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The Locomotory Index in diplegic and hemiplegic children: the effects of age and speed on the energy cost of walking

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Background. **The energy cost of locomotion (C) is a useful tool for quantifying the level of walking disability in the clinical evaluation of patients with cerebral palsy (CP). In addition to clinical condition, also age and velocity (v) can influence C, a fact that is often overlooked.**
Aim. **To show: i) that C differs in the clinical subtypes of CP (hemiplegia or diplegia) and ii) that C should be measured at comparable speeds in CP patients and controls (of the same age).**
Design. **Controlled study.**
Setting. **Pediatric Rehabilitation Unit of "E. Medea" Scientific Institute (Conegliano, TV); Exercise Physiology Lab of University of Verona.**
Population. **Forty-three CP children (32 diplegic: Dg; 11 hemiplegic: Hg) and 20 healthy children (Cg) with an age range of 4-14 years.**
Methods. **C was measured as the ratio of net oxygen uptake to walking speed (at v from 1 to 6 km·h^{-1}). The Locomotory index (LI) was calculated as the ratio of C in Dg/Hg and Cg (of the same age) at the same speed.**
Results. **C decreases with increasing speed in all groups but evolves differently in Hg and Dg: in the former C decreases by increasing age, becoming similar to that of Cg at 12-14 years; in the latter C does not change as a function of age being always larger than in Cg.**
Conclusion and clinical rehabilitation impact. **Our data highlight the reduction in C with increasing speed and suggest a better prognosis of locomotion for Hg compared to Dg.**

KEY WORDS: Cerebral palsy - Gait - Oxygen consumption - Energy metabolism - Locomotion.

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Cerebral palsy (CP) is a permanent disorder of movement development due to a non-progressive lesion of the immature brain occurring early in childhood or during the fetal period.[1] The heterogeneous alterations of the central nervous system produce the characteristics features that influence CP patients' quality of life, reducing their daily life activities, their motor independence and affecting their walking pattern. Indeed, the energy cost of locomotion (C), *i.e.* the amount of metabolic energy spent to cover a unit of distance, has been shown to be greater in this population compared to normally-developing peers and this is often combined with decreased stride length and walking velocity [2-10]. The low economy of walking in CP patients has been partially related to clinical symptoms, such as muscle's weakness and abnormal motor control,[11] but also to the presence of spasticity and co-contractions [9] and to a large mechanical work due to an increase of the vertical displacement of the body centre of mass.[10]

Even if the assessment of C is becoming a common clinical procedure for quantifying the economy of locomotion and for evaluating the effect of differ-

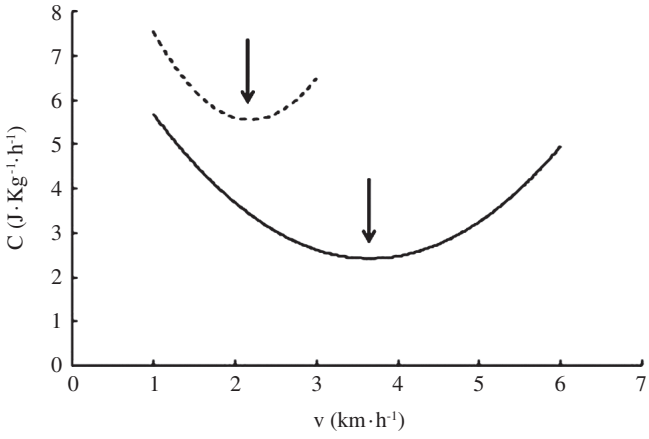


Figure 1.—Hypothetical relationship between energy cost (C) and walking speed (v) in healthy children (continuous line) and in CP children (dotted line);the arrows indicate the (hypothetical) self selected speed (sss) in the two cases. Were the values of C compared at the sss the difference in C would be overestimated, this difference being much lower when calculated at the same "absolute speed". The Locomotor Index proposed in this study indeed determines the C ratio (between CP patients and healthy children of the same age) at the very same speed. This Figure also indicates that more information about the walking disability could be obtained by measuring C at several speeds, i. e. by comparing the C vs. v relationship in CP children with that of an appropriate control group matched for age (instead by comparing one single couple of C values). See Text for details.

ent rehabilitation treatments, the fact that the energy cost of walking depends on factors additional to those directly connected with the physiopathological status of cerebral palsy (*e.g.* on speed and age) is not always taken in due consideration.

First, the C of walking is velocity-dependent: the relationship between C and speed is commonly described by means of a "U shaped curve" that, in healthy subjects, attains its minimum at the self selected speed.[12] Despite the velocity-dependent characteristics of C, most of the studies on the economy of pathological gaits assess C only at the self-selected speed.[1-4, 6, 10] However, this speed may be not necessarily the same in healthy subjects and in patients with locomotor disorders. As a consequence, it's not possible to exclude that the observed differences in C (*e.g.* in comparison to healthy subjects) may be simply ascribed to differences in speed (Figure 1) rather than to the effect of treatment or to the degree of locomotor deficit, thus leading to incorrect or inaccurate conclusions about the walking economy in patients with gait disorders.[13-15]

The second factor that can be expected to affect the economy of locomotion in CP patients is the age of the subjects (and the related differences in the anthropometric characteristics): the older the age, the longer is the leg length and of consequence the larger is the self selected speed.[16-19, 20]

To avoid that these two confounding factors (age and speed) could influence the correct determination of the impairement of the locomotory function, Fusi and co-workers[13] proposed to calculate the Locomotory Index, which is the ratio of the energy cost between a certain group (*e.g.* CP children) and a control group (of the same age) evaluated at the very same speed. This index was utilized by these authors to follow the changes in C before and after total knee and hip arthroplasty and proved to be a useful quantitative tool to evaluate ambulatory disability in patients. By means of this calculation is therefore possible to attribute the differences in C (between patients and healthy subjects) to the locomotor disorder itself and not to differences in speed or age (the two "confounding" factors).

In this study, we assessed the energy expenditure of locomotion (C) in a population of CP children of different ages and different clinical subtypes (diplegia or hemiplegia) as well as in a control group of healthy children at several comparable speeds, so to calculate the Locomotory Index. We aimed to show: 1) that C depends on the clinical forms of cerebral palsy (hemiplegia or diplegia); 2) that C is affected by the age of the subjects; and 3) that a complete assessment of the locomotion economy always requires the description of the C vs. v relationship, also in pathological populations.

Materials and methods

Participants

Forty-three children with an established diagnosis of CP (32 diplegia: Dg, and 11 hemiplegia: Hg) were recruited to volunteer for the study. They were patients under the care of the Pediatric Rehabilitation Unit of the "E. Medea" Scientific Institute. Inclusion criteria were: 1) clinical diagnosis of CP (spastic hemiplegia or diplegia), confirmed by neuroimaging; 2) age between 4 and 14 years; 3) independent ambulation with or without orthoses.

Exclusion criteria were: 1) inability to deambulate; 2) history of cardiovascular diseases; 3) previous treatment for spasticity in the 6 months before the evaluation; iv) previous orthopedic or neurological surgery.

Twenty able-body children (Cg) of comparable age were recruited as a control group. Within each group, three sub-groups on the basis of age were identified: A1 from 4 to 7 years old, A2 from 8 to 11 years old, A3 from 12 to 14 years old. No hemiplegic patients from 8 to 11 years old were recruited; hence, for these subjects only the A1 and A3 subgroups were formed.

All CP children were assessed by an experienced physical therapist according to the Gross Motor Function Measure (GMFM); this score ranges between 0 and 264 (the larger the score the larger the impairment). [21]

Ethical approval

The experiments were conducted with the understanding and the consent of the participants and of their parents. The protocol, the experimental design and the methods applied in the study were approved by the Institutional review board.

Experimental protocol

Breath-by-breath oxygen uptake ($\dot{V}O_2$) was measured by using a portable metabolimeter (Cosmed K4b², Rome, Italy). The equipment was calibrated according to the constructor's recommendations. The validity and reliability of the measurements obtained with this method are reported in several papers to which the reader is referred for further details.[22]

Each evaluation started with the subject standing at rest for 3 minutes on the treadmill. Subsequently, the children were asked to walk on a motor-driven treadmill at speeds that they were able to maintain safely and that ranged from 1 to 6 $km \cdot h^{-1}$. The speed was increased continuously by 1 $km \cdot h^{-1}$ every 3-4 minutes with no pauses in-between. Steady state conditions were checked by continuous online monitoring of the cardiopulmonary parameters and data corresponding to the last minute of each step (at steady state) were considered (and averaged) for further analysis. Healthy children practiced treadmill walking during a habituation session preceding the experiments and CP children were well acquainted with this procedure since they use treadmill walking during their physiotherapy sessions.

The experiments were performed in two different venues: Cg at the Exercise Physiology Lab of the University of Verona (Italy), whereas CP children were tested at the "F. Fabbri" Motion Analysis Laboratory of the "E. Medea" Scientific Institute of Conegliano (Italy). Two different treadmills were used (for healthy children: Saturn 200/300, H/P/Cosmos, Traunstain, Germany; for CP children: Galaxy MTC Climb, Runner, Italy); the accuracy of the velocity measurements was checked in both cases before each experimental run. All CP children were instructed to hold on the rails of the treadmill whereas healthy children always walked alone. There are evidences that walking on treadmill by holding handrails can help to maintain the balance [23] so that cardiovascular and metabolic responses may be attenuated.[24] However, we decided to allow CP children to hold on the rails for safety reasons.

During these experiments, O_2 uptake ($\dot{V}O_2$, $L \cdot min^{-1}$), CO_2 production ($\dot{V}CO_2$, $L \cdot min^{-1}$), heart rate (HR, bpm), pulmonary ventilation ($\dot{V}_E$, $l \cdot min^{-1}$) and respiratory exchange ratio (RER) were measured at steady state.

Data analysis

The cardiopulmonary data at rest and during walking were calculated as the average of the breath-by-breath values assessed in the last 2 minutes of rest and/or of exercise at each speed.

The energy cost of walking on level ground (C) was determined from the ratio of net $\dot{V}O_2$ ($\dot{V}O_{2net} = \dot{V}O_2 - \dot{V}O_{2rest}$ standing, $ml \cdot min^{-1} \cdot kg^{-1}$) to the speed of progression (v, $m \cdot min^{-1}$). Thereafter, C was expressed in Joules per meters per kilogram body mass on the assumption that 1 ml of O_2 consumed in the human body yields 20.9 J (a fact that is strictly true for RQ=0.93)

The ventilatory equivalent of oxygen at steady state was calculated as the ratio of $\dot{V}_E$ to $\dot{V}O_2$.

Finally, the locomotory index (LI) was calculated as proposed by Fusi and coworkers[13] for each CP children as the ratio of the energy cost in CP and the value of C in healthy children (of the same age) measured at the same speed.

TABLE I.—*Participants' characteristics.*

		Control group (Cg) mean (SD)	Diplegic group (Dg) mean (SD)	Hemiplegic group (Hg) mean (SD)
A1	Age (y)	6.6 (0.6)	5.3 (1.3)	5.3 (1.3)
(4-7 y)	n	7	18	6
	BM (kg)	28.0 (6.2)	20.6 (3.7)	23.2 (3.3)
	BH (m)	1.25 (0.08)	1.13 (0.08)	1.14 (0.06)
	M/F	5 M/2 F	10 M/8 F	3 M/3 F
	GMFM		230	250
A2	Age (y)	9.8 (1.1)	9.3 (1)	
(8-11 y)	n	8	8	
	BM (kg)	38.7 (8.0)	32.37 (9.05)	
	BH (m)	1.42 (0.08)	1.37 (0.11)	
	M/F	5 M/3 F	4 M/4 F	
	GMFM		217	
A3	Age (y)	12.5 (0.8)	12.6 (0.7)	13.7 (1.9)
(12-14 y)	n	5	6	4
	BM (kg)	52.8 (24.0)	44.8 (11.5)	53.80 (23.00)
	BH (m)	1.58 (0.13)	1.51 (0.11)	1.58 (0.15)
	M/F	2 M/3 F	5 M/1 F	3 M/1 F
	GMFM		213	252
All	Age (y)	9.3 (2.5)	7.5 (3.5)	8.3 (4)
	n	20	32	11
	BM (kg)	38.5 (15.9)	28.33 (11.9)	29.67 (10.6)
	BH (m)	1.40 (0.16)	1.24 (0.17)	1.29 (0.20)
	M/F	12 M/8 F	19 M/13 F	6 M/4 F
	GMFM		225	251

BM: body mass; BH: body height; n: number of subjects; M/F (males/females); GMFM: gross motor function measure.

Statistical analysis

The SPSS software (16.00 version) was used for the statistical analysis. All data violated the assumption of normal distribution (Shapiro-Wilk test). Hence, the natural logarithm transformation was applied. Anthropometric data and the GMFM scores, after the transformation, were analyzed by means of a two way ANOVA ($P<0.05$) where the two factors were "group" (Dg; Hg; Cg) and "age" (A1; A2; A3).

Data concerning the following physiological variables ($C,\dot{V}O_2 \cdot Kg^{-1}$, HR, LI) were analyzed, after transformation, by means of a Mix Models Analysis: velocity was considered as repeated factor, and group and age as fixed factors. For the post-hoc test the Bonferroni adjustment was used.

The data concerning the following variables (velocity, RER, $\dot{V}_E$, $\dot{V}_E \cdot \dot{V}O_2^{-1}$) didn't satisfy the assumption of normal distribution even after transformation. Therefore the original data were analyzed by a Kruskal-Wallis test and Dunn's post-hoc test.

Results

Anthropometric data

In the three groups (Dg, Hg and Cg) body mass (BM) and body height (BH) significantly increased with age (Table I). BM and BH were significantly different between Cg and Dg in the groups A1 and A2 ($P<0.05$). No differences in BH and BM were observed between Hg and Cg at all ages. The GMFM score was significantly larger in the hemiplegic with respect to the diplegic group ($P<0.05$).

Cardiorespiratory data

In Table II the average values (±SD) of HR, RER, $\dot{V}_E, \dot{V}O_2 \cdot Kg^{-1}, \dot{V}_E \cdot, \dot{V}O_2^{-1}$ measured in the three groups of subjects are reported for the three age sub-groups and at each tested speed. In Cg and Hg the highest tested speed was 6 $km \cdot h^{-1}$ whereas in Dg the maximal tested speed was 4 $km \cdot h^{-1}$. Data reported in Table II indicates that walking at these speeds corre-

TABLE II.—*Average values and standard deviation of heart rate (HR), respiratory exchange ratio (RER), expired ventilation ($\dot{V}_E$; L·min^{-1}), oxygen uptake ($\dot{V}O_2$·kg^{-1};ml·min^{-1}·kg^{-1}), ventilatory equivalent ($\dot{V}_E\dot{V}O_2^{-1}$) in hemiplegic (Hg), diplegic (Dg) and healthy children (Cg), for the three age groups (A1: 4-7 years; A2: 8-11 years; A3: 12-14 years) at the investigated walking speeds (1-6 km·h^{-1}).*

Hemiplegic group

		Condition-Speed						
		Standing	1 Km·h^{-1}	2 Km·h^{-1}	3 Km·h^{-1}	4 Km·h^{-1}	5 Km·h^{-1}	6 Km·h^{-1}
	n. subjects	6	6	6	5	3		
	HR	97 (15)	117 (12)	126 (11)	135 (12)	144 (21)		
A1	RER	0.95 (0.37)	0.84 (0.24)	0.86 (0.24)	0.85 (0.24)	1.05 (0.10)		
	$\dot{V}_E$	7.39 (1.20)	11.86 (2.63)	14.68 (2.69)	17.71 (3.32)	20.71 (7.46)		
	$\dot{V}O_2$·kg^{-1}	9.75 (1.95)	17.72 (3.80)	21.56 (4.84)	24.45 (5.24)	25.37 (9.92)		
	$\dot{V}_E\dot{V}O_2^{-1}$	33.5 (5)	29.3 (3)	30.1 (3)	31.3 (2)	33 (1)		
	n. subjects	4	4	4	4	4	3	2
	HR	85 (11)	91 (16)	103 (15)	116 (14)	128 (13)	138 (13)	144 (6)
A3	RER	0.90 (0.12)	0.91 (0.08)	0.95 (0.08)	1.00 (0.10)	1.04 (0.12)	1.15 (0.11)	1.19 (0.17)
	$\dot{V}_E$	8.01 (1.31)	8.51 (1.25)	15.02 (1.22)	21.53 (2.20)	28.04 (3.42)	36.54 (3.02)	41.68 (1.23)
	$\dot{V}O_2$·kg^{-1}	5.07 (1.03)	10.68 (3.09)	13.48 (3.35)	15.05 (2.22)	16.97 (1.14)	19.67 (0.94)	23.78 (2.32)
	$\dot{V}_E\dot{V}O_2^{-1}$	37 (7)	19.5 (6)	26.5 (5)	33.2 (4)	38.5 (7)	44.7 (2)	45.1 (1)

Diplegic group

		Condition-Speed				
		Standing	1 Km·h^{-1}	2 Km·h^{-1}	3 Km·h^{-1}	4 Km·h^{-1}
	n. subjects	18	18	18	11	3
	HR	104 (10)	120 (13)	136 (18)	138 (8)	114 (25)
	RER	0.82 (0.26)	0.75 (0.23)	0.81 (0.25)	0.80 (0.20)	0.88 (0.36)
A1	$\dot{V}_E$	8.59 (2.06)	11.92 (2.49)	16.25 (4.15)	17.72 (3.30)	12.38 (6.07)
	$\dot{V}O_2$·kg^{-1}	9.55 (2.86)	16.00 (4.56)	19.86 (5.38)	21.92 (4.95)	12.50 (8.44)
	$\dot{V}_E\dot{V}O_2^{-1}$	49.2 (25)	38.1 (11)	41.8 (13)	39.6 (11)	39.5 (13)
	n. subjects	8	8	7	5	2
	HR	102 (9)	125 (16)	138 (16)	141 (29)	111 (25)
	RER	0.80 (0.14)	0.82 (0.16)	0.85 (0.18)	0.84 (0.17)	0.69 (0.12)
A2	$\dot{V}_E$	8.63 (1.59)	14.89 (3.71)	21.61 (6.32)	22.65 (10.78)	13.07 (8.89)
	$\dot{V}O_2$·kg^{-1}	7.74 (2.30)	15.07 (5.08)	19.13 (5.29)	18.11 (8.39)	13.29 (9.72)
	$\dot{V}_E\dot{V}O_2^{-1}$	36.9 (7)	32.8 (7)	34.8 (10)	41.1 (11)	34.9 (5)
	n. subjects	6	6	5	3	2
	HR	103 (21)	127 (29)	138 (32)	127 (40)	118 (22)
	RER	0.73 (0.18)	0.71 (0.16)	0.73 (0.19)	0.83 (0.20)	0.73 (0.04)
A3	$\dot{V}_E$	10.74 (2.86)	19.52 (4.74)	26.22 (5.84)	28.05 (10.61)	26.89 (6.77)
	$\dot{V}O_2$·kg^{-1}	7.01 (2.44)	14.46 (4.22)	17.71 (4.15)	19.65 (3.65)	24.33 (3.12)
	$\dot{V}_E\dot{V}O_2^{-1}$	40.1 (11)	32 (8)	32.1 (5)	35.9 (3)	34.8 (8)

(Continued)

Healthy group

		Condition-Speed						
		Standing	1 Km·h^{-1}	2 Km·h^{-1}	3 Km·h^{-1}	4 Km·h^{-1}	5 Km·h^{-1}	6 Km·h^{-1}
	n. subjects	7	7	7	7	7	6	3
	HR	101 (8)	104 (11)	107 (9)	111 (12)	120 (11)	129 (10)	138 (7)
A1	RER	0.77 (0.1)	0.78 (0.09)	0.79 (0.09)	0.77 (0.14)	0.82 (0.08)	0.81 (0.10)	0.92 (0.17)
	$\dot{V}_E$	7.6 (1.08)	11.47 (2.83)	11.62 (2.5)	13.46 (3.61)	17 (3.38)	20.95 (4.51)	27.39 (1.07)
	$\dot{V}O_2$·kg^{-1}	6.08 (0.82)	10.41 (1.26)	11.97 (1.50)	14.18 (1.75)	18.09 (1.88)	23.62 (3.71)	30.64 (0.63)
	$\dot{V}_E\dot{V}O_2^{-1}$	45.5 (10)	40.1 (9)	35.5 (6)	33.9 (4)	34.1 (5)	32.6 (3)	33.9 (1)
	n. subjects	8	8	8	8	8	8	7
	HR	95 (13)	102 (14)	108 (13)	113 (116)	123 (15)	132 (18)	149 (23)
A2	RER	0.89 (0.21)	0.74 (0.06)	0.78 (0.08)	0.79 (0.06)	0.78 (0.06)	0.83 (0.07)	0.87 (0.07)
	$\dot{V}_E$	8.81 (1.98)	12.73 (2.95)	15.09 (3.49)	16.98 (3.6)	20.27 (4.57)	25.7 (5.33)	32.98 (7.07)
	$\dot{V}O_2$·kg^{-1}	5.96 (1.65)	10.42 (1.2)	12.69 (2.34)	15.14 (1.52)	18.47 (2.34)	23.78 (4.57)	28.21 (4.95)
	$\dot{V}_E\dot{V}O_2^{-1}$	40.3 (9)	31.7 (3)	31.3 (5)	29.3 (3)	28.7 (4)	28.8 (6)	29.8 (5)
	n. subjects	5	5	5	5	5	5	5
	HR	112 (11)	110 (12)	118 (13)	124 (13)	132 (15)	146 (17)	161 (19)
A3	RER	0.83 (0.09)	0.8 (0.06)	0.82 (0.02)	0.85 (0.04)	0.88 (0.05)	0.89 (0.05)	0.92 (0.11)
	$\dot{V}_E$	9.21 (2.55)	14.96 (3.08)	16.24 (4.25)	18.9 (5.21)	21.98 (6.27)	28.18 (7.47)	35.79 (9.18)
	$\dot{V}O_2$·kg^{-1}	4.9 (0.73)	10.13 (1.33)	11.12 (0.6)	13.23 (0.67)	15.13 (1.6)	19.36 (0.62)	23.5 (0.68)
	$\dot{V}_E\dot{V}O_2^{-1}$	37.5 (3)	30.1 (5)	29.3 (5)	28.5 (4)	29.1 (5)	29.1 (4)	30.6 (5)

sponded to a moderate intensity exercise performed in aerobic conditions, in all groups.

As far as the HR values are concerned, both the fixed effect of the factor "group" and the effect of the interaction between "group" and "age" were found to be significant ($F_{2,168.448}$=10.260; $F_{4,172.795}$=10.260 P<0.001). The Bonferroni test showed that in Dg HR was significantly larger than in Cg (P<0.001), whereas it was identical in Hg and Cg (P=1.000).

Considering the values of RER, these were significantly different between CP (Hg and Dg) and Cg (H=39,532; 2d.f.; P<0.001); the Dunn's test indicated that RER values were significantly larger in Hg than in Cg (P<0.05).

No differences in $\dot{V}_E$ were found between CP children and Cg (H=5,236; 2d.f.; P=0.073). For the $\dot{V}O_2$·Kg^{-1} data, the effect of the fixed factor "group" was found to be not significant ($F_{2,182.094}$=0.531; P=0.589), whereas the effect of the interaction between "group" and "age" was significant ($F_{4,28.792}$=28.792; P<0.001). The Bonferroni test showed that in Dg $\dot{V}O_2$·Kg^{-1} was significantly higher than in Cg (P<0.02) at each age. In Hg $\dot{V}O_2$·Kg^{-1} was significantly larger than in Cg only at A1 (P<0.001), whereas no differences are observed for the elder group of patients (A3; P=1.000).

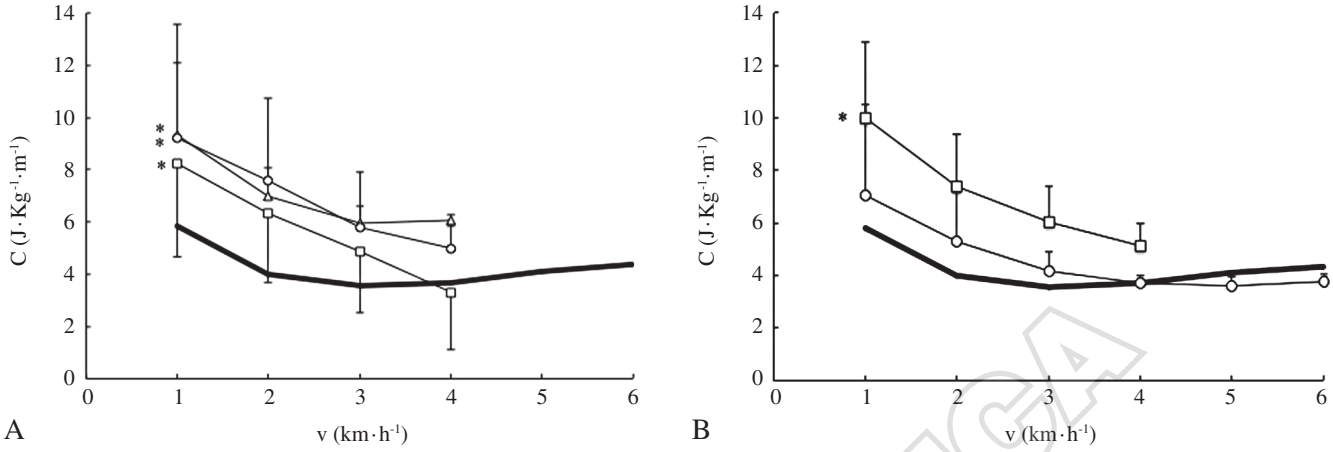
The $\dot{V}_E$·$\dot{V}O_2^{-1}$ ratio is significantly different in CP than in Cg (H=29,017; 2d.f.; P<0.001) and the post hoc test indicates that diplegic group has a significant larger $\dot{V}_E$·$\dot{V}O_2^{-1}$ compared to Cg (P<0.05).

Energy cost of walking

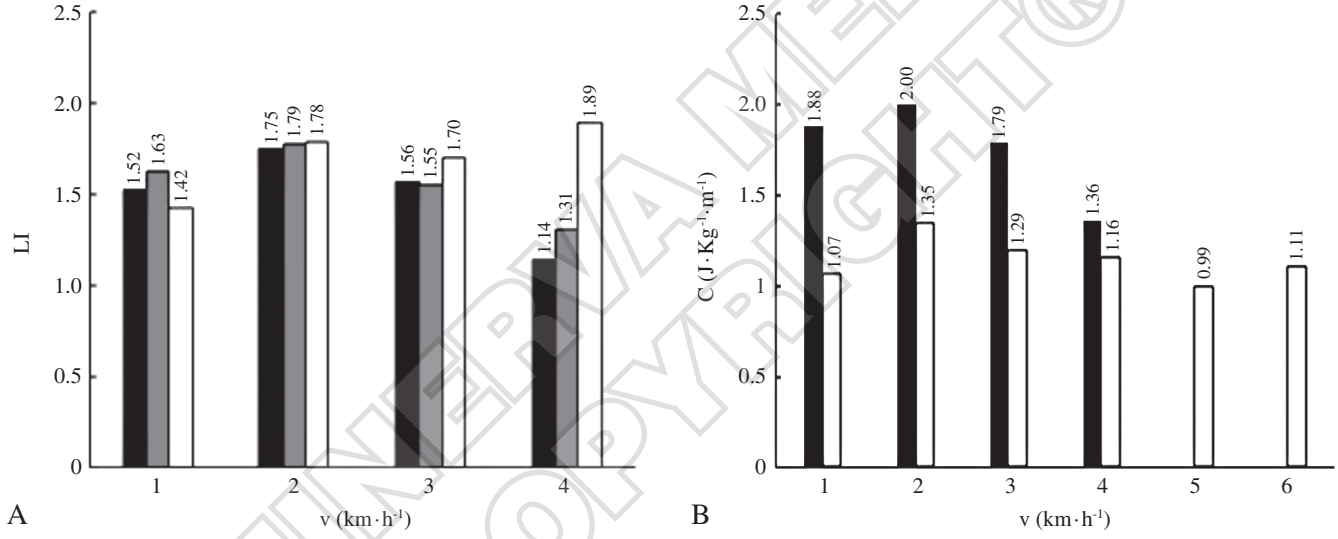
In Figure 2 the values of C are reported as a function of speed for all groups and at all ages. The data referring to healthy children were averaged regardless of the age since, in this group, C was not affected by growth (Bonferroni test, A1 vs. A2 P=0.832; A3 *vs.* A2 P=1.000, A3 *vs.* A1 P=0.523).

The function interpolating the C data as a function of speed in the control group ($C = 0.2473 \cdot v^2 - 1.9309 \cdot v + 7.2632$, R^2=0.8789) presents the typical "U shape pattern" and shows a minimum at 3.9 km·h^{-1}. Conversely, in hemiplegic and diplegic children, C tends to decrease monotonically with speed being the lowest at the highest tested speed.

The mixed models analysis showed that the fixed factor "group" and the interaction between "group" and "age" had a significant effect on C ($F_{2,151.129}$=34.848; $F_{4,157.875}$=7.821; P<0.001). The pairwise comparison (Bonferroni test) showed that: 1)



Figures 2.—A, B) Energy cost of walking (C) as function of speed (v) in CP children (Figure 2a:diplegic children;Figure 2b:hemiplegic children) and in healthy children (black continuous line), as function of speed (v) at different stages of development (A1: squares, 4-7 yrs; A2: triangles, 8-11 yrs; A3: circles, 12-14 yrs). Asterisks indicate significant differences between CP children and healthy children (P<0.05).



Figures 3.—A, B) Locomotory Index (LI , the ratio of C between CP children and healthy children) in diplegic (A) and hemiplegic (B) children as a function of speed (v). A1:black bars, 4-7 yrs; A2: grey bars, 8-11 yrs; A3: white bars, 12-14 yrs).

in Dg, C was significantly larger than in Cg at all ages (P<0.001); 2) in Hg, C was significantly larger than in Cg only for the A1 group (P<0.001), whereas no differences in C are observed for the A1 and A3 groups (P=0.055).

In Figure 3, the ratio of the values of C as assessed in CP children to the value of C as assessed in healthy children (of the same age) at the same speed (*i.e.* the locomotory index: LI) is reported. A locomotory index greater than 1 was found in Dg at all ages, whereas in Hg LI was found to be larger than 1 only in the youngest group (A1). The mixed models analysis for the LI data showed that the effect of the fixed factor "group" and of the interaction between "group" and "age" were significant ($F_{2,159.247}$=44.107; $F_{4,150.356}$=8.439; P<0.001). The pairwise comparison (Bonferroni test) showed that in the youngest group (A1) LI was not different between hemiplegic and diplegic children (P=0.087), whereas in

the eldest group (A3) it was significant larger in Dg compared with Hg ($p < 0.001$).

Discussion

Data reported in this paper confirm and extend previous findings since they indicate that C is significantly larger in CP children than in healthy children.[2-10] In addition, we underline the need of specifying at which speed C is assessed because of the influence of the speed on C and we propose LI as a tool to quantify the walking disability. Finally we showed that C and LI depend not only on the speed, but also on the different clinical subtypes of cerebral palsy and on the age of the patients.

Effect of speed on energy cost of walking and locomotory index

In the present study, we proposed a protocol with incremental speeds that started from a very low intensity (1 $km \cdot h^{-1}$) that can be maintained even by the most impaired children for the time required to attain steady state. Since C of walking is velocity-dependent, this method allows a comparison of C at different speeds. This is of paramount importance when comparing patients with locomotor impairment and healthy controls.

For instance, Zamparo *et al.*[15] showed that with increasing speed in hemiplegic adults, the percentage difference between C of healthy subjects and patients decreases. Also in the present study (Figures 2A, B), the differences between the CP groups and the healthy group tend to decrease with increasing speed. The explanation of this evidence could consist in a decreased efficiency of the compensation mechanism actuated by CP children (see discussion below) as well as to a larger presence of co-contractions to maintain balance and dynamic posture at the slowest walking speeds.

From the methodological standpoint, these data underline the importance of defining the speed at which data of C are measured in order to understand whether the differences in C are due to the locomotor impairment per se or are simply an artefact deriving from a different position on the "U shaped curve" of the couple of data collected in CP patients and in healthy controls (Figure 1).

The utility of comparing C at more than one speed is also confirmed by the progressive drop of LI that attains its minimum at the highest speeds. This index requires the parallel assessment of C, at the very same speed, both in patients and in healthy subjects of the same age. Indeed, differences in the anthropometrical features (in subjects of different size or biological ages) may have a confounding impact on the assessment of C.

In this study no statistical differences in BH and BM were observed between Hg and Cg at all ages whereas BM and BH were significantly larger in Cg than in Dg: for A2 (8-11 years) the average difference in BH is of 3.5% (and hence negligible), for A1 (4-7 yrs) is of 9.5%. These data indicate that comparing C among children of the same age not always allows take into account the "size effect".

A "safer" procedure would be to compare C at equivalent size-dependent speed values by means of the Froude number. Froude number is defined as the ratio between the square of the speed and the product of "pendulum length" (the distance from the foot at ground contact to the centre of mass of the body) and acceleration of gravity ($v^2 \cdot L^{-1} \cdot g^{-1}$);[16-19] it thus represents a speed normalized for body size. It goes without saying that a proper measure of L could only be done by means of an accurate kinematic analysis since the "pendulum length" is not necessarily equal to the leg length, this particularly so in CP children for which the vertical displacement of the body centre of mass was found to differ from that of healthy children [10]. For this reason, we preferred not to normalize the speed based on BH (or leg length) data, and we just took into account the effects of age, a "size-dependent normalization" that indeed seems appropriate for all groups (but for the A1 Dg children).

Effects of clinical subtypes and age on energy cost

Although the present study is cross-sectional and although our conclusions are based on a rather small set of data, especially at the higher walking speeds, our findings suggest that: 1- hemiplegic children become as economic as controls as they grow up whereas 2- no major changes in C were observed in diplegic children as a function of age. Thus, data presented in this paper confirms previous findings [2, 25] that indicate that hemiplegic children have a better prognosis of walking economy with respect to the diplegic.

Damiano *et al.* showed that the improvement of walking economy with age in Hg children holds true even if they are classified at the same GMFCS level.[25] Also in the present study C does not seem to be related to the GMFM scores: even if the GMFM score doesn't change with age in Hg, in the younger Hg patients C is larger than in healthy children (and similar to that of Dg) whereas in the "older" Hg patients C is closer to that of healthy children (of the same age).

The better economy of walking observed in hemiplegic children has been attributed to the "higher functions" [25] maintained in the preserved limb. A similar explanation has been proposed by Tesio *et al.*[14] in adults with acquired hemiplegia. In these subjects, because of the asymmetry of the stride, the transformation of kinetic to potential energy at each stride (which minimizes external work, as indicated by Cavagna and Kaneko [12]) is still possible and energy expenditure is not greatly affected. Thus, the reason of the better economy of hemiplegic children is likely related to the possibility to actuate compensatory mechanisms, whereas in diplegic children the impairment of both lower limbs prevents this compensation.

In a recent study conducted by Kerr and *et al.*[3] on a large group of CP children with different clinical subtypes, a deterioration of C by augmenting age was described. In the reported investigation the changes of C were observed at a distance of about 3 years and the distinction in clinical subtypes was not accounted for. These two factors could explain the different findings obtained in that study compared with ours. This further underlines the importance of treating data of different clinical subtypes separately, but also indicate that a longer follow up (from 4 to 14 yrs in this study) allows to gather more information on the changes in C in developing children.

Cardiorespiratory data

The majority of the studies reported in the literature so far for CP patients during walking deal just with values of C. However, the evaluation of the metabolic, cardiovascular and ventilatory responses to exercise offers important cues on the exercise performed and, in our opinion, should always be included in the evaluation of the economy of locomotion of CP children.

In analogy with the observed differences in C, diplegic children showed a larger $\dot{V}O_2$ at the same speed than control subjects at all ages, whereas only the younger hemiplegics patients have a larger oxygen uptake than healthy peers. Also HR showed a similar trend, as it is significantly larger in Dg (but not in Hg) than in the Cg.$\dot{V}O_2$ and HR data therefore suggest that, when walking, CP children perform a more intense exercise with respect to the control group, and this is particularly so for the diplegic ones.

RER is significantly larger in Hg compared to Cg. Although hemiplegic children walk as fast and as economically as healthy mates (but for the youngest age), they are likely affected by a substantial aerobic detraining.[26-28] Therefore, the same absolute intensity would correspond to higher fractions of $\dot{V}O_{2max}$.

The $\dot{V}E \cdot \dot{V}O_2^{-1}$ ratio was larger in Dg compared to Cg, whereas no differences between Cg and Hg were demonstrated as far as this parameter is concerned. A larger $\dot{V}E \cdot \dot{V}O_2^{-1}$ in children with spastic paresis at $\dot{V}O_{2max}$ has been found also by Hoofwijk [26] and Lunberg.[28] This evidence is an indication of inefficient ventilation, which per se may be one of the explanations of the observed reduced $\dot{V}O_{2max}$ in this population.[26, 28] A larger ventilatory equivalent of oxygen, however, may be also a clue indicating that children with CP show a high relative ventilatory strain during walking, eliciting higher blood lactate concentrations (the relative metabolic acidosis to be compensated with a larger ventilation).

Conclusions

The results of this investigation indicate that C tends to diminish with increasing speed both in hemiplegic and diplegic children (as in healthy children of comparable age). This evidence underlines the need to compare C at different paired speed in CP patients and controls. In this study we proposes the utilisation of the locomotory index (the ratio between CP and Cg values of C, at the same speed and age) as a useful tool to quantify the degree of impairment of walking function in CP patients.

Our data suggest that the energy cost of locomotion evolves differently in children with spastic hemiplegia and diplegia. In the former C tends to decrease by increasing age, becoming similar to that of healthy children at 12-14 years of age; in the latter C does not change as a function of age being

always larger than in normal developing children, at comparable speeds.

In conclusion, the results of our study strongly support: 1) the indication of evaluating C at several speeds both in pathological and in control subjects; 2) the importance of distinguishing between different clinical subtypes and different ages when evaluating the economy of locomotion; 3) the use of the LI as a useful tool to quantify the diseconomy of locomotion in CP patients.

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