PROBLEMS IN THE CLASSIFICATION OF CEREBRAL PALSY IN CHILDHOOD

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Cerebral palsy is a descriptive term applied to a group of motor disorders of young children, in whom full function of one or more limbs is prevented by paresis, involuntary movement, or incoordination. Tacit agreement has been reached that progressive diseases and those characterized by transient motor disturbances should be excluded as well as those primarily the result of spinal cord lesions. Unfortunately the many different classifications of the various forms of cerebral palsy that are in use lead to great confusion, and free interchange of ideas between different workers is impeded at a time when increasing attention to handicapped children gives some promise of real advances in management.

There is a need for a standard terminology and for a classification which can be employed successfully by all those engaged in the treatment of affected children. The present article presents a neurological classification which we have found useful and compares it to a number of older and contemporary classifications.

Terminology

Much unnecessary confusion has been caused in the past by the inaccurate use of neurological terms to describe the manifestations of cerebral palsy in childhood. Descriptive terms which refer to clearly defined features of neurological diseases in the adult patient have been applied indiscriminately to a variety of unrelated features of cerebral palsy in the child. For example, the term "athetosis" was originally used to describe slow writhing involuntary movements of the distal parts of the limbs, and especially the digits, which were frequently found in association with hemiplegia (Hammond, 1881; Gowers, 1876). The term is still used to describe a specific form of involuntary movement in adult neurology, but in cerebral palsy it may denote a great variety of dissimilar types of involuntary movements (Phelps, 1940, 1941; Asher and Schonell, 1950). To avoid such confusions of terminology we use descriptive neurological terms strictly in the sense in which they are used in adult neurology.

TABLE I.—*Classification of Cases of Cerebral Palsy in Childhood*

Neurological Diagnosis	Distribution	Severity
Hemiplegia	Right Left	Mild Moderately severe Severe
Double hemiplegia		Mild Moderately severe Severe
Diplegia:		
Hypotonic	Paraplegic	Mild
Dystonic	Triplegic	Moderately severe
Rigid or spastic	Tetraplegic	Severe
Ataxic diplegia:		
Hypotonic	Paraplegic	Mild
Spastic	Triplegic	Moderately severe
With contracture	Tetraplegic	Severe
Ataxia:		
Cerebellar	Predominantly unilateral	Mild
Vestibular	Bilateral	Moderately severe Severe
Dyskinesia:		
Dystonic	Monoplegic	Mild
Choreoid	Hemiplegic	Moderately severe
Athetoid	Triplegic	Severe
Tension	Tetraplegic	
Tremor		
Other types		

The Suggested Classification

This classification (Table I) was evolved during research into the prevalence and aetiology of cerebral palsy (Ingram, 1955a). It has also been used successfully in an advisory clinic supported by the Scottish Council for the Welfare of Spastics. It is based on an earlier neurological classification suggested by Freud, with modifications to bring it into line with more recent knowledge and to allow for the classification of cases by the severity and extent of their motor handicaps.

The various categories in our classification are considered in detail in the following text.

Hemiplegia

Hemiplegia is a unilateral paresis, usually associated with some spasticity and flexion contracture of the limbs. The upper limb is more severely affected than the lower. In most cases arising in early infancy, or from prenatal disorders, growth is retarded in the affected limbs, so that these are shorter and thinner. Vasomotor disturbances may occur, but are not parallel in severity to the dwarfing. Athetosis of the fingers and toes is a frequent finding, but even though the clinical picture may show some resemblance to unilateral dyskinesia we have not classified these cases in the latter group, as they clearly belong to the hemiplegic category. Sensory disturbances in the affected limbs are common; in certain cases an associated hemianopia may be demonstrated. It is inconvenient, however, to include sensory disorders in classification, though some account of them is possible in the assessment of the degree of disability.

In classification the side affected should be noted, as the disability tends to be greater if the master hand is involved, and there is a significantly higher prevalence of aphasia and speech disorders in right hemiplegia than in left. Severity is assessed by residual function. Mild cases are those able to use the affected hand independently for everyday activities; moderately severe cases are those able to use the affected hand to assist in bimanual activities; severe cases retain no useful function in the affected hand. It is unnecessary to consider the lower limb in assessing these cases. Athetosis and contracture are reflected in the classification by severity.

Double Hemiplegia

These patients show tetraplegic paralysis, greater in the arms than the legs, and there is usually moderate or severe flexion contracture and spastic increase in tone. Most are mentally defective and epileptic. They are further handicapped by dysphagia, severe dysarthria, and recurrent respiratory infections. A few of the less severely handicapped show slight athetosis.

It is necessary to separate this group from that of hemiplegia, as the disability exceeds the summation of two hemiplegic disorders: moreover, it appears likely that the majority are of prenatal origin and will eventually occupy a distinct aetiological group. Separation from the group of diplegic paresis is necessary because of the greater bulbar involvement and the greater disability in the arms. It will also be shown that the early history of patients with diplegia is often distinctive.

All cases are severely disabled: mildly affected cases retain some use of the arms; moderately severe cases have some use of the legs; and severe cases retain no proper use of their limbs, being bedridden and helpless.

Cerebral Diplegia

The term "diplegia" was introduced by Freud (1893) as a collective description of certain forms of more or less symmetrical paralysis. The term is not well derived and has been criticized on this account (Evans, 1948). It has the advantage that it does not simply enumerate the number of limbs affected, and therefore is not merely a topographical designation. It describes paraplegic, triplegic, and tetraplegic patients whose clinical features differ in degree and not in kind. There is need for a term that will do this, and "diplegia" has the backing of historical precedence for its use. The modern controversy over this usage dates from the conception of triplegia and tetraplegia as being examples of double hemiplegias (McNutt, 1885; Osler, 1889; Gowers, 1888). Most authors, even at that time, however, distinguished between paraplegia and double hemiplegia, and in modern work there has been a similar tendency to group patients with triplegic and tetraplegic paresis together in one category, and those without involvement of the upper limbs in another.

We have used the term diplegia to describe a more or less symmetrical paresis of cerebral origin, more severe in the lower limbs than the upper, and dating from birth or shortly afterwards. Fine movements of both the fingers and the toes are invariably impaired. The legs and pelvis are underdeveloped. Mental defect, epilepsy, and strabismus are often present, but bulbar paresis is rarely severe.

Approximately 40% of patients with congenital diplegia are born prematurely and the majority have a history of perinatal anoxia.

Several stages in the development of diplegia may be recognized (Ingram, 1955b). These proceed in a fixed order, but the duration of each stage varies from patient to patient, and the development of the condition may stop short of the final picture. In the first few weeks the child is often thought to be normal, but there is already marked poverty of movement and generalized hypotonia. This is usually noted by the parents when the disorder is severe, but may be ascribed to the premature birth by the medical attendant. After weeks or months the next stage begins. Sudden changes of position, or even sudden alarm, produce a forceful hyperextension of the back and head, so that the body, momentarily only, assumes a position of opisthotonos. This dystonic movement is most readily produced by sudden extension of the head. When unusually frequent, these attacks may be regarded as fits, and this confusion is legitimate, in that there often appears to be momentary impairment of consciousness.

After a variable period the stage of dystonia is succeeded by the development of rigidity and later by spasticity. At first a persistent increase of extensor tonus is noted; the limbs tend to assume stiff postures in extension and the righting reflexes are exaggerated. Deep reflexes are difficult to elicit; the plantar responses are flexor or infantile in type. At this stage the condition may therefore be described as a generalized rigidity without exaggeration of the reflexes. Further progression may cease at this stage. Usually, however, a spastic increase of tone gradually appears, and with it a tendency to assume flexor positions. The reflexes are now increased and the plantar responses are extensor.

Approximately 40% of a recent unselected series of children of school age with diplegia gave a history which indicated that there had been a period of hypotonia, and in approximately 50% a history of dystonia was elicited. Occasional cases are found in which dystonia persists until the age of 4 or 5 years, and these children are all severely affected. It is much commoner to find patients showing predominant rigidity in some limbs (those more severely affected), and spasticity in others (those less severely affected and capable of more mature motor activity). The picture of generalized spasticity with flexion contractures in all limbs must be regarded as the final result of diplegic paresis.

Since the stages of diplegia appear to be well defined in many patients, it is desirable to note the state of the development of the condition at the time of classification, though in many patients the subgroup will change as time elapses. It is also necessary to classify patients according to the extent and severity of their functional impairment in everyday life. They are therefore placed into groups of paraplegia, triplegia, and tetraplegia. Severity is best assessed by the efficiency in walking, since hand skills parallel the disability in the legs in the majority of patients. Mild cases are those whose gait is clumsy rather than disabled; moderately severe cases are those with an obviously impaired gait, including those who are unable to run; severe cases are those who can walk only with the aid of some support.

Ataxic Diplegia

Cases of ataxic diplegia have been described in the literature, originally under the name of diataxia (Ramsay Hunt, 1918), and reports of this condition have been reviewed by Kinnier Wilson (1940). All four limbs are involved neurologically, though the legs are always more severely affected and there may be relative sparing of one upper limb. Spastic increase of tone is present, but hypotonia may be demonstrated in some muscle groups, and rarely may be the chief finding. The gait is ataxic on a wide base; adductor spasm is always absent. The ataxia appears to be of cerebellar type clinically, but because of the severe paresis it is frequently impossible to assess its importance in causing functional impairment. These patients do not pass through a dystonic stage, so that they may be distinguished from diplegic patients by their history and clinical findings, and from ataxic patients by the presence of bilateral spastic paralysis.

A family history of neurological disorders, such as epilepsy, mental defect, or ataxic diplegia, is often present, and a history of perinatal anoxia is less frequent than in diplegic patients.

Mild cases are those without significant functional loss, though they are clearly affected and known to be clumsy; moderately severe cases have some limitation of normal activities; severe cases are those who need to be helped in all but the simplest everyday activities. It is useful, in classification, to note whether the muscles are predominantly hypotonic or spastic.

Ataxia

Patients in this category show incoordination of movement and impaired balance as their presenting clinical features. The ataxia may be symmetrical or chiefly unilateral. Mild cases have no significant loss of function, though they are recognized to be abnormally clumsy; moderately severe cases suffer some limitation of normal activity; severe cases require help in all but the simplest everyday activities.

Dyskinesia

Involuntary movements occur in a number of forms of cerebral palsy. Athetosis may be present in hemiplegic limbs, or in limbs showing the presence of spastic increase of tone in diplegia or ataxic diplegia. Choreoid movements may also occur occasionally in hemiplegia, and dystonia occurs for a time in about 50% of diplegic patients.

There is a group of patients, however, in whom similar involuntary movements occur, apparently without being associated with hemiplegia, diplegia, or diplegia with ataxia. To this group we have given the collective descriptive term "dyskinesia," after Perlstein (1952). It must at once be admitted that the group lacks the definition of the others, and the criteria are, in part, negative. The subgroups of dyskinesia are those suggested by Perlstein. Dystonic movements are those involving the trunk as well as the limbs so that walking is impossible in most cases. Choreoid movements involve the proximal limb muscles

more than the distal and are of such force that the limb is thrown from the trunk and balance is seriously disturbed. Athetoid movements involve chiefly the fingers and toes, but in severe cases they may spread to the wrist, forearm, and elbow. They are slow and writhing, they disappear at rest, and are accentuated by voluntary activity. Sudden involuntary variations of muscle tone may accompany any of the above disorders of motion. The whole limb stiffens during attempted movements and bizarre postures are assumed for short periods.

Opinions differ concerning the origin and nature of this disturbance. It has been suggested that it is an acquired adaptation to the disorder of motion—an attempt to steady the proximal parts of the limbs so that fine distal movements can be performed (Phelps, 1941). It seems more probable, however, that this variation of tone is involuntary, and that it is due to the sudden release of subcortically integrated postural reflex; it occurs most typically as a sudden involuntary extension of the limb during choreoid or dystonic movement.

By suitable combinations of these descriptive terms the various types of involuntary movement encountered in cases of cerebral palsy can be described with some accuracy, though it should be emphasized that usually more than one form of disorder of movement is found. It is exceptional to find patients with simple athetosis or simple chorea. Because of this, it is more convenient in practice to place patients suffering from dyskinesia in subgroups according to the distribution of their disorder before considering the forms of involuntary movement which they show. The severity of the condition is gauged as for ataxic patients.

Other Categories

Few cases fall outside the categories described, and most of these are known to be the result of specific post-natal diseases. We have included post-encephalitic syndromes and a few patients with arthromyodysplasia with associated cerebral palsy. Accurate symptomatological description of these cases is sufficient to ensure that there is no confusion when series are compared.

Occasional patients with cerebral palsy proper show features of more than one category. Thus a child with diplegia may have an associated hemiplegic dwarfing of one upper limb, or a child may show a combination of ataxia and hemiplegia following birth injury. These patients may usually be classified by their major disturbances, but the associated condition must be noted. They do not occur often enough to disturb the system of classification.

Previous Classification

The first attempts at classification of the various forms of cerebral palsy were on the basis of presumed pathology. Attempts were made to define clinical pictures which were diagnostic of particular cerebral lesions (Cazauvieilh, 1827; Henoch, 1842; Cruveilhier, 1862; Cotard, 1868; Strümpell, 1884; Jendrassik and Marie, 1885; Brissaud, 1886). These proved unsuccessful, for even with modern techniques it is impossible to obtain an accurate picture of underlying cerebral pathology in the living child, and many different pathological pictures may be associated with similar clinical syndromes (Freud, 1897).

Aetiological classification may be said to date from the work of Little (1862), the very title of whose paper, "On the influence of abnormal parturition, difficult labour, premature birth, and asphyxia neonatorum on the mental and physical condition of the child, especially in relation to deformities," implies an interpretation of the causes of cerebral palsy. An aetiological classification in which cases are classified according to whether their disorders appeared to result from prenatal, natal, or post-natal factors was proposed by Sachs (1891) and Sachs and Hausman (1926). It proved unsatisfactory because similar clinical disorders did not necessarily result from similar aetiological factors and multiple contributory causes of cerebral palsy were often recognized.

Freud looked forward to the day when it would be possible to make a comprehensive classification on the basis of pathology, aetiology, and clinical findings. He considered that until clinical syndromes could be defined in these terms classification by neurological syndromes was the most satisfactory method possible. He stressed that categories should be defined to emphasize the points of clinical similarity between cases, but he noted that this meant that many important and interesting but less significant clinical features might have to be ignored in classification. He proposed that patients showing predominantly unilateral disorders should be distinguished from those with bilateral cerebral palsy, and for the latter he coined the collective descriptive term "cerebral diplegia." In this category were included cases with paraplegic, triplegic, or tetraplegic paresis in which the lower limbs were more severely affected than the upper, cases of double hemiplegia, and cases showing involuntary movements as their presenting feature. He emphasized that advances in knowledge might necessitate the addition of other categories and a rearrangement of those which he had proposed.

Modern Classifications

The classification of Freud was in general use until the modern world-wide interest in the therapy of children with cerebral palsy arose. Attempts were then made to classify patients on the basis of clinical features most cogent to the therapist (Phelps, 1941; Hellebrandt, 1950–1). This type of classification attempts to define categories in terms of single clinical features, such as rigidity or spasticity (Table II).

TABLE II.—*Classifications Used for Some Recent Published Series of Cases of Cerebral Palsy*

Ingram (1955a)	Asher and Schonell (1950)	Evans (1948)	Hellebrandt (1950–1)
Hemiplegia: Right Left Double hemiplegia Diplegia: Paraplegic Triplegic Tetraplegic Ataxic diplegia: Triplegic Tetraplegic Ataxia: Bilateral Chiefly unilateral Dyskinesia: Monoplegic Hemiplegic Triplegic Tetraplegic Other types	Spastic paralysis: Symmetrical quadriplegia Asymmetrical quadriplegia Paraplegia Hemiplegia Others Athetosis Mixed spastic and athetoid Ataxia Flaccid cerebral palsy Tremor and rigidity	Monoplegia Hemiplegia Spastic paraplegia or quadriplegia Flaccid quadriplegia Athetoid Chorea Ataxia Mixed types	Spastic: A. Aspastic B. Spastic: (1) Monoplegia; (2) hemiplegia; (3) paraplegia; (4) triplegia; (5) quadriplegia C. Basilar Athetosis: A. Tension B. Non-tension C. Dystonic D. Flail E. Arm–neck F. Deaf G. Shudder H. Hemi-athetoid I. Cerebellar release J. Rotary K. Emotional release L. Tremor M. Unclassified: (1) paraplegia; (2) quadriplegia; (3) monoplegia; (4) recovered Rigidity: A. Intermittent B. Continuous C. Miscellaneous: (1) hemiplegia; (2) paraplegia; (3) triplegia; (4) quadriplegia Tremor: A. Intention B. Constant Ataxia: A. Cerebellar B. 8th nerve

Exception may be taken, however, to the fact that single clinical features, which are shared by a number of forms of cerebral palsy, are used to define categories. Thus in the category of rigidity will be found cases with stiffness of the limbs due to arthromyo-dysplasia associated with cerebral palsy, cases with rigidity due to increased extensor hypertonus, rigidity due to contracture, and rigidity of the Parkinsonian type. A patient with diplegia may have to be repeatedly reclassified as he passes through the stages of hypotonia, dystonia, and rigidity and spasticity.

Nevertheless this classification was evolved as a result of long experience in the treatment of affected children, and

many of the clinical descriptions are of great value. It fails as a scheme for general use because classification must be something more than a guide to therapy. It must emphasize useful distinctions between different forms of cerebral palsy, and this cannot be done by reference to a single scheme of therapy.

To meet some of these objections, further classifications have been proposed. In one of the most successful of these, cases are classified in parallel under the headings of clinical findings, distribution, muscle tone, severity, and probable aetiology (Perlstein, 1952). That author's claim that it permits the accurate description of almost every case of cerebral palsy appears justified, but the distinctions between different forms of cerebral palsy are not all valid. For example, under the topographical heading appear subgroups for both quadriplegia and bilateral hemiplegia, which is not a topographical distinction. There is no suitable category in which diplegic patients showing tetraplegic rigidity may be placed. These are minor criticisms, however, compared with the more important one that the informality of the system may encourage different interpretations by different workers. This classification is unlikely to facilitate comparison between clinics in respect of either the case material studied or the results of treatment of the various forms of cerebral palsy.

Comparison with other Contemporary Classifications

The present scheme is compared with other contemporary classifications in Tables II and III. In Table II three current classifications are listed against the present scheme to show the differences in terminology. We suggest that the present classification is sufficiently detailed to be used as a basis for future research into aetiology, pathology, and prognosis,

TABLE III.—*Comparison of Recent Published Series of Cases of Cerebral Palsy in Terms of the Proposed New Classification*

Category	Ingram (1955) a	Asher and Schonell (1950)	Evans (1948)	Hellebrandt (1950–1)
Hemiplegia ..	36%	29%	9%	24%
Diplegia ..	38%	—	—	—
Type not stated	0%	1%	2%	7%
Paraplegia ..	14%	26%		10%
Triplegia ..	11%	5%		4%
Tetraplegia ..	13%		42%	
Double hemiplegia	4%	22%		19%
Ataxic diplegia ..	6%			
Ataxia	7%	1%	3%	8%
Dyskinesia ..	8%	10%	40%	27%
Other	1%	1%	—	1%
Mixed forms ..	0%	6%	4%	0%
Total cases reported	208	349	115	531

that it contains no misleading groupings, and that it will simplify analysis for the numerous non-medical auxiliaries engaged in the management of patients with cerebral palsy.

Comparison of different series is easier if published reports are reclassified in terms of the present scheme (Table III). Real differences between the types of case seen at different clinics are shown clearly. The series collected at a screening clinic for a residential school (Evans, 1948) contains an excess of cases of dyskinesia and a deficit of hemiplegic disorders compared with the unselected series (Ingram, 1955a, Asher and Schonell, 1950). Comparison of the frequency of diplegia, double hemiplegia, and ataxic diplegia is more difficult, but an attempt has been made in Table III after careful study of the other classifications. Systematic groups which could not be separated are shown joined by brackets overlapping the groups concerned. It is possible, by this means, to obtain a rough estimate of the frequency, in other series, of the groups in the present scheme, and with a little more information it would be possible to make a complete comparison. We suggest, therefore, that the present scheme, based on neurological syndromes, could be used by other workers without discarding the further subdivisions which they have found useful for their own therapeutic programme. In particular, we suggest that some of the terms introduced by Phelps (1943) to describe certain forms of dyskinesia are useful to the physiotherapist, and that they can be used without disturbing the new scheme we have proposed.

Summary

A standard simplified terminology and classification of cases of cerebral palsy would facilitate comparison of the work done at different centres, and facilitate exchange of information between workers with different points of view.

A classification which has been found useful for research purposes and for routine clinical practice is described. It is based on well-defined neurological syndromes and permits more accurate description of cases than other classifications which are based on presumed pathology, aetiology, or single clinical features.

This classification has not been influenced by any therapeutic programme. It may therefore be recommended to those medical auxiliaries whose approach to patients is different from that of the surgeon or physiotherapist, whose chief concern is to give the patient some means of locomotion.

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The World Health Organization has published its fifth year book of *Annual Epidemiological and Vital Statistics* (Geneva, price £2 10s.). The series is a continuation of the annual epidemiological reports published by the Health Organization of the League of Nations during 1922–38. The current volume deals with the year 1952, and its two main parts refer respectively to statistics of causes of death, and the cases of and deaths from communicable diseases. Special tables are included showing deaths from tuberculosis and malignant neoplasm according to site, sex, and age. Deaths of children under 5 from diseases prevalent in that age group are classified according to age and sex. Statistics are not available for the causes of death for the greater part of the world's population: countries with the medical and administrative resources to organize returns are in the minority. The notification of diseases is even less complete. W.H.O. is struggling to obtain a greater accuracy in all epidemiological data so that health activities in underdeveloped countries may be as profitably directed as possible.