NEW CEREBRAL PALSY REHABILITATION ORIENTED CLASSIFICATION

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The term diplegia should be enhanced. Part I: a new rehabilitation oriented classification of cerebral palsy

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The classification systems for cerebral palsy (CP) need to be continuously updated, according to specific aims and to significant changes observed over the years in the panorama of CP. A simplification of CP categories, abandoning the use of the term diplegia, has been recently suggested. Conversely, in this paper a new proposal for classification of CP is briefly presented, where special attention is given to diplegia which is suggested to be divided into four main clinical forms, according to the patterns of walking observable in these subjects. The proposed classification was applied to a large population of 213 subjects with diplegia, among 467 cases of CP admitted to two reference centres for this disorder. The relative incidence of the four forms is reported. The adopted classification criteria, based on a primary ability of professionals working in rehabilitation, i.e. observation capacity, makes this approach simple and easy to use at all levels of the rehabilitation services for CP.

KEY WORDS: Cerebral palsy, classification - Cerebral palsy, diagnosis - Cerebral palsy, therapy.

Different opinions about the classification of cerebral palsy (CP),[1] the problem of greatest interest among all child motor disabilities for its frequency and severity,[2] still exist. In fact, besides a clear definition of CP as "a group of disorders of development of movement and posture",[3] a likewise common classification system of different clinical forms of spastic syndromes has not yet been achieved.[4]

A CP classification system must be functional to one, or if possible, multiple aims; it is highly unlikely that an instrument useful for epidemiological data collection can also be helpful in the clinical area, especially when we intend to measure the results of therapeutic treatments. The general tendency in the last years has been to simplify the categories in which CP can be divided, in order to meet the needs of epidemiologists. The most recent indication of the international epidemiological supervision of CP (Surveillance Cerebral Palsy Europe, SCPE) [4] also proceeds in this direction, suggesting a classification system that minimizes the clinical categories (bilateral or unilateral) within the spastic forms of CP. In this way they overcome the historical distinction among tetraplegia, diplegia, triplegia, hemiplegia and monoplegia. Colver and Sethumadhavan [5] state that "*the term diplegia should be abandoned because the use of this term has*

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served to confuse classification and obscure interpretation of epidemiological and clinic studies". Morris and Rosenbaum [6] declare that "*the term diplegia has limited clinical value as a way to communicate about children with CP and the use of the term should generally be discouraged".* These authors underline however that the SCPE system can not provide, if used alone, a reliable measure of child functional abilities and has to be integrated, both for epidemiological and clinical aims, with other disability measures, such as the Gross Motor Function Classification System for Cerebral Palsy.[7]

Towards a kinematic classification

In Colver and Sethumanhavan's article,[5] an interesting historical overview of the use of the term diplegia was carried out. It can be agreed with these authors that over the last 150 years this term has been used with different meanings and often with scarce clarity in scientific articles and textbooks. In any case it has always referred to the topographic distribution of motor deficit. In none of the named classification systems reference to functional aspects has been made, and specifically to walking architecture. Since the whole problem is considered only from the topographic point of view, the quoted authors[5] declare that "*there is no justification for separating diplegia and tetraplegia*". They openly support the SCPE proposal of including both these forms in a single macro category, the bilateral cerebral palsy, a term not completely new, since it had already been used by Freud [8] in his CP organization. The proposed solution, however, includes in the same category seriously compromised children, who are not able to achieve any autonomy, not even with the best individually tailored, instrumental and environmental therapeutic facilitation, and children whose only observable defect consists of an inversion of the gait pattern. Due to the necessity to express the prognosis and measure real results achieved through the rehabilitation treatment, this proposal is questionable. In the Authors' opinion, in order to classify CP forms, they have to link to the criterion of the topographic distribution of motor impairments also at least the evaluation of one adaptive meaningful function.

Studying for more than twenty years the development of adaptive functions from a suitable sample of CP children, coming from all over Italy and distributed over a wide range of age (from a few months to adulthood), our groups have been able to identify and classify recurrent and sufficiently pathognomic motor patterns, also through the use of scheduled videotapes made according to standardized, common and ratified protocols.[9] These motor patterns can be favourably used in order to classify CP forms, even when expressed with the possible variations, which characterise the individuality of every single child. The CP classification system suggested by the Authors divides the spastic forms according to the architecture of three guiding functions: postural control, walking and manipulation.[10] In this taxonomic system, the terms tetraplegia, diplegia and hemiplegia are not only closely connected with the topographic distribution of motor deficits as in the past, but they are also related to the above-mentioned guiding functions, respectively postural control (antigravity organisation) for tetraplegia, walking patterns for diplegia and manipulation abilities for hemiplegia. The architecture of these functions contemporarily provides the guiding criterion for the diagnosis of clinical form and offers many useful indications for prognosis and therapeutic decisions. For tetraplegic CP children the antigravity organization has been chosen as the guiding function since the achievement of a suitable sitting position, as the basis for any possible autonomy, is the main goal of rehabilitative treatment. For diplegic CP children it is better to use walking, because this function, which can have different patterns and is not always achievable spontaneously or maintainable until the adulthood, is the activity to which the main therapeutic engagement (physiotherapy, orthosis, drugs, functional surgery) is directed. For hemiplegic CP children walking capability is spontaneously achieved, so that manipulation becomes the main core of rehabilitation.

Moreover, within the categories of tetraplegia, diplegia and hemiplegia, it is possible to distinguish further subtypes on the basis of the architecture of explored function.

As for the diplegic forms, some subtypes by can be identified observing the walking pattern.[11]

Outline of four clinical forms of diplegia

This approach derives mainly from the path suggested by Karel and Bertha Bobath,[12] the motoscopic examination proposed by Milani Comparetti [13] and

the results of modern optoelectronic technology (gait analysis) [14-16] and has been used for many years and for hundreds of CP patients in the reference centres of Pisa and Reggio Emilia. According to the Authors, it is possible to separate the macro category "diplegia" into four main subgroups, in which the architecture of walking function shows meaningful clinical and prognostic differences. In each clinical form, the motor and perceptual disturbances are expressed in a different way, both influencing the functional prognosis.[10] For example, in the first form ("forward leaning propulsion") the influence of perceptual disturbance is higher than that of motor deficit. The same thing happens to a larger extent in the third form ("tight rope walker"). In the fourth form ("dare devils") the disturbance is almost exclusively motor.

The aims of this paper were to provide a clinical sketch of the main walking pattern for each of these forms of diplegia, and to show preliminary data on relative incidence of each form in a large hospital based population of subjects with diplegia.

Subjects and methods

The population eligible for inclusion in the study consisted of subjects with CP admitted for developmental check-up and/or specific treatments from January 2005 to December 2006 to the Department of Developmental Neurosciences of the IRCCS Stella Maris (Pisa, Italy) or to the Unit of Children Rehabilitation of S. Maria Nuova Hospital (Reggio Emilia, Italy). In addition to clear-cut diagnosis of CP,[3, 4] an inclusion criterion was the age older than two years. An epidemiological chart to classify impairment was administered to each child which included assessment according to SCPE,[4] modified for further subdividing the spastic bilateral forms into diplegic and tetraplegic types according to Hagberg et al.[17, 18] and the classification of walking in diplegia, described below.[10]

Classification of walking patterns in children with diplegia

All subjects affected by spastic diplegia were observed and evaluated for the walking pattern according to its kinematics feature.[11] Trained doctors of the two centres were asked to allocate, if possible, each child to one of the four clinical forms, differentiated according to the following description.

First form of diplegia ("forward leaning propulsion")

The evolved walking pattern of this form is characterised by antepulsed trunk, constant support on four point canes, which are placed in front and laterally to the trunk, lower limbs with the classic pathologic pattern of flexion, adduction and internal rotation of thighs and equinus-valgus-pronation of feet (scissor pattern). The pelvis can be more or less antepulsed and the lumbar rachis have increased lordosis consequently. The head is usually held in slight retroflexion in order to neutralize the trunk antepulsion. A distinguishing sign is a specific type of balance, which occurs on toe tips, even when wearing ankle foot orthosis (AFO) or after triceps surae surgical release. The need to project the vertical of the overall barycentre forward is the consequence of a visual-kinesthetic conflict, which forces the child to be forwardly unbalanced (controllable direction) for fear of possible falling backward. The walking pattern is like a subsequent parachute reaction of the lower limb. From this motor behaviour derives the term "propulsive", which gives the name to this clinical form. The steps follow one after another in a slow and sequential way. The four point canes perform a defensive task against any possible forward fall, rather than a support function. In fact the load on upper limbs usually remains limited. Since the child does not have to support himself, but only to hold himself up, he maintains the upper limbs flexed. The four point cane advancement occurs in alternate way following the four phases pattern. The walking speed remains modest and the progression slow and tiring, also due to the early exhaustion of the support reaction. The walking pattern is characterised by an increased hip internal rotation in mid stance (jump gait with or without stiff knee, in accordance to the motor behaviour of rectus femoris [15]). The foot contact on the ground always occurs in equinus-valgus-pronation as a consequence of hip and knee flexion, even if the tibio-tarsal joint does not exceed 90° (apparent equinus [16]). The foot clearance during the swing phase is limited. The knee, already flexed, is progressively stressed toward valgus deformity, with serious torsion conflict caused by the presence upstream of an internal rotation of the thigh, produced by the antiversion of femur neck, and downstream by an external torsion of the tibia. The hip of the loaded lower limb maintains some flexion also in terminal stance. The pelvis as a consequence of

adductor interference usually shifts towards the loaded lower limb.

Second form of diplegia ("tight skirt")

The child walks keeping the head moderately projected forward and the trunk in a vertical position. The four point canes are maintained slightly to the side of the trunk, with semi extended elbows, and advanced in a four phase pattern. Even if the canes are in constant contact with the ground, as a consequence of the vertical trunk-feet alignment, they are only slightly loaded. Their use is more comparable to direction tasks than to anterior defensive aims. The lower limbs maintain the flexion pattern already observed in the first form, but with a more pronounced knee flexion. The distinguishing sign of this form is, in fact, represented by the loaded knee behaviour, which increases flexion at the moment in which the opposite lower limb crosses the zenith and comes in contact with the ground (therefore the term tight skirt). The pelvis oscillates in anti – retropulsion at every step as a consequence of hamstrings strain, with a knee flexion synchronised swing and a subsequent increased – decreased lumbar lordosis. The loaded hip never completely extends in terminal stance and can have a limited flexion also in terminal swing. In monopodalic phase the pelvis shifts towards the loaded lower limb. The trunk swings prevalently in the sagittal plane. A swing phase equinus can be present, followed by an initial and then full contact one. Instead there is no push-off equinus. In order to complete the tight skirt scheme, besides the antiretroversion pelvis swing and loaded knee flexion, it is necessary that the loaded foot achieves a dorsal flexion. This movement can take place at the ankle, in case of primary or secondary weakness of triceps surae, or in mid foot (rocker bottom) as a consequence of a pathological breakdown of plantar muscles and a collapse of the longitudinal arch (hidden equinus). An AFO, able to sustain body weight, could contribute to protect the foot and improve walk.[16] Both in case of dorsal flexed or spastic flat foot and of AFO employment, the load is kept on the forefoot (apparent equinus [16]). The steps are short and frequent and forward speed is reduced. The presence of a weak or excessively lengthened triceps surae, combined with a subpatellar ligament failure, reducing the extensor couple power (knee extensors-foot plantarflexors), can lead, before adolescence, to the development of a crouch gait,[15] the most frequent cause of loss of walk in this category of diplegic children.

Third form of diplegia ("tightrope walkers")

These children are able to walk without canes. Those who adopt these devices, in reality, use them only at the moment of stopping and in order to maintain the standing position, preferring to lift the canes when walking. In this form of diplegia the motor deficit is actually rather limited, but the perceptual disorder is so significant that many patients employ a numbers of years to achieve walking ability, or are able to walk only if someone follows them while touching them on the back with a finger (posterior space confinement). In order to walk, these children adopt the speed up strategy and for this reason it is better that they do not use AFOs, but rather flexible laced shoes. The head and the trunk are tilted forward in order to favour a more efficient advancement. The upper limbs, laterally raised, swing in the frontal plane contributing to obtain balance. During these movements, the position of the hands can reach or even surpass the shoulder level. The lower limbs show the typical pattern of hip flexion, tight adduction and internal rotation, knee flexion and talipes equinus. In this form it is easier to see foot varus-supination rather than valgus-pronation, typically present in the previously mentioned forms. At the moment of walk start, the children begin to swing upper limbs and trunk in the frontal plane, leaning forward, accentuating equinus and launching a series of steps in rapid acceleration. At walk stop, they often bump against something or someone, always arriving too quickly and heavily, or they slow down walking by executing, before completely stopping, a sort of walking in place. The step sequence usually is short and rectilinear (step cluster). In order to modify the progression trajectory, these diplegic children look for a momentary contact on an opportune support. The foot makes contact with the ground with the toes (swing equinus) and increases the plantar flexion at the end of the stance phase (push off equinus). Foot lifting can be made easier though a placing reaction, which facilitates a secondary contraction of ankle dorsal flexors and toe extensors, recruited in an inversed kinetic chain. In these diplegic children, the improvement of perceptual control is testified by the progressive lowering of upper limbs in the frontal pendulum and by the

reduction of walking speed, in addition to the capacity to walk through open spaces.

FOURTH FORM OF DIPLEGIA ("DARE DEVILS")

The children belonging to this form are the most capable in motor performances, since their defect is mainly if not exclusively of a motor type. Walking is precociously acquired and preserved forever, differently from what can occur in the other three forms. The pathologic scheme can be limited to the inversion of step pattern or be extended in a proximal direction to involve the knees (in flexion) and the hips (in flexion, adduction and more or less internal rotation, in accordance with the degree of femur neck antiversion). During walking, the head is vertical and the trunk slightly antepulsed, shoulder and hips swinging in countertendency and elementary rotation between girdles are possible. The hip flexion and pelvis antiversion justify the increase of lumbar lordosis. The upper limbs are kept slightly abducted laterally to the trunk and can produce elementary pendulum movements, generally asymmetrical. The wrists are semi extended, while the hands remain open. The adductor interference is present, but rarely become excessive. The motor behaviour of lower limb is hardly ever symmetrical, especially at lower speeds. It tends toward symmetry when the child speeds up walk velocity or begins to run. Also in upright standing position these diplegic children show difficulties in weight balancing on the lower limbs and prefer to maintain the load on either one foot or the other, frequently shifting sides. The loaded lower limb lines up, while the counterlateral one shows hip and knee flexion and foot equinus. In order to bring the heel in contact with the ground, the child emphasises hip flexion and trunk antepulsion and pushes the knee in hyperextension. The distinguishing sign of this diplegic form certainly is represented by the onset of talipes equinus just as the walk is about to start and by its reduction just as the child returns to standing position. During walking there are frequently swing, contact (generally with varus) and push-off talipes equinus phenomena. In order to facilitate foot clearance, the child in terminal stance can carry out a placing reaction of ankle dorsal flexors and toe extensors. Instead in the less compromised children a stepping reaction can also occur. Just as the body load is accepted on the supporting foot, it is possible to show a jerky knee movement. The dynamic balance is acceptable, especially if one considers the reduction of support basis produced by the equinus foot. When they are very young, diplegic patients of this form, since they are unable to stop walking, have no hesitation to drop down on their knees with hip adduction and internal rotation. Successively they do not have any difficulty in getting up either with the half kneeling manoeuvre or with simultaneous extension of knees and hips, and to set on walking again. Over time, they can learn to abruptly stop walking without the necessity of taking further steps after receiving an order to stop. Their endurance is on the whole good enough.

The main features of the four forms of gait are reported in Table I, while the Authors can refer to other publications for a more complete description.[10]

Doctors in charge for the classification procedures in the centres strictly followed the criteria described above. They periodically met during the study, analysing videotapes of subjects with diplegia, to maintain a high interobserver reliability.

The study was approved by the Ethical Committee of the Stella Maris Scientific Institute.

Results

In the two year period of this study, 467 subjects with CP were recruited in the two centres: their age ranged from 2.0 to 21.7 years (mean 7.8±4.1); 262 were male. A large prevalence of spastic CP was found (434 subjects, 93%), *versus* dyskinetic (24 subjects, 5%) and ataxic CP (9 subjects, 2%) (Table II).

Spastic diplegia was diagnosed in 213 subjects (115 male, mean age 7.9±3.9, range 2.0-21.7 years). This group was the largest in this series (46%), whereas the subjects with tetraplegia were 115 (25%) and those with hemiplegia 106 (22%). The frequency of the four forms of diplegia, distinguished according to the proposed walking classification, is reported in Table III. Eighteen subjects were not classified because they were too young to walk, or they had just started to do it. The IV form (42%, 89 subjects of the diplegic sample) more often occurred, followed by form I (22%, 47 subjects), III and II (15 and 12%, 33 and 26 subjects, respectively).

More details on the motor features and associated disorders in the diplegic and tetraplegic groups of this samples are reported elsewhere.[19]

TABLE I.—*Main characteristics of the four clinical forms of spastic diplegia.*

Segments	1st form	2nd form	3rd form	4th form
Upper limbs	Devices used for protection 4 phases constant support	Devices used for direction 4 phases constant support	Tilted laterally used as stabilizers 2 phases, with no constant support	No devices
Trunk	Forward leaning With or without lordosis	Vertical	Forward leaning with hyperlordosis	Vertical or slightly for-ward leaning
Swing	Combined mainly sagittal	Combined, mainly sagittal	Frontal, related to shoulders, pelvis and upper limbs	Combined, mainly frontal Countertendency between shoulders and pelvis
Pelvis	Antiversed and unstable, translated toward the supporting limb with homolateral elevation	Sagittal swinging in antiversion/retroversion and translation towards the supporting limb	Contralateral elevation, translation toward the lower limb in swing phase	Valid proximal fixation and slightly expressed inter girdle rotation
Progression	Rotation on supporting hip	Flexion on supporting knee	Propulsion and pivot on foot	Hip intrarotation
Foot	Talipes equines in contact, full support and swing phase	Contact talipes equinus/ dorsiflexion/ drop foot	Contact and push off talipes equinus, pivot/ placing	Contact and push off talipes equinus, possible placing reaction Drop foot
Fulcrum	Toe balancing	Lumbar hinge/knee	Trunk/foot	Hip/foot
Distinguishing signs	Hip/knee Trunk forward leaning and toe balancing	Knee flexion in mid stance	Frontal trunk swinging	Increasing talipes equinus at start of walking

TABLE II.—*Distribution of the main types of cerebral palsy, classified according to Hagberg et al.*[17, 18]

Type of cerebral palsy		N (%)
Spastic CP		434 (93%)
Tetraplegia	115 (25%)	
Diplegia	213 (46%)	
Hemiplegia	106 (22%)	
Dyskinetic CP		24 (5%)
Dystonic	17 (3.5%)	
Choreo-athetotic	7 (1.5%)	
Ataxic CP	9 (2%)	
Total		467

TABLE III.—*Distribution of the subjects with diplegia according to the suggested classification.*

	N. (%)
I form (Forward leaning propulsion)	47 (22%)
II form (Tight skirt)	26 (12%)
III form (Tight rope walkers)	33 (15%)
IV form (Dare devils)	89 (42%)
Unclassified	18 (9%)
Total	213 (46%)

Discussion

Among a large population of spastic forms of CP, we classified 213 children with diplegia of our series into four clinical forms analysing the walking pattern (guiding function). This proposal is not completely new, since Berta and Karel Bobath [12] had already distinguished children with diplegia into two categories analyzing the walking function ("pigeon walk" and "duck walk").

Furthermore, in a recent systematic review of gait classification in CP children, Dobson *et al.*[11] have shown how this methodological approach is shared and adopted to assist in diagnosis, clinical decision-making and communication.

The classification system proposed here allowed us to distinguish the 91% of the diplegic children group into the four diplegic forms, supporting the clinical existence of these forms, with a different prevalence, within the hospital based sample, which includes all subjects with CP who have attended two large centers for disabilities at the Hospitals of Pisa and Reggio Emilia.

In two other related articles the Authors summarized the results obtained by systematically applying

the classification of diplegia to subjects admitted to our two CP reference centres, in order to confirm the difference in the clinical profile between diplegic and tetraplegic forms of CP, to compare the results of this new classification proposal with other methods, already available and standardized, to match up the clinical description of the walking patterns for the different forms, with those obtainable through gait analysis, 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 and visual disorders and, lastly, to demonstrate the confidence in the observers and the clinical and research applicability, to assess the interobserver reliability of experienced observers *versus* professionals who attended a one-day training course of the application of this classification. (Cioni G, Lodesani M, Coluccini M, Sassi S, Paolicelli PB, Pascale R, Perazza S, Ferrari A. The term diplegia should be enhanced: contribution to the validation of a new classification system. Eur Medicophys [submitted]; Pascale R, Perazza S, Borelli G, Bianchini E, Alboresi S, Ferrari A, Cioni G. Reliability of a classification of spastic diplegia: inter-observer agreement in 50 cases. Eur Medicophys [submitted])

Conclusions

The sustained results of the above mentioned investigation, based on clinical meaningful categories, indicate that this proposed classification of CP is easily utilizable, both for diagnostic and therapeutic aims, in clinical environments at every institutional level. Its value is based on the fact that it does not require any sophisticated or specific instruments, except for the recommended videotaping, being substantially supported by guided observational criteria. As further investigation, through this classification system it will be possible to evaluate the possible relationship between walking organization and type of brain lesions observable by brain magnetic resonance imaging (MRI). Moreover, the consistency over time of the classification system proposed in this work has to be reviewed, analysing the natural history of children with diplegia, in order to assess the reliability of the form diagnosis for the same subject at different ages. Another important aspect to investigate is the time employed by children belonging to the four diplegic forms to start walking, alone or with upper limb devices (guiding function), to see whether there is a strict correlation between each form and walk acquisition age. This classification can also be a starting point to explore activity limitation and consider participation and to investigate the effective needs at different ages of available therapeutic interventions (physiotherapy, orthosis, drugs, functional surgery) within the four groups.

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