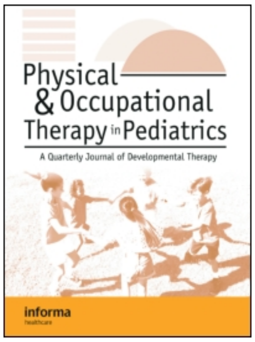


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# Reliability and Construct Validity of the 6-Minute Racerunner Test in Children and Youth with Cerebral Palsy, GMFCS Levels III and IV

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**ABSTRACT.** *Aim:* To determine the test–retest reliability and construct validity of a novel 6-Minute Racerunner Test (6MRT) in children and youth with cerebral palsy (CP) classified as Gross Motor Function Classification System (GMFCS) levels III and IV. The racerunner is a step-propelled tricycle. *Methods:* The participants were 38 children and youth with CP (mean age 11 y 2 m, SD 3 y 7 m; GMFCS III, $n$ = 19; IV, $n$ = 19). Racerunner capability was determined as the distance covered during the 6MRT on three occasions. The intraclass correlation coefficient (ICC), standard error of measurement (SEM), and smallest detectable differences (SDD) were calculated to assess test-retest reliability. *Results*: The ICC for tests 2 and 3 were 0.89 (SDD 37%; 147 m) for children in level III and 0.91 for children in level IV (SDD 52%; 118 m). When the average of two separate test occasions was used, the SDDs were reduced to 26% (104 m; level III) and 37% (118 m; level IV). For tests 1 to 3, the mean distance covered increased from 345 m (SD 148 m) to 413 m (SD 137 m) for children in level III, and from 193 m (SD 100 m) to 239 m (SD 148 m) for children in level IV. *Conclusions:* Results suggest high test–retest reliability. However, large SDDs indicate that a single 6MRT measurement is only useful for individual evaluation when large improvements are expected, or when taking the average of two tests. The 6MRT discriminated the distance covered between children and youth in levels III and IV, supporting construct validity.

**KEYWORDS.** Cerebral palsy, GMFCS level III, GMFCS level IV, racerunner, reliability, construct validity, fitness, exercise

Children and youth with cerebral palsy (CP) classified as Gross Motor Function Classification System (GMFCS) levels III and IV experience difficulties with walking, or are not able to walk, even when using assistive mobility devices such as

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canes, crutches, or hand-held walkers (Palisano et al., 1997). They have independent mobility when using assistive mobility devices for short distances but may use a wheelchair for community mobility, whereas children classified as GMFCS levels I and II walk independent without using assistive mobility devices (Palisano et al., 1997). Research suggests that children with CP in GMFCS levels I, II, and III tend to have poorer physical fitness than children with no physical disabilities (Hoofdwijk et al., 1995; Verschuren & Takken, 2010; Balemans et al., 2013b), which may lead to a lower health status and higher risk for developing secondary conditions (Fowler et al., 2007). Little is known about the fitness levels of children and youth classified as GMFCS level IV. It is assumed that they have poorer physical fitness because individuals who use an assistive mobility device or wheelchair in daily live are less active than individuals who walk without devices (Maltais et al., 2010). Due to motor impairments children and youth classified as GMFCS levels III and especially IV are unable to perform usual exercise modes like cycling and running and therefore testing and training their physical fitness is more complicated and alternative training modes are needed.

An alternative training device for children and youth with limited walking ability is the racerunner (Petra-bike, Connie Hansen, Stenlose, Denmark). This device consists of a 3-wheeled frame, with handlebars, saddle and a trunk support, similar to a tricycle (Donnell et al., 2010). Rather than using a pedaling system, children and youth propel themselves forward by stepping their feet on the ground (Figure 1). An advantage of the racerunner is that children and youth reach higher levels of speed because the racerunner is light and has a more aerodynamic shape in comparison with existing gait trainer devices. This makes the racerunner more suitable for (outdoor) sports activities, and as a consequence the racerunner could be an appropriate device for improving the physical fitness of these children and youth. Propulsion in a manual wheelchair is a more functional skill in comparison with racerunning, but children and youth with limited walking ability only use their arms in a wheelchair. With the racerunner children and youth use their legs for propulsion instead of their arms, and therefore physical training with the racerunner provides a higher training stimuli compared to arm training with a wheelchair. It is important to develop a field-based test to measure the individual progress of these children and youth after a training period with the racerunner. Reliable and valid field-based tests are developed for children with CP who are able to walk, like the shuttle run test (Verschuren et al., 2006) and the muscle power sprint test (Verschuren et al., 2007). There are however fewer tests available for children without walking ability (Verschuren et al., 2013). In a clinimetric review of fitness tests for children with CP, it was emphasized that further research is required on reliability, validity and clinical utility of field-based tests to assess physical fitness in children with CP classified as GMFCS levels III to V (Balemans et al., 2013a).

We therefore developed a novel test to measure racerunner capability based on the 6-minute walking test (6MWT) (American Thoraxic Society, 2002), which is a valid and reliable test for measuring walking capability in children with CP who are able to walk (Maher et al., 2008; Thompson et al., 2008; Nsenga Leunkeu et al., 2012). During the 6-Minute Racerunner Test (6MRT) children use the racerunner to propel themselves forward for 6 min on an oval track. The outcome of the

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**FIGURE 1.** A picture of a boy using the racerunner.

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test is the distance covered in 6 min. The 6MRT measures racerunner capability, where capability is described as "what a person can do in his/her environment" (Holsbeeke et al., 2009). The aim of the present study was to assess the test–retest reliability of the 6MRT for evaluating racerunner capability in children and youth with CP classified as GMFCS levels III or IV.

## *METHOD*

### *Participants*

The participants were 38 children and youth, 5 to 20 years of age, with CP; 19 were classified as GMFCS III and 19 were classified as GMFCS level IV. We included children and youth with the ability to understand simple instructions and perform the 6MRT. Exclusion criteria were contraindications for intensive physical training, unstable seizures or severe behavioral problems, recent (< 3 months) Botulinum Toxin A treatment or surgery, and participants with injuries. We excluded children and youth with pain when they were not able to perform the 6MRT. The Medical Ethical Board of the VU University Medical Center Amsterdam reviewed the study protocol and determined that the study did not fall under the scope of the Medical Research Involving Human Subjects Act (WMO). Characteristics of the participants are presented in Table 1. There were no significant differences in the characteristics of children and youth classified as GMFCS level III and IV for age, body height, weight and BMI.

**TABLE 1. Patients Characteristics ($n = 38$)**

| Patients characteristics | GMFCS III ($n = 19$) | GMFCS IV ($n = 19$) | $p$ |
|---|---|---|---|
| Male/Female | 10/9 | 12/7 | 0.743 |
| Age (y, mo), mean (SD; range) | 11.9 (4.10; 5.2–19.4) | 11.0 (3.5; 6.3–17.11) | 0.487 |
| Body height (cm), mean (SD; range) | 147.2 (20.4; 112.0–177.0) | 140.6 (19.9; 100.0–172.0) | 0.322 |
| Body mass (kg), mean (SD; range) | 40.7 (17.8; 17.0–82.0) | 36.9 (13.3; 17.0–62.0) | 0.470 |
| Body mass index (kg/m$^2$), mean (SD; range) | 17.8 (3.5; 13.6–26.4) | 18.3 (4.3; 12.9–29.8) | 0.724 |
| Type of cerebral palsy | | | — |
| *Spastic* | 12 | 15 | |
| *Ataxic* | 3 | 0 | |
| *Dystonia* | 4 | 4 | |
| MACS level I/II/III/IV/V missing | 6/5/2/0/0<br>6 | 5/0/4/5/2<br>3 | — |
| FMS | | | |
| *5 m 1/2/C* | 1/16/2 | 14/5/0 | — |
| *50 m 1/2/C* | 4/15/0 | 18/1/0 | |
| *500 m 1/2/C* | 16/3/0 | 19/0/0 | |

Note: GMFCS, gross motor function classification system; MACS, manual ability classification system; FMS, functional mobility scale; FMS 1, child uses wheelchair; FMS 2, child uses a walker or a frame; FMS C, child crawls for mobility.

### *Procedure*

Before testing, each participant practiced at least two times with the racerunner, supervised by their physical therapist. In these practice sessions participants propelled with the racerunner for at least 10 minutes and the correct settings for the racerunner, such as height of the saddle, were established. The measurements took place in six rehabilitation centers on school days (parents were not present). Body height (m), weight (kg), GMFCS level, Manual Ability Classification System (MACS) level (Eliassen et al., 2006) and Functional Mobility Scale (FMS) (Graham et al., 2004) were determined. Body height was assessed in an upright standing position. Racerunner capability was determined with the 6MRT on three occasions on separate days; the second and third 6MRT were performed within 2 weeks after the first test and the minimum amount of days between the tests was two.

During the tests heart rate was registered with a flexible heart rate monitor (Polar FT7, Kempele, Finland). Participants who wore orthopedic shoes or orthotics in daily life, wore them during the tests. The tests were performed on an inside, oval track using the racerunner (Petra-bike, Connie Hansen, Stenlose, Denmark). The length of the track (between 25 and 40 m) was dependent on the size of the gymnasium, but all participants used the same track on all test occasions. There were no sharp turns included in the track to avoid steering problems. The participants were instructed to propel as far as possible in 6 min. Rest was not allowed, participants were encouraged to continue if the stopped moving. The test leader walked behind the participants for safety reasons. Children and youth were given maximal verbal encouragement and the three tests, for each participant, were administered by the same person.

The number of tracks completed and the distance covered in the final uncompleted track (measured with a measurement wheel) were recorded. All participants

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were able to complete all three tests. There were only mild complaints that the children and youth expressed during the tests like saddle pain.

### *Data Analysis*

The data were analyzed using SPSS 20.0 (SPSS, Chicago, IL, USA). The outcome was the distance covered (m) in 6 min. A second outcome of the 6MRT, the total cost index was calculated combining the walking distance with the heart rate during the 6MRT. The total cost index is calculated as the quotient of the mean heart rate (beats/min) during the last two minutes of the test divided by walking speed (m/min), and is expressed in heart beats per meter (beats/m) (Bratteby Tollerz et al., 2011). To assess the differences between baseline characteristics for participants in GMFCS level III and IV, the chi-square test was used for dichotomous data, and the independent sample *t*-test for the normally distributed continuous data.

### *Test–Retest Reliability*

Test–retest reliability was determined for the distance covered and for the total cost index during the 6MRT. The data for the distance covered between tests 1 and 2 and between tests 2 and 3 were plotted using Bland–Altman plots; a graphical representation of the individual participant differences between two tests, plotted against the individual average of the two tests (Bland & Altman, 1986). The plots were visually inspected to check for heteroscedasticity (Brehm et al., 2012).

The intraclass correlation coefficient (ICC), the standard error of measurement (SEM) and smallest detectable difference (SDD) were determined based on the variance components separately for children and youth in level III and IV. The sources of measurement variability were determined by an analysis of variance, with a random design using the restricted maximum likelihood with 1 factor: measurement occasion (3 levels). First, three components of variance were estimated: the patients ($var_p$), the occasions ($var_o$), and the error variance ($var_r$). ICC was calculated as $var_p/(var_p + var_o + var_r)$ (Streiner & Norman, 2008; Vet et al., 2011). Acceptable reliability criteria for ICC values were values $> 0.75$ (Portney & Watkins, 2009). Besides the ICC, we calculated the two measures of agreement: the SEM and SDD (Vet et al., 2006). The SEM is an absolute measure and quantifies the precision of individual scores within subjects instead of among subjects like the ICC. The SEM was computed as: $SEM = \sqrt{var_o + var_r}$ (Streiner & Norman, 2008). The SDD reflects the smallest within person change in a score that ($p < 0.05$) cannot be attributed to measurement error. The SDD was calculated as: $1.96 \text{ x } \sqrt{2} \times SEM$ (Streiner & Norman, 2008). The SEM and SDD were reported in the actual units of measurement (m and beats/m) and the SDD's were also expressed as a percentage of the average group value (Streiner & Norman, 2008).

For the SEM and SDD of the distance covered during the test we assessed the implications for evaluating changes when taking the average distance covered over two separate test occasions. This was done by dividing the variance components by the number of tests (two) (Streiner & Norman, 2008; Vet et al., 2011).

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### *Construct Validity*

Construct validity was measured using the "known groups" method. We hypothesized that children and youth classified as level III would propel further than children and youth classified as level IV. The results of test 3 were used for these analyses. A multiple linear regression analysis was performed to adjust for the influence of body height. A significance level of $p < 0.05$ was used.

## *RESULTS*

6MRT results for all three test occasions are shown in Table 2. The distance covered during the tests increased (despite familiarization sessions) from test 1 to test 3 in both GMFCS levels, indicating a learning effect with the racerunner. Similarly, the total cost index decreased. There were 25 participants (66%) who reached a heart rate of 180 beats per minute (bpm) or higher indicating that these children and youth reached a (near) maximum effort during the test.

Bland–Altman plots were created for the distance covered between tests 1 and 2 (Figure 2A) and between tests 2 and 3 (Figure 2B). Visual inspection of these plots showed heteroscedasticity for tests 1 and 2, this was not observed between tests 2 and 3.

Table 3 presents the test–retest reliability data of the distance covered and the total cost index. Because heart rate data during one of the three tests was missing for nine participants, the total cost index was calculated for 29 participants. For children and youth in level IV, the ICC and SEM values improved when the data of tests 2 and 3 were taken compared to values for the tests 1 and 2. For children

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**TABLE 2. Outcomes of the Walking Distance, Heart Rate (Mean and Maximum) and Total Cost Index (TCI) of the 6-Minute Racerunner Test (6MRT)**

| | Total ($n = 38$) | GMFCS level III ($n = 19$) | GMFCS level IV ($n = 19$) |
|---|---|---|---|
| Mean walking distance in m (SD; range) | | | |
| *Test 1* | 268 (166; 51–727) | 345 (148; 117–727) | 193 (100; 51–361) |
| *Test 2* | 299 (170; 40–822) | 385 (161; 187–822) | 214 (134; 40–550) |
| *Test 3* | 326 (166; 30–702) | 414 (136; 225–702) | 239 (148; 30–631) |
| Mean heart rate (SD; range) | | | |
| *Test 1* | 161 (23; 120–196) | 164 (24; 127–196) | 159 (22; 120–196) |
| *Test 2* | 153 (25; 111–193) | 160 (23; 118–191) | 147 (25; 111–193) |
| *Test 3* | 156 (23; 113–198) | 160 (20; 128–195) | 151 (26; 113–198) |
| Maximum heart rate (SD; range) | | | |
| *Test 1* | 174 (23; 125–214) | 179 (22; 137–214) | 170 (23; 125–203) |
| *Test 2* | 168 (26; 116–212) | 176 (24; 132–212) | 160 (26; 116–201) |
| *Test 3* | 174 (23; 124–217) | 181 (20; 151–217) | 167 (25; 124–205) |
| TCI last two minutes* (SD; range) | | | |
| *Test 1* | 5.7 (4.4; 2.5–18.2) | 3.6 (1.9; 1.6–9.6) | 7.2 (5.0; 3.1–18.2) |
| *Test 2* | 4.8 (3.5; 1.8–17.3) | 3.2 (1.3; 1.4–6.5) | 6.0 (4.2; 1.8–17.3) |
| *Test 3* | 4.4 (4.0; 1.5–24.0) | 2.9 (1.0; 1.9–5.0) | 5.6 (5.0; 1.5–24.0) |

Note: GMFCS, gross motor function classification system; TCI, total cost index; *data are analyzed over 29 children (GMFCS level III, $n = 13$; IV, $n = 16$).

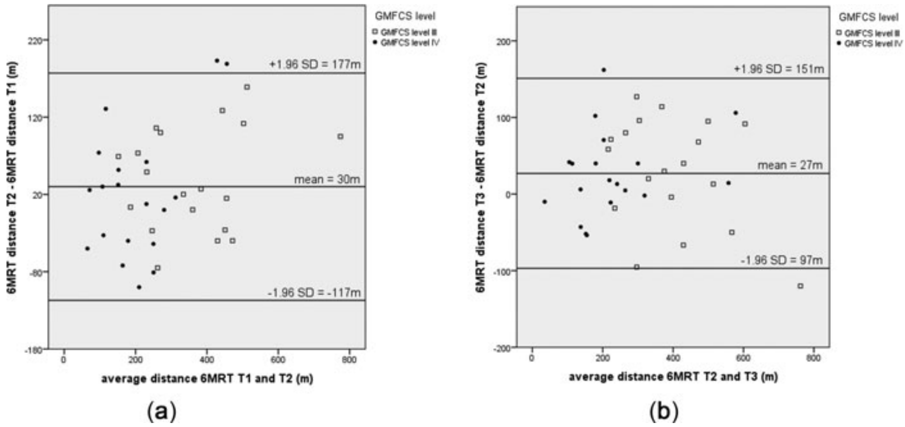


**FIGURE 2. a) Bland–Altman plot of distance covered during test one (T1) and test two (T2) of the 6MRT. b) Bland–Altman plot of distance covered during test one (T2) and test two (T3) of the 6MRT. GMFCS, Gross Motor Function Classification System.**

and youth in level III, the ICC and SEM values were similar for test 1-2 and test 2-3. Findings were similar for the total cost index data. The SDD for tests 2 and 3 was 37% (147 m) for children and youth in level III and 52% (118 m) for children and youth in level IV (Table 3). When the average of two separate test occasions (tests 2 and 3) was used, the SDD was 26% (104 m) and 37% (83 m), for children and youth in level III and IV, respectively. Corrected for body height there was a significant difference in the mean distance covered between children and youth in level III and IV (151 m, SE 41 m, $p < 0.01$).

## *DISCUSSION*

The purpose of this study was to evaluate the test–retest reliability and construct validity of a novel 6-Minute Racerunner Test (6MRT) for evaluating racerunner capability in children and youth with CP classified as GMFCS levels III and IV. The ICC's for the distance covered between tests 2 and 3 in GMFCS level III (0.89) and GMFCS level IV (0.91) indicate acceptable test–retest reliability for group level (Portney & Watkins, 2009), whereas the ICC's for the total cost index show less than acceptable ICC values (< 0.75). Therefore we advise the use of the distance covered and not the total cost index (expressed in heart beats per meter), as an outcome measure for the 6MRT. Alternatively, measurements of agreement including the standard error of measurement (SEM) and smallest detectable difference (SDD), for the distance covered were large and caution is necessary when evaluating changes over time for individual patients. Children and youth classified as level III were able to propel significant further than children and youth classified as level IV, supporting construct validity.

The results provide evidence that the 6MRT is a reliable test for children and youth with CP classified as GMFCS levels III and IV. These results are in agreement with studies evaluating the 6MWT in children with CP who are able to walk (Nsenga Leunkeu et al., 2012). However, the distance covered with the racerunner

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**TABLE 3. Test–Retest Reliability Statistics 6-Minute Racerunner Test (6MRT) for the Distance Covered and the Total Cost Index (TCI)**

| | | ICC | | SEM (m) | | SDD (m) | | SDD in% of the mean | |
|---|---|---|---|---|---|---|---|---|---|
| | | GMFCS III | GMFCS IV | GMFCS III | GMFCS IV | GMFCS III | GMFCS IV | GMFCS III | GMFCS IV |
| Distance covered on 6MRT (m) | Over test 1–3 | 0.86 | 0.78 | 59.9 | 62.4 | 166.0 | 172.9 | 43.6 | 80.6 |
| | Over test 1–2 | 0.89 | 0.73 | 52.8 | 60.4 | 146.3 | 167.3 | 40.1 | 82.6 |
| | Over test 2–3 | 0.89 | 0.91 | 52.9 | 42.5 | 146.6 | 117.8 | 36.6 | 52.1 |
| TCI (b/m) | Over test 1–3 | 0.74 | 0.57 | 0.76 | 3.28 | 2.11 | 9.09 | 62.7 | 144.3 |
| | Over test 1–2 | 0.81 | 0.50 | 0.72 | 3.40 | 2.00 | 9.45 | 56.1 | 142.7 |
| | Over test 2–3 | 0.85 | 0.88 | 0.44 | 1.63 | 1.23 | 4.52 | 38.7 | 81.0 |

Note: 6MRT, 6-Minute Racerunner Test; ICC, intraclass correlation coefficient; SEM, standard error of measurement; SDD, smallest detectable differences; GMFCS, Gross Motor Function Classification System; m, meters; TCI, total cost index; b/m, beats per meter.

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systematically increased every test, indicating a learning effect when performing the 6MRT. These improvements in the distance covered, even after two familiarization sessions with the racerunner, are probably caused by the improved efficiency of propelling the racerunner. This improved efficiency can be considered as part of a training effect, implying that it is not possible to distinguish whether improved efficiency or improved physical fitness lead to better 6MRT capability after a training period with the racerunner. The high ICC values can be explained by the variability within patients (Vet et al., 2006). One child in our study propelled the racerunner for only 30 m while another child propelled 822 m, and therefore the heterogeneity of our sample was large.

The variability in the distance covered within the individual patients was also large, explaining the large SEM and SDD values. These values are important when evaluating change over time for individual patients. The large SDDs for children and youth in level III (147 m, 37% for test 2-3) and IV (118 m, 52% for test 2-3) indicate that the value of a single 6MRT for the individual and clinical evaluation of these children and youth is only useful when large improvements after a training period can be expected. In addition, the use of the 6MRT for individual children and youth improves by taking the average of two tests (Streiner & Norman, 2008; Vet et al., 2011). For children and youth classified as GMFCS level III the SDD decreases from 147 (or 37%) to 104 m (or 26%) when the average of tests 2 and 3 is used to determine 6MRT outcome. By taking the average of tests 2 and 3 for GMFCS level IV, the SDD decreases from 118 (or 52%) to 83 m (or 37%). Pilot studies with the racerunner show substantial improvements in the distance covered during the 6MRT after a training period and therefore improvements of 83 and 104 m seem achievable. Although these improvements seem clinically relevant from a mobility perspective, further studies using an external criterion are needed to determine the clinically meaningful change of the 6MRT.

Motivation probably explains a part of the variability in distance covered. The children and youth were given maximal verbal encouragement during the 6MRT instead of the standardized verbal encouragements as described for the 6MWT (American Thoraxic Society, 2002) because of intellectual disabilities and limited motivation. Despite these maximal verbal encouragements some children and youth demonstrated large variability in their mean and maximum heart rate, indicating that motivation probably explains a part of the variability in distance covered. For example, one of the children covered 184 and 187 m in the first and second test and 258 m during the third test. His maximum heart rate was respectively 150 bpm during the first test, 157 bmp during the second test and 187 bpm during the third test. Children and youth with learning and behavioral problems may experience more motivational issues. We did not exclude these participants from our study, to improve the generalizability of the 6MRT. Further research might focus on the differences in test–retest reliability for the 6MRT between subgroups, such as children and youth with and without learning and behavioral problems or children and youth with other diagnosis.

An important advantage of using the racerunner is that children and youth with limitations in walking are able to achieve high heart rates. The American College of Sports Medicine recommends exercising at heart rates above 55% of the maximum heart rate for moderate to vigorous aerobic training. When using 194 bpm as an estimate of maximum heart rate as described by Verschuren et al. (2011), all participants were able to reach this adequate training zone for increasing aerobic fitness during the test (above 107 bpm). There were even 25 participants who reached a heart rate of 180 bpm or higher and therefore, an intervention period with the racerunner seems a feasible and useful intervention strategy to improve the physical fitness for these children and youth.

There are many personal and environmental barriers for children with CP to participate in physical activity (Verschuren et al., 2012). Reducing sedentary time and encouraging light-intensity activities become more and more important (Verschuren et al., 2014). Sedentary time is higher in children with CP (Obeid et al., 2014), especially in those who depend on assistive mobility devices and wheelchairs. Apart from training periods and sport activities with the racerunner, children and youth could use the racerunner to be active in their home environment, and as such reduce their sedentary time. For example, they can play outside with the racerunner or walk with their dog. Lauruschkus et al. (2015) described how to stimulate participation in physical activities, after individual interviews and focus groups in children with CP. Two important implications are: "children want to be physically active together with friends or others" and "children want to have fun and enjoy the sensation of speed when being physically active." The racerunner could provide all these components and further research could focus on training programs with the racerunner, specifically on (1) improving the aerobic capacity during a training period (2) using the racerunner for activities in daily life, and (3) improving participation.

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## *CONCLUSION*

The 6-Minute Racerunner Test (6MRT) is a reliable test for evaluating racerunner capability in children and youth with CP classified as GMFCS levels III and IV and discriminated the distance covered between children and youth in levels III and IV, supporting construct validity. The large smallest detectable differences (SDD) and observed learning effect of propelling the racerunner indicate that, a single 6MRT measurement is only useful when large improvements are expected. Accuracy of the 6MRT for individual children and youth can be improved by taking the average of two tests.

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***Declaration of interest:*** The authors report no conflicts of interest. The authors alone are responsible for the content and writing of this article.

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