Physiological Response to the 6-Minute Frame Running Test in Children and Adults With Cerebral Palsy

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Purpose: To determine the physiological response and association to peak oxygen uptake of the 6-minute Frame Running test (6-MFRT) in persons with cerebral palsy (CP).
Methods: Twenty-four participants with CP, Gross Motor Function Classification System II/III/IV, performed the 6-MFRT. Distance, peak heart rate (HR_{peak}), peak respiratory exchange ratio (RER_{peak}), and peak oxygen uptake ($\dot{V}O_{2peak}$) were measured.
Results: HR_{peak} ranged from 146 to 201 beats per minute, RER_{peak} from 0.94 to 1.49, 6-MFRT distance from 179 to 1220 m and $\dot{V}O_{2peak}$ from 0.62 to 2.18 L/min. HR_{peak} was achieved in 63%, RER_{peak} in 71%. A strong correlation was observed between 6-MFRT and $\dot{V}O_{2peak}$.
Conclusions: The 6-MFRT represented a (near) maximum effort for 75% of the participants and the 6-MFRT can be used to estimate oxygen consumption on an individual basis. (Pediatr Phys Ther 2022;34:529–534)
Key words: cardiorespiratory fitness, cerebral palsy, exercise test, oxygen consumption, physiology, sports for persons with disabilities

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Drs von Walden and van Schie contributed equally.

This was a collaborative research, which was conducted at 2 sites, thereby we have 9 authors who made significant contribution.

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- 75% of participants with CP achieved (near) maximum effort during the 6-MFRT
- Peak $\dot{V}O_2$ correlates with distance covered on the 6-MFRT.
- The 6-MFRT can serve as an estimation of oxygen consumption.

Cerebral palsy (CP) is a neurological disorder defined as a group of permanent disorders of movement and posture, attributed to nonprogressive disturbances in the developing fetal or infant brain.[1] Depending on the severity of the neurological disorder, activity limitations vary.[1] In terms of functional mobility, CP can be classified using the Gross Motor Function Classification System (GMFCS).[2] Individuals classified as GMFCS level I can generally walk without restrictions, although they are limited in more advanced motor skills. Individuals classified as GMFCS level V have difficulties with maintaining posture and are wheelchair users in all settings.[2] Limitations in daily activities and mobility possibly explain the lower physical fitness of individuals with CP compared with peers without disabilities.[3,4] One aspect of physical fitness is cardiorespiratory fitness (ie, aerobic capacity), preferably measured as the peak oxygen uptake ($\dot{V}O_{2peak}$) during a progressive maximal exercise test.[5-7] Previous studies have measured the $\dot{V}O_{2peak}$ of individuals with CP (mainly GMFCS I-II) during maximum exercise tests on a treadmill or cycle ergometer. According to these studies, the $\dot{V}O_{2peak}$ of children and young adults with CP is reduced by 14% to 29% and 9% to 46%, respectively, compared with peers developing typically.[3,7,8] However, a measure of activity that reflects aerobic fitness is yet to be identified in more severely affected persons with CP, as exercise tests on a treadmill or cycle ergometer have limitations in this population.[9,10] Therefore, it is necessary to develop alternative tests and exercise devices to enable measurement of aerobic fitness in individuals with CP, especially at higher GMFCS levels.

The running frame (formerly known as RaceRunner) is an alternative running device for individuals who have limited walking ability. This tricycle without pedals can be used for locomotion in daily life, as a form of health-related exercise and for competitive sports. Frame Running (formerly known as RaceRunning) capability can be measured using the 6-minute Frame Running test (6-MFRT).[11] The outcome of the 6-MFRT is the distance covered with a running frame in 6 minutes. Even though the 6-MFRT was not developed as a maximum exercise test, many participants achieve (near) maximum heart rates (HRs) during the test. However, the wider physiological response, such as oxygen uptake and ventilation, has not been investigated. Furthermore, Bolster et al[11] observed an acceptable test-retest reliability of the 6-MFRT at group level, but the relationship between the 6-MFRT and oxygen uptake (ie, aerobic fitness) has not been investigated. Given this background, the first aim of our study was to investigate gas exchange, ventilation, rated perceived exertion, and HR during the 6-MFRT. The second aim was to investigate the relationship between $\dot{V}O_{2peak}$ and distance covered during the 6-MFRT. We hypothesized that the included athletes in this study would reach a near maximum physiological response during the 6-MFRT, and that there would be a relationship between $\dot{V}O_{2peak}$ and distance covered during the 6-MFRT.

METHODS

Participants

Trainers of possible participants received an e-mail with information about the study and were asked to inform the athletes and their parents about the study. When the athletes indicated that they were interested, they received an e-mail with general information about the study and an appointment was made to take the measurements. We included participants with CP, 8 years and older, GMFCS levels II to IV, with the ability to exercise for at least 10 minutes, to independently complete a lap on a standard 400-m athletics track with a running frame, and able to understand simple instructions. All participants had been accustomed to the sports for at least 2 months before the 6-MFRT. We excluded participants who had seizures, or who had undergone selective dorsal rhizotomy or single-event multilevel surgery in the past year, or botulinum toxin treatment in the past 3 months, when there were contraindications for intensive physical training, and when participants were uncomfortable with the measurement equipment or had pain during the 6-MFRT.

Twenty-six participants were recruited. Two participants were excluded; 1 due to communication failure and clear submaximal performance, as HR was highest during warm-up. The other participant was excluded because of incomplete data due to technical error. Due to a limited report, from the company Metamax, the data of minute ventilation/carbon dioxide production ($\dot{V}_E/\dot{V}CO_2$) for participant 3 were missing and raw data were accidentally deleted. Since this value ($\dot{V}_E/\dot{V}CO_2$) was the only missing value in the protocol for this participant and not any of the primary outcomes, we decided to include the trial of participant 3 in the study. The included participants ($n = 24$) were 15 children (<18 years) and 9 adults, 8 to 37 years of age, 24 with CP (spastic = 15, dyskinetic = 7, and ataxic = 2). Eight participants were classified as GMFCS level II, 3 as GMFCS level III, and 13 as GMFCS level IV. We checked for potential bias between the Swedish and Dutch participants, and there were no statistically significant differences in the age, height, weight, gender, and GMFCS level ($P > .05$). Participant characteristics are in Table 1.

Written informed consent was obtained from participants older than 12 years and from the parents of the participants younger than 18 years after written and verbal explanation about participation in this study.

Test Protocol and Materials

Measurements in the Netherlands took place at outdoor track and field facilities (400 m), where the participants attended Frame Running training in 2017. The measurements in Sweden took place at 4 different track and field facilities (indoor: 163, 200, and 400 m; outdoor: 400 m) during 2017-2019. During the 6-MFRT, continuous breath-by-breath data on oxygen uptake ($\dot{V}O_2$), carbon dioxide production ($\dot{V}co_2$), and ventilation ($\dot{V}_E$) were recorded with a portable gas analysis system (Cortex Metamax 3B, CORTEX Biophysik GmbH, Leipzig, Germany), and corresponding software (MetaSoft Studio). In addition, HR monitors were used (Dutch participants: Polar FT7, Kempele, Finland; Swedish participants: Garmin Edge 25, Garmin, United States). The equipment was first used during a resting period of 5 to 10 minutes. Then the Swedish participants performed a low-intensity warm-up period of 5 to 10 minutes. During the 6-MFRT, the participants were instructed to cover as much distance as possible in 6 minutes on their running frame without stopping. Continuous verbal encouragement was given by the test leader who ran behind or beside the running frame if needed for safety and/or motivation. With the encouragement, they were pushed to continue the test at the highest speed as possible and the test leader motivated them to keep exercising until the 6-MFRT was completed.

To measure 6-MFRT distance, a GPS (Nike+ Running, version 1.7.9) was used in the Netherlands, and by counting laps and adding distance of incomplete laps, measured with a measuring wheel in Sweden.

Outcome Measures

The highest HR averaged over a period of 10 seconds during the 6-MFRT was determined as the HR_{peak}. Aerobic

TABLE 1

Characteristics of the Participants

	Total (24) Mean ± SD	GMFCS II (8) Mean ± SD	GMFCS III (3) Mean ± SD	GMFCS IV (13) Mean ± SD
Male/female, n	14/10	3/5	3/0	8/5
Age, y	18.6 ± 8.9	24.1 ± 10.3	18.0 ± 8.7	15.4 ± 6.8
Height, cm	156.0 ± 15.7	164.8 ± 6.4	156.7 ± 20.8	150.5 ± 17.0
Weight, kg	49.8 ± 17.6	59.4 ± 15.3	60.0 ± 27.8	41.5 ± 12.8
Body mass index, kg/m^2	19.9 ± 4.6	21.6 ± 4.5	23.5 ± 7.2	17.9 ± 3.2
CP motor type (spastic/dyskinetic/ataxic), n	15/7/2	6/0/2	3/0/0	6/7/0
FR experience, mo	27.0 ± 24.7	14.1 ± 14.9	19.3 ± 8.7	36.6 ± 28.3
FR, h/wk	1.4 ± 0.7	1.5 ± 0.8	1.5 ± 0.5	1.4 ± 0.7

Abbreviations: CP, cerebral palsy; GMFCS, Gross Motor Function Classification System; FR, Frame Running; SD, standard deviation.

parameters were calculated over 30 seconds during the 6-MFRT and consisted of (1) $\dot{V}O_{2peak}$ (mL/kg/min), (2) $\dot{V}O_{2peak}$ (L/min), (3) $\dot{V}_{Epeak}$ (L/min), (4) peak respiratory frequency (Rf_{peak}) (breaths/min), (5) $\dot{V}_{Epeak}/\dot{V}O_{2peak}$, and (6) $\dot{V}_{Epeak}/\dot{V}CO_{2peak}$.[8,12] The data of $\dot{V}CO_{2peak}$ and $\dot{V}O_{2peak}$ were used to calculate the peak respiratory exchange ratio (RER_{peak}) ($=\dot{V}CO_{2peak}/\dot{V}O_{2peak}$). All participants rated their perceived exertion after completing the 6-MFRT. Dutch participants used the children's OMNI Scale of Perceived Exertion (OMNI Scale), category range: 0 to 10. The Borg Rating of Perceived Exertion Scale (category range: 6-20) was used for Swedish participants. In addition, we collected data on age, body weight (kg), height (m), CP motor type (spastic, ataxic, or dyskinetic), and GMFCS level.

Criteria for Maximum Aerobic Performance

The definition of a maximum physiological response during the 6-MFRT was set according to objective HR and RER criteria. For the pediatric sample the criteria were HR 95% or more of 195 beats per minute (bpm) (ie, ≥185 bpm) and RER 1.0 or more.[13] For adults the criteria were set at HR 85% or more of the age-predicted HR_{max} and RER 1.1 or more.[5] The HRmax of an adult participant was predicted with the equation: 220 − age = HRmax. The HR_{peak} was then expressed as a percentage of the predicted HRmax.

Statistical Analysis

The data were analyzed using SPSS 26.0 (SPSS Inc, Chicago, Illinois). Descriptive statistics were used to summarize the characteristics of the participants and to describe the results of the 6-MFRT. The relationship between physiological parameters and 6-MFRT was calculated with Spearman correlation coefficients (ρ), due to small sample size and nonnormally distributed data.

Correlation coefficient ρ 0 to 0.39 was considered as negligible to weak, 0.40 to 0.69 as moderate, 0.70 to 0.89 as strong, and 0.90 to 1.0 as very strong or perfect.[14] Significance level was set at $P < .05$. Data are reported as mean ± standard deviation (SD) and 95% confidence interval (CI) for correlation coefficients.

RESULTS

All participants completed the 6-MFRT and the peak results are in Table 2. The $\dot{V}O_{2peak}$ values ranged from 0.62 to 2.18 L/min (mean 1.40; SD 0.48) or 19.0 to 49.8 mL/kg/min (mean 29.3; SD 7.3). The HR_{peak} ranged from 146 to 201 bpm (mean 180.7; SD 14.3). The RER_{peak} values ranged between 0.94 and 1.49 (mean 1.16; SD 0.16). On average, peak values in $\dot{V}O_2$ were observed after 3:35 min and for HR after 4:15 min.

TABLE 2

Peak Results of the 6-Minute Frame Running Test

	Total (24) Mean ± SD	GMFCS II (8) Mean ± SD	GMFCS III (3) Mean ± SD	GMFCS IV (13) Mean ± SD
$\dot{V}O_{2peak}$, mL/kg/min	29.3 ± 7.3	25.4 ± 2.8	36.2 ± 12.5	30.1 ± 7.0
$\dot{V}O_{2peak}$, L/min	1.40 ± 0.48	1.50 ± 0.40	1.94 ± 0.39	1.22 ± 0.45
HR_{peak}, bpm	180.7 ± 14.3	183.0 ± 11.1	181.3 ± 6.0	179.2 ± 17.6
RER_{peak}	1.16 ± 0.16	1.21 ± 0.16	1.11 ± 0.03	1.14 ± 0.17
$\dot{V}_{Epeak}$, L/min	62.8 ± 23.9	66.4 ± 20.4	86.4 ± 21.4	55.2 ± 23.7
Rf_{peak}, breaths/min	63.2 ± 12.9	59.4 ± 6.8	73.3 ± 14.2	63.2 ± 15.0
$\dot{V}_{Epeak}/\dot{V}O_{2peak}$	43.5 ± 11.7	44.6 ± 13.6	44.9 ± 7.5	42.5 ± 11.9
$\dot{V}_{Epeak}/\dot{V}CO_{2peak}$	39.7 ± 8.6	39.9 ± 10.0	41.3 ± 7.1	39.1 ± 8.5
OMNI scale (n = 12)	6.8 ± 1.1	6.3 ± 1.2	7.0 ± 0	7.4 ± 0.9
Borg scale (n = 12)	16.9 ± 3.4	17.0 ± 2.8	20 ± 0	16.4 ± 3.7
6-MFRT distance, m	707 ± 244	871 ± 249	729 ± 152	600 ± 210

Abbreviations: bpm, beats per minute; GMFCS, Gross Motor Function Classification System; HR_{peak}, peak heart rate; RER_{peak}, peak respiratory exchange ratio; Rf_{peak}, peak respiratory frequency; 6-MRT, 6-minute Frame Running Test; $\dot{V}_{Epeak}$, peak minute ventilation; $\dot{V}_{Epeak}/\dot{V}CO_{2peak}$, relationship between the peak minute ventilation and the peak carbon dioxide production; $\dot{V}_{Epeak}/\dot{V}O_{2peak}$, relationship between the peak minute ventilation and the peak oxygen uptake; $\dot{V}O_{2peak}$, peak oxygen uptake.

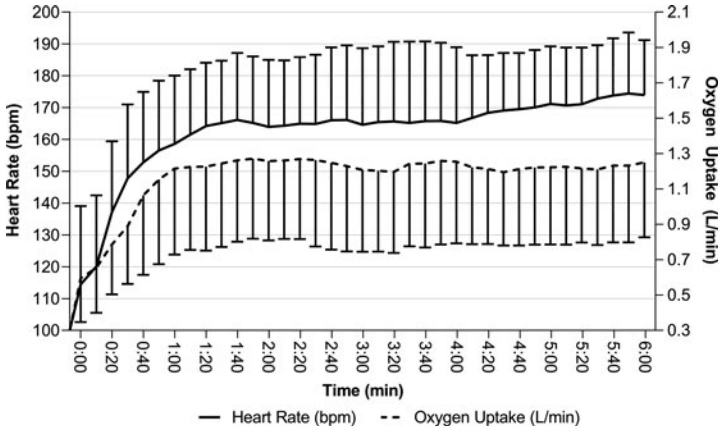

Fig. 1. Average heart rate (beats per minute) (——) and oxygen uptake (L/min) (- -) with error bars (SD) of all participants during the 6-minute Frame Running test.

The total distance of the 6-MFRT ranged from 179 to 1220 m (mean 707; SD 244).

In general, a rapid rise in $\dot{V}O_2$ and HR was found in the first minutes of the 6-MFRT (Figure 1). Due to individual differences and variations in intensity caused by the nonincremental, uncontrolled characteristics of the 6-MFRT, individual fluctuations in HR and gas analysis were found (see Supplemental Digital Content 1, available at: http://links.lww.com/PPT/A410).

Thirteen of 24 participants (54%) reached both criteria for maximum intensity based on peak values for HR and RER during the 6-MFRT. In addition, 5 participants reached either the HR or RER criteria and were close to fulfilling the other. This means that 18 participants (75%) reached near maximum effort. The $\dot{V}O_{2peak}$ for these 18 participants was similar to the whole group (29.4 ± 7.3 mL/kg/min vs 29.3 ± 7.3 mL/kg/min). HR criteria were reached in 15 of 24 participants (63%) and RER criteria in 17 participants (71%). One participant (GMFCS IV) reached the RER criterion but was far from the HR criterion. The remaining 5 participants did not meet the criteria for a maximum effort (n = 1: GMFCS II; and n = 4: GMFCS IV) (see Supplemental Digital Content 2, available at: http://links.lww.com/PPT/A411).

Distance covered during the 6-MFRT correlated significantly to oxygen uptake measured in L/min (Spearman $\rho = 0.75$; 95% CI: 0.50-0.89; Figure 2). The same relationship was not seen when normalizing to body weight (mL/kg/min). A positive correlation was observed between parameters HR_{peak} and RER_{peak} (Spearman $\rho = 0.70$; 95% CI: 0.41-0.86). Moderate correlations were found between HR_{peak} and 6-MFRT (Spearman $\rho = 0.41$; 95% CI: 0.01-0.70) and RER_{peak} and 6-MFRT ($\rho = 0.48$; 95% CI: 0.10-0.74).

DISCUSSION

The purpose of this study was to determine the physiological response of children and adults with CP classified as GMFCS levels II to IV to the nonincremental 6-MFRT and determine whether the 6-MFRT can be used as an estimation of oxygen consumption. Our data show that the 6-MFRT represented a near maximum aerobic performance in 75% of participants. Furthermore, there was a strong positive correlation between the distance covered during the 6-MFRT and the absolute $\dot{V}O_{2peak}$ L/min, indicating that the 6-MFRT can be used to estimate aerobic capacity in the individual Frame Running athlete.

Our findings support previous research by Bolster et al,[11] who report that two-thirds of the participants achieved an HR above 180 bpm during the 6-MFRT. To our knowledge, no previous data have been reported for respiratory parameters, making our study the first to assess oxygen uptake during the 6-MFRT in individuals with CP. Moreover, information about aerobic capacity in persons with CP classified as GMFCS levels III to IV is sparse, and currently reliable reference data for $\dot{V}O_{2peak}$ are only available for individuals with less severe CP (GMFCS I-III).

In the current study, the average HR_{peak} of all participants (180.7 ± 14.3 bpm) was lower but RER_{peak} (1.16 ± 0.16) was higher as compared with other studies.[6-8,15,16] There is however a discrepancy in severity of CP (more severe in our study) and the exercise test (6-MFRT vs graded max tests on bike/treadmill) compared with these studies. Frame Running seems to be more similar to cycling (as a non-weight-bearing activity) since mean HR_{peak} (187.5 ± 7.7) in the participants (75%) that reached near maximum effort was comparable to studies[6,8,16] using a cycle ergometer, but lower when compared with studies using a treadmill.[7,15] However, mean RER_{peak}

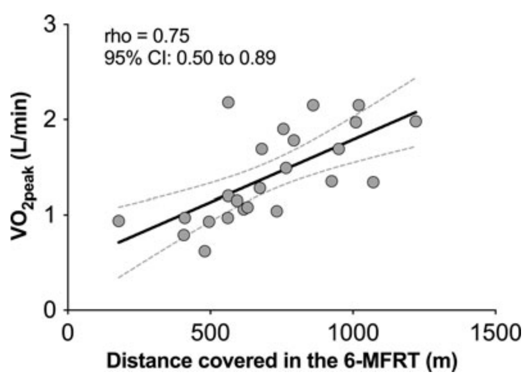

Fig. 2. Scatterplot with $\dot{V}O_{2peak}$ (L/min) and distance covered in the 6-minute Frame Running test (m). The dotted lines represent 95% confident interval (95% CI: 0.50-0.89).

(1.22 ± 0.13) in this study was higher compared with all other studies.

Our results confirm previous studies that suggest that the more severe the GMFCS level, the lower the $\dot{V}O_{2peak}$. Our participants had equivalent $\dot{V}O_{2peak}$ levels, as those from a study by Balemans et al,[8] that included children with GMFCS III. Studies that include children and adolescents with GMFCS levels I and II generally have higher $\dot{V}O_{2peak}$ levels.[6-8,15,16] Compared with children developing typically and adults, more than 90% fell in the 10th percentile or lower.[17,18] Altogether, this supports that improving aerobic fitness is warranted in this population.

In the current study, 6 participants did not meet maximal physiological response (n = 1: GMFCS II; and n = 5: GMFCS IV). In addition to the difficulty of conducting the testing, it may also be more difficult to achieve criteria for maximum cardiorespiratory performance at the more severe GMFCS levels. Tentative reasons for not reaching criteria are more severe motor limitations such as spasticity, poor muscle strength and control, as well as having difficulties in planning the race and run at a steady and maximum speed in the participants at higher GMFCS levels. Indications that the cardiorespiratory system is not the limiting factor for performance among some individuals with CP classified as GMFCS levels III and IV have been observed previously[19] with the 10-m Shuttle ride test in wheelchairs.

In persons with severe physical limitations, little is known about the physiological response to maximal exercise. Considering, 5 participants who reached either the RER or the HR criteria were just below the threshold value for RER (n = 2) or for HR (n = 3). It remains unclear whether $\dot{V}O_{2peak}$ values were truly achieved with the nonincremental 6-MFRT, yet these participants were possibly very close to maximal aerobic capacity. However, less strict criteria are often used to determine whether maximum aerobic capacity has been reached in individuals with CP, as often not all criteria are met during a maximal exercise test in this population.[6-8,16,17] Adding participant signs of exhaustion (out of breath, sweating, facial flushing, and clear unwillingness to continue despite strong verbal encouragement) as a third criterion, and using 2 out of 3 criteria is a recommendation for future research. In addition, we used different criteria for children and adults, since the physiological response to exercise is influenced by age.[5]

Regarding the second aim of our study, to investigate whether the distance covered on the 6-MFRT correlates to $\dot{V}O_{2peak}$, it is relevant to compare with other field tests appropriate for children with CP; the submaximal 6-minute walk test (6-MWT); the maximal incremental Shuttle Run Tests (SRT; 10-m SRT-I, SRT-II) and 7.5-m SRT-III.[20-23] The 6-MWT has been used to evaluate the effect of interventions in both children and adults with CP.[21,24,25] Nsenga Leunkeu et al[24] observed close agreement between measurements of $\dot{V}O_{2peak}$ on the cycle ergometer and during the 6-MWT for children with CP GMFCS levels I and II; however, it is not clear whether the 6-MWT can be used to predict $\dot{V}O_{2peak}$ in individuals with CP.[26] Moreover, the Shuttle Run Tests (SRT-I, SRT-II) were found to represent a maximal effort (RER >1.0, HR >180 bpm) and correlate strongly to $\dot{V}O_{2peak}$, as validated by graded treadmill test.[22] Similarly, the SRT-III (GMFCS III, using a walker) promotes HR more than 180 bpm in the majority of participants. However, the validity of the SRT-III as a proxy for oxygen consumption is currently unknown. Collectively, current field tests either exclude children with more severe CP (GMFCS III-IV) or lack information on oxygen consumption, underlining the importance of continuous development of new feasible and inclusive field tests.

Given that the 6-MFRT represented a near maximum effort for the majority of the participants in the present study, we investigated the relationship between distance covered during the 6-MFRT and $\dot{V}O_{2peak}$. We observed a strong significant correlation between the 6-MFRT distance (m) and absolute $\dot{V}O_{2peak}$ (L/min). This suggests that the 6-MFRT can be used as an estimation of oxygen consumption for the individual Frame Running athlete to assess gains in aerobic capacity after a training intervention. As expected, the same relationship was not seen when normalizing to body weight (mL/kg/min), as the validity of $\dot{V}O_{2peak}$ normalized to body weight has been strongly criticized in children and adolescents developing typically, as body fat is highly variable in growing individuals.[27] This is relevant to our study population as it was heterogeneous with respect to body weight, ranging from underweight to overweight, as previously described for the CP population.[28] Lean body mass would have been a more suitable normalization parameter, as it is considered a more appropriate measure of body size and composition when testing $\dot{V}O_{2max}$ in individuals younger than 18 years[29]; however these data were not available in the current study.

Our results should be viewed in the light of the following limitations. The measurements were performed at indoor and outdoor locations and thereby with variable environmental conditions, which could influence RER and HR.[30] The small sample size with large age range in this study reduces the generalization of the results to a larger population.

CONCLUSIONS

This study provides new information about aerobic capacity and the physiological response during the 6-MFRT in children and adults with CP. Eighteen out of 24 participants (75%) reached a near maximum aerobic performance during the 6-MFRT. Although more research is needed, in terms of a progressive maximum exercise test on a running frame to validate the result, it should be noted that currently no other exercise tests are available for individuals with CP, classified as GMFCS III/IV, that can evoke a cardiorespiratory response of this intensity. Moreover, field tests like the 6-MFRT are practical and provide results that are specific and of direct interest to the sports. Nevertheless, as distance covered during the 6-MFRT was strongly related to the oxygen uptake, we suggest that the 6-MFRT can be used as an estimation of oxygen consumption for the individual Frame Running athlete to assess gains in aerobic capacity after a training intervention.

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