Gross and fine motor function and accompanying impairments in cerebral palsy

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The aim of this study was to describe and analyze gross and fine motor function and accompanying neurological impairments in children with cerebral palsy (CP) born between 1991 and 1998 in western Sweden. A population-based study comprised 411 children with a diagnosis of CP ascertained at 4 to 8 years of age. Gross Motor Function Classification System (GMFCS) levels were documented in 367 children (205 males, 162 females). Bimanual Fine Motor Function (BFMF) classification levels of 345 of the children and information on learning disability*, epilepsy, visual and hearing impairments, and hydrocephalus from 353 children were obtained. For spastic CP, a new classification according to the Surveillance of Cerebral Palsy in Europe of uni- and bilateral spastic CP was applied. GMFCS was distributed at Level I in 32%, Level II in 29%, Level III in 8%, Level IV in 15%, and Level V in 16%. The corresponding percentages for BFMF were 30.7%, 31.6%, 12.2%, 11.9%, and 13.6% respectively. Learning disability was present in 40%, epilepsy in 33%, and severe visual impairment in 19% of the children. Motor function differed between CP types. More severe GMFCS levels correlated with larger proportions of accompanying impairments and, in children born at term, to the presence of adverse peri/neonatal events in the form of intracranial haemorrhage/stroke, cerebral infection, and hypoxic–ischaemic encephalopathy. GMFCS Level I correlated positively to increasing gestational age. We conclude that the classification of CP should be based on CP type and motor function, as the two combine to produce an indicator of total impairment load.

*North American usage: mental retardation.

Cerebral palsy (CP), the most common cause of motor disability in children, occurs in around 2 per 1000 liveborn children in western Sweden.[1] In recent years, interest in the motor function of children with CP has increased.[2–4] The now widely used Gross Motor Function Classification System (GMFCS) has proved to be a valid, reliable, and useful prognostic tool.[5–8] A new classification of Bimanual Fine Motor Function (BFMF) has been developed that is designed to match the five levels of the GMFCS.[3] In addition, aspects of functioning, disability, and health other than motor aspects have attracted increasing interest.[9,10] The definition of CP as a pure motor impairment is under debate[11] and a new definition has been proposed.[12]

In many international studies, the Swedish classification of types of CP has been used over the years.[13] However, the definitions of and delineation between spastic tetraplegia and spastic diplegia, and the distinction between severe diplegia and tetraplegia in particular, have been difficult to agree upon internationally.[14] The Surveillance of Cerebral Palsy in Europe (SCPE) has therefore decided to use a simplified classification for spastic children of uni- or bilateral CP.[15,16] The SCPE has also developed a reference manual with a hierarchal diagnostic tree for CP and its subtypes,[16] which offers relatively good reliability (kappa 0.63; Surman G, personal communication 2003). Moreover, the SCPE constructed the concept of 'severe CP' by combining severe motor disability, i.e. inability to walk, with severe learning disability, i.e. IQ<50.[16]

In this paper, gross and fine motor function, as well as accompanying neurological impairments, in a population-based series of children with CP, assessed at age 4 to 8 years, is described. Motor function as an indicator of outcome is analyzed and discussed. To explore the possibility of finding a better way to describe and delimit spastic CP, the new classification of CP, according to SCPE, into uni- and bilateral spastic CP combined with levels of GMFCS, is applied in parallel.

Method

The study comprised 411 children with CP (born between 1991 and 1998) from the western health care region of Sweden, which had a total population of 2.1 million inhabitants and 202 095 live births during the study period.[1,17] All the children had a diagnosis of CP ascertained by the local paediatric neurologist at 4 to 8 years of age. Controversial cases were seen by one or both of the neuropaediatric authors (mainly KH). Eighteen parents among the group of 411 children did not give their consent for reviewing the records, and for 26 children information on GMFCS was missing, leaving 367 children (89%; 205 males, 162 females) for analysis. The missing 44 children did not differ from those included for CP type, sex, or gestational age. The documentation of BFMF levels was available in 345 of the 367 children (94%), whereas documentation about accompanying impairments was available for 353 (96%). Of the 367 children, 154 (41%) were born preterm, defined as birth before 37 completed weeks of gestation, and 213 at term, none of whom were born after more than 42 weeks.

The definition of CP was that agreed on at an international consensus meeting in 1990, i.e. 'an umbrella term covering a group of non-progressive, but often changing, motor impairment syndromes secondary to lesions or anomalies of the brain arising in the early stages of development',[13] which is also compatible with the definition formulated by Bax.[18] The Swedish classification of CP types was applied.[13,19] Children with spastic CP were also classified according to the SCPE into unilateral or

bilateral spastic CP.[16] Peri/neonatal was defined as the period from the onset of labour leading to delivery through to the 28th day of life. Excluding cases with major prenatal anomaly or lesion, cases of verified intracranial haemorrhage/stroke, cerebral infection (viral or bacterial meningitis/meningoencephalitis), and hypoxic-ischaemic encephalopathy (HIE) were judged as peri/neonatal adverse events. The criteria of HIE were that two of the following symptoms/signs should be present in children born at no less than 34 weeks of gestation: Apgar scores less than 4 at 1 or 5 minutes, need for respirator treatment in the first week, and neonatal seizures before day 3. The GMFCS[5] and the BFMF classifications[3] were performed when the child was 5 to 6 years old (Table I). The GMFCS has been reported to be relatively stable over time and, in general, a child will stay at the same level of the GMFCS from age 1 to 2 years to 6 to 12 years.[20]

Severe CP was defined as severe learning disability combined with an inability to walk, corresponding to GMFCS Levels IV to V. The following neurological impairments were recorded: learning disability, defined as mild in children with an estimated or measured IQ of 50 to 70 and severe if the IQ was less than 50 (IQ measured by Wechsler[27,28] or Griffith[29] scales or estimated from clinical observation); epilepsy, defined as a diagnosis of active epilepsy at 4 to 8 years of age; visual impairment, defined as functional blindness or an acuity after correction of refraction errors of no more than 0.3 (20/60) in the better eye; hearing impairment, defined as the need for a hearing aid or no hearing. Infantile hydrocephalus was defined as a diagnosis of surgically treated expansive hydrocephalus in the first year of life.

STATISTICS

The χ^2 test for contingency tables and the χ^2 test for trend were used. All the statistics and *p*-values have been interpreted in a descriptive manner.

ETHICS

The study was approved by the Ethics Committee at the Medical Faculty at the University of Göteborg. Informed consent was obtained for all 367 children included in the study.

Results

GROSS MOTOR FUNCTION

The gross motor function expressed as GMFCS was at Level I in 116 (32%), at Level II in 108 (29%), at Level III in 30 (8%), at Level IV in 56 (15%), and at Level V in 57 (16%) of the 367 children (Fig. 1; Table II). The distribution differed significantly between CP types ($p<0.001$).

In children with spastic hemiplegia, 127 of 134 (95%) were classified at GMFCS Levels I to II, with most (66%) at Level I. In children with spastic diplegia, 78 of 144 (54%) were classified at GMFCS Levels I to II, with most (38%) at Level II, whereas 45 (31%) performed at Levels IV and V. All 23 children with spastic tetraplegia had severe motor impairments: two at GMFCS Level IV, and the remaining 21 at Level V. The only two children at Level IV in the tetraplegic group were characterized by spasticity and dyskinesia in combination. Among the children with dyskinetic CP, only 2 of 51 (4%) were assigned to GMFCS Level I, whereas 26 (50%) were assigned to Level V. All but one child with ataxia were affiliated to GMFCS Levels I to II, with most (12/14) at Level II.

The distribution of GMFCS levels by gestational age in the 367 children is shown in Figure 2. The proportion of children with the mildest motor impairment, i.e. GMFCS Level I, increased with gestational age ($p<0.001$).

In children born at term, the distribution of GMFCS levels was significantly associated with the occurrence of adverse peri/neonatal events, i.e. intracranial haemorrhage/stroke, cerebral infection, and HIE. Thirty of 72 such children (42%) were able to walk independently (GMFCS Levels I–II) as opposed to 107 of 141 children (76%) without these complications

Table I: Criteria for five levels of Gross Motor Function Classification System (GMFCS)[5] and Bimanual Fine Motor Function (BFMF)[3], relevant for ages studied

GMFCS	*BFMF*
Level I Walks without restrictions, limitations in more advanced gross motor skills	Level I One hand: manipulates without restrictions. The other hand: manipulates with restrictions or limitations in more advanced fine motor skills.
Level II Walks without restrictions, limitations walking outdoors and in the community	Level II (a) One hand: manipulates without restrictions. The other hand: only ability to grasp or hold. (b) Both hands: limitations in more advanced fine motor skills.
Level III Walks with assistive mobility devices, limitations walking outdoors and in the community	Level III (a) One hand: manipulates without restrictions. The other hand: no functional ability. (b) One hand: limitations in more advanced fine motor skills. The other hand: only ability to grasp or worse.
Level IV Self-mobility with limitations, children are transported or use power mobility outdoors and in the community	Level IV (a) Both hands: only ability to grasp. (b) One hand: only ability to hold. The other hand: only ability to hold or worse.
Level V Self-mobility is severely limited, even with the use of assistive technology	Level V Both hands: only ability to hold or worse.

($p<0.001$). This difference originated especially from the subgroup of children with a HIE, of whom only 21 of 56 (38%) were able to walk without aid.

BIMANUAL FINE MOTOR FUNCTION

The distribution of BFMF levels in 345 children is shown in Figure 3: 106 (30.7%) performed at level I, 109 (31.6%) at level II, 42 (12.2%) at level III, 41 (11.9%) at level IV, and 47 (13.6%) at level V. The distribution differed significantly between types of CP ($p<0.001$) and largely followed the pattern of GMFCS. The BFMF level was consistent with that of GMFCS in 198 of 345 (57%) children (Table II). Hemiplegia dominating in the arm with the BFMF level exceeding the GMFCS level was found in 55 of 130 (42%) children. The correlation found between GMFCS levels and gestational age at birth could not be demonstrated between BFMF levels and gestational age. However, as was the case for gross motor function, there was a corresponding correlation between peri/neonatal compromise and BFMF levels ($p<0.001$).

ACCOMPANYING IMPAIRMENTS

Learning disability was present in 141 of 353 (40%) children, epilepsy in 118 (33%), severe visual impairment in 66 (19%), hearing impairment in seven (2%), and hydrocephalus in 25 (7%).

The proportion of children with accompanying impairments increased significantly with GMFCS levels ($p<0.001$; Fig. 4). In children with motor function at GMFCS Level I, 91 of 115 (79%) had no accompanying impairment, contrasting with 3 of 54 (6%) of those at GMFCS Level V. In children performing at GMFCS Level V, 48 of 54 (89%) had two or more accompanying impairments.

The distribution of accompanying impairments by type of CP is shown in Table III. Types of CP characterized by milder impairments of gross motor function, i.e. hemiplegia and ataxia, had fewer accompanying impairments. At least one impairment was present in 42 (31%) of 134 children with hemiplegia, in 81 of 137 (59%) of those with diplegia, and in all the children with tetraplegia. Learning disability was milder in children with hemiplegia and diplegia than in the other types of CP. Epilepsy was most frequent in tetraplegia (18/20; 90%), followed by dyskinetic CP (26/50; 52%). Hydrocephalus was predominantly found in children with diplegia (16/137; 11%) and tetraplegia (3/20; 15%).

Table II: Association between Gross Motor Function Classification System (GMFCS)[5] and Bimanual Fine Motor Function (BFMF)[3] in 367 children with cerebral palsy (CP)

CP type	*BFMF*						*Total*
	I	*II*	*III*	*IV*	*V*	*BFMF unknown*	
GMFCS I							
Hemiplegia	41	39	5	–	–	4	89
Diplegia	21	2	–	–	–	1	24
Dyskinetic CP	–	2	–	–	–	–	2
Ataxia	–	1	–	–	–	–	1
Subtotal	62	44	5	–	–	5	116
GMFCS II							
Hemiplegia	9	18	11	–	–	–	38
Diplegia	26	22	2	–	–	4	54
Dyskinetic CP	–	3	1	–	–	–	4
Ataxia	2	9	1	–	–	–	12
Subtotal	37	52	15	–	–	4	108
GMFCS III							
Hemiplegia	–	–	3	–	–	–	3
Diplegia	5	7	4	–	–	5	21
Dyskinetic CP	–	–	4	1	–	–	5
Ataxia	–	–	1	–	–	–	1
Subtotal	5	7	12	1	–	5	30
GMFCS IV							
Hemiplegia	1	–	–	–	–	–	1
Diplegia	1	6	7	19	2	3	38
Tetraplegia	–	–	–	–	2	–	2
Dyskinetic CP	–	–	–	12	2	1	15
Subtotal	2	6	7	31	6	4	56
GMFCS V							
Hemiplegia	–	–	2	1	–	–	3
Diplegia	–	–	–	4	3	–	7
Tetraplegia	–	–	–	2	19	–	21
Dyskinetic CP	–	–	1	2	19	4	26
Subtotal	–	–	3	9	41	4	57
Total	106	109	42	41	47	22	367

Children with unknown BFMF levels ($n=22$) are shown by GMFCS level. Difference between CP types was significant. Levels II–III and IV–V were combined. GMFCS: $\chi^2=232.4$, degrees of freedom (df)=8, $p<0.001$; BFMF: $\chi^2=195.5$, df=8, $p<0.001$.

The distribution of accompanying impairments by gestational age is shown in Table IV. The children born before 28 weeks of gestation had the highest proportion of impairments. In this group, 19 of 37 had a learning disability with an IQ<70 and 12 had epilepsy, 10 were severely visually impaired, and nine had hydrocephalus. Children born at term were less frequently affected in every aspect except for epilepsy.

The distribution of accompanying impairments in relation to adverse peri/neonatal events in children born at term is shown in Table V. Accompanying impairments were present in 49 of 71 of the children (69%) with peri/neonatal compromise, and 29 (41%) had two or more. In children without peri/neonatal compromise, 49 of 136 (36%) had accompanying impairments and 29 (21%) had two or more. The proportions of learning disability, epilepsy, and severe visual impairment were higher in children with peri/neonatal compromise than in those without.

When the SCPE concept of 'severe CP' was applied to the children in this study, it was found that 65 of 353 (18%) fulfilled the combined criteria for non-ambulatory and severe learning disability. The group comprised three children with unilateral spastic CP, 41 children with bilateral spastic CP, and 21 children with dyskinetic CP. An additional 37 children were non-ambulatory with normal cognitive function or mild learning disability, and 10 ambulatory children had a severe learning disability.

TYPES OF CP

The new SCPE classification of spastic CP was applied. When the CP types spastic diplegia and spastic tetraplegia were combined into one entity, that of bilateral spastic CP, 24 of 167 (14%) performed at GMFCS Level I, 54 (32%) at Level II, and 21 (13%),

Table III: Distribution of accompanying impairments by cerebral palsy (CP) type in 353 children with CP

	Hemiplegia (n=134) n (%)	*Diplegia (n=137) n (%)*	*Tetraplegia (n=20) n (%)*	*Dyskinetic CP (n=50) n (%)*	*Ataxia (n=12) n (%)*	*Total (n=353) n (%)*
Learning disability	22 (17)	64 (49)	20 (100)	30 (62)	5 (50)	141 (40)
Mild	15	37	0	9	5	66
Severe	7	27	20	21	0	75
Epilepsy	30 (23)	43 (34)	18 (87)	26 (52)	1 (7)	118 (33)
Severe visual impairment	10 (8)	26 (21)	17 (83)	12 (27)	1 (8)	66 (19)
Number of accompanying impairments						
0	92 (66)	56 (41)	0	14 (28)	7 (50)	169 (48)
1	23 (17)	36 (26)	0	11 (22)	3 (36)	73 (21)
≥2	19 (15)	45 (32)	20 (100)	25 (50)	2 (14)	111 (31)

Difference between types of CP was significant (χ^2=83.6, degrees of freedom=8, p<0.001).

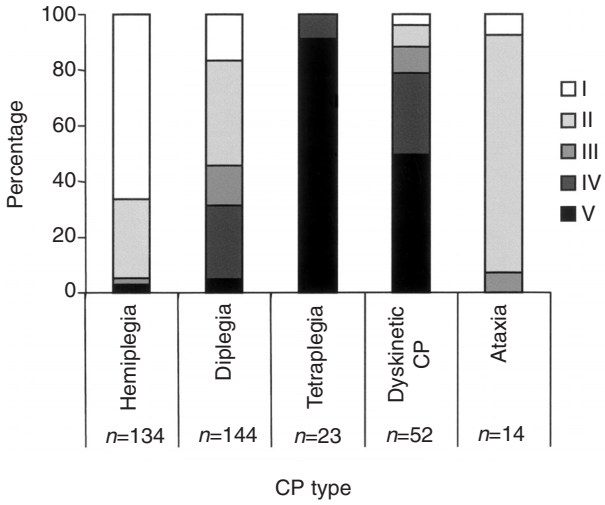

Figure 1: *Distribution of Gross Motor Function Classification System levels by type of cerebral palsy (CP) in 367 children with CP.*

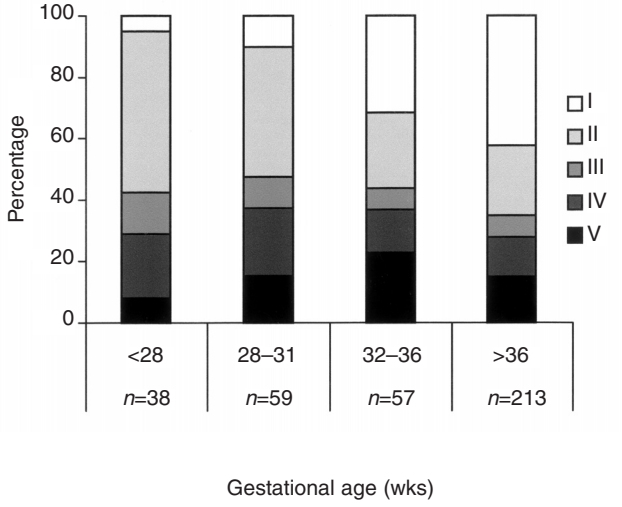

Figure 2: *Distribution of Gross Motor Function Classification System (GMFCS) levels by gestational age in 367 children with cerebral palsy. Proportion of children with GMFCS Level I increased significantly with gestational age (χ^2_{trend}=34.78, degrees of freedom=1, p<0.001).*

40 (24%), and 28 (17%) at Levels III, IV, and V respectively. A comparison between the Swedish classification of diplegia/tetraplegia and the classification of bilateral CP according to SCPE revealed that, at GMFCS Levels I, II, and III, all 99 cases were classified as diplegia, at Level IV 38 of 40 (95%), and at Level V seven of 28 (25%). The CP type tetraplegia comprised 21 of 23 at GFMCS level V and the remaining two at level IV, both with fine motor function at BFMF level V.

Discussion

The definition of CP as a pure motor impairment is under debate[11] as the various brain lesions causing the motor dysfunction often also directly or indirectly impair sensation, vision, cognition, communication, and behaviour and may cause epilepsy. A definition of CP including the naming of some accompanying impairments was put forward by the participants at an international workshop on the definition and classification of CP, held in Washington, USA, in July 2004.[12] Consensus in classification, for the CP concept and its different syndromes,[16,21] is important to avoid misconceptions about the aetiology and severity of disability. Population-based series are necessary for international comparisons and epidemiological studies of trends.[4] Only in well-defined populations can aspects such as prevention, the prevalence of perception and behavioural problems, as well as participation[3] and the provision of health care be studied. In western Sweden, population-based studies of CP of this kind have monitored not only prevalence and aetiology but also functional aspects of CP and its accompanying impairments for many years in succession.[1,19,22] The development of new, reliable, and valid

Table IV: Accompanying impairments by gestational age (GA) completed weeks in 353 children with cerebral palsy

	GA<28wks (n=37) n (%)	*GA 28–31wks (n=55) n (%)*	*GA 32–36wks (n=55) n (%)*	*GA>36wks (n=206) n (%)*	*Total (n=353) n (%)*
Learning disability	19 (51)	25 (44)	24 (43)	73 (37)	141 (40)
Mild	11	14	10	31	66
Severe	8	11	14	42	75
Epilepsy	12 (32)	16 (28)	17 (30)	73 (36)	118 (33)
Severe visual impairment	10 (27)	17 (30)	13 (20)	26 (14)	66 (19)
Number of accompanying impairments					
0	13 (35)	23 (42)	24 (44)	109 (53)	169 (48)
1	9 (24)	13 (24)	12 (22)	39 (19)	73 (21)
≥2	15 (41)	19 (35)	19 (35)	58 (28)	111 (31)

Proportion of children with accompanying impairments increased significantly with lower gestational age (χ^2_{trend} 5.47, degrees of freedom=1, $p<0.05$).

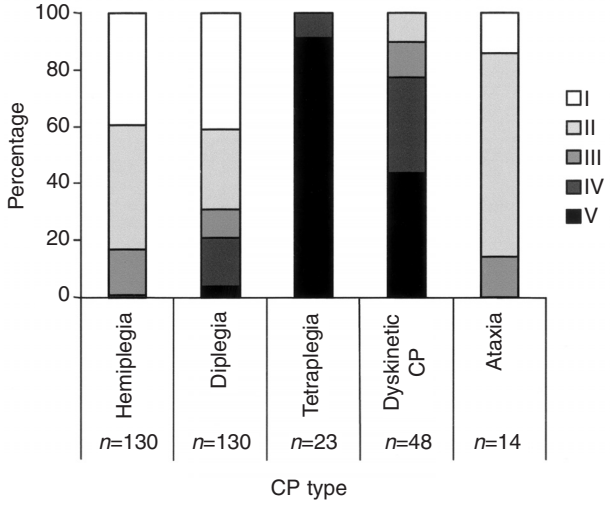

Figure 3: *Distribution of Bimanual Fine Motor Function levels by type of cerebral palsy (CP) in 345 children with CP.*

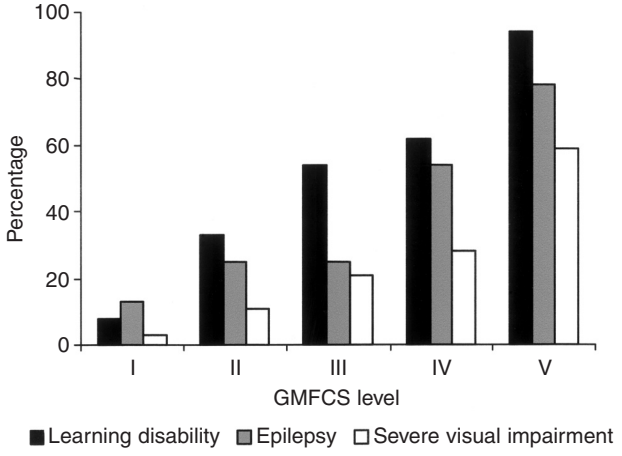

Figure 4: *Proportion of learning disability, epilepsy, and severe visual impairment by Gross Motor Function Classification System (GMFCS) levels in 353 children with cerebral palsy. The proportion increased significantly with GMFCS levels (χ^2_{trend} for learning disability=127.14, degrees of freedom [df]=1, $p<0.001$; χ^2_{trend} for epilepsy=77.99, df=1, $p<0.001$; and χ^2_{trend} for severe visual impairment=73.59, df=1, $p<0.001$).*

measures of function has facilitated the recording of gross motor function[5,6] and, in accordance with an emerging interest,[4] fine motor function was recently added.[3] The focus on accompanying impairments is also growing, as we learn more about perception and specific learning difficulties and how to assess them. As a result of this more comprehensive way of looking at CP, the definition of CP itself has been questioned[11,12] but too much attention must not be drawn from the motor impairment, which is still the core feature of CP.

This article, which covers the birth years 1991 to 1998, reveals that the distribution of gross motor function, expressed as GMFCS levels, was similar to that reported by Nordmark et al.,[23] with 75% at Levels I to III and 25% at Levels IV to V, corresponding to 70% and 31% in this study. The level of GMFCS was found to correlate strongly to at least three accompanying impairments present in CP, namely learning disabilities, visual impairment, and epilepsy. The GMFCS can, therefore, be used as an indicator of total disability load.

A factor of importance for gross motor function in this study was gestational age, where a positive trend was found between increasing gestational age and the percentage of children with the mildest motor impairment. When accompanying impairments were considered, the children born before 28 weeks' gestation had the highest percentages of all impairments. This is in accordance with Marlow et al.,[24] who found that cognitive and neurological impairments were very common in children born before 26 weeks' gestation.

Children born at term with peri/neonatal compromise (intracranial haemorrhage/stroke, cerebral infection, and HIE) appeared to be particularly burdened when it came to the severity of the resulting fine and gross motor impairment, as well as the accompanying impairment load.

Although standardized classification systems and the subgrouping of children with CP are frequently discussed and used, there is still some confusion about the distinction between severe diplegia and tetraplegia; the latter is sometimes also called quadriplegia, tetraparesis, or double hemiplegia. In this case, the SCPE classification of bilateral spastic CP can provide structural benefit and simplify diagnosis. By combining the new CP classification of bilateral spastic CP with GMFCS, it was possible in this study to identify the most disabling subgroup of spastic CP according to the Hagberg classification,[19] namely spastic tetraplegia, as 21 of 23 of these children were classified as bilateral spastic CP at GMFCS Level V. The two remaining children with tetraplegia were classified at GMFCS Level IV and performed at BFMF Level V, thus fulfilling the criterion for tetraplegia defined as severely impaired motor function with fine motor function equal to or worse than the gross motor function. A further sub-classification of CP based on limb distribution has been proposed, but Gorter et al.[25] showed that this did not add prognostic value to classification with the GMFCS.

A concept with the focus on function, created by the SCPE and not previously used in CP studies in western Sweden, was that of 'severe CP', defined as non-ambulant CP corresponding to GMFCS levels IV to V, combined with severe learning disability. In this study, the percentage of those with severe CP would have been 18%, compared with 20% in the SCPE survey.[16] However, a concept of this kind should be used with care, as some children with an isolated motor impairment may be extremely disabled. Consequently, the number of children with a severe disability may be underestimated if the SCPE definition of severe CP is used.

In their recent review of CP research, Koman et al.[26] reported that more than 50% of children with CP in the 8- to 17-year age range could walk without aid, 25% could not walk, and 30% were cognitively impaired. Our corresponding findings were 61% independent walkers, 31% unable to walk, and 40% with learning disabilities. As Bax pointed out,[4] these differences may occur when wide or undefined age ranges are used. Our study focused on the 4- to 8-year age range, an age at which a diagnosis of CP can be reliably certified and most paediatric accompanying impairments can be identified. Furthermore, these preschool and early school ages are important for planning and providing support for the optimal schooling of the child with CP.

In conclusion, the classification of CP should be based on CP type and motor function, as the two combine to produce a relevant indicator of the total impairment load. In addition, gestational age and peri/neonatal morbidity can supply prognostic information. The concept of uni- and bilateral spastic CP, combined with GMFCS, adds structure and comprehension to the classification of CP.

Table V: Accompanying impairments by adverse peri/neonatal events (intracranial haemorrhage/stroke, cerebral infection, or hypoxic–ischaemic encephalopathy) in 207 children with cerebral palsy born at term

Adverse peri/neonatal events	*Present n=71 n (%)*	*Not present n=136 n (%)*	*Total n=207 n (%)*
Learning disability[a]	37 (52)	36 (26)	73 (35)
Mild	13	18	31
Severe	24	18	42
Epilepsy[b]	36 (51)	37 (27)	73 (35)
Severe visual impairment[c]	13 (18)	13 (10)	26 (13)
Number of accompanying impairments[d]			
0	22 (31)	87 (64)	109 (53)
1	20 (28)	20 (15)	40 (19)
≥2	29 (41)	29 (21)	58 (28)

Differences between groups: [a]$\chi^2=13.44$, degrees of freedom (df)=1, $p<0.001$; [b]$\chi^2=11.28$, df=1, $p<0.001$; [c]$\chi^2=3.25$, df=1, $p=0.07$; [d]$\chi^2=13.44$, df=2, $p<0.001$.

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