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Cardiopulmonary exercise data during quadriceps isometric contraction sustained to fatigue in children with cerebral palsy

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Abstract. The aims of this study were to: 1) study the cardiopulmonary responses during exhaustive isometric contraction in cerebral palsy (CP) children vs. apparently normal (AN) children 2) to study the relationship between muscle endurance and maximal O_2 uptake (peak VO_2) in CP children. Eight CP children (GMFC 1 to 2) and 8 AN children underwent a graded cycle exercise test and a quadriceps force and endurance assessment with cardiopulmonary measurement on an isokinetic dynamometer. During isometric contraction, muscle endurance did not differ between the groups but a higher O_2 uptake was shown in the CP group. Moreover in this group, peak VO_2 was correlated with muscle strength ($R = 0.77$, $P < 0.05$) and endurance ($R = 0.68$, $P < 0.05$). Cerebral palsy children have a higher O_2 uptake during isometric endurance testing despite a lower absolute isometric force but their quadriceps endurance is not impaired after normalization with muscle mass and time. Additionally, quadriceps strength and endurance in CP children were in relationship with their peak VO_2. This higher O_2 uptake is probably due to coactivation and/or cocontraction during exhaustive quadriceps isometric contraction.

Keywords: Children, cerebral palsy, cardiorespiratory data, isometric contraction, quadriceps

1. Introduction

Cerebral palsy (CP) is a chronic neurological disorder that affects movement and postural control [18]; in children this disorder is associated with muscle weakness leading to a reduced functional capacity [8,21,22]. In addition, CP children show evidence of exercise intolerance and early fatigability [17]. Recently, there has been a renewed interest in strength testing and in the use of strength training in rehabilitation programs for individuals with CP or other spastic motor disorders [4,7,26]. Assessing the physical capacity of CP children with is important for two reasons: it can yield information on the evolution of functional capacity and the natural history of the disease [24] and may assist in assessing the effects of therapy and conditioning [2,8].

Cerebral palsy children demonstrate a reduced physical capacity relative to children who apparently develop normally [7,10,15]. However, the precise causes of exercise intolerance in this population are not clearly known. Little data is available regarding cardiorespiratory performance during exercise in CP children [17]. The oxygen cost of physical activity in CP children appears to be high [15] possibly accounting for their

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Table 1
Clinical characteristics and chest wall deformity in CP children

Subject number	CP Classification	GMFC level	Chest wall deformity
1	Sd	2	yes
2	Sd	2	no
3	Sd	2	yes
4	Sh	1	yes
5	Sh	1	yes
6	Sh	1	no
7	Sh	1	no
8	Sd	2	no

CP: Cerebral palsy, Sd = Spastic diplegic, Sh = Spastic hemiplegic, GMFC: gross motor function classification system.

early fatigability [3]. Several studies have demonstrated that these children have decreased muscle strength compared to age-matched controls [1,6,23]. It has been suggested that high antagonist muscle activation and high mechanical power may partially explain part this high oxygen cost and the ensuing easy fatigability [3].

The need for VO_2 max assessment includes the measurement of the potential improvements in the economy of locomotion in the child who undergoes surgical intervention, uses ankle-foot orthoses or undergoes rehabilitation. The measurement of VO_2 max should be considered a useful adjunct to other modes of exercise testing such as muscle strength and endurance evaluation. Two studies [12,13] seemed to indicate that the more aerobically fit CP children are better with their functional abilities.

Cerebral palsy children have decreased aerobic capacity [13] that could be related to their skeletal muscle function. Little information is available on the potential link between peak VO_2 and skeletal muscle function in CP children. Such information could help in better understanding the physiological levels of exercise intolerance (cardiopulmonary and muscular levels) [5]. Therefore, the aims of our study were to investigate quadriceps skeletal muscle endurance during isometric exercise and the associated cardiopulmonary exercise responses in CP children compared to age-matched typically developing children as well as examine the possible relationship between peak VO_2 and skeletal muscle force and endurance in CP children.

2. Methods

2.1. Subjects

Eight CP children (6 boys, 2 girls) with normal intelligence to mild mental retardation (recorded from medical chart) and 8 aged-matched AN children (8 boys) free of any cardiopulmonary or cerebral palsy pathologies took part in this study. The affected children were attending elementary school at the Motor Education Institute in Amiens, France. All were spastic; 4 were classified as diplegic (legs and feet more affected than arms and hands) and 4 were classified as hemiplegic (involvement of one side of the body). Two children with spastic hemiplegia wore ankle-foot orthoses. All CP children possessed mild motor abnormalities with a GMFC classification of 1 or 2. Four children were affected by chest wall deformity (physician-evaluated). None were taking medications for spasticity. Children and their parents were informed of all aspects of the study and written consent was obtained. The study was in accordance with the ethical standards of the Helsinki Declaration of 1975. All children were assigned randomly to a cardiopulmonary exercise testing on cycloergometer and an assessment of skeletal muscular function on an isokinetic dynamometer with gas exchange analysis. All tests were performed in the morning (between 9 and 12 AM) in the same place following the same protocol. Clinical characteristics of CP children are given in Table 1.

2.2. Anthropometrics measurements

For normalization purposes, anthropometric measurements (weight, height, lean body mass and muscle volume of lower limb) were carried out before the beginning of the tests with the measurement of the skin folds (4 skinfolds: biceps, triceps, subscapular and supra-iliac) and circumferences of the lower limb (limb less affected by spasticity was chosen) according to previously published methodology [16]. Anthropometric measurements for both groups are given in Table 2.

Table 2
Anthropometric data values in children developing typically (control) and CP children

Parameter	Control children ($n = 8$)	CP children ($n = 8$)	P values
Age (years)	14 ± 1	14 ± 1	ns
Weight (kg)	54 ± 11	45 ± 10	ns
Height (cm)	165 ± 8.7	149 ± 12†	†

Means $\pm$ standard deviation. † = $P < 0.01$. CP: cerebral palsy.

2.3. Cardiopulmonary exercise testing

Cardiopulmonary exercise testing (CPET) was performed on a cycloergometer (Monark 824 E, Vansbro, Sweden). After a familiarization procedure [11,16,24], children underwent an incremental protocol. In healthy control children, initial power was 30W and increased by 15W every minute with a pedaling rate fixed at 60 rpm [16]. In CP children, the same load increase was used (0.5 kg/min) on the scale of the ergometer. CP children were asked to maintain a regular pedaling cadence; a 60 rpm pedaling rate was not used because this rate cannot be maintained by most CP children [13, 17,25]. Vigorous verbal encouragement was given throughout the tests to achieve maximal effort. Gas exchange was measured breath-by-breath using an open circuit technique with the Cosmed K4b^2 gas analyser (Cosmed Srl, Rome, Italy). Children breathed through a rubber mask connected to a bi-directional flow-meter. The calibration of the flow module was accomplished by introducing a calibrated volume of air at several flow rates with a 3-liter pump. Each gas analyser was calibrated before each test using a standard certified commercial gas preparation (16% for O_2 and 5% for CO_2). Data were averaged every 15 s for minute ventilation (VE, l.min^{-1} BTPS), O_2 uptake (VO_2, l.min^{-1} STPD), CO_2 production (VCO_2, l.min^{-1} STPD), respiratory exchange ratio (R.E.R), ventilatory equivalent for O_2 (VE/VO_2), and CO_2 (VE/VCO_2), and breathing frequency (f). CPET lasted until the attainment of three of the four following maximal criteria in healthy control children: 1. a plateau of VO_2 despite an increased workload; 2. maximal heart rate (maximal predicted heart rate $\pm$ 5%); 3. respiratory exchange ratio > 1.1; 4. inability to maintain the pedaling frequency at 60 rpm (3). For CP children, the test lasted until fatigue (inability to pedal) and/or the appearance of symptoms (dyspnea and/or fatigue) that limited and/or terminated the exercise. Maximal VO_2 value reached during the last 15 s of the CPET was considered as the peak VO_2 for both groups. Exercise tolerance was assessed with cardiorespiratoy parameters measured at peak exercise for the last 15 s of the exercise phase. At the end of the test, subjects were allowed 5 min of passive recovery.

2.4. Skeletal muscle strength and endurance assessment

Skeletal muscle strength and endurance were measured for the quadriceps muscle on an isokinetic apparatus (Cybex Norm II, Ronkonkoma, USA). Control children were seated and positioned with the knee at 60° and hip at 120°. In CP children, the hip angle was set at 120°. The respective position of CP children depended on their available knee joint range (CP children: $68 \pm 12°$ vs 60° for control children, $P = 0.06$).

Following a familiarization procedure, skeletal muscle strength and endurance were measured using maximal voluntary isometric force (MVIF), and the isometric endurance time (IET) at an intensity level of 50% of the MVIF. Three sets of 5 repetitions were given to the children to develop a maximum force. During MVIF measurement, each child could visualise his/her generated muscular force on a computer screen; moreover, encouragement was given to the children during this maximal test in order to obtain the best possible performance. Each repetition was separated by 20 s recovery and each set was separated by a 1 min recovery period. The best values of each set were retained and averaged; this average value was considered as the MVIF. For the IET measurement, children were asked to maintain an isometric contraction at 50% of MVIF as long as possible. IET reflects skeletal muscle fatigue because the skeletal muscle blood flow is restricted. A visual reference mark corresponding to 50% of the MVIF was placed on the computer screen; children could control as best as possible their intensity of contraction. Children were asked to maintain this intensity of contraction near the visual reference mark during the test. As soon as the produced force reached this reference mark, measurement of IET (using a stopwatch) ensued. Children were encouraged during the measurement of the IET while feedback regarding the elapsed time was regularly given every 15 s. IET test stopped when children were unable to maintain the contraction intensity near the visual reference mark (IET test ended when a 5% drop of the produced force dropped occurred).

Because endurance time (in seconds) was different among children, we normalized time (in %) to allow comparison between groups. In Fig. 1, the time between 2 data points respresents approximately 4 s for both groups. During IET, VO_2 uptake (normalized

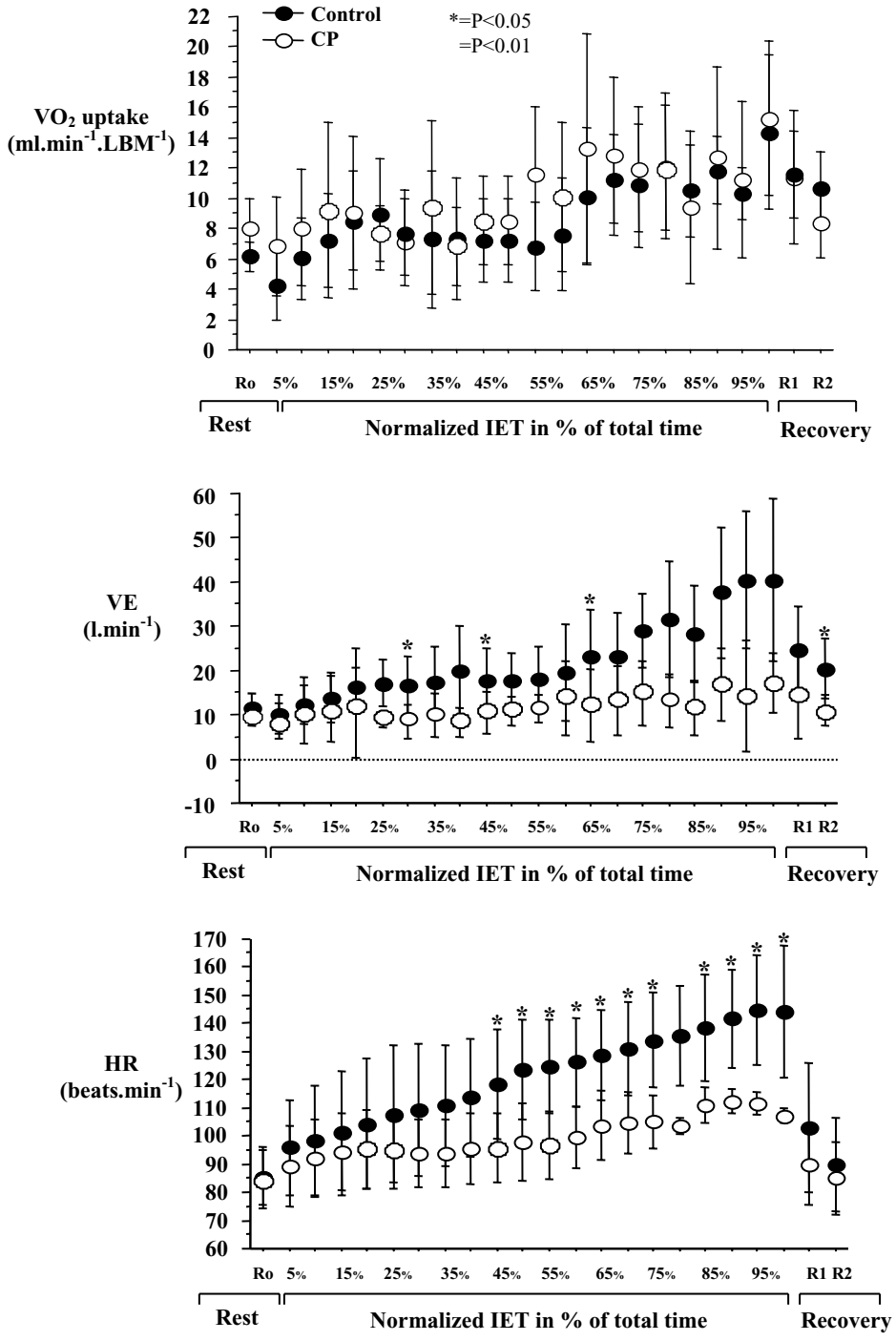

Fig. 1. Cardiorespiratory kinetics during isometric endurance time (IET) in children developing typically (control) and in cerebral palsy (CP) children . Mean values ± SD. Isometric endurance time (IET) is normalized in % of total time in each group. NB: Average isometric force output (50% of maximal force) during IET is 24 Nm forCP children and 44 Nm for children developing typically.

with lean body masss: $ml.min^{-1}.LBM^{-1}$), ventilation (VE: $l.min^{-1}$) and heart rate (HR: $beats.min^{-1}$) were measured and represented in Fig. 1. Because VO_2 uptake is dependant on lean body mass, VO_2 uptake was normalized with LBM to allow a more realistic comparison between the groups (CP children had a leaner

body mass in our study). The force output was kept constant in each group (50% of the MVIF), however we expected an increase of VO_2, VE and HR during the progressive muscle fatigue installation.

2.5. Statistical methods

Anthropometric, skeletal muscle and CPET data at peak effort were compared using a one-way (group) analysis of variance (ANOVA). A post hoc test (Games and Howell's test) was used to localize the differences. Cardiorespiratory data evolution during IET between control and CP children were compared and analyzed using an analysis of variance with repeated measures (ANOVA, by Statview version 5.0 (SAS Institute Inc, Cary, USA). The relationship between peak VO_2 and skeletal muscle function data was performed using regression analyses and Pearson correlation coefficients. All values are expressed as mean $\pm$ SD unless otherwise indicated. A P-value < 0.05 was considered statistically significant.

3. Results

3.1. Skeletal muscle data

Cerebral palsy children exercised at an absolute lower 50% isometric torque compared to AN children (50% of MVIF: 44 ± 11 vs. 24 ± 18 Nm, $P < 0.05$) but their level of muscle endurance was not different after normalization with muscle volume (liter) and exercise time (seconds) (0.13 ± 0.51 vs. 0.21 ± 0.10 Nm.l^{-1}.sec^{-1}, $P = 0.08$).

3.2. Cardiorespiratory data evolution during isometric endurance time

No inter-group differences were found for VO_2 uptake during IET ($P = 0.34$). CP children with CP had lower VE and HR compared to healthy control children ($P < 0.001$) during IET. Cardiorespiratory data evolution assessed during IET in CP children and AN children are presented in Fig. 1.

3.3. Relationship between peak VO_2 and skeletal muscle force and endurance

Peak VO_2 value (normalized with lean body mass) on ergocyle was 31.27 ± 12.04 ml.min^{-1}.LBM^{-1} in CP children. In this group peak VO_2 correlated with MVIF (R = 0.77, P <0.05), and isometric endurance time (R = 0.68, $P < 0.05$).

4. Discussion

The precise causes of exercise intolerance remain poorly documented in CP children [24]. Investigating cardiorespiratory function during exercise has the potential to enhance our understanding of the underlying mechanisms of poor exercise tolerance in this population. Moreover, an increasing number of clinicians use peak VO_2 measurement to evaluate the efficacy of interventions and rehabilitation programs in children with CP [24]. Poor functional capacity is an important clinical problem in such individuals, having the potential to affect quality of life and the ability to carry out activities of daily living.

Regarding the performance of CP children, the principal findings of our study are the following: 1) during quadriceps muscle endurance testing, they have a higher O_2 uptake despite their significantly lower absolute isometric force output. 2) Quadriceps strength and endurance are correlated with peak VO_2 3) they are not impaired in terms of quadriceps endurance when values are normalized by muscle volume and exercise time. Nevertheless, the muscular and cardiorespiratory responses identified in this group suggest that reduced peak aerobic capacity is more related to a peripheral motor limitation (correlated with reduced strength and endurance). In addition, for any given activity level, the O_2 demand during isometric exercise was higher in the affected children. It has been documented that higher coactivation and/or co-contraction was responsible of more important working muscle mass in CP children [24]. Concomitantly, compensatory or adaptive mechanisms appear to increase O_2 demand during local muscular isometric exercise (see below).

Our study showed that CP children have preserved muscle endurance associated with higher O_2 demand (uptake) during quadriceps isometric contraction despite a lower absolute force output (24 vs. 44 Nm). During isometric contraction which is maintained until exhaustion, aerobic metabolism is an important contributor to endurance time via the oxidative phosphorylation and the metabolic economy [20]. That is why cardiorespiratory gas exchanges (and particularly VO_2 uptake) were measured breath by breath during IET with the Cosmed K4b^2 gas analyzer to measure the O_2 uptake during contraction. Differences in skeletal muscle endurance observed between the groups has been shown in previous investigations [2,5,9,10,19]. However, the difference in muscle mass must be taken into account when comparing strength and endurance with children developing typically. Engsberg et al. [10] and

Ross et al. [19] have shown that CP children were significantly weaker than their peers without disability. In fact, coactivation of antagonist muscles and muscle cocotraction are potential mechanisms that could affect muscle endurance and higher cost of contraction during isometric contractions [2,5,9]. Increased stretch reflexes and muscle tone can limit the clearance of muscle metabolites especially during isometric contraction, reducing local muscle efficiency in CP children. Additionally, an earlier recruitment of fast fibers during isometric contraction could contribute to earlier muscle fatigue. Damiano et al. [6] indicated that in CP children, skeletal muscle fatigue might arise from impaired central nervous command and from abnormalities of the neuromuscular junction

In the present study, peak VO_2 correlated with muscle force and endurance in CP children. Reduced muscle strength is a commonly reported finding in this group of children when absolute values are used [2,5–7, 9,10,17] and CP affects strength development by reducing muscle mass [2,9]. Also, in these children, atrophy of type II fibers caused by upper motor neuron lesions, resulting in a greater proportion of type I fibers has been suggested [24]. Reduced non-normalized strength would appear to be the result of a smaller muscle mass in our study [9]. Additionally, compromised reciprocal synchronism in muscle groups could influence strength during voluntary contraction, thus limiting movement and reducing the ability to exert high degrees of muscular force [9,11,27]. Cerebral palsy children have been shown to possess a lower peak VO_2 compared to control children [11,14,17,24,25]. Previous available peak VO_2 data in children with CP were obtained during exercise using a cycloergometer [14], arm ergometry [25] or treadmill [11,24]. Factors that may have influenced VO_2 values include muscle weakness as shown by our results but also lower breathing efficiency and chest wall abnormalities that were present in 4 children in our study. High muscle tone, which reduces venous return and inhibits muscle lactate clearance during exercise, and thereby increases muscle fatigue, is also a possible mechanism contributing to lower VO_2 max values [11].

There are several limitations inherent to this study. First, skeletal muscle strength and endurance were assessed on the limb affected by spasticity; future studies will require testing of both limbs. Second, our sample size was small and mainly composed of boys with mixed CP. Future work will require larger sample sizes, a greater proportion of girls, and the inclusion of specific types of CP (diplegic vs. hemiplegic) in order to generalize the results and to identify any differences between sexes. Third, the work rate (power) developed by children with CP during exercise could not be calculated because children were not able to maintain a regular cadence at 60 rpm. This phenomenon which is already documented [17,25], means that work rates have to be interpreted as relative values compared to able-bodied peers [25].

In conclusion, our study indicates that muscle endurance of CP children was not different from children developing typically. This higher O_2 uptake is probably due to coactivation and/or cocontraction during exhaustive quadriceps isometric contraction. Because daily physical activities require a higher energetic cost for CP children (as suggested by our data), resistance training may also be recommended (in addition to aerobic training) to improve skeletal muscle strength and endurance and favorably improve their functional capacity [7].

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