Applied exercise physiology in rehabilitation of children with cerebral palsy

Balemans, A.C.J.

2014

Physical Therapy
Journal of the American Physical Therapy Association and de Fysiotherapeut
Royal Dutch Society for Physical Therapy

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PHYS THER. 2014; 94:121-128.
Originally published online September 12, 2013
doi: 10.2522/ptj.20130197

Reliability of a Progressive Maximal Cycle Ergometer Test to Assess Peak Oxygen Uptake in Children With Mild to Moderate Cerebral Palsy

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Background. Rehabilitation research in children with cerebral palsy (CP) is increasingly addressing cardiorespiratory fitness testing. However, evidence on the reliability of peak oxygen uptake ($\dot{V}O_2$peak) measurements, considered the best indicator of aerobic fitness, is not available in this population.

Objective. The objective of this study was to establish the reliability of a progressive maximal cycle ergometer test when assessing $\dot{V}O_2$peak in children with mild to moderate CP.

Design. Repeated measures were used to assess test-retest reliability.

Methods. Eligible participants were ambulant, 6 to 14 years of age, and classified as level I, II, or III according to the Gross Motor Function Classification System (GMFCS). Two progressive maximal cycle ergometer tests were conducted (separated by 3 weeks), with the workload increasing every minute in steps of 3 to 11 W, dependent on height and GMFCS level. Reliability was determined by means of the intraclass correlation coefficient (ICC [2,1]) and smallest detectable change (SDC).

Results. Twenty-one children participated (GMFCS I: n=4; GMFCS II: n=12; and GMFCS III: n=5). Sixteen of them (9 boys, 7 girls; GMFCS I: n=3; GMFCS II: n=11; and GMFCS III: n=2) performed 2 successful tests, separated by 9.5 days on average. Reliability for $\dot{V}O_2$peak was excellent (ICC=.94, 95% confidence interval=.83–.98). The SDC was 5.72 mL/kg/min, reflecting 14.6% of the mean.

Limitations. The small sample size did not allow separate analysis of reliability per GMFCS level.

Conclusions. In children with CP of GMFCS levels I and II, a progressive maximal cycle ergometer test to assess $\dot{V}O_2$peak is reliable and has the potential to detect change in cardiorespiratory fitness over time. Further study is needed to establish the reliability of $\dot{V}O_2$peak in children of GMFCS level III.

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[Brehm M-A, 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;94:121–128.]

Published Ahead of Print: September 12, 2013
Accepted: August 30, 2013
Submitted: May 22, 2013

Rehabilitation programs for children with disabilities, including cerebral palsy (CP), are increasingly aimed at improving physical fitness. Given the decreased levels of physical fitness in children with CP[1–5] and the concomitant reduction in physical activity,[6–8] this is an important intervention strategy, especially because lack of optimal fitness and physical activity may contribute to the development of secondary complications related to a sedentary lifestyle, such as cardiovascular risk and osteoporosis.[9]

According to the American College of Sports Medicine, physical fitness can be divided into 5 components: cardiorespiratory fitness, muscular fitness, body composition, flexibility, and neuromotor fitness.[10,11] For decades, fitness testing in CP has mainly focused on the muscular component,[12,13] with less attention being paid to cardiorespiratory fitness (ie, aerobic capacity). Nevertheless, adequate fitness programs aimed at improving both strength and aerobic capacity may contribute to maintaining or enhancing fitness levels in people with CP[14,15] and prevent further inactivity and loss of mobility.[9]

Aerobic capacity is defined as the ability to deliver oxygen to the muscles and to utilize it to generate energy during exercise at the maximal capacity of the cardiorespiratory system.[16] In children, the peak oxygen uptake ($\dot{V}O_2$peak) during progressive maximal exercise to the point of volitional termination as the result of exhaustion is considered the single best indicator of aerobic capacity.[17] For children who are able to walk independently, the most suitable way to assess their $\dot{V}O_2$peak is by means of a progressive maximal walking or running exercise test, tested either in the laboratory (treadmill test) or in the field (shuttle run).[2,18] Walking and running exercise tests are functional modes of regular daily mobility that are assumed to help children with CP to maintain a physically active lifestyle. A disadvantage of these tests, however, is that they are less suitable for children who have disturbances in coordination and balance (Gross Motor Function Classification System [GMFCS] II), and for children dependent on assistive devices for walking, such as crutches and walkers (GMFCS III and IV). Assessment of $\dot{V}O_2$peak during a progressive maximal cycle ergometer exercise test may be more appropriate for these children,[4] with the additional advantage that a cycle ergometer also allows the workload to be determined.

To date, 3 studies have used progressive maximal cycle ergometer testing to assess the $\dot{V}O_2$peak in children with CP.[19–21] In essence, the test procedures used in these studies were all based on the McMaster protocol, which is recommended for fitness testing in young people.[22] The McMaster protocol divides people into 4 groups on the basis of height, with the workload increasing every 2 minutes, ranging from 12.5 W to 25 W and 50 W.[23] Although this protocol is valid in people who are healthy, the large differences in disease severity in CP may require a more individualized approach to the incremental exercise test. Such an approach has been shown to be useful in children with cardiac and respiratory diseases[24,25] and was recently introduced for children with CP.[1] In the latter study, the exercise test workload was increased by small steps every minute, dependent on the child's height and GMFCS level. A clear advantage of adapting workload increments to the level of motor ability is that smaller increments are better endured, leading to an optimal test duration, higher $\dot{V}O_2$peak values, and improved test validity.

The Bottom Line

What do we already know about this topic?

Cardiorespiratory fitness levels in children with cerebral palsy (CP) are significantly reduced; however, these fitness levels can be improved by adequate fitness training. In order to determine the effects of fitness training on peak oxygen consumption ($\dot{V}O_2$peak), which is considered the best indicator of cardiorespiratory fitness, it is important to use reliable assessment tools.

What new information does this study offer?

In children with CP who are ambulatory, $\dot{V}O_2$peak can be assessed reliably with the use of a progressive cycle ergometer graded exercise test. Furthermore, when evaluating the effects of fitness training on $\dot{V}O_2$peak, individual treatment responses of greater than 15% can be detected.

If you're a patient/caregiver, what might these findings mean for you?

Fitness training aimed at improving $\dot{V}O_2$peak in children with CP can be evaluated reliably with the use of cycle ergometer testing.

In addition to validity, reliability is another important clinimetric property, especially when $\dot{V}O_2$peak is used to determine individual changes or when the effects of interventions at group level must be evaluated. Currently, the reliability of aerobic cycle ergometer tests to assess $\dot{V}O_2$peak in children with CP has been insufficiently established.[26] One study was found in adults with CP that indicated good reliability and moderate precision for $\dot{V}O_2$peak[27] on the basis of the currently recommended intraclass correlation coefficient (ICC) and standard error of measurement (SEM).[28] However, exercise responses change with growth and maturation,[29] requiring the reliability of measurement protocols to be specifically established in children. Also, protocols used for adults contain rather large workload increments that cannot be applied in children. A study that did establish the reliability of cycle ergometer testing in children reported only on peak power output with the use of the Spearman correlation[30] and not on $\dot{V}O_2$peak. Because $\dot{V}O_2$peak is considered the best indicator of aerobic capacity, and especially in view of its increasing use in evaluating rehabilitation fitness programs in children with CP, information on the reliability of this measure in this patient group is important.

The purpose of our study was to establish the test-retest reliability of a progressive maximal cycle ergometer test when assessing $\dot{V}O_2$peak in ambulant children with spastic CP and classified as GMFCS level I, II, or III.

Method

Participants

The study was conducted at the Department of Rehabilitation Medicine of the Free University Hospital in Amsterdam between 2008 and 2012. Inclusion criteria were children diagnosed with spastic CP; 6 to 14 years of age; classified as GMFCS level I, II, or III; and cognitively capable of following simple instructions. Furthermore, participants had no contraindications for fitness testing, no surgical procedures in the previous 6 months or treatment with botulinum toxin in the previous 3 months, and no other diseases that could influence fitness or mobility. Relevant institutional review board approvals were obtained before study commencement, and all participants and their parents provided written informed consent.

Measurement Equipment

Cardiorespiratory fitness testing was performed on an upright cycle ergometer, specifically adapted for children with CP (adjustable cranks, shoe fixation, pediatric saddle) (Corival, Lode BV, Groningen, the Netherlands). During testing, heart rate (HR) was recorded with a polar HR monitor (Polar Vantage XL, Polar Electro, Nederland BV, Almere, the Netherlands) and pulmonary gas exchange was measured breath-by-breath with an accurate[31] and reliable[32] gas analysis system (VmaxST, Sensormedics, Houten, the Netherlands). The system was calibrated with certified calibration gasses before each measurement.

Procedure

Participants were tested twice by use of the same test protocol, with a maximum of 3 weeks between the 2 visits.[33] Test and retest measures were performed at the same time of the day, by the same trained researcher, in a laboratory with an environmental temperature of 19°C to 22°C and a relative humidity of 41% to 50%. The researcher who performed the retest was blinded to the results of the initial test. After anthropometric measurements were taken, a warm-up was performed, followed by a submaximal cycle test and a progressive maximal cycle test. Children who normally wore orthopedic shoes or orthoses were allowed to wear them during the test.

Test Protocol

After a 5-minute rest measurement, a warm-up period of 2 to 3 minutes of steady-state pedaling (60–70 rpm) was used to estimate each child's level of cycling ability and to familiarize the child with the cycle ergometer. The target HR during the warming up was 100 to 115 bpm. The warm-up was followed by a 5-minute submaximal steady-state cycling test at a constant pedaling speed (60–70 rpm), providing additional familiarization. The submaximal workload (1.75 × warm-up load) had a target HR of 60% to 70% of the maximal estimated HR.[34] When HR was not between 60% to 70% of the maximal estimated HR, some resistance (5–10 W) was either added or removed. Separated by a 1-minute rest period, the progressive maximal cycle test was performed. The workload of the submaximal test was chosen as the initial load, and this load was increased every minute in steps of approximately 3 to 10 W (Pinc), according to height and GMFCS level (Appendix).[1] If, depending on the individual's ability, alterations to Pinc were needed, workload increments corresponding to either a lower or higher GMFCS level were chosen (Appendix). The required pedal revolution was 60 to 70 rpm.

Throughout the test, children were provided with auditory and visual feedback on target and cycling performance, and they were verbally encouraged to continue cycling until exhaustion. At this point, the children's perception of exhaustion was scored with the use of the Children's OMNI Scale of Perceived Exertion (OMNI scale).[35] A test was considered maximal and was included for analysis if 2 of 3 of the following criteria were achieved: (1) HR >180 bpm,[36] (2) respiratory exchange ratio (RER) >1, or (3) subjective

Table 1.
Participants' Characteristics[a]

Variable	Values
Boys/girls, n	9/7
Type of CP (unilateral/bilateral), n	2/14
GMFCS levels I/II/III, n	3/11/2
Age (y), $\overline{X}$ (SD) [range]	10.5 (2.1) [7.2–14.2]
Weight (kg), $\overline{X}$ (SD) [range]	37.9 (12.5) [18–67]
Height (cm), $\overline{X}$ (SD) [range]	146.5 (14.7) [119–171]
BMI (kg/m^2), $\overline{X}$ (SD) [range]	17.2 (2.8) [12.7–23.7]

[a] BMI=body mass index, CP=cerebral palsy, GMFCS=Gross Motor Function Classification System.

Table 2.
Results for Effect Parameters on the Progressive Maximal Cycle Test[a]

Variable	Test	Retest	Mean Test-Retest	Mean Difference Retest−Test
$\dot{V}O_2$peak (mL/kg/min)	39.8 (7.7)	38.8 (8.8)	39.3 (8.2)	−1.08 (2.8)
POpeak (W/kg)	2.19 (0.80)	2.17 (0.81)	2.18 (0.80)	−0.024 (0.21)
HRpeak (bpm)	190 (11)	188 (15.8)	189 (12.8)	−2 (9.5)
RERpeak	1.05 (0.14)	1.02 (0.10)	1.035 (0.11)	−0.025 (0.13)
OMNI score	8.50 (2.01)	8.50 (1.35)	8.50 (1.27)	0 (2.51)
Test duration (min)	9.06 (1.65)	9.00 (1.4)	9.03 (1.51)	−0.0625 (0.77)

[a] Values are presented as mean (SD). $\dot{V}O_2$peak=peak oxygen uptake, POpeak=peak power output, HRpeak=peak heart rate, RERpeak=peak respiratory exchange ratio, OMNI=Children's OMNI Scale of Perceived Exertion.

signs of exhaustion (out of breath, sweating, fatigued, or OMNI scale score >8) or unable to continue the test. A recovery phase of 2 minutes finished the test protocol and consisted of cycling at 10 W until HR had decreased to the HR reached in the submaximal phase.

Data Analysis

A 30-second moving-window average was calculated for $\dot{V}O_2$peak (mL/kg/min), peak power output (POpeak, W/kg), and peak heart rate (HRpeak, bpm). Furthermore, peak respiratory exchange ratio (RERpeak, defined as the ratio between carbon dioxide production [$\dot{V}CO_2$] and $\dot{V}O_2$), total test duration [min], and OMNI scale scores were registered. Reliability was determined only for $\dot{V}O_2$peak, POpeak, and HRpeak.

Descriptive statistics were used to characterize the study population. Dependent on the distribution of the data, parametric or nonparametric analyses were applied.

Test-retest reliability[28] was expressed with the ICC and 95% confidence intervals (CI). The ICC was calculated as $\sqrt{(\sigma^2_s/[\sigma^2_s + \sigma^2_o + \sigma^2_e])}$, where σ^2_s represents the variance among participants, σ^2_o is the variance caused by measurement occasions, and σ^2_e represents the error variance. Variance components were estimated by means of analysis of variance, with the use of a 2-way random-effects model (ICC [2,1]), with a restricted maximum likelihood method. An ICC value >.70 is considered to indicate acceptable reliability, and a lower bound of the 95% CI of the ICC >.75 indicates excellent reliability.[37]

Measurement error[28] was expressed by means of the SEM and 95% limits of agreement (LoA). The SEM was calculated as $\sqrt{(\sigma^2_o + \sigma^2_e)}$. Because the use of the SEM implies that data are not heteroscedastic, Bland-Altman plots were visually inspected to rule out the presence of heteroscedasticity (ie, the SEM not being independent of the mean[38]). From the SEM, the smallest detectable change (SDC) was calculated, which represents the minimum difference (in individual patients) that is considered to be a real change between measurements with 95% certainty.[28] The SDC was calculated as SDC=1.96 × SEM × $\sqrt{2}$. Analyses were performed with the use of SPSS (version 20.0, IBM Corp, Armonk, New York).

Results

Twenty-one children with CP participated (11 boys and 10 girls: GMFCS 1 [n=4], GMFCS II [n=12], and GMFCS III [n=5]). Because 2 children could not take part on the second occasion (because of time constraints) and 3 children (all GMFCS III) were not able to accomplish the maximal cycle test (ie, they were not able to maintain a 60- to 70-rpm pedaling rate with increasing workloads), analyses were performed on only 16 children: GMFCS I (n=3), GMFCS II (n=11), and GMFCS III (n=2). All children included in the full analysis had prior cycling experience. Their characteristics are presented in Table 1.

All 16 children included in the full analyses successfully performed both the initial test and the retest, fulfilling previously defined criteria. During both tests, 7 children (44%) reached the endpoint on the basis of all 3 criteria (subjective signs of exhaustion, HR >180 bpm, and RER >1.0). Nine children (56%) successfully completed the tests on the basis of 2 of the 3 criteria (either subjective signs of exhaustion and a target HR >180 bpm [n=5] or subjective signs of exhaustion and an RER >1.0 [n=4]). The median interval between test and retest was 9.5 days (range=1–18). The mean (SD) $\dot{V}O_2$peak and POpeak values were 39.3 (8.2) mL/kg/min and 2.18 (0.80) W/kg, respectively (Tab. 2).

Table 3.
Results for Reliability[a]

Variable	ICC (95% CI)	SEM	95% LoA	SDC
$\dot{V}O_2$peak (mL/kg/min)	.94 (.83–.98)	2.06 (5.3)	−6.6 to 4.4	5.72 (14.6)
POpeak (W/kg)	.97 (.91–.99)	0.145 (6.6)	−0.44 to 0.39	0.402 (18.4)
HRpeak (bpm)	.76 (.44–.91)	6.6 (3.5)	−20.6 to 16.6	18.5 (9.8)

[a] Standard error of measurement (SEM) and smallest detectable change (SDC) values are presented as absolute (% of the mean). ICC=intraclass correlation coefficient, 95% CI=95% confidence interval, LoA=limits of agreement, $\dot{V}O_2$peak=peak oxygen uptake, POpeak=peak power output, HRpeak=peak heart rate.

Reliability

Differences between the means of outcomes on the 2 tests were rather small, varying between 1% (HRpeak) and 8% (POpeak), with no learning effects observed. The results of test-retest reliability (ICC [2,1]) are presented in Table 3. Reliability for $\dot{V}O_2$peak was excellent (ICC [2,1]=.94, 95% CI=.83–.98). The ICC values for the other outcomes ranged from .76 (95% CI=.44–.91) for HRpeak to .97 (95% CI=.91–.99) for POpeak.

Results for measurement error are also presented in Table 3. Because heteroscedasticity was not observed for any of the effect parameters, the SEM, 95% LoA, and SDC were used to express measurement error. The SEM for $\dot{V}O_2$peak was 2.06 mL/kg/min, reflecting 5.3% of the mean; the LoA values were −6.6 and 4.4 mL/kg/min (Figure); and the SDC was 5.72 mL/kg/min (14.6%). Measurement error for the other outcomes also was within acceptable boundaries (Tab. 3).

Discussion

The purpose of this study was to establish the test-retest reliability of a progressive maximal cycle ergometer test when assessing cardiorespiratory fitness in children with mild to moderate CP. We found that $\dot{V}O_2$peak, as the best indicator of cardiorespiratory fitness, is reliable in children of GMFCS levels I through III and can be used as an evaluative measure to detect changes after interventions in this population. This finding is clinically relevant, given that fitness levels in children with CP are reduced, especially in children of GMFCS levels II and III[1,5] and probably in levels IV and V, although testing in these children often is limited by functional abilities.

Because this study was the first to assess the reliability of $\dot{V}O_2$peak in children with CP, our results could not be directly compared. A comparison with the reliability of $\dot{V}O_2$peak obtained in adults with CP[27] showed similar results (ICC=.98), although the 95% CIs of the ICC were not reported on in this study. A comparison of POpeak results with the only other study in children with CP that assessed the reliability of a progressive cycle ergometer test showed that our ICC was slightly higher (.97) compared with the Spearman correlation coefficient found by van den Berg-Emons and colleagues (.92).[30] However, methods of calculating and reporting POpeak differed. Our POpeak outcome was normalized for body weight (W/kg) and calculated

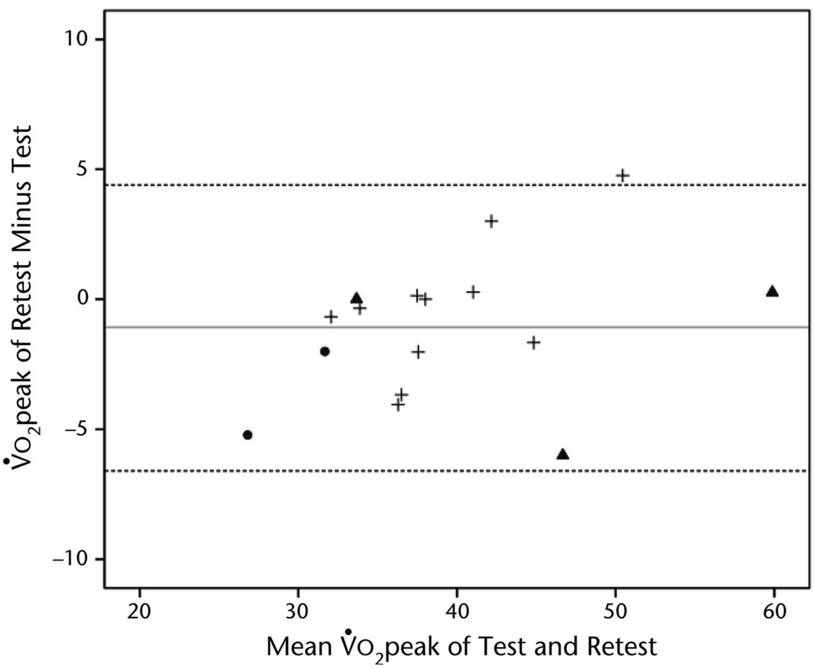

Figure.
Bland-Altman plot of the achieved peak oxygen uptake ($\dot{V}O_2$peak, mL/kg/min) during test and retest (n=16). Gross Motor Function Classification System (GMFCS) levels: ▲=GMFCS I, +=GMFCS II, ●=GMFCS III.

over the 30 seconds with the highest sustained load, whereas van den Berg-Emons et al defined POpeak as the highest power output over the 2 minutes of a workload, expressed in watts. Although these differences should be considered, the high reliability coefficients of both studies indicate that a progressive cycle ergometer test can be used to reliably measure power output. However, when using power output as a substitute for $\dot{V}O_2$peak in assessing aerobic fitness, differences in mechanical efficiency among individuals with different levels of motor ability should be considered.

Regarding measurement error, the SEM of $\dot{V}O_2$peak was fairly small (5.3%) compared with the SEM reported by De Groot et al[27] in trained adults with CP (7.4%). When using a cycle ergometer test to assess cardiorespiratory fitness in clinical practice, the reported measurement error should be considered. We, therefore, calculated the SDC, which is an important reliability measure when interpreting individual changes after an intervention. Our SDC for $\dot{V}O_2$peak shows that changes larger than 15% can be detected when cycle ergometry is used to evaluate the effects of aerobic training in individual children with CP classified as GMFCS I through III. Although this finding implies satisfactory sensitivity, it remains unclear whether the reported measurement error is adequate to detect training effects. Measurement error can be considered adequate if the SDC is smaller than the minimal important change (MIC), which is the minimal amount of change in score that patients or clinicians consider to be important.[39] Unfortunately, no information is available on the MIC of $\dot{V}O_2$peak in children with CP. Therefore, we related our SDC to the established training effects for $\dot{V}O_2$peak in CP, which was reported in only 1 study. That study showed an increase of 7.8 mL/kg/min (19.8% of the mean) in $\dot{V}O_2$peak after 8 weeks of training,[40] which suggests that the test as used in the present study appears to be adequate and sufficiently sensitive to detect individual changes in cardiorespiratory fitness over time in this population.

Although not our primary outcome, measurement error of HRpeak corresponded to the 95% LoA described in the literature.[18] Furthermore, both mean HRpeak and mean RERpeak values met our criteria of reaching at least 180 bpm and 1.0, respectively, which indicates that children with CP are able to achieve their maximal aerobic capacity on a cycle ergometer. A similar finding also was reported by Balemans et al.[1] However, in their study, 8 of 10 children classified as GMFCS level III were able to meet the criteria, whereas in our study, 3 of 5 children with GMFCS level III could not perform the cycle test. These participants may have differed in important aspects of motor ability. One example could be the score on the Functional Mobility Scale (FMS),[41] because children in the present study who successfully completed the test had a higher FMS score compared with children who did not complete the test, especially with respect to medium-long distances (eg, 50 m); score 2 (uses a walker or frame to cover this distance) for successful children versus score 1 (uses a wheelchair) for children who did not successfully complete the test. Aspects of muscle function such as strength and selectivity might have played a role in these findings, and further investigation is needed to determine in which children with GMFCS level III a progressive graded cycle test or a graded arm exercise test[42] may be more feasible and valid.

The ability to perform a cycle test also depends on the cycling experience of tested individuals, which is a disadvantage of cycle ergometry in countries where cycling is less common. However, the advantages of cycle ergometry include the lack of restriction on children achieving maximal performance caused by ambulatory balance problems and the possibility of measuring standardized external power output.[1] A novel aspect of the current study was the individualized approach to workload increments during cycling, which showed that equal $\dot{V}O_2$peak levels were achieved within a shorter test duration compared with treadmill running[3] or shuttle test running.[2,18] These are important advantages that should support the use of cycle ergometer testing for assessing cardiorespiratory fitness.

Limitations

A limitation of this study was that 24% of the included children did not complete the study, which resulted in a small number of children included in the analyses according to the criteria defined by Cosmin.[33] Furthermore, the number of children in GMFCS levels I and III was much smaller than the number in GMFCS level II. This finding was partly because 3 of 5 children in GMFCS level III were unable to perform the cycle test. As noted, these participants had lower levels of motor ability than did the participants in GMFCS III who completed the test. Therefore, further evaluation of the influence of motor ability on the feasibility of test performance, as well as a more extensive reliability analysis in children in GMFCS level III, is needed.

Conclusion

We found that $\dot{V}O_2$peak, as measured with a progressive maximal cycle ergometer test, could be reliably assessed in children with CP of GMFCS levels I and II. Furthermore, when evaluating the effects of cardiorespiratory training on $\dot{V}O_2$peak, individual treatment responses of greater than 15% can be detected, and the detection of much smaller changes is possible when studying groups of children with CP. Future studies should establish the feasibility and reliability of $\dot{V}O_2$peak in a larger group of children with GMFCS level III.

Dr Brehm, Dr Becher, and Dr Dallmeijer provided concept/idea/research design. Dr Brehm, Ms Balemans, and Dr Dallmeijer provided writing and data analysis. Dr Brehm and Ms Balemans provided data collection. All authors provided project management. Dr Becher provided fund procurement and study participants. Dr Dallmeijer provided institutional liaisons. Ms Balemans, Dr Becher, and Dr Dallmeijer provided consultation (including review of manuscript before submission). The authors thank all of the children who participated in this study. They also acknowledge Peggy Geerdinck and Jantine van der Vlist for their help with data collection.

DOI: 10.2522/ptj.20130197

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Appendix.

Guideline Recommendations for Workload Increments per Minute (Pinc, in Watts) During the Progressive Maximal Cycle Ergometer Test (Based on Balemans et al[1])[a]

Height (cm)	GMFCS I	GMFCS II	GMFCS III
120	3	3	3
140	7	6	5
160	10	9	7

[a] GMFCS=Gross Motor Function Classification System.

Physical Therapy
Journal of the American Physical Therapy Association and de Fysiotherapeut
Royal Dutch Society for Physical Therapy

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PHYS THER. 2014; 94:121-128.
Originally published online September 12, 2013
doi: 10.2522/ptj.20130197