HEALTH-RELATED PHYSICAL FITNESS OF AMBULATORY ADOLESCENTS AND YOUNG ADULTS WITH SPASTIC CEREBRAL PALSY

Carla Nooijen, MSc¹, Jorrit Slaman, MSc¹, Wilma van der Slot, MD, PhD¹,², Henk J. Stam, MD, PhD, FRCP¹, Marij E. Roebroeck, PhD¹, Rita van den Berg-Emons, PhD¹ and Learn2Move Research Group

From the ¹Department of Rehabilitation Medicine, Research Lines MoveFit & Transition and Lifespan Research, Erasmus MC, University Medical Center and ²Rijndam Rehabilitation Center, Rotterdam, The Netherlands

INTRODUCTION

Cerebral palsy (CP) describes a group of disorders of movement and posture that are attributed to non-progressive disturbances in the developing brain (1). Movement and posture problems in CP include walking and balance, gross and fine motor control, and muscle spasticity (2). These problems may lead to impairments in physical fitness and physical activity levels in persons with CP (3, 4). Reduced physical fitness and physical activity can interact to cause a cycle of de-conditioning; low physical fitness might result in high physical strain during activities of daily living (ADL) (5, 6), possibly leading to a reduction in activity, and, consequently, further decreasing physical fitness. Moreover, physical fitness is known to contribute to health and quality of life of persons with chronic conditions (7).

Improving physical fitness during adolescence is important, because at this stage many changes influence adult lifestyle (8). Moreover, childhood fitness and habitual participation in activity track into adulthood (9). Knowledge of health-related physical fitness at this age can help in optimizing recommendations and treatments to improve physical fitness in adolescents and young adults with CP. Furthermore, by comparing physical fitness between subgroups with specific CP characteristics, recommendations and treatments to improve physical fitness can be further optimized and tailored to a person’s characteristics.

Caspersen et al. (10) defined several health-related components of physical fitness, including cardiopulmonary fitness, muscle strength and body composition. In addition, lipid profile is an important objective indicator for the risk of cardiovascular disease (11). We found 7 previous studies describing health-related physical fitness components in adolescents and young adults with CP (12–18). Most of these studies focus on only one component, e.g. cardiopulmonary fitness (12–15), muscle strength (16) or body composition (17). One study assessed both cardiopulmonary fitness and body composition (18). To our knowledge there has been no study of lipid profiles in adolescents and young adults with CP. Furthermore, the sample sizes in the previous studies were small (range 5–19), and the study groups were heterogeneous with regard to level of motor functioning (19). Moreover, most of the studies were old (date range 1976–2003) (12, 13, 15, 17, 18), with 4 of the 7 studies being over 20 years old. The approach to paediatric rehabilitation has changed and developed over the years, and this may have influenced current physical fitness levels in adolescents and young adults with CP (2). The goal of the present study was therefore to describe in detail the health-related physical fitness of adolescents and young adults with spastic CP aged 16–24 years. Multiple components of health-related fitness were assessed, including cardiopulmonary fitness, body composition, cholesterol, (ii) muscle strength, (iii) body mass index and waist circumference, (iv) skin-folds, and (v) lipid profile.
fitness, muscle strength, anthropometric measures, skin-folds and lipid profile. Data were compared with reference values for able-bodied persons, and differences were explored within the CP group related to Gross Motor Functioning Classification System (GMFCS) (20) and distribution of CP.

METHODS

Participants

Ambulatory adolescents and young adults with spastic unilateral or bilateral CP, aged 16–24 years, were recruited from 6 rehabilitation centres and rehabilitation departments at university hospitals throughout the western and central regions of The Netherlands, and by the Association of Physically Disabled Persons and their Parents.

This study is a retrospective cross-sectional analysis, as part of the longitudinal, multi-centre, randomized controlled trial Learn2Move, which evaluates an intervention that aims to promote daily physical activity and sports participation, reduce sedentary behaviour, and improve physical fitness among adolescents and young adults with spastic CP (21).

Exclusion criteria were: (i) disabilities other than CP that affect physical activity or cardiopulmonary fitness; (ii) contraindications to (maximal) exercise; or (iii) severe cognitive disorders or insufficient comprehension of the Dutch language. All participants provided written informed consent. The study was approved by the medical ethics committee of the Erasmus Medical Center and local approval was granted by all participating centres.

Procedures

Data collection was carried out at 3 different centres using consistent testing protocols. In the present study, baseline data of the longitudinal study were analysed. All tests were performed on the same day, and sufficient rest periods were provided between the different tests. A rehabilitation physician screened subjects for contraindications to exercise and all participants completed the Physical Activity Readiness Questionnaire (22).

Outcome measures

The health-related physical fitness outcome measures were: (i) cardiopulmonary fitness; (ii) muscle strength; (iii) anthropometric measures, including body mass index (BMI) and waist circumference; (iv) skin-folds; and (v) blood lipid profile.

Cardiopulmonary fitness. All participants performed a maximal exercise test consisting of a progressive ramp protocol (23) on electronically braked cycle ergometers (Jaeger ER800, Jaeger Toemmes, Breda, The Netherlands, or Corival V2 Lode B.V., Groningen, The Netherlands). The test started with a 3-min warm-up without resistance. After this warm-up period, the resistance was increased every 12 s in steps of 2–6 W, depending on GMFCS level and gender. The target pedal rate during the test was 60–70 rpm. Participants were verbally encouraged to continue as long as possible. The test ended when the participant stopped voluntarily due to exhaustion or when they were unable to maintain the target pedal rate. During the test, gas exchange and heart rate (HR) were measured continuously. Two comparable breath-by-breath analysis systems were used in the different centres (Oxycon Pro, ViaSys Healthcare, Houten, The Netherlands; and Quark CPET system, Cosmed, Rome, Italy). The highest mean oxygen uptake over a period of 30 s was defined as VO\textsubscript{peak}. VO\textsubscript{peak} was expressed in l/min and in ml/kg/min and as a percentage of the reference values of Jones et al. (24), taking into account gender, age, height and body mass. Furthermore, peak ventilation (VE\textsubscript{peak}), ventilatory equivalent ratio for oxygen (VE\textsubscript{O2}/VO\textsubscript{2peak}), and highest power output (PO\textsubscript{peak}) for a period of 30 s were determined. In addition, ventilatory anaerobic threshold (VAT, in l/min and %VO\textsubscript{2peak}) was estimated by the ventilatory equivalent method (25): when VE/VO\textsubscript{2} and the end-tidal O\textsubscript{2} partial pressure (PET\textsubscript{O2}) increased while the ventilatory equivalent for CO\textsubscript{2} (VE/\textsubscript{CO2}) and end-tidal CO\textsubscript{2} partial pressure (PET\textsubscript{CO2}) remained stable.

In addition, HR\textsubscript{peak} and peak respiratory exchange ratio (RER\textsubscript{peak}) were determined and used as objective criteria for maximal exercise, set at HR\textsubscript{peak} ≥ 175 bpm (90% of predicted maximum HR) (26) or RER\textsubscript{peak} ≥ 1.1 (27). When these criteria were not met, the results of the maximal exercise test were excluded.

Muscle strength. Muscle strength of the hip flexors, hip abductors and knee extensors was measured with a hand-held dynamometer (Micro-Fet, Hoggan Health Industries Inc., West Jordan, UT, USA) using the “break” method (28). The dynamometer applicator was held against the distal part of the limb segment, and participants were asked to exert their maximum force against it. When maximum was reached the examiner applied sufficient resistance to overcome the force exerted by the participant. Both the left and right sides were measured 3 times, with 1 min rest in between. The mean value of the 3 repetitions was determined. The leg with the highest muscle strength was categorized as the stronger leg, and the other leg as the weaker leg. The mean muscle strength of both legs was compared with able-bodied Dutch reference values for muscle strength, measured with the break testing method in a group of 16-year-olds (29).

Anthropometric measures. Height (cm) and body mass (kg) were measured, and BMI, kg/m\textsuperscript{2} was determined. Waist circumference (cm) was measured mid-way between the lowest rib and the iliac crest while standing. For BMI and waist circumference, Dutch reference values for persons aged 30–39 years from the project “NLdeMaat 2009–2010” were used (30). Similar to the general population, a BMI < 18.5 was considered underweight, ≥ 18.5 and < 25 normal weight, ≥ 25 and < 30 overweight, and ≥ 30 obese. In males, waist circumference was categorized as: ≤ 79 cm underweight, ≥ 79 and < 94 cm normal weight, ≥ 94 and < 102 cm overweight, and ≥ 102 cm obese. In females, waist circumference < 68 cm was considered underweight, ≥ 68 and < 80 cm normal weight, ≥ 80 and < 88 cm overweight, and ≥ 88 cm obese.

Skin-folds. The thickness of 4 skin-folds (biceps, triceps, subscapular and suprailiac) was measured twice on the left side of the body with a Harpenden calliper (Burgess Hill, UK), and mean values were determined (31).

Lipid profile. Non-fasting blood samples were taken, and total serum cholesterol (TC), high density lipoprotein cholesterol (HDL) and TC/HDL ratio were determined. TC and HDL are known to be minimally altered when measured in fasting or non-fasting blood samples (32, 33). Dutch reference values for persons aged 30–39 years were used (30). TC was considered unfavourable when ≥ 6.5 mmol/l and HDL unfavourable when < 0.9 mmol/l.

Statistical analyses

Participants’ characteristics and the components of health-related fitness are presented as means (standard deviation; SD). Furthermore, mean values (SD) for subgroups regarding GMFCS level and distribution of CP were determined. Regression analyses were performed to test whether there were differences in age and gender between the subgroups regarding GMFCS level and distribution of CP. To test for differences in health-related fitness between subgroups, separate linear regression models were made, all corrected for age and gender. Statistical analyses were performed using SPSS 20 (SPSS Inc, Chicago, IL, USA). The significance level was set at \( p \leq 0.05 \).

Percentages of reference values were determined for VO\textsubscript{2peak}, mean muscle strength of both legs for hip flexion, hip abduction and knee extension, BMI, waist circumference, TC and HDL. VO\textsubscript{2peak} was predicted per person (24) and the measured VO\textsubscript{2peak} was expressed as a percentage of the predicted VO\textsubscript{2peak}. A paired \( t \)-test was used to test for differences between the measured VO\textsubscript{2peak} and the predicted VO\textsubscript{2peak}.

Muscle strength, BMI, waist circumference, TC and HDL, the mean value of the participants with CP was expressed as a percentage of the reference mean. When values were found to be less than 75% of reference they were marked considerably lower. For VO\textsubscript{2peak} and mean muscle strength of both legs we described the number of participants.
with an outcome < 50% of reference and < 75% of reference. For BMI and waist circumference we described the number of participants in the following categories: underweight, normal weight, overweight and obese. For lipid profile, we described the number of participants with favourable and unfavourable outcomes.

RESULTS

A total of 55 adolescents and young adults with CP were measured for attributes of health-related physical fitness. Of these 55, only 5 were GMFCS level III. Since this subgroup was small, we excluded these persons from further analyses. We included 50 participants with GMFCS levels I or II.

Table I shows participants’ characteristics and their cardiopulmonary fitness and muscle strength. The distribution of CP was unknown in one participant. Participants with bilateral CP were significantly older than those with unilateral CP ($p=0.03$).

Results of the maximal exercise test are given for 41 participants. Nine participants were excluded because they did not meet the objective criteria due to lack of motivation. Muscle strength data for 46 participants were available. Data for 4 participants were missing either because a participant could not perform the test or for logistical reasons.

Corrected for age and gender, persons with GMFCS level II had significantly lower PO peak compared with persons with GMFCS level I ($p=0.02$). When comparing persons with unilateral and bilateral CP, corrected for age and gender, we found significant differences between groups for VO peak, VE peak, PO peak, HR peak and VO peak/HR peak. Furthermore, participants with unilateral CP had significantly higher hip abduction muscle strength compared with persons with bilateral CP ($p<0.01$). Table II shows the comparison with references for cardiopulmonary fitness and muscle strength. Mean VO peak was 62% of reference, which was a significant difference ($p<0.01$). Hip

### Table I. Participant’s characteristics, cardiopulmonary fitness and muscle strength, and differences related to the Gross Motor Functioning Classification System (GMFCS) level and distribution of cerebral palsy (CP)

<table>
<thead>
<tr>
<th>Participants’ characteristics</th>
<th>GMFCS</th>
<th>Distribution of CP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>All</td>
<td>I</td>
</tr>
<tr>
<td>Participants, $n$</td>
<td>50</td>
<td>30</td>
</tr>
<tr>
<td>Age, years, mean (SD)</td>
<td>20 (3)</td>
<td>20 (3)</td>
</tr>
<tr>
<td>Males/females, $n$</td>
<td>25/25</td>
<td>14/16</td>
</tr>
<tr>
<td>Cardiopulmonary, mean (SD)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Participants, $n$</td>
<td>41</td>
<td>24</td>
</tr>
<tr>
<td>VO peak, l/min</td>
<td>2.46 (0.77)</td>
<td>2.57 (0.69)</td>
</tr>
<tr>
<td>VO peak, ml/[kg/min]</td>
<td>35.08 (9.37)</td>
<td>35.85 (9.37)</td>
</tr>
<tr>
<td>VE peak, l/min</td>
<td>97.71 (32.68)</td>
<td>101.17 (32.61)</td>
</tr>
<tr>
<td>VE peak/VO peak, l/min</td>
<td>40.04 (7.13)</td>
<td>39.47 (7.65)</td>
</tr>
<tr>
<td>VAT, l/min</td>
<td>1.62 (0.60)</td>
<td>1.65 (0.13)</td>
</tr>
<tr>
<td>VAT, %VO peak</td>
<td>65.23 (13.24)</td>
<td>64.12 (2.54)</td>
</tr>
<tr>
<td>PO peak, Watt</td>
<td>180.37 (66.14)</td>
<td>195.21 (55.57)</td>
</tr>
<tr>
<td>HR peak</td>
<td>188.62 (12.34)</td>
<td>189.43 (11.90)</td>
</tr>
<tr>
<td>HR peak, % of predicted max</td>
<td>94.49 (6.55)</td>
<td>94.71 (6.19)</td>
</tr>
<tr>
<td>VO peak/HR peak</td>
<td>13.14 (3.88)</td>
<td>13.79 (3.62)</td>
</tr>
<tr>
<td>RER peak, VOpeak/VO2</td>
<td>1.16 (0.09)</td>
<td>1.18 (0.09)</td>
</tr>
</tbody>
</table>

### Table II. Cardiopulmonary fitness and muscle strength compared with reference

<table>
<thead>
<tr>
<th>% reference</th>
<th>$p$-value</th>
<th>n&lt;sub&gt;d&lt;/sub&gt;</th>
<th>&lt;50% reference</th>
<th>$p$-value</th>
<th>n&lt;sub&gt;d&lt;/sub&gt;</th>
</tr>
</thead>
<tbody>
<tr>
<td>VO peak</td>
<td>62</td>
<td>&lt;0.01*</td>
<td>30/41</td>
<td>16/41</td>
<td></td>
</tr>
<tr>
<td>Hip flexion</td>
<td>65</td>
<td>33/46</td>
<td>16/46</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hip abduction</td>
<td>85</td>
<td>19/46</td>
<td>6/46</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Knee extension</td>
<td>65</td>
<td>33/46</td>
<td>13/46</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

*p<0.05.

*Regression analyses studying differences in cardiopulmonary fitness and muscle strength between GMFCS level I and II and between unilateral and bilateral CP were corrected for age and gender.

SD: standard deviation; VO$_2$: oxygen uptake; VE peak: ventilation; VAT: ventilatory anaerobic threshold; PO: power output; HR: heart rate; RER: respiratory exchange ratio.

*J Rehabil Med* 46
Health-related physical fitness of persons with CP

Flexion and knee extension strength were considerably lower than reference values (both 65% of reference value).

The 9 persons who did not reach maximal exertion, due to lack of motivation, (3 males/6 females, 6 gMFCs level I/3 gMFCs level II) had a mean HR peak of 165 beats/min (sD 6), RER peak of 1.04 (sD 0.04), and VO_{2peak} of 1.69 l/min (sD 0.45). Independent t-tests showed that these 3 measures were significantly lower than those of persons who met objective criteria for maximal exercise ($p < 0.01$). The excluded persons did not differ significantly in terms of age (mean 19 years (sD 3)) and body mass (mean 59 kg (sD 9)).

Table III shows the results regarding anthropometric measures, skin-folds and lipid profile and results of the subgroup analysis. No significant differences on these outcomes were found between subgroups with different CP characteristics.

Table IV presents comparisons of anthropometric measures and lipid profile with reference values. Mean BMI and mean waist circumference were comparable to references. According to BMI, 6% of participants (3 of 50) were underweight, compared with 0.9% of the reference sample. Sixteen percent of participants (8 out of 50) were overweight, compared with 21% of the reference sample. Furthermore, 12% (6 out of 50) had a BMI indicating obesity, compared with 10% of the reference sample. Waist circumference indicated obesity in 24% of males (6 out of 25), compared with 12% of the reference sample, and in 24% of females (6 out of 25), compared with 26% of the reference sample. None of the 23 persons had unfavourable TC, compared with 8.5% of the reference sample. Two out of 23 participants (8.7%) had unfavourable HDL, compared with 7.9% of the reference sample. Twenty-one blood samples were missing.

**DISCUSSION**

This is the first study describing multiple aspects of health-related physical fitness in a substantial sample of ambulatory adolescents and young adults with spastic CP (gMFCs levels I and II). Distribution of CP (unilateral vs bilateral) seems to affect physical fitness more than gMFCs level (I vs II) does. Persons with bilateral CP had lower cardiopulmonary fitness and lower hip abduction muscle strength compared with persons with unilateral CP. Persons with gMFCs levels I and II had almost comparable outcomes, except for the larger waist circumference found in males. Almost all outcomes for lipid profile were favourable with respect to norm values.

Although the approach to paediatric rehabilitation has evolved over the past decades, the results for cardiopulmonary fitness in the present study are comparable to those in stud-
ies performed 30 years ago (12, 13, 15). This suggests that there is a need to change the approach, or that more attention should be paid to physical fitness in young persons with CP. Adolescence and young adulthood is a critical stage of life with great influence on adult lifestyle, and this may therefore be an important period for intervention. Previous research in children with CP has shown that exercise programmes focusing on lower extremity muscle strength, cardiovascular fitness, or a combination of these, lead to increases in cardiopulmonary fitness and muscle strength (34, 35). In maintaining a favourable level of physical fitness, there is initial evidence that behavioural interventions are of added value (36). Further longitudinal studies on the effects of such interventions on health-related physical fitness are necessary.

Hip flexion and knee extension strength were considerably lower (< 75%) in participants with CP compared with reference values. Mean hip abduction was 85% of the reference value. However, persons in the bilateral subgroup had significantly lower hip abduction strength than those with unilateral CP. Therefore, attention should be paid to hip abduction muscle strength in persons with bilateral CP. The comparison of muscle strength with reference values must be regarded with caution, since values were available only for 16-year-olds, possibly leading to an overestimation of the percentage of reference values (29), since it is known that muscle strength is still developing at that age. However, no reference values for these 3 muscle groups measured with the break testing method were available for the age range 16–24 years. It is difficult to compare our results with the results of a previous study of adolescents with CP, because the earlier study used a different method: electromyography while seated in an adjustable chair, strapped in a fixed position (16).

Based on BMI, the number of participants categorized as overweight or obese was comparable to reference values. However, regarding waist circumference, 6 out of 25 males had a waist circumference indicating obesity, which is a propensity twice as high as the reference value. There were also 6 out of 25 females with a waist circumference indicating obesity. However, the propensity to obesity in the reference female population was comparable. Comparisons with reference values should be interpreted with caution, since the reference population was somewhat older (age range 30–39 years). Further research should compare persons with CP with sex- and age-matched able-bodied controls. Mean outcomes on anthropometric measures were comparable to those in previous studies in adolescents and young adults with CP (17, 18). A BMI indicating underweight was found in 2 persons with GMFCS level I, and in 1 person with GMFCS level II. Three out of 50 is a higher propensity for underweight compared with reference. However, although the propensity was higher it only included 3 persons, and therefore further research is necessary. BMI should be interpreted with caution, since BMI in persons with CP has been reported to be less valid than in able-bodied persons (37, 38).

This is one of the first studies to assess lipid profiles in a group of high-functioning adolescents and young adults with CP. The results indicate that, both at the group and individual levels, there is no indication for increased cardiovascular risk based on lipid profile for persons with GMFCS level I and II at this age. This is in line with the results of a previous study in adolescents (9–16 years) with CP, which concluded that arterial health was not different from that of a control group (39). In a study of adults with bilateral CP (age range 25–45 years) lipid profile was slightly less desirable than in the present study (40). There is a limitation in power when studying subgroup differences in lipid profile when correcting for both age and gender in the analyses, since data for only 23 persons were available. Furthermore, larger amounts of data for females were missing, partly explained by a larger proportion of females who did not give consent for taking a blood sample. Therefore, our sample is limited to data for only 6 females. More research, especially longitudinal studies, on this important indicator for cardiovascular disease, is required. Furthermore, it would be interesting to study the correlation between lipid profile and other health-related physical fitness measures.

A significant difference was found in age between our subgroups of persons with unilateral CP and bilateral CP. However, the difference in the mean age of persons with unilateral CP and bilateral CP was only 2 years (mean ages of 19 and 21 years). This difference of 2 years is not likely to have a large influence on physical fitness. Moreover, we corrected for age in our regression analyses. Although this is a limitation of our study, we believe that this difference in characteristics did not influence our conclusions.

The present results are limited to the description of health-related physical fitness of ambulatory adolescents and young adults with GMFCS levels I and II. Further research in persons with GMFCS levels III and IV is necessary. Furthermore, criteria for maximal exercise were not met by 9 participants; however, since their GMFCS level, age and weight were comparable to the group that did meet the criteria, their exclusion is thought to have had minimal influence on the generalizability of the results. Skin-fold thickness was measured on the left side of the body. However, regarding the asymmetry of topography, it is possible that an impaired side would not have the same anthropometric features as a non-impaired side.

Ambulatory adolescents and young adults with CP have impaired cardiopulmonary fitness, hip flexion and knee extension strength. Fitness outcomes in persons with bilateral CP were lower than those with unilateral CP. This stage of life has a great influence on adult lifestyle, and therefore may be an important interventional period for interventions regarding physical fitness.

ACKNOWLEDGEMENTS

Financial support. Financial support was received from ZonMW (no. 60-61300-98-003), den Haag, The Netherlands, and Phelps Stichting voor Spastici (no. 2008039), Bussum, The Netherlands.

The authors declare no conflicts of interest.

REFERENCES

1. Rosenbaum P, Paneth N, Leviton A, Goldstein M, Bax M, Dami-


