Neurology

The term diplegia should be abandoned

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Use of the term has served to confuse classification and obscure interpretation of epidemiological and clinical studies

From the writings of Freud onwards there is broad agreement on the definition of cerebral palsy (CP) but attempts to classify it, based on brain pathology, timing of postulated insult, aetiology, or clinical syndrome, taking account of one or more of neurological findings, distribution, and associated impairments, have been less successful.

Any syndrome must be clearly defined, meaningful, reliable, and used consistently by different people. A few CP syndromes such as choreoathetosis with deafness caused by bilirubin encephalopathy and ataxia caused by hydrocephalus have stood the test of time and are reviewed by Ingram.[1] However, we think that diplegia is not a description of a valid category or syndrome and use of the term has served to confuse classification and obscure interpretation of epidemiological and clinical studies. We draw on historical papers and our own new analyses of recent published epidemiological papers to argue that the term diplegia should be abandoned.

HISTORICAL OVERVIEW OF USE OF TERM DIPLEGIA

Table 1 summarises important classifications of the past 150 years which we shall discuss with respect to their use of the term diplegia.

William Little[2] first described the syndrome complex of cerebral palsy in 1862. His paper proposed a link between abnormal parturition, difficult labour, premature birth, asphyxia neonatorum, and physical deformities, which he described lucidly. He did not use the term diplegia.

In 1890, Sachs and Peterson proposed a classification which linked clinical syndrome to timing of the insult,[3] and introduced diplegia and paraplegia as separate categories.

In 1893, Freud considered cerebral palsy to be caused not just at parturition but also sooner in pregnancy because of "deeper effects that influenced the development of the foetus".[4] Freud was the first to use the term "cerebral diplegia", which covered all bilateral cerebral palsies, including non-spastic types.

Further classifications appeared in the 1950s.[5 6] As table 1 shows, Minear's[6] was more a descriptive grid than a classification, and for him diplegia was "paralysis affecting like parts on either side of the body".

In Ingram's classification of 1955,[7 11] spastic diplegia was described "as a condition of more or less symmetrical paresis of cerebral origin more severe in the lower limbs than the upper and dating from birth or shortly thereafter". He qualified the disorder with an association with prematurity and lesser incidence of mental retardation, pseudobulbar palsy, and seizures compared to quadriplegics. He also described its clinical evolution through the stages of hypotonia, dystonia, and rigidity.[12] Later in his 1966 review article "The neurology of cerebral palsy", he re-emphasised what he regarded as the unequivocal distinction between spastic diplegia and other bilateral cerebral palsies.[13]

In 1959 in England,[8] The Little Club presented a definition and classification of cerebral palsy: "In diplegia there is affection of the muscles of all four limbs. The lower limbs are the more affected." The classification had an additional category "atonic diplegia", but the reasoning presented in the article for this category and other aspects of the classification are difficult to follow. By 1964 the problems were apparent, and an annotation[14] recommended that description should be based on clinical features and that attempts to define certain syndromes combining clinical aetiological and pathological features should be avoided. In particular the idea of a diplegic syndrome should be avoided.

In their first epidemiological report in 1975, the Hagbergs[9] used a classification with a definition of diplegia similar to Ingram's. Hypertonic cases were diplegic where the lower limbs were more affected than the upper ones. Cases were regarded as diplegic even when they exhibited severe generalised damage and "might have been classified as tetraplegia by other investigators". Their classification had an additional category of "ataxic diplegia" where the children had diplegia with ataxic traits, especially dysynergia and intention tremor in the upper limbs. However by 1989, Hagberg[15] appreciated the limitations of his own classification—particularly the unsatisfactory nature of the dividing line between diplegia and quadriplegia and the fact that many children change categories as they grow older.

In 2000, the European Collaboration SCPE[10] did not include diplegia in its classification because of the inherent ambiguities.

Not surprisingly, all this confusion is mirrored in the standard paediatric textbooks.

Forfar and Arniel[16] regard diplegia as a clear entity in which the legs are more affected than the arms; the more affected the upper limbs, the lower is intelligence. It is linked to specific cerebral pathologies, which are themselves linked to premature delivery or asphyxia. In Nelson,[17] diplegia is spasticity of just the legs, with good prognosis for intelligence and seizures. The lesions in the brain are similar in diplegia and quadriplegia, except for more extensive necrotic degeneration of white matter which coalesces into cystic cavities in quadriplegia. In Rudolph,[18] diplegia requires the legs to have greater spasticity and weakness than the arms. The children are frequently preterm and the deficit is more apparent in wrist extensors and activities of daily living such as self feeding, drawing, or writing. Quadriplegia is determined by symmetric impairment of all four extremities; growth retardation, mental retardation, language disorders, and seizures are common. Avery[19] describes spastic diplegia as restricted to bilateral spastic involvement of the lower extremities, often associated with normal cognitive function. Quadriplegia is spastic involvement of the extremities, often with orobuccal musculature rendering swallowing and talking difficult.

THE PRESENT SITUATION

The most recent International Classification of Diseases[20] has a category "spastic diplegia" which it does not further define and another category "diplegia of upper limbs"! However the common ground in the papers we have just discussed appears to be that diplegia is a spastic form of cerebral palsy with lower limbs more affected than upper limbs.

It is however unclear whether presence or absence of prematurity, seizures, or mental retardation is relevant to the definition or just an association. For example, if a child has severe four limb involvement with upper limbs slightly less affected than lower limbs, does presence or absence of the above features such as mental retardation determine whether this is classified as diplegia or quadriplegia?

Even more importantly, what does "more affected" mean and should the comparison be based on clinical signs or function? If comparison is based on

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Table 1 Classifications of cerebral palsy

Reference	Year	Classification		
Little[2]	1862	Hemiplegic rigidity		
		Paraplegic rigidity		
		Generalised rigidity		
		Disordered movements without rigidity		
Sachs and Petersen[3]	1890	Paralysis of intrauterine origin	Diplegia	
			Paraplegia	
			Hemiplegia	
		Birth palsies	Diplegia	
			Paraplegia	
			Hemiplegia	
			Diataxia (ataxia)	
		Acute acquired palsies	Hemiplegia	
			Paraplegia	
			Diplegia	
			Choreo-athetoid	
Freud[4]	1893	Unilateral disorders—hemiplegia	Right or left	
		Bilateral disorders—diplegia	Generalised rigidity	
			Paraplegic rigidity	
			Bilateral hemiplegia	
			Choreo-athetosis	
			Others	
Wyllie[5]	1951	Congenital symmetric diplegia		
		Congenital paraplegia		
		Quadriplegia or bilateral hemiplegia		
		Hemiplegia with additional qualifications referring to all categories	Choreo-athetoid cerebral palsy	
			Mixed forms of cerebral palsy	
			Ataxic cerebral palsy	
			Atonic diplegia	
Minear(6)	1956	A. Physiological	Spasticity, athetosis, rigidity, ataxia, tremor, atonia, mixed, unclassified	
		B. Topographical	Monoplegia, diplegia, paraplegia, hemiplegia, triplegia, quadriplegia	
		C. Aetiological	Prenatal, natal anoxia, postnatal, cause described	
		D. Trauma	Cause described	
		E. Supplemental	Psychological evaluation	
			Physical status, convulsive seizures, posture and locomotive behaviour pattern, eye-hand behaviour pattern, visual status, auditory status, speech disturbances	
		F. Neuroanatomical		
		G. Functional capacity	Class I–IV	
		H. Therapeutic	Class I–IV	
		Neurology	*Extent*	*Severity*
Ingram[7]	1955	Hemiplegia	Right or left	Mild
				Moderate
				Severe
		Double hemiplegia		Mild
				Moderate
				Severe
		Diplegia		
		Hypotonic	Paraplegia	Mild
		Dystonic	Triplegia	Moderate
		Rigid or Spastic	Tetraplegia	Severe
		Ataxia		
		Cerebellar	Unilateral	Mild
		Vestibular	Bilateral	Moderate
				Severe
		Ataxic diplegia		
		Hypotonic	Paraplegia	Mild
		Spastic	Triplegia	Moderate
			Tetraplegia	Severe
		Dyskinesia		
		Dystonic	Monoplegia	Mild
		Choreoid	Hemiplegia	Moderate
		Athetoid	Triplegia	Severe
		Tension	Tetraplegia	
		Tremor		
		Other		
Little Club[8]	1959	Spastic cerebral palsy	Hemiplegia	
			Diplegia	
			Double hemiplegia	
		Dystonic cerebral palsy		
		Choreo-athetoid cerebral palsy		
		Mixed forms of cerebral palsy		
		Ataxic cerebral palsy		
		Atonic diplegia		

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Table 1 continued

Reference	Year	Classification	
Hagberg[9]	1975	Spastic	Hemiplegia Tetraplegia Diplegia
		Ataxic	Congenital Diplegia
		Dyskinetic	Dystonic—mainly Choreo-athetotic
SCPE[10]	2000	Spastic	Bilateral spastic Unilateral spastic
		Ataxic	
		Dyskinetic	Dystonic Choreo-athetotic

clinical signs, tone, tendon reflexes, and power may be easy to separate at the extremes but very difficult when similar. Signs also vary day to day and with the mood of the child. Further, there is a considerable inter- and intraobserver diagnostic variability in the assessment of clinical signs.[21] Ashworth developed a scale, tested for reliability—but this validation was in adults and just for hemiplegia.[22] If comparison is based on function, again this may be straightforward at the extremes but otherwise very difficult. For example, does "walking with difficulty and needing walking aids" mean legs are more or less affected than arms where the child "is not able to write tidily and needs assistance to go to the toilet"?

The North of England Collaborative Cerebral Palsy Survey[23] found such confusion in the application of clinical signs and assessment of functional severity. A local audit of classification of cases of cerebral palsy revealed 98 with diplegia and 130 with quadriplegia. Of those with diplegia, 58 had moderate to severe functional involvement of upper limbs at age 5 but lower limb function was always the same or worse. Five of these children had severe mental retardation. Of those with quadriplegia, six had nil to mild involvement of the upper limbs, but these six had severe mental retardation, suggesting that the clinician used the presence of severe mental retardation to assign the cerebral palsy to the category quadriplegia.

The term diplegia continues to be used in studies which assume its meaning is understood and uniformly applied. For instance, it is claimed that diplegia is associated with and may be caused by premature birth.[24] In this study, Powell *et al* even postulate the existence of a "diplegia factor" which tends to cause premature labour; and impair growth and development if the fetus remains in utero. Their study does not define diplegia and uses the clinical diagnoses from cases notes of children looked after by many paediatricians. In another more recent study, it is suggested that chorioamnionitis may be the cause of spastic diplegia in low birth weight infants—especially in babies with a normal neonatal ultrasound—but again diplegia is not defined.[25]

NEW ANALYSES OF EPIDEMIOLOGICAL STUDIES

We shall show that the confusion, which is clear from our review of papers describing the definitions and practical problems in making sense of them, is born out by our review and further analysis of recent epidemiological studies.

Table 2 shows the overall numbers of cases of cerebral palsy and percentages of different subtypes in studies reported from about 1980—which we take as the date by which neonatal intensive care had been introduced in the reporting regions. We have only included studies where all types and severities of cerebral palsy are reported, and where a whole population is reported either on the basis of birth cohort prevalence or population prevalence. It shows that spastic cases as

Table 2 Proportions of cerebral palsy (CP) subtypes

Reference	Study	Duration	Number of cases of CP	Number of spastic cases of CP	Spastic cases as % of all cases	Unilateral spastic as % of spastic cases	Diplegia as % of spastic cases	Bilateral spastic as % of spastic cases	Diplegia as % of bilateral spastic cases
26	North Italy*	1980–89				35	45	65	69
27, 28	Denmark	1979–90	908	734	81	23	62	77	81
29	North England	1991–96	537	499	93	37	23	63	36
23	Northeast England	1980–96	380	355	93	42	23	58	40
30	Avon, England	1979–88	237	196	83	39	39	61	63
31	Mersey, England	1984–89	497	460	93	39	23	61	38
32	Atlanta, USA	1985–87	204	166	81	34	19	66	28
33, 34, 35	Sweden†	1979–90	545	470	86	40	51	60	84
36	England and Scotland	1984–89	1649	1334	81	35	22	65	33
37	Slovenia	1981–90	768	651	85	33	43	67	64
38	Western Australia	1980–94	819	639	78	45	37	55	68
39	Rome, Italy	1977–96	282	213	76	33	27	67	40
40	Northern Ireland‡	1977–92	960	572	87	43	21	57	36
41	Norway	1980–89	46	39	85	49	33	51	65
42	Oxford, England §	1984–95	967	806	83	40		60	
	Averaged percentage				85	38	33	62	53
	Range				76–93	23–49	19–62	51–77	28–84

*Derived from rates in fig 4 of reference 26.
†Includes 11 acquired cases 1979–82.
‡CP type only available in two thirds of cases.
§Includes 68 acquired cases.

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Table 3 Proportions of cerebral palsy subtypes in low birth weight babies

Reference	Study	<2.5 kg			<1.5 kg		
		Unilateral spastic as % of spastic cases	Diplegia as % of spastic cases	Bilateral spastic as % of spastic cases	Unilateral spastic as % of spastic cases	Diplegia as % of spastic cases	Bilateral spastic as % of spastic cases
26	North Italy	30	55	70			
28	North England	26	32	74	26	36	74
23	Northeast England	30	31	70	35	31	65
31	Mersey, England	30	35	70	33	35	67
32	Atlanta, USA	25	30	75			
36	England and Scotland	25	31	75	26	31	74
38	Western Australia	41	49	59	45	48	55
39	Rome, Italy	25	31	75	22	29	78
42	Oxford, England	30		70	30		70
	Averaged percentage	29	37	71	31	35	69
	Range	25–41	30–55	59–75	22–45	29–48	55–78

a proportion of all cases average 85% (range 76–93%).

Diplegic cases as a proportion of spastic cases average 33% (range 19–62%); whereas diplegic plus quadriplegic cases as a proportion of spastic cases average 62%, with a much smaller range of 51–77%. If one looks just at bilateral spastic cases, the proportion of diplegic cases varies from 28% to 84%—a more than threefold difference. Such a large difference is very unlikely to be a real one and is almost certainly a result of the way in which different centres interpret diplegia.

Table 3 shows data from the studies which report by birth weight. In those less than 2.5 kg, diplegic cases as a proportion of spastic cases average 37% (range 30–55%), only very slightly higher than the average of 33% in all birth weights; and for those less than 1.5 kg, the average is 35%. These findings argue against diplegia being especially associated with either low birth weight or very low birth weight.

DISCUSSION

Semantically, the word "diplegia" must mean two limbed or two sided weakness. If two limbed, it is bizarre that often three or four limbs are involved. If two sided, it is preferable to use a term such as bilateral with no implication about number of limbs involved. This was Freud's original concept—simple but consistent and understandable.

There is no justification for separating diplegia and quadriplegia. While it is possible that the term diplegia as used by Ingram[12] may have represented a distinct clinical syndrome before the introduction of neonatal intensive care, we are confident that it does not now. Our analysis of published epidemiological studies has shown that confusion about and inconsistent application of the term diplegia is embedded unknowingly in the studies. When diplegia and quadriplegia are treated separately, there are very unlikely differences between countries which largely disappear when treated together. Further, the supposed association of diplegia with low birth weight is not seen.

Recent advances in imaging support this position. In her magnetic resonance imaging (MRI) studies of children with spastic cerebral palsy,[43 44] Krageloh-Mann found that periventricular leucomalacia and parasaggital lesions were the commonest lesion in preterm infants and term infants. Very similar MRI appearances were responsible for quadriplegic, diplegic, and triplegic syndromes. Severity of motor involvement was related to severity of white matter reduction but the authors could find no justification for separating these syndromes and therefore used the phrase "bilateral spastic cerebral palsy".

In epidemiological studies, more will be gained from an agreed simple logical classification which is uniformly applied than from a complex one which may have internal inconsistencies, be inconsistently applied and influenced by clinical judgement. Our own preference is for that developed by the European Collaboration.[10]

In clinical studies, there may be a need to define more refined groups of children, but these should be described by their neurological and functional deficits, not by shorthand terms such as diplegia or terms such as "more" or "less affected", when like is not being compared with like. An Oxford group addressed this issue in the 1980s.[45 46] Their systematic description would allow the detail necessary for evaluating the effects of different drugs, operations, or physical therapies. An alternative would be to build on the European classification.[10] For example, the description:

> "spastic, bilateral, neurological signs in four limbs, minimal functional involvement of upper limbs, all cognitive abilities"

would describe reliably a group of children if the term "minimal" was defined for the age group(s) of children being studied.

ACKNOWLEDGEMENTS

We are grateful to all members of Surveillance of Cerebral Palsy in Europe (SCPE) for discussions on this subject over four years.

Arch Dis Child 2003;**88**:286–290

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