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Metabolic, cardiorespiratory, and neuromuscular fitness performance in children with cerebral palsy: A comparison with healthy youth

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The aim of this study was to assess metabolic, cardiorespiratory, and neuromuscular fitness parameters in children with spastic cerebral palsy (CP) and to compare these findings with typically developing children. 40 children with CP (21 males, 19 females; mean age, 11.0±3.3 yr; range, 6.5–17.1 yr; Gross Motor Function Classification System levels 1 or 2) and 40 healthy, age- and sex-matched children completed a test battery that consisted of 8 tests and 28 measures that assessed cardiorespiratory fitness, energy expenditure, anaerobic endurance, muscle strength, agility, stability and flexibility. Children with CP had significantly lower performance ($P<0.05$) on most cardiorespiratory and metabolic tests than those of healthy children, Differences in neuromuscular measures of muscular strength, speed, agility, anaerobic endurance, and flexibility between groups were most apparent. Grouped differences in cardiorespiratory variables revealed a 25% difference in performance, whereas grouped differences in metabolic and neuromuscular measures were 43% and 60%, respectively. The physical fitness of contemporary children with CP is significantly less than healthy, age-matched children. Significant differences in neuromuscular measures between groups can aid in the identification of specific fitness abilities in need of improvement in this population.

Keywords: Cerebral palsy, Strength, Agility, Flexibility, Physical fitness, Child

INTRODUCTION

Spastic cerebral palsy (CP) is a chronic, neuromuscular disorder which is characterized by increased muscle tone and spasticity, muscle weakness, poor coordination, muscle strains, bone deformities, and muscle rigidity. Due to these impairments, many individuals with CP have difficulty walking independently, navigating uneven terrain, and performing daily physical functioning (Damiano and Abel, 1998). Even in highly functional youth with CP, movements are often characterized by excessive knee and hip flexion which are characteristic of muscle weakness in the lower extremities (Unnithan et al., 1998). Consequently, children with CP tend to engage in less regular physical activity than able-bodied peers and this can adversely affect their long-term health and well-being (Van den Berg-Emons et al., 1995).

The capacity to improve the physical fitness of youth with CP and enhance their daily functioning is of interest to therapists and researchers because youth with CP tend to have relatively low levels of cardiorespiratory fitness (Hoofwijk et al., 1995), a reduced metabolic reserve (Unnithan et al., 1996) and pervasive muscle weakness

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(MacPhail and Kramer, 1995). While the available data indicate that youth with CP may benefit from exercise training (Verschuren et al., 2008), non–intervention-specific testing has been used to assess the effectiveness of training interventions in children with CP and the methodological quality of some exercise studies has been deemed low (Verschuren et al., 2008; Verschuren et al., 2011a). Moreover, the available data on anaerobic testing and training on youth with CP is scanty which is surprising since most childhood activities are characterized by short bursts of high intensity activity followed by brief periods of rest as needed (Bailey et al., 1995).

To our knowledge, no published research has included a comprehensive test battery that was specifically designed to assess cardiorespiratory, metabolic, and neuromuscular fitness performance in children with CP. To address this gap in the research literature, the current study compared cardiorespiratory fitness, energy expenditure, anaerobic endurance, muscle strength, agility, stability, balance, and flexibility in children with CP with healthy, age- and sex-matched children. This study reports the cardiorespiratory, metabolic, and neuromuscular fitness abilities of children with CP for providing a quantitative assessment that has the potential to be used by therapists and researchers to establish baseline measures, define objective treatment outcomes, and identify specific fitness abilities in need of improvement in this clinical population.

MATERIALS AND METHODS

Participants and procedures

All subjects with CP attended a local rehabilitation service (Niño Jesús Children's Hospital, Madrid, Spain). The inclusion criteria were that each subject (a) was aged between 6 yr and 15 yr old; (b) had been diagnosed as a spastic diplegic by a medical practitioner; (c) had a Gross Motor Function Classification System of level I or II determined by a medical pratitioner; (d) had not been subject to any orthopedic surgery for the year preceding the start of the study; (e) had not been subject to any botulinum toxin injection to the lower extremities for three months preceding the start of the study; and (f) had nonaffected cognitive abilities. Healthy able-bodied children were recruited from two schools in the local community. All participants lived in or near Madrid, Spain.

Forty children with CP (21 males and 19 females) and 40 healthy children (21 males and 19 females) participated in the study. Descriptive data for both groups is presented in Table 1. Verbal assent was obtained from each participant and written parental permission approved by the local ethics committee at the hospital was obtained from the parents. All participants with CP maintained their normal physical therapy treatments throughout the study period.

Table 1. Physical characteristics

Variable	Healthy group (n=40)	CP group (n=40)
Age (yr)	11±3.6	11±3.3[a)]
Mass (kg)	40.7±15.0	38.7±14.0[a)]
Height (cm)	147.2±18.1	143.5±17.0[a)]
Body mass index (kg/m^2)	18.0±3.0	18.4±3.2[a)]

Values are presented as mean±standard deviation.
CP, cerebral palsy.
[a)]No significant difference between groups.

Study design

Children in both groups participated in 3 familiarization sessions on nonconsecutive days of the week prior to testing in order to practice each test protocol and become familiar with the equipment. Participants practiced each test at submaximal levels and received guidance and instruction from the research staff. Following the familiarization period, participants completed all study-related procedures during two testing sessions on nonconsecutive days. Fitness performance was closely supervised and assessed by a physician, a physical therapist, and a clinical exercise physiologist. Research assistants provided technical support and encouragement throughout the study period. The same team of researchers evaluated all participants throughout the study period.

Measurements

Body weight (BW) was measured on a calibrated electronic scale and height was measured using a stadiometer. Resting heart rate (HR) was measured with a HR monitor (Polar Electro OY, Kempele, Finland) and the lowest value observed during a 5-min seated rest period was recorded. Tests were conducted in the same

Table 2. Tests order

Test	Variable measured
Body mass, height, and resting heart rate	Body mass, height, and resting heart rate
6-min free walk	Energy cost
Graded exercise test	Aerobic capacity and power
Sit & reach and knee flexion	Flexibility
Stability test	Stability
Time up & go, time up and down stairs	Agility
Power squat	Maximal power
Leg press	Maximal strength
3 nonstop time up and down stairs (3×TUDS)	Anaerobic endurance

order for all participants and a 5-min rest interval between each test was permitted (Table 2). Since the lingering effects of fatigue from the global test battery could influence energy cost and metabolic measures, these tests were performed first. Also, power was assessed before strength to maximize performance. Anaerobic performance was performed last since these fatiguing tests consist of relatively short-term, high intensity exercise bouts.

The majority of the tests used in the present investigation have been used by other investigators (Ahlborg et al., 2006; Darrah et al., 1999; Unnithan et al., 1998). Additional tests such as stability or 3×TUDS (time up and down stairs) for anaerobic endurance were included to provide a global fitness assessment in our study population. Bar-Or (1986) noted that anaerobic endurance was the best indicator of functional capacity in children with neural disorders since most of their tasks fall into this metabolic milieu. More recently, a panel of experts identified a core set of exercise tests for children with CP that included aerobic and anaerobic measures (Verschuren et al., 2011a). Of potential relevance, Verschuren et al. (2011b) noted the need for a systematic strength assessment in this clinical population since previous research that examined 'anaerobic' performance in youth with CP did not include measures of strength or power.

Metabolic tests

The first metabolic test consisted of a 6-min walk over 5 m. Every minute, data for HR, oxygen consumption (VO_2), walking speed, and step frequency was recorded. Walking speed was calculated from the time it took to perform 3 contacts with the same foot and step length was calculated from frequency and speed. Using the average HR and speed, the energetic cost index (energy expenditure index with heart rate, EEIHR) and the walking energy efficiency (WEE) were calculated. The second metabolic test was used to calculate a fatigue index and consisted of climbing up and walking down twelve stairs as fast as possible. Subjects performed three continuous timed attempts. Lap times were recorded every time they reached the first step. The time difference between 1st and 3rd trials was used to determine a fatigue index which represented a decrement in performance as a percentage of time lost. Subjects performed only one trial of this test.

Cardiorespiratory test

The cardiorespiratory test was performed after a short rest from the 6-min walk or when the participants' HR was below 100 beats/min. Each participant walked on a treadmill (Technogym Run Race 1400 HC, Gambettola, Emilia-Romagna, Italy) and performed a graded exercise test until voluntary exhaustion. Initial speed was set at 2.0 km/hr and grade was 0.5%. Both speed and grade were increased every 15 sec (0.1 km/hr and 0.5% respectively). Due to the peripheral limitations in youth with CP and related safety concerns during exercise testing, all participants were allowed to hold the handrail of the treadmill with one hand and a research assistant stood on the frame of the treadmill behind each participant. In addition, the portable oxygen uptake system was supported by a research assistant during the test to reduce the weight bearing load on the participants.

Throughout the graded exercise test, HR was recorded continuously with a HR monitor (Polar Electro OY) and a maximal HR was estimated using the Gellish et al. formula (207–[0.7×age]). Gas exchange data was collected continuously using a portable system (Medical Graphics Corp., VO2000, St. Paul, MN, USA) and the following variables were measured: oxygen uptake (VO_2), pulmonary ventilation (VE), ventilatory equivalent for oxygen ($VE{\cdot}VO_2^{-1}$) and carbon dioxide ($VE{\cdot}CO_2^{-1}$), and end-tidal partial pressure of oxygen ($P_{ET}O_2$) and carbon dioxide ($P_{ET}CO_2$). Maximal oxygen uptake (VO_2max) was recorded as the highest VO_2 value obtained for any continuous 1-min period during the test. At least two of the following criteria were required for the attainment of VO_2max: (a) a plateau in VO_2 values despite increasing velocity; (b) a respiratory exchange ratio ≥1.15; or (c) the attainment of a peak HR value above 95% of the age-predicted maximum. The ventilatory threshold (VT) was determined using the criteria of an increase in both $VE{\cdot}VO_2^{-1}$ and $P_{ET}O_2$ with no increase in $VE{\cdot}VCO_2^{-1}$, whereas the RCT was determined using the criteria of an increase in both $VE{\cdot}VO_2^{-1}$ and $VE{\cdot}VCO_2^{-1}$ and a decrease in $P_{ET}CO_2$. Two independent researchers determined VT and RCT. If there was disagreement, the opinion of a third researcher was obtained (Lucía et al., 2000). Since the progressive workload was determined by speed and grade, the variable time was used for VT, RCT, and peak VO_2 comparisons between healthy youth and participants with CP.

Neuromuscular tests

All subjects performed a series of tests to assess strength, power, agility, stability, and flexibility. The first neuromuscular test was a power half squat. Starting from a seated position with 90º knee flexion and hands on the side of seat, participants stood up as fast as possible wearing a weighted vest with 20% body mass. During the test, subjects supported a light plastic stick on their shoulders which was connected to an encoder system to measure power (Real Power, Globus, Codognè, Italy). Using total load (body mass plus

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overload) and the average velocity of the best lifting performance of three trials, mean power (w) was calculated. Only the concentric phase of the movement was used for mean power calculations.

A lower body repetition maximum (RM) test was used to estimate maximal dynamic muscular strength using a child-size variable resistance machine (Strive Inc., Bethlehem, PA, USA). Following dynamic calisthenics and 2 warm-up sets (8 repetitions with 50% of the estimated 1 RM and 3 repetitions with 70% of estimated 1 RM), participants performed the maximum number of repetitions to failure (less than 7 repetitions) on the leg press using proper exercise technique. The selected loads for RM testing were based on data collected during the familiarization period. The 1 RM was estimated using the Brzycki formula (1 RM = weight lifted/1.0278 – (0.0278 × No. of repetitions).

Subjects performed two tests to assess agility which have been used by our clinical group in pediatric populations. The first test was the time up and go (TUG) whereby participants started from a seated position with their hands crossed over their chest (Podsiadlo and Richardson, 1991). Subsequently, they stood up and walked as fast as possible to a 3-m mark and then returned to the seat. The second agility test was the TUDS and consisted of climbing up and walking down 12 stairs as fast as possible (Zaino et al., 2004). Subjects were allowed to hold onto the handrails for safety and a research assistant was nearby. Each agility test was performed twice with a 1-min rest period between trials and the best time was recorded to the nearest 0.1 sec.

Subjects performed the knee flexion and sit and reach tests to assess flexibility. Knee flexion was measured in a prone position with the shoulders, hips, and knees in contact with the floor. Knee flexion was measured in each leg and was defined as the lowest angle reached with the passive force of a therapist without any modification of the initial position. The sit and reach test was performed following standard procedures with both feet at 23-cm mark on the flexibility box. The best score of two trials was measured to the nearest cm.

A seven stage exercise test adapted from Kibler et al. (2006) was used to assess stability. Participants attempted to mimic and maintain total body stability in a series of positions or movements while standing on one leg. A 0 score was recorded if the participant was unable to maintain the proper position and 1 point was recorded if they had difficulties but were able to maintain the desired position for 2 to 10 sec. If subjects were able to maintain the desired position or movement for 10 sec for exercises 1 to 6 and 30 sec for exercise 7 then 2 points were recorded. Left and right sides were evaluated independently and the difference between them was calculated. Global stability for each leg and relative differences between legs were used for data analysis.

Statistical analysis

Student *t*-tests for independent samples were used for matching comparisons and for conditioning differences. All data were expressed as mean ± standard deviation. Significance level was set at $P < 0.05$ and data analyses were performed with IBM SPSS Statistics ver. 20.0 (IBM Co., Armonk, NY, USA).

RESULTS

Healthy subjects had superior performance on most of the performance measures used in this investigation. Although several cardiorespiratory and metabolic variables showed minimal differences between groups, differences in neuromuscular performance between healthy participants and youth with CP were most apparent.

Metabolic tests

No significant differences between healthy youth and participants with CP were found in the energy cost of 6-min free walk-

Table 3. Data from 6-min walk test

Variable	Healthy group (n = 40)	CP group (n = 40)	*P*-value
Energy cost (mL/kg/min)	12.1 ± 2.8	12.02 ± 3.7	
Energy cost speed (m/min)	77.4 ± 27.4	47.5 ± 11.8	< 0.05
Economy (mL/kg/m)	0.167 ± 0.05	0.265 ± 0.09	< 0.05
EEIHR (beats/m)	0.45 ± 0.2	0.71 ± 0.3	< 0.05
WEE (m/beats)	2.5 ± 1.0	1.7 ± 0.8	< 0.05
Step length (cm)	70.4 ± 23.9	45.7 ± 9.2	< 0.05
Length/speed (cmsec/[m/sec])	0.9 ± 0.1	1.0 ± 0.2	

Values are presented as mean ± standard deviation.
CP, cerebral palsy; EEIHR, energy expenditure index with heart rate; WEE, walking energy efficiency.
$P < 0.05$, significant difference between groups.

ing (t_{78} = -0.103; P > 0.05) (Table 3). However, walking speed was significantly higher in healthy youth as compared to youth with CP (t_{78} = -6.334; P < 0.05 [P = 0.001]). Thus, the energy cost at a given speed (i.e., economy), was significantly better in healthy children (t_{78} = 5.728; P < 0.05 [P < 0.001]). HR vs speed indexes were also significantly higher in healthy children as compared to youth with CP EEIHR (t_{78} = -4.01, P < 0.05 [P < 0.001]) and WEE (t_{78} = -6.09, P < 0.05 [P < 0.001]). Average step length during the 6-min walk was longer in healthy children (t_{78} = 4.778, P < 0.05 [P < 0.005]). However, the shorter step length in children with CP was related to the slower speed since the step length/speed rate showed no significant difference (t_{78} = 1.866, P > 0.05 [P = 0.066]).

Cardiorespiratory tests

Healthy subjects had better performance in time to exhaustion during the graded test as compared to youth with CP (t_{78} = -8.104, P < 0.001 [P < 0.001]). Table 4 illustrates these findings and other cardiorespiratory data. Both peak VO_2 (t_{78} = -4.852, P < 0.05 [P < 0.0001]) and RCT (t_{78} = -6.865, P < 0.05 [P = 0.001]) appeared later in healthy children. Only threshold HR percentages were the same between healthy and CP children (t_{78} = 0.592, P > 0.05 and t_{78} = -1.070, P > 0.05, respectively). Peak VO_2 in healthy children was higher (t_{78} = -6.354, P < 0.05 [P < 0.001]). However, healthy children were closer to reach estimated maximal HR than youth with CP (95% vs 90%, respectively; t_{78} = -4.852, P < 0.05 [P < 0.001]).

Neuromuscular tests

Neuromuscular performance was significantly better in healthy children as compared to youth with CP with the exception of the fatigue index between 1st and 3rd repetition in the 3-TUDS (t_{78} = -1,167, P > 0.05 [P = 0.06]). Neuromuscular data are presented in Table 5.

Table 4. Graded exercise results

Variable	Healthy group (n = 40)	CP group (n = 40)	*P*-value
VT (%peakVO_2)	52.2 ± 10.9	58.8 ± 13.0	< 0.05
VT (% estimated maximal HR)	70.7 ± 6.9	70.0 ± 6.3	
RCT (%peakVO_2)	75.9 ± 10.0	81.6 ± 8.3	< 0.05
RCT % (estimated maximal HR)	83.0 ± 5.8	81.6 ± 6.1	
Time to RCT (sec)	517 ± 122	341 ± 106	< 0.05
Peak VO_2 (mL/kg/min)	40.2 ± 9.0	28.7 ± 7.0	< 0.05
Peak VO_2 (% estimated maximal HR)	95.5 ± 4.9	90.0 ± 5.3	< 0.05
Time to peak VO_2 (sec)	761 ± 200	445 ± 143	< 0.05

Values are presented as mean ± standard deviation.
CP, cerebral palsy; VT, ventilatory threshold; VO_2, oxygen consumption; HR, heart rate; RCT, respiratory compensation threshold.
P < 0.05, significant difference between groups.

Table 5. Neuromuscular tests

Variable	Healthy group (n = 40)	CP group (n = 40)	*P*-value
Stability (%)	95.6 ± 8.0	26.2 ± 23.4	< 0.05
Stability (legs % dif)	3.2 ± 6.0	34.5 ± 32.8	< 0.05
1RM leg press (kg)	101.0 ± 46.7	38.3 ± 21.8	< 0.05
Maximal power 90° squat (w)	496 ± 320	262 ± 194	< 0.05
Maximal power 90° squat (w/kg)	11.5 ± 3.8	6.7 ± 2.7	< 0.05
Time up & go (sec)	3.5 ± 0.5	5.7 ± 1.8	< 0.05
TUDS (sec)	5.8 ± 1.8	12.7 ± 7.5	< 0.05
3-TUDS (sec)	20.1 ± 3.2	45.0 ± 26.5	< 0.05
3-TUDS (% 1st a 3rd)	-12.9 ± 7.7	-15.5 ± 11.8	
Knee flexion (degrees)	36.5 ± 9.5	53.3 ± 15.5	< 0.05
Sit and reach (cm)	26.3 ± 9.1	15.6 ± 6.4	< 0.05

Values are presented as mean ± standard deviation.
CP, cerebral palsy; RM, repetition maximum; TUDS, time up and down stairs.
P < 0.05, significant difference between groups.

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DISCUSSION

The major finding from this study was that previously reported differences in cardiorespiratory fitness between healthy children and children with CP were also observed in other fitness measures including muscular strength, speed, agility, anaerobic endurance, flexibility, and balance. To our knowledge, this is the first trial with a substantial sample size to compare healthy youth with participants who have CP over a wide range of cardiorespiratory, metabolic, and neuromuscular performance parameters. These data add to our understanding of the limitations in fitness performance in youth with CP as compared to age-matched healthy controls, and can be used as initial references values for identifying specific fitness abilities in need of improvement in this clinical population.

From a metabolic perspective, energy expenditure during the 6-min walk was similar between groups. However, walking speed was significantly higher in healthy children which highlights their better walking economy. Other studies found a 14% to 45% difference for WEE between healthy youth and participants with CP which is similar to our observations of a 40% difference in performance (Unnithan et al., 1998, Van den Berg-Emons et al., 1996). Further, previously reported differences of 25% to 100% for EEIHR are also consistent with our findings of 75% (Raja et al., 2007; Suzuki et al., 2001). Differences in peak VO_2 between healthy youth and participants with CP has been found to range from 56% to 15% (Verschuren and Takken, 2010), whereas we observed group differences of 29%. Testing protocols and, more particularly, sample differences in motor limitations could explain these differences.

During the cardiovascular assessment, metabolic thresholds were at the same estimated percent maximal HR. This observation, along with the lower peak HR in children who have CP, may reflect the noteworthy importance of muscle endurance limitations during graded exercise testing in youth with CP. As such, the lower peak VO_2 and shorter time to exhaustion in children and adolescents with CP would be partially dependant on peripheral limitations (both muscular and mechanical) as previously reported (Hoofwijk et al., 1995; Unnithan et al., 1998). An evaluation of neuromuscular performance indicated that differences in strength and power between healthy youth and participants with CP ranged from 35% to 60% which was consistent with observed differences in flexibility. When expressing 1 RM leg press strength relative to body mass, we found youth with CP had a relative strength measure of 1.0-kg/kg BW whereas the relative strength for healthy participants was 2.6-kg/kg BW. Although few leg press data in pediatric populations are available for comparison, Scholtes et al. (2010) reported a relative leg press strength measure of approximately 1.3-kg/kg BW in 6- to 13-yr-old participants and Faigenbaum et al. (2003) noted a relative strength measure of 1.5-kg/kg BW in healthy untrained 6- to 12-yr-old children (Faingenbau et al., 2003). Although Liao et al. (2007) assessed 1 RM weighted vest sit-to-stand in 5-12 yr-old children with CP, it is not possible to compare our findings with this report due to notable differences in testing methodology.

In the present investigation, the most notable differences between healthy youth and participants with CP were found in measures of stability and anaerobic endurance. For example, we found the greatest differences between groups were related to measures of static and dynamic stability since the TUG, TUDS and stability tests required participants to turn around, climb up, climb down, stand and balance. These observations are supported by findings from Schalow and Jaigma (2005) who found that the development of neuromuscular control with this type of training and fundamental movement skill development is essential for youth with CP.

Differences for maximal power assessments in the present study were not so apparent between healthy children and children with CP. These observations may be due to the difficulty of the test as well as the testing methodology. The power assessment did not require an ankle plantarflexion action while agility, stability, and anaerobic endurance tasks take into account ankle plantarflexor power generation, the specific power assessment used in our investigation did not take this into account. Future studies involving youth with CP should define a more functional test of muscular power Notwithstanding this limitation, it is important to note that muscular power generation directly influences performance in tests of stability and anaerobic endurance In addition, these tests take into account the stretch-shortening cycle, key for gait efficiency, which should be addressed in future studies as plyometric (Moreau et al., 2012) and eccentric strength training (Reid et al., 2010) is developing in the scientific literature regarding rehabilitation in CP.

It appears that as the neuromuscular demands of a selected performance measure increase, the differences in performance between healthy participants and youth with CP become more apparent. For example, performance on the TUG test which required agility and dynamic balance revealed a 63% difference in performance between groups whereas differences in performance on the TUDS test which required a higher stability, exceeded

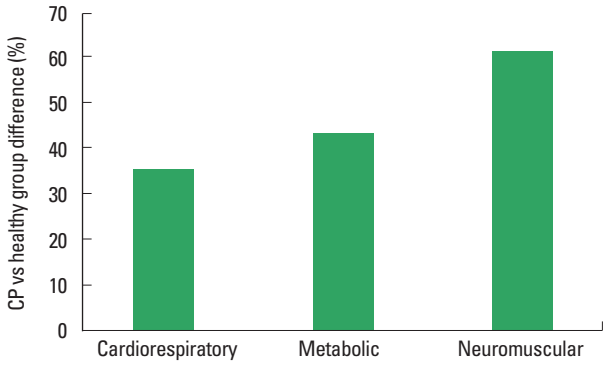

Fig. 1. Percentage differences in metabolic, cardiorespiratory, and neuromuscular grouped parameters in youth with cerebral palsy (CP) vs healthy, age-matched controls.

100% with healthy youth outperforming participants with CP. Percent differences in cardiorespiratory, metabolic, and neuromuscular performance between youth with CP and age-matched healthy participants are illustrated in Fig. 1.

There were certain limitations associated with this study which should be considered when evaluating these data. First, youth with CP and healthy participants were matched for age, height, and body mass, although subjects in our investigation included children and adolescents (age range, 6 to 15 yr). Second, youth with CP were receiving usual physiotherapy treatment during the study period. These observations may affect the generalizability of our findings to other populations with CP.

Furthermore, the aim of the treatment administered would also determine test selection, thus future studies should broaden fitness assessments to comply with the different rehabilitations goals proposed for children with CP.

In conclusion, our data indicate that differences previously reported in cardiorespiratory fitness between healthy youth and participants with CP are also evident in selected measures of metabolic and neuromuscular performance. These findings highlight the importance of identifying specific fitness abilities in need of improvement in youth with CP, and emphasize the importance of developing targeted training interventions to enhance functional fitness performance in this clinical population. Consequently, integrative fitness training which addresses several elements of fitness including neuromuscular performance is indicated for youth with CP.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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