

The term diplegia should be enhanced. Part II: contribution to validation of the new rehabilitation oriented classification

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Recent proposals of classification for cerebral palsy (CP), mainly revised for epidemiological purposes, suggest to abandon the use of the term diplegia. Conversely, in this paper data are presented to support the proposal to maintain the distinction between spastic tetraplegia and diplegia, and to subdivide this latter according to four main clinical patterns of walking observable in these children. This proposal of classification was validated by testing a group of 467 subjects with CP, of whom 213 with diplegia and 115 with tetraplegia, consecutively admitted between January 2005 and December 2006 to two national reference centers for this disability. The results were compared with findings obtained by other methods of classifying gross and fine motor function and associated disorders. The subjects with tetraplegia strongly differ from those of diplegia, both for motor functions and for other disabilities. The four main walking patterns of spastic diplegia were easily recognizable and observers were able to assign most of the subjects to one form of the classification. Significant correlations between walking forms of diplegia and distribution of Gross Motor Function Classification System (GMFCS) levels were found. Some of the forms significantly differ also for fine motor and mental disability. These findings suggest that in clinical practice the category of diplegia not only can be kept as a separate form of CP, but it may be enhanced, through the identification

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of different subcategories of children, divided according to their walking patterns.

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Recently, a simplification of the categories to be included in the classification of cerebral palsy (CP) has been proposed, abandoning the term "diplegia".^{1,2} This proposal has advantages, when the aim of classification is mainly for epidemiology, but may also have significant limitations in the clinical practice, for prognosis and planning of rehabilitation programs. The group of Authors has suggested another classification approach to the spastic forms of CP, based on observation of motor and perceptual features,^{3,4} which maintains the distinction between tetraplegia and diplegia, and enhances it. According to this proposal children with the spastic forms of CP can be classified according to the architecture of three guiding functions: type of postural organization for tetraplegia, walking patterns for diplegia and manipulating class-

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es for hemiplegia, respectively. Four main clinical patterns (forms) of walking have been suggested in subjects with spastic diplegia.⁴ (Ferrari A, Alboresi S, Muzzini S, Pascale R, Perazza S, Cioni G. The terms diplegia should be enhanced (I): around the problem of classification of cerebral palsy. *Eur J Phys Rehabil Med* [submitted]). The motor and perceptual disorders are expressed in a different way in these forms, with a strong influence on the functional prognosis. A main feature of walking can be identified for each form, namely trunk forward leaning and toe balancing for the first form ("forward leaning propulsion"), knee flexion in mid stance for the second form ("tight skirt"), frontal trunk swinging for the third form ("tight rope walkers"), increasing talipes equinus at start of walking for the fourth form ("dare devils").

This paper reports the results obtained by applying systematically the classification of diplegia to an hospital-based series of subjects admitted to two reference centers for CP, with the following specific aims:

- 1) to confirm the difference in the clinical profile between diplegic and tetraplegic forms on a large groups of subjects with CP, studied in clinical settings;
- 2) to explore the possibility to use, in the same subjects, a new classification system of diplegia, based on the clinical assessment of children's walking;
- 3) to compare the results of this new classification with other methods, already available and standardized;
- 4) to explore whether the different forms of diplegia correspond to different profiles for the severity of gross and fine motor disability, and for other disabilities such as mental retardation, epilepsy, visual disorders;
- 5) to compare the clinical description of the walking patterns for the different forms, with those obtainable with quantitative methods, i.e. instrumental clinical gait analysis.

Subjects and methods

Application of various classification criteria

The subjects included in this study were all children, adolescent and young adults with CP admitted to the Unit of Children Rehabilitation of S. Maria Nuova Hospital (Reggio Emilia, Italy) or to the Department of Developmental Neurosciences of the IRCCS Stella

Maris (Pisa, Italy) from January 2005 to December 2006. Inclusion criteria were diagnosis of CP according to international definitions,⁵⁻⁷ age older than two years and available data concerning possible associated disorders (see below). An epidemiological chart to classify impairment was administered to each subject which included assessment according to Surveillance Cerebral Palsy in Europe (SCPE)², modified for further subdividing the spastic bilateral forms into diplegic and tetraplegic types according to Hagberg *et al.*^{8,9} and the above indicated classification of walking in diplegia.⁴ (Ferrari A, Alboresi S, Muzzini S, Pascale R, Perazza S, Cioni G. The terms diplegia should be enhanced (I): around the problem of classification of cerebral palsy. *Eur J Phys Rehabil Med* [submitted]).

Type of CP (spastic in its various type – see above, dyskinetic, ataxic), severity of lower and upper limb impairment and associated impairments (seizures, hearing and visual problems, mental retardation) were assessed for each patient. The global motor impairment was classified using the Gross Motor Function Classification System (GMFCS),¹⁰ while the ability to handle objects was evaluated by means of systems suitable to grade manual motor abilities, *i.e.* till early 2006 by the Bimanual Fine Motor Function scoring system (BFMF)¹¹ and subsequently by the Manual Ability Classification System (MACS).¹²

Subjects affected by spastic diplegia were also classified in the same epidemiological chart, according to their walking characteristics, into the suggested four forms of diplegia. The four form main features of gait have been described in a joined paper, (Ferrari A, Alboresi S, Muzzini S, Pascale R, Perazza S, Cioni G. The terms diplegia should be enhanced (I): around the problem of classification of cerebral palsy. *Eur J Phys Rehabil Med* [submitted]) whereas the Authors refer to other publications for a more complete description.⁴

Videos including samples of children's classification were discussed between Pisa and Reggio Emilia groups in periodical meetings, in order to maintain a good intergroup reliability among the doctors of the two groups responsible of the classification procedure.

Correlation between clinical forms and impairment was evaluated by statistical analysis. χ^2 test was used to determine statistical differences for incidence of epilepsy, mental and visual impairment between tetraplegia and diplegia and in each diplegic form. One way-analysis of variance (ANOVA) was conducted in order to establish significant differences

among groups (tetraplegia *versus* diplegia, one form of diplegia *versus* the others). Furthermore, multiple comparisons among each group were processed by Bonferroni *post hoc* range analysis. The level of statistical significance was set at $P < 0.05$.

Gait analysis

A retrospective analysis of gait data collected from children with CP at Movement Analysis Laboratory of Stella Maris Scientific Institute from March 2005 to September 2006 was carried out.

According to the standard protocol of the laboratory, the kinematic of pelvis and lower limbs during gait was acquired using an optoelectronic motion analysis system (Elite BTS, Italy) which is equipped with 8 infrared cameras (sampling rate 100 Hz). Ground reaction force was measured using one force platform (AMTI, USA). Surface ElectroMyoGraphy (SEMG) recordings were acquired by a BTS TeEMG (8 channel, sampling rate 1 000 Hz).

For each subject, 17 reflective markers were placed in correspondence of usual specific anatomical landmarks according to Davis's protocol.¹³ SEMG electrodes were placed on tibialis anterior, gastrocnemius medialis, rectus femoris, and semitendinosus muscles. SEMG electrode placement was based on SENIAM recommendations.¹⁴

The children with spastic diplegia walked barefoot at self-selected speed along a straight level rectilinear walkway approximately 10 m long. After the first settling-in period, a set of 15-20 trials was recorded for each child. Short periods of rest were granted between each trial. The first and final few trials were excluded from further analysis in order to avoid the bias that may be possibly associated with the environment novelty and the fatiguing effects. The subjects were asked to walk at their normal speed and at least five bare foot walking trials were recorded for each child. During each acquisition, synchronized-video recordings of the gait was also performed both on sagittal and frontal planes.

Results

Clinical classification of subjects with cerebral palsy

Final sample group consisted of 467 subjects, aged 2.0-21.7 years (mean 7.8 ± 4.1). Two hundred and sixty-two were male and 205 female.

The CP distribution shows a large prevalence of spastic CP, with a percentage of 93% (434 subjects) *versus* dyskinetic and ataxic CP, 5% (24 subjects) and 2% (9 subjects), respectively. The subjects with tetraplegia were 115 (25% of spastic CP); 64 males, 51 females; mean age 7.8 ± 4.5 (range 2.0-20.2).

The subjects with diplegia were 213 (46% of spastic CP); 115 were male, 98 female; mean age was 7.9 ± 3.9 (range 2.0-21.7 years). They were divided into the four forms according to the proposed walking classification. The IV form (42% of diplegic sample, 89 subjects) more often occurred than the other forms, followed by the I form (22%, 47 subjects) and finally by III and II ones (15% and 12%, 33 and 26 subjects respectively). Eighteen subjects were not classified because they were too young to walk, or they had just started to do it.

The results of the assessment for motor functions and associated impairments of subjects with tetraplegia and diplegia are described in Tables I, II respectively. The subjects with tetraplegia were more severely impaired than those with diplegia; impairment differences were statistically significant ($P < 0.001$). Generally, the sample of diplegic subjects had a normal mental development and did not have seizures.

The greater involvement in the visual and mental abilities was found in I and III forms; a significant correlation with severe mental impairment was found for form I (χ^2 , $P = 0.023$). Moreover, GMFCS and MACS/BFMF levels showed a decreasing severity of impairment from form I to IV. The GMFCS level distribution was statistically significant: I form and IV form had lower and higher values, respectively, II and III forms together had an intermediate position. BFMF level distribution showed a minor involvement in the IV form. However, two subjects of IV form presented considerable manual impairment (BFMF level V): one had a severe mental retardation ($IQ < 20$), whereas the other had no mental impairment but abnormal visual functions and an asymmetric distal involvement.

Gait analysis

Gait patterns of 64 children affected by spastic diplegia were recorded in the laboratory in the study period. Their age ranged from 4.1 to 16.8 years with a mean age of 11.2 years.

The description of the kinematic patterns of the different forms of diplegia, reported in the previous

TABLE I.—Main features of 213 subjects with diplegia and 113 with tetraplegia.

Type of cerebral palsy		Diplegia Subjects N (%)	Tetraplegia Subjects N (%)	Test	P
Mental development	Normal	105 (49%)	14 (12%)	χ^2	0.001
	Abnormal	89 (42%)	95 (82.5%)		
	Missing	19 (9%)	6 (5.5)		
Total ss		213	115		
Epilepsy	Present	22 (10.5)	54 (47%)	χ^2	0.001
	Absent	191 (89.5)	61 (53%)		
	Total ss	213	115		
Visual abilities	Normal	94 (44%)	24 (21%)	χ^2	0.001
	Abnormal	105 (49%)	89 (77.5%)		
	Missing	14 (7%)	2 (1.5%)		
Total ss		213	115		
GMFCS	Level 1	54 (25.5%)	2 (1.5%)	ANOVA. Bonferroni post hoc	0.001
	Level 2	51(24%)			
	Level 3	57(26.5%)	3 (2.5%)		
	Level 4	33 (15.5%)	34 (29.5%)		
	Level 5	4 (2%)	72 (62.5%)		
	Missing	14 (6.5%)	4 (3.5%)		
	Total ss	213	115		
BFMF	Level 1	41 (33.5%)		ANOVA. Bonferroni post hoc	0.001
	Level 2	68 (56%)	7 (10%)		
	Level 3	9 (7.5%)	11 (15%)		
	Level 4	2 (1.5%)	22 (30%)		
	Level 5	2 (1.5%)	32 (45%)		
	Total ss	122	72		
MACS	Level 1	18 (31%)		ANOVA. Bonferroni post hoc	0.001
	Level 2	22 (38%)			
	Level 3	15 (26%)	7 (17%)		
	Level 4	3 (5%)	13 (32%)		
	Level 5		21 (51%)		
	Total ss	58	41		

Ss: subjects; GMFCS: Gross Motor Function Classification System; BFMF: Bimanual Fine Motor Function; MACS: Manual Ability Classification System.

pages, was supported by the results of gait analysis. Representative samples of the four gait patterns are reported in Figures 1-4.

Figure 1 illustrates the findings obtained in a girl classified as I form of diplegia who walks using devices for upper limbs.

In the “virtual” reconstruction of body segments of upper limbs, lower limbs and trunk reported in the fig-

ure, it can be observed that the distinguishing sign of the gait of this child is the constant forward trunk lean.

Examining the kinematic profiles of joint angles of trunk, pelvis, hip, knee and ankle in the sagittal plane (Figure 1A), it can be noted that the hip is constantly flexed for the whole gait cycle (the flexion attitude in fact does not disappear during the terminal stance

TABLE II.—*Main features of the 213 subjects belonging to the four forms of diplegia.*

Forms of diplegia		Subjects N (%)				
		Form I	Form II	Form III	Form IV	Unclassified
Mental development (Ss 213)	Normal	13 (27.5%)	18 (69%)	14 (42%)	51 (57.5%)	9 (50%)
	Abnormal	29 (62%)	5 (19%)	16 (48%)	31 (34.5%)	8 (44.5%)
	Missing	5 (10.5%)	3 (12%)	3 (10%)	7 (8%)	1 (4.5%)
Epilepsy (Ss 213)	Present	6 (13%)	3 (12%)	1 (3%)	12 (13.5%)	
	Absent	41 (87%)	23 (88%)	32 (97%)	77 (86.5%)	18 (100%)
Visual abilities (Ss 213)	Normal	16 (34%)	13 (50%)	16 (48.5%)	40 (45%)	9 (50%)
	Abnormal	29 (62%)	12 (46%)	17 (51.5%)	38 (42.5%)	9 (50%)
	Missing	2 (4%)	1 (4%)		11 (12.5%)	
GMFCS (Ss 213)	Level 1		5 (19%)	2 (6%)	46 (51.5%)	1 (5.5%)
	Level 2	2 (4%)	7 (27%)	11 (33.5%)	30 (33.5%)	1 (5.5%)
	Level 3	21 (45%)	8 (30%)	15 (45.5%)	6 (7%)	7 (39%)
	Level 4	22 (47%)	3 (12%)	2 (6%)	1 (1%)	5(28%)
	Level 5	2 (4%)				2 (11%)
	Missing		3 (12%)	3 (9%)	6 (7%)	2 (11%)
	BFMF (Ss 122)	Level 1	4 (13.5%)	8 (42%)	3 (15%)	26 (49%)
	Level 2	21 (70%)	11 (58%)	15 (75%)	21 (39%)	
	Level 3	5 (16.5%)		2 (10%)	2 (4%)	
	Level 4				2 (4%)	
	Level 5				2 (4%)	
MACS (Ss 58)	Level 1		2 (50%)	2 (17%)	14 (48.5%)	
	Level 2	5 (38.5%)	1 (25%)	6 (50%)	10 (34.5%)	
	Level 3	8 (61.5%)	1(25%)	3 (25%)	3 (10%)	
	Level 4			1 (8%)	2 (7%)	
	Level 5					

Ss. subjects; GMFCS: Gross Motor Function Classification System; BFMF: Bimanual Fine Motor Function; MACS: Manual Ability Classification System.

phase). The knee is bilaterally in rigid flexion. The joint range of motion (ROM) is reduced. The reduction of the normal ROM is evident also at the ankle joint, which is in excessive dorsal flexion for the whole duration of the stance phase (there is no increase of plantar flexion during terminal stance).

Analysing the gait kinematics in coronal and transverse planes (Figure 1B), a fixed pelvis obliquity during the whole gait cycle (the anterior superior iliac spine -ASIS- of right side of the pelvis is constantly elevated in respect to the lower left ASIS) can be observed. An excessive adduction of the right hip during the stance phase, in respect to the left one, is also present. It can be also observed an increase of the pelvis ROM in the transversal plane and a fixed asymmetry of rotations (the right ASIS is constantly in internal rotation, while the left one is in external rotation). This pattern is related to an asymmetry of advancement (only the left side of the body remains in a fixed backward alignment in respect to the right one) which allows for the progression of gait. The foot (simplified by a rigid segment) in respect to the line of the pro-

gression of walking is bilaterally in excessively externally rotated (severe valgus-pronation of forefoot).

The results of gait analysis in a subject of the II form of diplegia with no need of devices for upper limbs are reported in Figure 2.

In this form of diplegia the global pattern of gait is characterized by the excessive flexion of the knee in the stance phase. This subject strikes the ground with a flat foot contact, the knee and ankle joints are in excessive flexion. Consequently, there is no heel rocking. This introduces a passive and excessive dorsal flexion as the lower limb is loaded (the tibia is excessively advanced because of an inadequate eccentric contraction of the plantar flexors muscles-soleus). However, in terminal stance phase a reduction of the plantar flexion can be observed; it is necessary to provide a force for body progression (lifting of the heel from ground is reduced). This evidence is confirmed by the information obtained from the study of gait kinetics, showing the reduction of the power "generated" at the ankle joint by the plantar flexor muscles during push-off. Also in this subject,

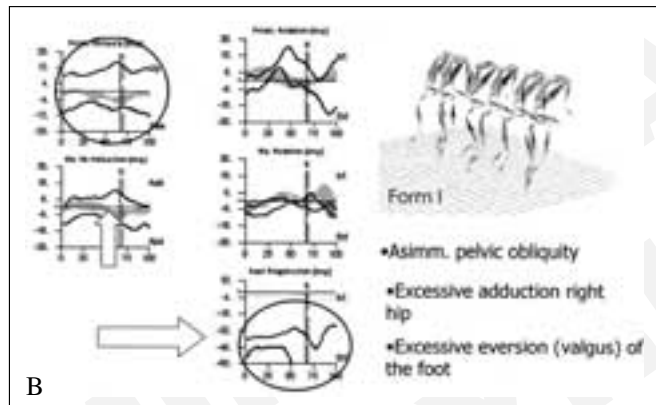
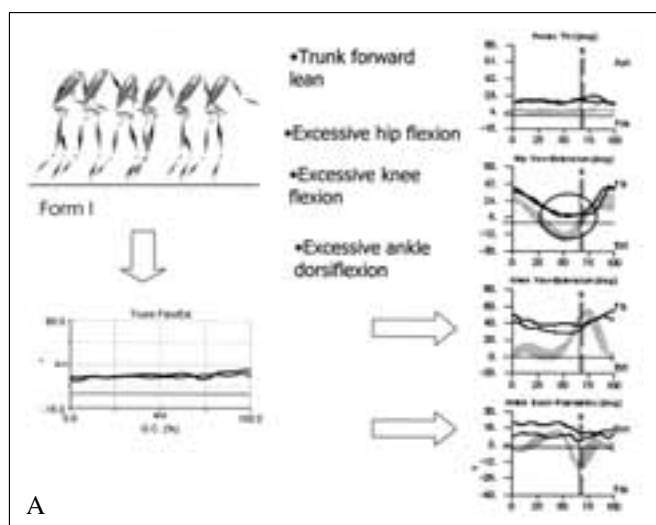


Figure 1.—Example of gait analysis in form I of spastic diplegia in lateral and coronal plane. For description of the figure, see text.

the knee remains constantly flexed for the whole gait cycle because of an increased external flexor “moment”; an excessive effort of the extensor muscles of the knee is required.

The results of gait analysis in a subject belonging to the III form of diplegia with no need of devices for upper limbs are reported in Figure 3.

In this form the gait pattern is characterized by an excessive lateral trunk lean in the coronal plane (frontal pendulum) (Figure 3A), with an increase in swinging amplitude of the trunk in the coronal plane in respect to that of the pelvis. The arms are maintained in slight abduction, the elbows are flexed and the hands are maintained at shoulder height.

This subject shows also an excessive internal rota-

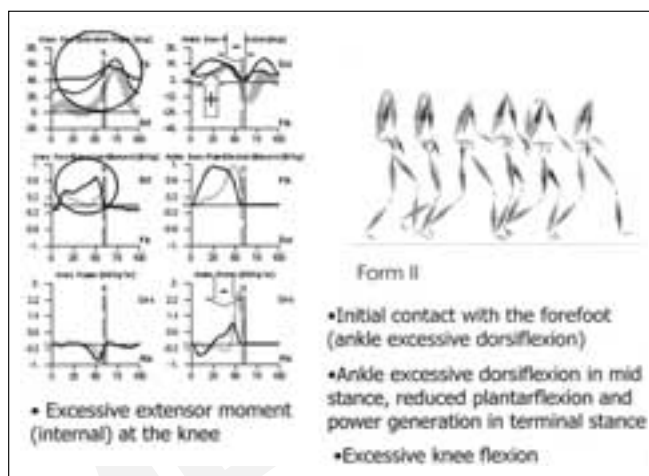


Figure 2.—Example of gait analysis in form II of spastic diplegia in lateral plane. For description of the figure, see text.

tion of the right hip more evident during the contact phase.

On the lateral plan (Figure 3B) at pelvis level it can be observed a pattern characterized by an increase of the anterior tilt, which occurs twice during the gait cycle, one in stance and one in swing phase. Generally the first peak of anterior tilt occurs during the single support stance and the second during the double support stance phase. In this subject an asymmetry in the angular profile of hip and knee in the sagittal plane can also be observed (the left hip and knee are more extended in respect to the right hip and knee).

The foot contacts the ground with the forefoot; immediately after the contact the ankle goes in excessive dorsal flexion but, contrary to the subjects of the previous forms, the ankle joint is rigid in stance phase and plantar flexion at the moment of push-off is efficient.

Figure 4 reports the results of gait analysis in a subject of the IV generalized form of diplegia. In this form the gait pattern is characterized by the equinus of the foot which begins, or becomes more evident, even before the start of gait.

Generally in this category of subjects there is a “double bump” pattern of the kinematics and of the kinetics of the ankle (Figure 4A). The foot contacts the ground with the forefoot with excessive flexion of the knee and variable degrees going from the neutral position to the plantar flexion of the ankle. Immediately after the contact, there is a rapid dorsal flexion of the ankle and an extension of the knee.

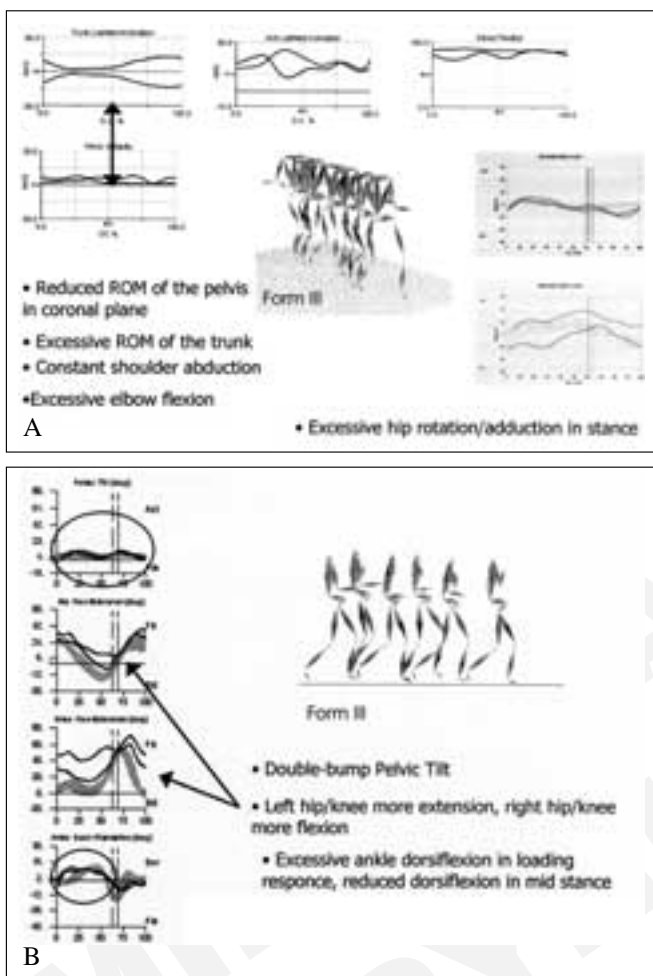


Figure 3—Example of gait analysis in form III of spastic diplegia in coronal (A) and lateral (B). For description of the figure, see text.

This couple of forces determines a rapid stretching of the gastrocnemius muscle at both ends. This muscle responds with a premature contraction which usually produces excessive plantar flexion of the ankle. After passing the vertical, the knee flexes again and the ankle presents a dorsal flexion. In the terminal stance phase, there is again an increase of the plantar flexion necessary to arise the heel and to favour body progression. Joint kinetic data show biphasic activity, and there is a first peak of power generation at the initial stance phase (concentric action of the plantar flexor muscles) and a second one at the terminal stance phase (characteristic of the push-off), both of which can be reduced in absolute values.

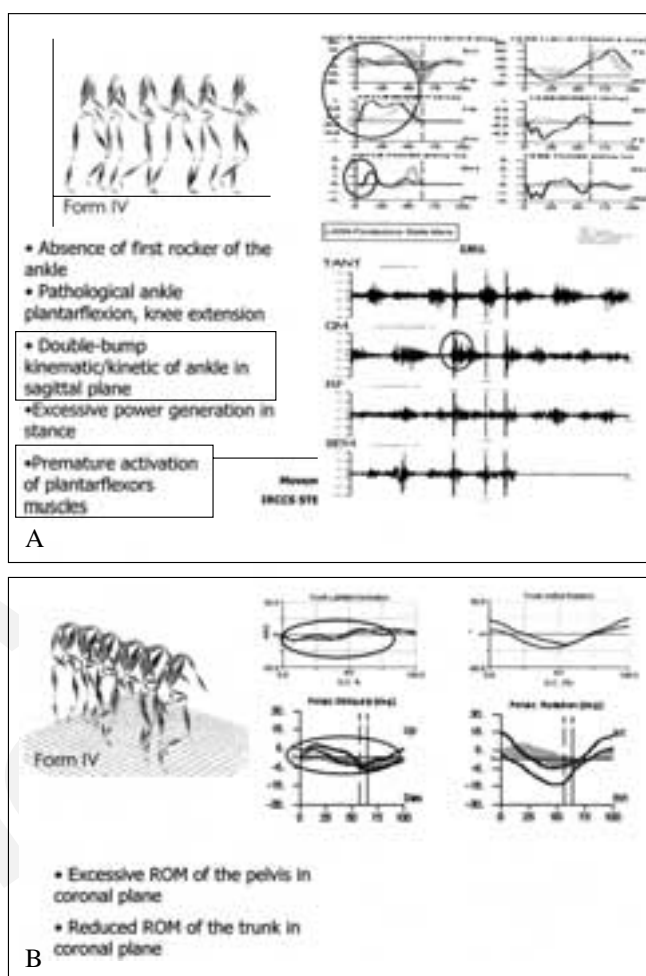


Figure 4A, B.—Example of gait analysis in form IV of spastic diplegia in lateral (A) and coronal plane (B). For description of the figure, see text.

In contrast to the subject belonging to the III form of diplegia, in this case the swinging amplitude in the coronal plane is increased at pelvis level and reduced at trunk level (Figure 4B).

Discussion

The definition and the use of one or more CP classification systems remains an important problem in children rehabilitation, not only for diagnosis but also and mainly for treatment (physiotherapy, drugs, orthosis and functional surgery). Among the spastic forms that include the largest group of children with CP,

93% in this sample, it is possible to distinguish children with diplegia and tetraplegia, not only according to a different level of motor impairment, as demonstrated with the assessment scales applied in this study (GMFCS, BFMF, MACS), but also according to other disabilities associated to the paralysis (mental, visual impairment, seizures). According to Morris and Rosenbaum,¹⁵ the use of the GMFCS underlines the existence of two different categories, as most subjects present with diplegia mainly in level I-III and with tetraplegia in level IV e V (Table I).

The findings of the present study are in agreement with the results of a recent multicentre European study of CP, where clinical and neuroradiological features of children with diplegia and quadriplegia were compared.¹⁶ Significant differences in the clinical picture and in brain magnetic resonance imaging (MRI) were reported between the two groups. The distinction between diplegia and tetraplegia is important to measure appropriately the rehabilitation needs and the sanitary and social costs associated to these two categories, as a consequence of the different motor impairment and of overall disability. According to the Authors, the terms tetraplegia and diplegia remain useful clinical descriptors.

Differently from the idea of Colver¹ and of the SCPE group² to create the macrocategory of bilateral CP, the Authors underline the need for professionals who work in the rehabilitation field to distinguish diplegia from tetraplegia. In fact it can be deceiving to put into the same category children whose highest reachable target in lifetime is the control of sitting position and children who walk, run, jump, from the first years of life. From an epidemiological point of view the Authors believe that the single category of bilateral CP forms can give more homogeneous data collected in different geographic areas. However, it has probably less useful application for health policies in programming and organizing sanitary intervention.

They also suggest the possibility to distinguish different forms among the subjects with spastic diplegia. The 213 children of the series with diplegia (46% of the whole sample) were classified into four clinical forms analysing the walking pattern (guiding function). Berta and Karel Bobath¹⁷ already distinguished two categories of children with diplegia, *i.e.* "pigeon walk" and "duck walk", analyzing their walking function. More recently, gait classification in children with CP has being object of several studies and many proposals (see Dobson *et al.*¹⁸ for a review). However, so

far the suggested classifications have limitations, and more evidences for their validity and reliability are needed.

The classification system here proposed allowed the Authors to allocate 91% of the subjects with diplegia to one of the four forms, supporting the clinical existence of these forms with a different prevalence within our large hospital based sample. This taxonomic system allows to assess and reliably classify the different clinical forms also from videotapes (Pascale R, Perazza S, Bianchini E, Alboresi S, Borrelli G, Ferrari A, Cioni G. Reliability of a classification of spastic diplegia: inter-observer agreement in 50 cases. *Eur Medicophys* [submitted]) on condition that these have been realized in accordance with standardized criteria.²⁰

The Authors have compared the degree of limb impairment and other disabilities, between subjects with tetraplegia and diplegia, and between the different forms of the latter. Within the diplegic group it was observed a gradual increase in impairment and disability both for the motor disturbances (GMFCS, BFMF, MACS) and the non motor ones from the I to IV form, enhancing the idea that inside the subjects with diplegia there are different groups of patients. The GMFCS is able to distinguish the most severe form (I) and the less severe one (IV), but it cannot significantly separate the two intermediate forms. These two categories of diplegic children are in clinical practice very different, not only for their kinematic patterns of walking, but also for physiotherapeutic needs, most suitable types of AFO, modality of antispastic drugs administration (*i.e.* timing and sites of botulinum toxin injections), surgery planning. For example, the triceps surae release does not modify balancing on toes in the first form, it may favour crouch gait in the second (due to progressive weakness), it may delay walking acquisition and above all the ability to stop in the third (due to the impossibility to adopt a velocity strategy), whereas it penalizes running and strength endurance in the fourth.

Finally, the existence of four different diplegic categories was confirmed by gait analysis, which allowed us to outline the kinetic and kinematic features embodying each clinical form.

Thinking about Colver and Sethumanhavan's¹ proposal, the Authors wonder whether the term diplegia should really be abandoned and if this choice could further increase the knowledge of CP. According to the Authors, this term not only has to be kept, because it

is possible to define the peculiar features of diplegia, and distinguish this form from tetraplegia and hemiplegia, but also it has to be enhanced and developed, because it is even possible to recognize within it various subsequent subcategories, which are very useful both from the prognostic and therapeutic point of view.

If the use of the term “bilateral cerebral palsy” can effectively simplify the collection of epidemiological data, it may confuse classification and obscure interpretation of clinical studies and therapeutic interventions. The CP panorama is not only very broad and complex, but also in constant evolution and transformation. Therefore it is necessary to have a simple classification system, which periodically could be revised and updated in order to adapt to the transformation of patient clinical features, which today are quite different from those of 30 years ago and will be soon different from those of today.

Conclusions

Preliminary results on the use of the classification system for children with spastic diplegia applied in this study are encouraging, although further validations are needed. The relationship between walking organization and type of brain lesions as observable by brain MRI has to be studied, as well as the consistency over time of the classification of the same subjects at different ages. The natural history of the different forms has also to be described, with the needs and the most appropriate therapeutic interventions.

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