute to higher walking-related PAL. Earlier findings showed that decreases in anaerobic exercise responses are more strongly related to the severity of CP, in contrast to the aerobic exercise responses, indicating that aerobic exercise responses are determined to a greater extent by other factors, such as the amount of physical exercise.[4] This supports the stronger relationship we found between aerobic fitness and walking-related PAL. It should be noted, however, that the large decrease in anaerobic capacity compared to peers (-55%) and the short, intermittent activity patterns that characterize physical activity of children indicate that this physical fitness component also remains important in enhancing physical fitness and walking-related PAL.[2;4] In addition, anaerobic training can contribute to improvement in both aerobic and anaerobic fitness in children with CP.[43]

With respect to the anaerobic threshold, a change of 1 $ml \cdot kg^{-1} \cdot min^{-1}$ was related to a change of 70 strides per day for children with bilateral CP. The anaerobic threshold might be a restricting factor in walking-related PAL, since the anaerobic threshold appears at lower exercise intensity when compared to reference values.[4] The average oxygen consumption of walking in children with CP, classified as GMFCS level I (uni- and bilateral involved) (19.7 $ml \cdot kg^{-1} \cdot min^{-1}$)[15], is at the same level as the anaerobic threshold (19.4 $ml \cdot kg^{-1} \cdot min^{-1}$).[4] As a result, children with CP walk at an intensity at or above the anaerobic threshold, requiring anaerobic glycolysis, which hampers walking for longer periods.[15] If the anaerobic threshold is improved it enables performance of activities at a higher absolute exercise intensity.

In children with unilateral CP, we uncovered a significant relationship between functional muscle strength and fatigue. This association was not found for children with bilateral CP or with any of the other fitness parameters. The ability to perform one additional repetition might have resulted in the higher score on the fatigue scale (less fatigue) of 0.32. In the

TABLE 7.3 Relations of Physical Fitness with Physical Activity and Fatigue

Dependent variables	Physical activity level						Fatigue		
	Strides per day			SR > 30 [Min]					
Independent variables	B	95% CI	*p*	B	95% CI	*p*	B	95% CI	*p*
VO₂peak [ml·kg⁻¹·min⁻¹]									
Overall	**75[1]**	**20 to 131**	**0.01**	**1.26[1]**	**0.42 to 2.10**	**0.01**	-0.04	-0.56 to 0.48	0.87
Bilateral	**98[1]**	**39 to 158**	**0.01**	**1.50[1]**	**0.58 to 2.43**	**0.01**	0.00	-0.62 to 0.62	1.00
Unilateral	2[1]	-75 to 79	0.96	0.21[1]	-1.01 to 1.43	0.73	-0.08	-0.94 to 0.78	0.85
AT [ml·kg⁻¹·min⁻¹]									
Overall	**70**	**10 to 130**	**0.02**	**1.03**	**0.05 to 2.01**	**0.04**	-0.28	-0.92 to 0.35	0.38
Bilateral	**90**	**15 to 164**	**0.02**	1.14	-0.09 to 2.37	0.07	-0.10	-0.90 to 0.70	0.80
Unilateral	20	-64 to 105	0.63	0.49	-0.89 to 1.86	0.49	-0.60	-1.59 to 0.38	0.23
P20mean [W·kg⁻¹]									
Overall	**316[1]**	**41 to 591**	**0.03**	**5.13[1]**	**1.10 to 9.17**	**0.01**	0.98	-1.34 to 3.30	0.40
Bilateral	**542[1]**	**236 to 848**	**≤0.001**	**8.03[1]**	**3.59 to 12.47**	**≤0.001**	-0.47	-3.62 to 2.69	0.77
Unilateral	-318[1]	-713 to 77	0.11	-4.63[1]	-10.62 to 1.36	0.13	3.50	-0.45 to 7.42	0.08
Knee ext [Nm·kg⁻¹]*									
Overall	**1047[1]**	**55 to 2040**	**0.04**	**23.6[1]**	**8.1 to 39.0**	**0.01**	4.2	-5.2 to 13.6	0.38
Bilateral	**1425[1]**	**231 to 2619**	**0.02**	**27.3[1]**	**9.2 to 45.4**	**0.01**	4.0	-8.1 to 16.1	0.52
Unilateral	59[1]	-1417 to 1534	0.94	6.5[1]	-16.9 to 29.9	0.59	4.7	-10.6 to 20.0	0.54
Hip abd [Nm·kg⁻¹]*									
Overall	**1664[1]**	**394 to 2935**	**0.01**	**26.8[1]**	**7.3 to 46.4**	**0.01**	5.2	-6.6 to 17.1	0.38
Bilateral	**2429[1]**	**894 to 3965**	**0.01**	**39.0[1]**	**16.8 to 61.1**	**0.01**	4.7	-11.0 to 20.5	0.55
Unilateral	199[1]	-1663 to 2061	0.83	-3.2[1]	-32.8 to 26.4	0.83	6.7	-13.1 to 26.7	0.50
Functional strength [rep]									
Overall	**37.6[1]**	**19.0 to 56.2**	**≤0.001**	**0.60[1]**	**0.32 to 0.87**	**≤0.001**	**0.19**	**0.02 to 0.35**	**0.03**
Bilateral	**54.5[1]**	**31.7 to 77.3**	**≤0.001**	**0.81[1]**	**0.46 to 1.15**	**≤0.001**	0.14	-0.10 to 0.38	0.25
Unilateral	-6.9[1]	-34 to 20	0.62	-0.05[1]	-0.46 to 0.37	0.82	**0.32**	**0.04 to 0.61**	**0.03**

Analysis revealed a significant interaction effect of bilateral or unilateral CP (indicating different relationships for bilateral than for unilateral CP) for all analyzed relationships between fitness and walking-related PAL, except for AT, isometric knee extension strength and hip abduction strength. Therefore, the regressioncoefficients (B) are presented for the whole group and separated for unilateral and bilateral CP. The regressioncoefficient reflects the strength of the relationship between fitness (the independent variable) and PAL or fatigue (the dependent variable) for the intra-individual change and the inter-individual differences. Abbreviations: B: unstandardised regression coefficient; SR > 30: Stride rate > 30 strides·min^{-1}(high stride rate); Knee ext: isometric knee extensor muscle strength; Hip Abd: isometric hip abductor muscle strength; rep: repetitions; na: not applicable; [1] corrected for height; ***:** *of the non-dominant leg only. Significant results are marked in bold text.*

knowledge that a previous study showed that children and adolescents with CP (bilateral and unilateral) are able to improve their functional muscle strength by 20% as a result of training, this would correspond to an improvement of 3.4 points (< 5%) in fatigue.[43] With a group mean score of 72 in unilateral CP, compared to 85 in peers, this 3.4 point increase does not seem clinically relevant.[41] An explanation for the lack of a relationship between physical fitness and fatigue may lie in the actual construct that the multidimensional fatigue scale measures.[7;13;40] The PedSQL measures general fatigue, while peripheral muscle fatigue might be the restricting factor for walking-related PAL.[41] Another explanation may lie in our assumption that the child experiences early fatigue during walking, whereas the child might already have reduced their walking-related PAL to prevent fatigue. Interestingly, an earlier study showed that parents gave a more severe rating to the fatigue experienced by their child than the child themselves[40], indicating that children do not experience high levels of fatigue.

Study limitations

Although random coefficient analysis can be used to investigate longitudinal relations between parameters, a causal relationship cannot be determined. The actual direction of the relation between physical fitness and walking-related PAL therefore remains inconclusive. Nevertheless, it seems that focusing on either factor contributes to improvements in the other. An activity-specific mode of exercise testing (running) would be preferred in children who are able to walk. However, a cycle ergometer test was performed because a cycle ergometer was experienced to be more suitable for children who have disturbances in balance and for children being dependent on assistive devices for walking.

CONCLUSIONS

It can be concluded that changes in physical fitness are related to changes in walking-related PAL in children with bilateral CP. In contrast, no relation was found between physical fitness and walking-related PAL in children with unilateral CP. Functional muscle strength was significantly (but without clinical relevance) related to fatigue. While children with bilateral spastic CP might benefit from better physical fitness and consequently an increased walking-related PAL (or vice versa), this is not the case in children with unilateral CP. The role of physical fitness in reducing fatigue remains unclear in all children with CP. Interventions aimed at improving walking-related PAL should be differently targeted in children with bilateral CP compared to children with unilateral CP.

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