


REGULAR ARTICLE

One-third of school-aged children with cerebral palsy have neuropsychiatric impairments in a population-based study

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Keywords

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ABSTRACT

Aim: To describe motor function and associated impairments, particularly autism spectrum disorder (ASD) and attention-deficit/hyperactivity disorder (ADHD), in school-aged children with cerebral palsy (CP).

Methods: Population-based study of all children with CP born 1999–2006 from the county of Västra Götaland, Sweden; 264 children (141 males, 123 females). Information was obtained from the CP Register of western Sweden (data collected at 4–8 years of age) and all medical records at 10–17 years of age.

Results: Cerebral palsy was spastic in 76%, dyskinetic in 17% and ataxic in 7% of all children. Sixty-three per cent were independent walkers. Associated impairments were present in 75%. Vision was impaired in 19%, hearing in 8% and speech in 54%. Intellectual disability (ID) was present in 53% and epilepsy in 41%. ID had increased from 42% to 53% since preschool-age. Neuropsychiatric impairments were present in 32% of the children; ASD in 18%; and ADHD in 21%. All impairments, except for ASD and ADHD, increased with more severe motor impairment.

Conclusion: Three in four school-aged children with CP have associated impairments, underscoring the need to broadly assess every child. The high rate of ASD and ADHD points to the importance of in-depth studies of such impairments in CP.

INTRODUCTION

Cerebral palsy (CP) is a clinical entity with a very variable functional profile. Early disturbances in the developing brain result in the motor disorder of CP, which is 'often accompanied by disturbances of sensation, cognition, communication, perception and/or behaviour, and/or by a seizure disorder', as emphasised in the most recent definition of CP (1). These disturbances are sometimes more disabling than the motor disorder (2–4). Some impairments become apparent with age, since some abilities, such as reading, only develop later in childhood.

Intellectual disability (ID) is reported in 30–50% of children with CP (5,6). Earlier studies from the CP Register of western Sweden have shown ID in 40–45%, with a clear

association with severity of gross motor impairment and epilepsy (2,3), concurrent with other studies (7).

Visual impairment is common in children with CP, including problems with visual acuity, perception and eye motility (2,3,5). Hearing may also be impaired in CP. Epilepsy is reported in 25–40% of children with CP (2,3,7).

Speech disorders occur in more than half of all children with CP (8,9). Communication problems have come into focus during the last decade with the development of new classification scales for both speech and communication (10,11).

Abbreviations

ADHD, Attention-deficit/hyperactivity disorder; ASD, Autism spectrum disorder; BFMF, Bimanual fine motor function; BSCP, Bilateral spastic cerebral palsy; CP, Cerebral palsy; DSM, Diagnostic and statistical manual of mental disorders; GMFCS, Gross motor function classification system; ICD, International classification of diseases and related health problems; ID, Intellectual disability; IQ, Intelligence quotient; MACS, Manual ability classification system; USCP, Unilateral spastic cerebral palsy; VSS, Viking speech scale.

Key notes

- Associated impairments in cerebral palsy (CP) are common, but the occurrence of autism spectrum disorder (ASD) and attention-deficit/hyperactivity disorder (ADHD) is less studied.
- One-third of school-aged children with CP had been diagnosed with ASD and/or ADHD, with rates (ASD 18% and ADHD 21%) higher than described in earlier population-based studies.
- ASD and ADHD may still be underdiagnosed in CP and should be assessed in all children with CP.

Behaviour problems are also common in children with CP (12,13), including neuropsychiatric disorders such as autism spectrum disorder (ASD) and attention-deficit/hyperactivity disorder (ADHD). During the last decade, there has been growing awareness particularly of ASD being overrepresented in children with CP, reported from 3% to 16% (14–21), depending on the group studied and the methods used. In a European multicentre CP study by Delobel-Ayoub et al, male sex, epilepsy, ID and better walking ability were associated with ASD in children with CP (20).

There are also data suggesting that attention problems may be more frequent in children with CP than in the general population. In a Norwegian study, 50% of children with CP at Gross Motor Function Classification System (GMFCS) (22) level I-IV would meet the criteria for ADHD (21).

Associated impairments are insufficiently studied in older children with CP. In particular, there is a need for more population-based knowledge on ASD and ADHD in CP.

The aims of this study were (i) to describe motor function and the occurrence of associated impairments, particularly ASD and ADHD, (ii) to examine the associations between CP type, motor function and associated impairments in a population-based group of school-aged children with CP and (iii) to compare with the occurrence of associated impairments in the same children at preschool-age.

PATIENTS AND METHODS

The study population comprised eight birth-year cohorts of children and adolescents with CP born 1999–2006 from the CP Register of western Sweden (23,24), but limited to the county of Västra Götaland, Sweden. Of 281 children and adolescents, 11 had died, three had moved abroad and three children were no longer considered having CP, leaving 264 children for the present study. Function and associated impairments in the children born 1999–2002 (120 out of 264 children) have earlier been reported at a younger age (3).

Gestational age groups were considered: extremely preterm (birth occurring at less than 28 completed gestational weeks), very preterm (28–31 weeks), moderately preterm (32–36 weeks) and term (more than 36 weeks).

Information about CP type, gross motor function, fine motor function, gestational age and associated impairments at 4–8 years of age was taken from the CP register. All available medical and habilitation records were scrutinised to retrieve updated information about intellectual level, vision, hearing, epilepsy, speech ability, language disorder, ASD and ADHD. Data collection took place from January to March 2017 regarding the conditions on 31 December 2016, when the study population was 10–17 years of age (median 13 years 8 months). The data extraction procedure was the same as for the CP register.

Cerebral palsy types were classified according to the Surveillance of Cerebral Palsy in Europe (SCPE); into unilateral spastic CP (USCP), bilateral spastic CP (BSCP), dyskinetic CP and ataxic CP. Gross and fine motor function

was classified with the GMFCS, the Bimanual Fine Motor Function (BFMF) classification and the Manual Ability Classification System (MACS), respectively (22,25,26).

Intellectual level was defined as normal if intelligence quotient (IQ) was ≥ 85 , and borderline intellectual functioning if IQ was 70–84. Intellectual disability, ID (term according to Diagnostic and Statistical Manual of Mental Disorders (DSM)-5), was defined according to International Classification of Diseases and Related Health Problems – Tenth Revision (ICD-10); mild (IQ 50–69), moderate (IQ 35–49), severe (IQ 20–34) and profound (IQ < 20). IQ, or developmental quotient, had been measured by Wechsler scales or Griffith developmental scales or estimated on the basis of clinical observation. The results from psychological tests had been complemented with results from adaptive behaviour scales.

Visual impairment was defined as an acuity of not more than 0.3 in the best eye with correction, and severe visual impairment was defined as an acuity of not more than 0.1 in the best eye with correction or the presence of functional blindness. Hearing impairment included sensorineural impairment or deafness, unilateral or bilateral. Epilepsy was defined as epilepsy under treatment according to the medical records.

Speech was classified with the Viking Speech Scale (VSS) (11); level I not affected speech, II imprecise speech, III unclear speech and IV no understandable speech. Children at level III and IV were regarded having a severe speech impairment. Language disorder diagnoses (F80 according to ICD-10) in records were documented.

Neuropsychiatric diagnoses in this study included ASD and ADHD. These were derived from the medical records and had been coded according to ICD-10. In the clinical setting, the diagnostic criteria used were those of the DSM-IV or, more recently, DSM-5. Three autism spectrum diagnoses were found: autistic disorder (F840), atypical autism (F841) and Asperger syndrome (F845). ASD is used here for all three categories. ADHD diagnoses had been coded with F90, and subtypes were noted if mentioned. The neuropsychiatric diagnoses had been made by child psychiatrists or paediatric neurologists.

Ethics

The study was approved by The Regional Ethical Review Board in Gothenburg.

Statistical analysis

For contingency tables, the χ^2 test for independence was used. In the comparison between cohorts, the Cochran–Armitage χ^2 test for trends was used. Spearman's rank correlation (ρ) was used to analyse the relation between classification scales.

RESULTS

Gross and fine motor function, associated impairments, sex distribution and gestational age are presented by type of CP in Table 1.

Table 1 Distribution of sex, gestational age, motor function and associated impairments by type of cerebral palsy (CP) in a population-based group of 264 school-aged children from western Sweden born 1999–2006

CP type	Unilateral spastic CP n = 103 n (%)	Bilateral spastic CP n = 98 n (%)	Dyskinetic CP n = 45 n (%)	Ataxic CP n = 18 n (%)	Total n = 264 n (%)
Sex					
Male	58 (56)	52 (53)	23 (51)	8 (44)	141 (53)
Female	45 (44)	46 (47)	22 (49)	10 (56)	123 (47)
Gestational age					
w23-27	7 (7)	17 (17)	1 (2)	1 (5)	26 (10)
w28-31	10 (10)	16 (16)	3 (7)	1 (6)	30 (11)
w32-36	13 (12)	26 (27)	4 (9)	2 (11)	45 (17)
w37-42	73 (71)	39 (40)	37 (82)	14 (78)	163 (62)
GMFCS					
I	93 (90)	25 (26)	1 (2)	8 (44)	127 (48)
II	10 (10)	17 (17)	6 (13)	7 (39)	40 (15)
III	0 (0)	14 (14)	4 (9)	2 (11)	20 (8)
IV	0 (0)	26 (27)	8 (18)	1 (6)	35 (13)
V	0 (0)	16 (16)	26 (58)	0 (0)	42 (16)
BFMF					
I	58 (56)	32 (33)	1 (2)	6 (33)	97 (37)
II	31 (30)	22 (23)	4 (9)	10 (56)	67 (25)
III	14 (14)	18 (18)	7 (16)	2 (11)	41 (16)
IV	0 (0)	11 (11)	13 (29)	0 (0)	24 (9)
V	0 (0)	15 (15)	20 (44)	0 (0)	35 (13)
MACS					
I	58 (56)	28 (29)	1 (2)	6 (33)	93 (35)
II	30 (29)	16 (16)	3 (7)	10 (56)	59 (22)
III	13 (13)	14 (14)	7 (15)	1 (5)	35 (13)
IV	2 (2)	21 (22)	9 (20)	1 (6)	33 (13)
V	0 (0)	19 (19)	25 (56)	0 (0)	44 (17)
Visual impairment					
No	99 (96)	66 (67)	30 (67)	18 (100)	213 (81)
Yes, not severe	3 (3)	11 (11)	4 (9)	0 (0)	18 (7)
Yes, severe	1 (1)	21 (22)	11 (24)	0 (0)	33 (12)
Hearing impairment					
No	96 (93)	89 (91)	42 (93)	16 (89)	243 (92)
Sensorineural	7 (7)	9 (9)	3 (7)	2 (11)	21 (8)
Intellectual level					
Normal	57 (55)	26 (27)	8 (18)	7 (39)	98 (37)
Borderline	17 (17)	8 (8)	0 (0)	1 (5)	26 (10)
Mild ID	24 (23)	21 (22)	7 (15)	5 (28)	57 (22)
Moderate ID	2 (2)	10 (10)	4 (9)	3 (17)	19 (7)
Severe ID	3 (3)	17 (17)	10 (22)	2 (11)	32 (12)
Profound ID	0 (0)	16 (16)	16 (36)	0 (0)	32 (12)
VSS					
I	84 (82)	34 (35)	1 (2)	3 (17)	122 (46)
II	16 (15)	27 (27)	5 (11)	10 (55)	58 (22)
III	1 (1)	6 (6)	7 (16)	3 (17)	17 (7)
IV	2 (2)	31 (32)	32 (71)	2 (11)	67 (25)
Epilepsy					
No	70 (68)	48 (49)	10 (22)	9 (50)	137 (52)
Previous	8 (8)	6 (6)	3 (7)	1 (6)	18 (7)
Active	25 (24)	44 (45)	32 (71)	8 (44)	109 (41)
ASD	15 (15)	23 (23)	4 (9)	5 (28)	47 (18)
ADHD	28 (27)	12 (12)	7 (16)	8 (44)	55 (21)
ASD and ADHD	7 (7)	6 (6)	1 (2)	4 (22)	18 (7)
No identified associated impairment	44 (43)	16 (16)	4 (9)	1 (6)	65 (25)

ADHD = Attention-Deficit/Hyperactivity Disorder; ASD = Autism Spectrum Disorder; BFMF = Bimanual Fine Motor Function; GMFCS = Gross Motor Function Classification System; ID = Intellectual Disability; MACS = Manual Ability Classification System; VSS = Viking Speech Scale.

The group of 264 children comprised of slightly more boys than girls. A majority was born at term. Spastic CP was found in 201 children (76%). The unilateral spastic CP was right-sided in 47 and left-sided in 56 children.

Gross and fine motor function

According to the GMFCS, 63% were at level I-II, that is independent walkers. The distribution of GMFCS levels varied between CP types. Children with dyskinetic CP were the most severely impaired with 76% at GMFCS level IV-V. In contrast, all children with USCP were at GMFCS level I or II. There was no change in distribution of GMFCS levels from preschool till school age.

Fine motor function according to the BFMF and MACS was at corresponding levels in 75% ($\rho = 0.91$; $p < 0.001$). Fine motor function corresponded with GMFCS levels in 55% using BFMF, and 61% using MACS.

The severity of gross motor impairment did not differ between children born at term and preterm, while the distribution of CP types did; BSCP was more common in

children born preterm, while dyskinetic CP was most prevalent in those born at term.

Associated impairments according to the definition of cerebral palsy

Proportion of associated impairments are presented by GMFCS levels in Figure 1A and by type of CP in Figure 1B.

Cognition

Intellectual level had been tested in 204 of the 264 children on one to five occasions. Tests performed at the latest testing (at two to 17 years of age) were Wechsler Intelligence Scale for Children (WISC)-IV in 100 children, Wechsler Preschool and Primary Scale of Intelligence (WPPSI)-III in 72 and Griffith Mental Development Scales in 32. In 60 children (23%), the intellectual level had been estimated through clinical observation; as normal in 36 children, severe ID in six and profound ID in 18 children.

Intellectual level was normal or borderline in 124 (47%) of the 264 children, and 140 (53%) had ID. The proportion

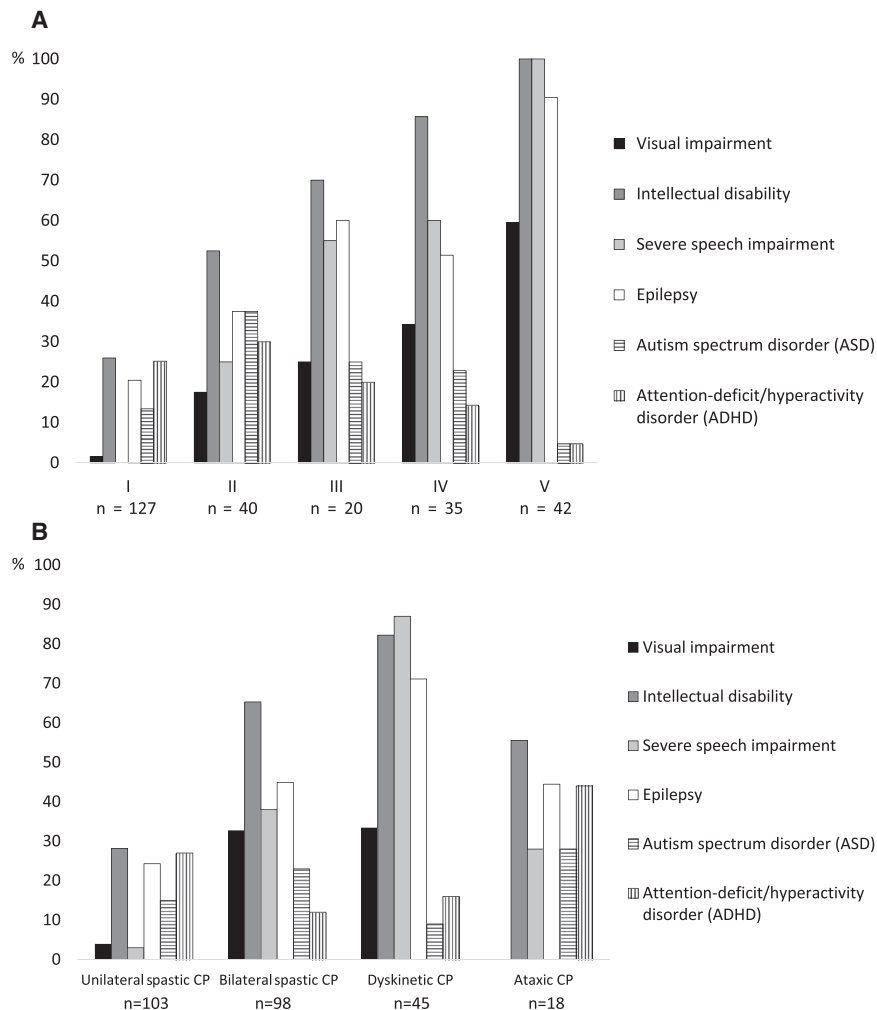


Figure 1 Proportion of associated impairments in a population-based group of 264 children with cerebral palsy (CP) at the age of 10-17 years, presented by (A) Gross Motor Function Classification System (GMFCS) and (B) type of CP.

of ID increased with increasing GMFCS level ($\rho = 0.71$; $p < 0.001$). At 4 to 8 years of age, 110 (42%) were diagnosed with ID, and the increase was statistically significant ($\chi^2 = 6.84$; $p < 0.001$).

Sensation

Visual impairment was found in 51 children and was severe in 33 children. Visual impairment was more prevalent in BSCP and dyskinetic CP; and more prevalent at severe GMFCS levels ($\chi^2_{\text{trend}} = 72.67$; $p < 0.001$). In the group with severe visual impairment, 29 of 33 children were at GMFCS level IV and V. The occurrence of severe visual impairment had not changed since the children were 4 to 8 years old. Hearing impairment was rare (8%).

Seizure disorder

Epilepsy was present and under treatment in 109 children (41%). The occurrence of epilepsy was highest in dyskinetic CP and became more common at more severe GMFCS levels ($\chi^2_{\text{trend}} = 64.00$; $p < 0.001$). There was also a strong association between epilepsy and more severe ID ($\chi^2_{\text{trend}} = 103.59$; $p < 0.001$). The occurrence of epilepsy was unchanged compared to when the children were 4 to 8 years old.

Communication

Speech ability was classified according to the VSS in all 264 children. Severe speech impairment (VSS levels III and IV) was present in 84 children (32%). VSS and GMFCS levels were associated ($\rho = 0.80$; $p < 0.001$). All children at GMFCS level I were understandable to unfamiliar listeners (VSS I and II), while only one at GMFCS level V had any speech. In children with dyskinetic CP, 87% had a severe speech impairment (VSS III and IV). More severe ID also correlated with less speech ability ($\rho = 0.72$; $p < 0.001$).

Language disorder was diagnosed in 27 children (10%); generalised type was most common (16/27). Of these 27 children, 17 had USCP (right-sided in ten and left-sided in

seven), and five had ataxic CP. All 27 children were at GMFCS level I and II; and none had more than mild ID.

Behaviour

Neuropsychiatric disorders, ASD or ADHD or both, had been diagnosed in 84 (32%) of the 264 children. Forty-seven (18%) of the 264 were diagnosed with ASD and 55 (21%) of the 264 with ADHD, hence 18 (7%) had both ASD and ADHD.

In the 47 children with ASD, 38 had autistic disorder, four had atypical autism and five had Asperger syndrome. Age at ASD diagnosis varied between three and 15 years, and did not differ between CP types or GMFCS levels. There was no difference in occurrence between boys and girls ($\chi^2 = 1.58$; $p = 0.21$). Fifteen children had USCP and 23 BSCP (Fig. 2). Children born before 28 gestational weeks more likely had ASD than children born after 28 gestational weeks ($\chi^2 = 5.57$; $p = 0.018$). The extremely preterm children constituted 10% of the total CP group and accounted for 20% of the ASD group.

The majority of children with ASD (32/47) were at GMFCS level I and II. ASD was found at all intellectual levels, increasing with more severe ID ($\chi^2_{\text{trend}} = 17.86$; $p < 0.001$), profound ID not included. ASD was also found at all levels of speech ability. Epilepsy was more common in the children with ASD (27/47), than in the children without ASD ($\chi^2 = 6.16$; $p = 0.013$).

ADHD was diagnosed in 55 children; 28 specified as combined type, 20 as inattentive type, while seven were not specified. Age at ADHD diagnosis varied from four to 15 years. Half of the children with CP and ADHD were boys (27/55). USCP (28/55) was more frequent than BSCP (12/55) in children with ADHD ($\chi^2 = 7.03$; $p = 0.008$) (Fig. 2). Children with ADHD were found in all gestational age groups.

The majority of children with ADHD (44/55) was at GMFCS level I and II. ADHD was rarely diagnosed in severe ID (3/55) and not found in profound ID. ADHD was found at all levels of speech ability, but in few (5/55)

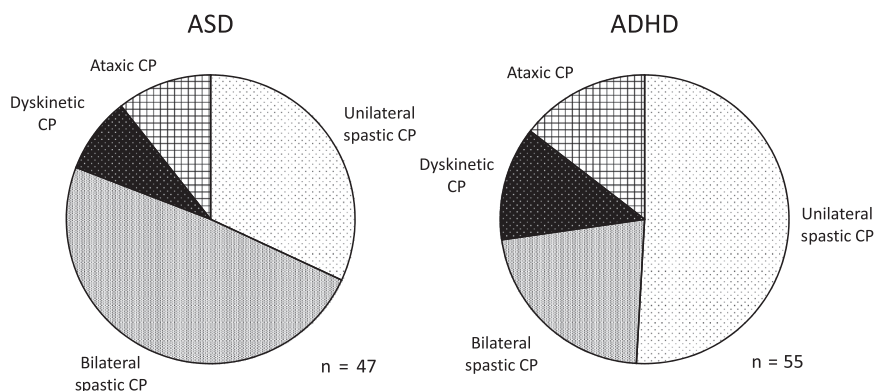


Figure 2 Distribution of diagnosed autism spectrum disorder (ASD) and attention-deficit/hyperactivity disorder (ADHD) by type of cerebral palsy (CP), in a population-based group of 264 children with CP at the age of 10–17 years.

without understandable speech. In contrast to ASD, epilepsy was not overrepresented in ADHD.

Both ASD and ADHD were present in 18 children (9 boys, 9 girls). The majority were diagnosed with ASD and ADHD at the same occasion. All CP subtypes were found in this group, and most children (14/18) were at GMFCS level I or II. The group with both ASD and ADHD had an impairment profile similar to the group with ADHD only, associated with lower occurrence of other impairments, except for epilepsy which was similar to the ASD only group (Fig. 3).

Children with no identified associated impairment

Sixty-five children (25%) (32 boys, 33 girls) had no identified impairment besides the motor impairment, which also was mild; 55 children were at GMFCS level I, 48 at BFMF level I and 46 at MACS level I. Forty-one were at level I in all three motor classifications. Most of children without other impairments (44/65) had USCP. The proportion of children without identified associated impairment decreased with age.

DISCUSSION

The vast majority of children with CP in this study had associated impairments. At the age of 10–17 years, only 25% had no identified other impairment than the motor impairment, mild in most cases. Neuropsychiatric impairments were frequent; ASD about ten times more common and ADHD almost four times more common than in the general population (27,28). The occurrence of ID and neuropsychiatric impairments had increased with age, which was not the case for other recorded impairments.

ID was strongly associated with more severe GMFCS level, concurrent with other studies (2,3,5,6). The increase in ID from 42% to 53% in the same children from

preschool-age may have several explanations. More children had been tested over time, and some abilities develop later in childhood and cannot be assessed until school age. Further, the brain lesion may hamper the intellectual development compared to typically developing children (29).

Visual impairment was present in 19%, compared to 15% in an Icelandic study with a definition similar to ours (5). Epilepsy, present in 41%, had not increased with age supporting early onset in most cases, as shown in earlier studies (7,30). Epilepsy was as expected strongly associated with more severe motor and intellectual impairment (4–7).

Communication is essential for the participation and independence of an individual. Of all children, 46% had no speech problems while 25% had no understandable speech, consistent with earlier studies (8,9). Speech impairment was strongly associated with more severe motor and intellectual impairment, concordant with epilepsy and visual impairment. In contrast, language disorder was associated with less severe motor impairment and less intellectual problems, which reflects the possibility to investigate language functions more thoroughly in these particular children.

More children were diagnosed with ASD and ADHD with age. ASD has previously been reported from partly the same population, but at a younger age, by Delobel-Ayoub et al with a prevalence of 15% (20). Some more children had been investigated and diagnosed since that study. The 18% reported in the present study is higher than in previous population-based studies (14,18–21). Impairments are easier to detect at older age, and there is an increasing awareness of these impairments among clinicians, which may explain the higher rates in the present study. No differences in the occurrence of ASD and ADHD were seen between boys and girls, suggesting that the brain lesion per se is involved in the pathogenesis, rather than genetic causes (31).

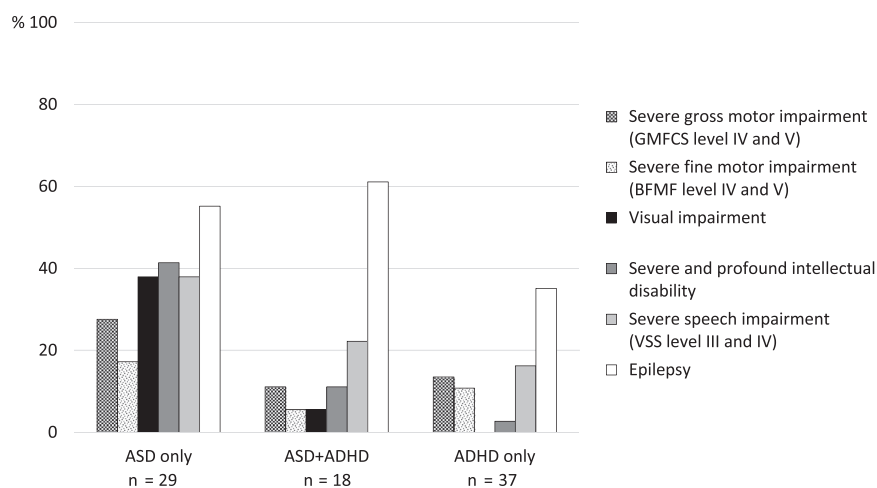


Figure 3 Proportion of children with other impairments in 84 children with cerebral palsy (CP) and neuropsychiatric diagnoses; autism spectrum disorder (ASD) only, attention-deficit/hyperactivity disorder (ADHD) only or both ASD and ADHD. The 84 children were identified in a population-based group of CP at the age of 10–17 years. BFMF = Bimanual Fine Motor Function; GMFCS = Gross Motor Function Classification System; VSS = Viking Speech Scale; visual impairment defined as an acuity of not more than 0.3 in the best eye with correction.

Studies on ADHD in children with CP are scarce. ADHD may be hard to diagnose in children with speech problems and severe motor impairment. To our knowledge, there is only one population-based study on the occurrence of ADHD in CP. In this Norwegian study, where 15% had an ADHD diagnosis or were under diagnostic investigation, Bjørngaas estimated the occurrence of ADHD to 50% in children at GMFCS level I-IV using a parental interview (21).

In contrast to other associated impairments, the occurrence of both ASD and ADHD decreased with more severe GMFCS levels, from level II to level V. The rates of at least ASD may be expected to be higher in children with more severe CP, similar to epilepsy and ID, both known to be associated with ASD (32). It can be speculated that ASD and ADHD are underdiagnosed especially in children at GMFCS level III and IV, while the most severely disabled children at GMFCS level V often have several other impairments hampering neuropsychiatric diagnostics. Moreover, they may be at too low intellectual level for neuropsychiatric diagnoses to be made.

The children with ASD and ADHD showed different patterns regarding other impairments. Children with ASD only had more severe and more often combined impairments, than the children with ADHD. ASD, regardless of ADHD, was strongly associated with epilepsy. This is in line with the population-based study of children with epilepsy by Reilly et al, where ASD was associated with ID and epilepsy, while ADHD was not (32). It can be noted that 18% of these children with epilepsy also had CP.

The varying impairment profile in children with CP suggests that every child should be assessed broadly and early to identify strengths and difficulties. Early diagnosis and adequate interventions may give a better prognosis for children with ASD (33), as well as ADHD (34). Parents may also benefit from early support with more knowledge about how to take care of their child, creating less stress. From a community perspective, it is important to recognise neuropsychiatric impairments in children with CP, in order to provide better services.

Having one neurodevelopmental impairment is a strong risk factor for having other impairments. The co-existence of disorders is the rule rather than the exception in developmental medicine and child psychiatry. The concept ESSENCE (Early Symptomatic Syndromes Eliciting Neurodevelopmental Clinical Examinations) has been coined to emphasise this (35) and may be applied for children with CP.

A strength of this study is it being population-based, covering the spectrum of CP. The age of the children (10–17 years) made it more likely that impairments identifiable first at school age had been detected. There was good access to medical records from both hospitals and habilitation units. A limitation is the descriptive nature of the study with all data taken from medical records where there may have been differences in how systematically impairments had been looked for and diagnosed by clinicians. The generalisability of the present results may be limited to other high-income countries, which are more likely to have a similar

panorama of CP. However, traditions in diagnosing neuropsychiatric impairments may differ between countries (20), which may affect the possibility to compare different populations.

CONCLUSION

Associated impairments are found in three out of four in school-aged children with CP. Some impairments become more identifiable with age, most obvious in the increase of ID from 4–8 years to 10–17 years. Neuropsychiatric impairments were found in one-third of the children and were more commonly diagnosed than previously described in population-based studies of children with CP; ASD was diagnosed in 18% and ADHD in 21%. However, the pattern of ASD and ADHD decreasing with increasing motor impairment may suggest that ASD and ADHD are underdiagnosed in children with more severe CP. Even though our results indicate very high rates of associated impairments in CP, we believe that, if anything, they represent minimum estimates. There is a need for further research with active assessment of neuropsychiatric impairments in children with CP.

CONFLICT OF INTEREST

The authors have no conflict of interests to declare.

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