

# Psychiatric disorders in children with cerebral palsy

Is there a need for mental health screening?

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## **Scientific environment**

Research groups with which the project has been affiliated:

The Research Group for Paediatric Follow-up Studies, Haukeland University Hospital, Bergen.

Regional Centre for Child and Youth Mental Health and Child Welfare, Uni Research Health, Bergen.

The Research Group for Women and Child Health at Stavanger University Hospital, Stavanger.

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The desire to study mental health in children with Cerebral Palsy (CP) was sparked by clinical encounters with many parents of children with CP. The parents expressed concern for the future of their child in terms of happiness, participation and living a life to the best of their abilities. As a clinician involved in families affected by CP, I was left with a desire to look for possible answers to the many questions asked by the parents. I am very grateful to Prof. Irene Elgen who shared this concern, and took the challenge of leading me step by step, from my comfort zone of interacting directly with the families, to the academic world of numbers and statistics. The journey has created more questions and a curiosity to keep looking for useful interventions. My hope is that the knowledge we have gained through this thesis can be integrated into the regular follow-up services for children with CP to address the concerns raised by parents prior to the study.

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## Abstract

Cerebral palsy (CP) is one of the most prevalent neuro-motor disorders in childhood. Previous studies using mental health questionnaires, had found mental health problems in up to every second child with CP. Peer problems were among the most common, often co-existing with other mental health problems. Whether a high prevalence of mental health problems found when using screening questionnaires reflected a high prevalence of psychiatric disorders when using stringent diagnostic criteria, was unknown. Neither was the impact of co-existing medical conditions on the prevalence of psychiatric disorders known, nor the feasibility of using screening questionnaires to identify psychiatric disorders in children with CP.

The aims of the present thesis were to ascertain the prevalence of psychiatric disorders in children with CP, and to assess to what extent psychiatric disorders were associated with co-existing medical conditions. Further, the aim was to assess the prevalence of autism spectrum symptoms in children with CP, using the Autism Spectrum Screening Questionnaire (ASSQ). Likewise, we aimed to compare mental health problems in children with CP to a population based sample using the Strengths and Difficulties Questionnaire (SDQ), and to evaluate the SDQ as a screening instrument in children with CP.

The thesis is based on a population including children with CP diagnosed according to ICD-10 criteria under G.80.0-G80.9, born in 2001-2003 and living in the Western Health Region of Norway. Children were assessed at school starting age, and parents of all children taking part in the study were interviewed using the child psychiatric diagnostic instrument, the Kiddie-SADS. Psychiatric disorders were diagnosed according to DSM-IV criteria. Mental health problems were assessed using the SDQ, and problems with social functioning were assessed using the ASSQ. Medical information was gathered through medical records and a medical examination.

Psychiatric disorders were found in almost one in two children with CP according to Kiddie-SADS. The burden of mental health problems was even higher, with two in three children scoring above a population cut-off at 90<sup>th</sup> percentile when using the SDQ. Peer problems were most common, with nine in ten children scoring above the 90<sup>th</sup> percentile. When using the ASSQ, one in five children scored above a population cut-off at 98<sup>th</sup> percentile. There was a considerable co-occurrence of mental health problems. Using the Kiddie-SADS as a gold standard, results from SDQ total difficulties score in children with CP were compared. Sensitivity was 0.85, and specificity was 0.55.

*Conclusion:* A high prevalence of psychiatric disorders in children with CP was found. Autism spectrum symptoms, possibly representing autism spectrum disorders, were highly prevalent. Co-existing medical conditions and co-occurring mental health problems were common, representing a challenge when diagnosing psychiatric disorders in children with CP. Mental health screening is recommended at school starting age in children with CP.

## List of publications

Bjorgaas, H. M., Hysing, M., & Elgen, I. (2012). Psychiatric disorders among children with cerebral palsy at school starting age. *Res Dev Disabil*

Bjorgaas, H. M., Elgen, I., Boe, T., & Hysing, M. (2013). Mental health in children with cerebral palsy: does screening capture the complexity? *Scientific World Journal*

Bjorgaas, H. M., Elgen, I., Ryland, H. K., & Hysing, M. (2014). Autism spectrum symptoms in children with cerebral palsy: Prevalence and co-occurring conditions. *Research in Autism Spectrum Disorders*

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## List of abbreviations

AD/HD	Attention Deficit /Hyperactivity Disorder (attention deficit disorder with or without hyperactivity)
ASD	Autism Spectrum Disorders
ASSQ	Autism Symptoms Screening Questionnaire
CBCL	Child Behaviour Checklist
CP	Cerebral Palsy
DAWBA	Development and Well-Being Assessment
GMFCS	Gross Motor Function Classification System
ID	Intellectual Disability
Kiddie-SADS	The Schedule for Affective Disorders and Schizophrenia for School- Aged Children: Present and Lifetime Version (6-18)
MACS	Manual Ability Classification System
OCD	Obsessive Compulsive Disorder
ODD	Oppositional Defiant Disorder
PVL	Periventricular Leucomalacia
SDQ	Strengths and Difficulties Questionnaire
SDQ-TDS	Strengths and Difficulties Questionnaire-Total Difficulties Score
TDS	Total Difficulties Score



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## 1. Introduction

Cerebral palsy (CP) is one of the most common neurological disorders in childhood, and the condition is often diagnosed before the age of two years (Andersen et al., 2008). Although it is a neuro-motor disorder, requiring distinct motor symptoms to meet criteria for a diagnosis (Bax et al., 2005), a number of co-existing conditions are associated with the disorder (Novak, Hines, Goldsmith, & Barclay, 2012). For some children, the co-existing conditions may even represent a symptom burden going beyond that of the motor symptoms. During the last years, focus of care for children with CP has shifted from a main emphasis on motor function, towards participation and minimizing limitations of activity, in line with the World Health Organization (WHO) framework; the International Classification of Functioning, Disability and Health (ICF). In this framework, participation is referred to as involvement in life situations across functional domains, such as self-care, relationships, education and employment (McDougall, Wright, & Rosenbaum, 2010; Rosenbaum & Stewart, 2004). An expanded childhood disability follow-up model, with emphasis on quality of life and participation is being advocated for (McDougall et al., 2010), and is likely to become the framework in which future services will be organized (Rosenbaum & Gorter, 2012; Rosenbaum & Stewart, 2007). Medical issues still remain important factors in enabling children with CP to participate across functional domains. However a broader approach is encouraged, encompassing a number of other aspects, of which one is mental health (Ramstad, Jahnsen, Skjeldal, & Diseth, 2012). Mental health is defined as a state of well-being in which every individual realizes his or her own potential, can cope with the normal stresses of life, can work productively and fruitfully, and is able to make a contribution to her or his community” (WHO, 2014). The definition of mental health incorporates an understanding that this is a necessary factor for enabling participation and contribution in society. Perhaps early detection and intervention, addressing issues such as anxiety and behavioural problems, could increase participation alongside peers, similar to the increased accessibility and participation seen for persons with motor disability after removing physical obstacles.

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Previous studies using mental health screening tools, have found a prevalence of mental health problems ranging from one in four children, to every second child with CP (Parkes et al., 2008; Parkes, White-Koning, McCullough, & Colver, 2009; Sigurdardottir et al., 2010). Despite having found a high prevalence of mental health problems when using screening instruments, little was known regarding the prevalence of psychiatric disorders in these children when using stringent diagnostic criteria. In this thesis, we aimed to ascertain the prevalence of psychiatric disorders in children with CP, and to assess the extent to which psychiatric disorders were associated with co-existing medical conditions. Further, we aimed to assess the prevalence of autism spectrum symptoms in children with CP, and the prevalence of mental health problems in children with CP using the Strengths and Difficulties Questionnaire (SDQ). The SDQ results were compared to a large population based sample (the Bergen Child Study). Further, we aimed to validate the SDQ against the child psychiatric diagnostic instrument Kiddie-SADS.

### ***1.1 History of cerebral palsy (CP)***

Cerebral palsy (CP) was first identified by William John Little (1810-1894). In his monograph from 1853 “On the Nature and Treatment of the Deformities of the Human Frame”(Little, 1843). He described generalized spasticity, associated with prematurity, difficult labour or birth asphyxia. By the end of the 19<sup>th</sup> century, spastic diplegia was called Little’s disease, and professor William Osler (1849-1919) further refined the diagnosis, and in 1889 published “The cerebral palsies of children“(Osler, 1889). He defined CP as a specific group of non-progressive neuromuscular disabilities in children, and related these conditions strongly to intracranial haemorrhage as a cause. Sigmund Freud (1856-1939) further classified the disorders on the basis of clinical phenotypes (Freud, 1897), which has been taken further in the International Classification of Disorders ICD (ICD-10, 2014). Andreas Peto (1893-1967) a Hungarian physician and neuropsychologist seems to be one of the first to

emphasize integration. He developed conductive education, with a focus on training to enable children with CP to walk in order to participate in regular education (Darrah, Watkins, Chen, & Bonin, 2004). From the mid 1950's, early physiotherapeutic interventions were developed, attempting to improve the outcome of the brain lesion, leading to multidisciplinary approaches becoming the norm. In 1997 the *Gross Motor Function Classification System* (GMFCS) (Appendix 1) was introduced as an improvement over the previous assessments of limitation, classified as either mild, moderate or severe (Palisano et al., 1997). Attempting to incorporate the variability in both aetiology and phenotypes of CP, the Surveillance of Cerebral palsy in Europe (SCPE) developed a definition of CP in 2000 emphasizing the nature of the CP diagnosis being an umbrella term, incorporating a number of phenotypes. Further, the SCPE definition emphasizes the permanent but not unchanging nature of CP, as well as the non-progressive nature of the interference/lesion or abnormality and its impact on the developing brain (Platt, Krageloh-Mann, & Cans, 2009).

## ***1.2 Definition, prevalence, pathophysiology and aetiology***

### *1.2.1 Definition*

One of the most used and internationally approved definitions of CP was originally proposed by Bax et al in 1964. It was further developed by the Executive Committee for the Definition of Cerebral Palsy, resulting in the following proposed definition and classification of CP published in 2005 (Bax et al., 2005); “Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture, causing activity limitations that are attributed to non- progressive disturbances in the developing foetal or infant brain. The motor disorders of CP are often accompanied by disturbances of sensation, cognition, communication, perception, and behaviour, in addition to epilepsy, and secondary musculoskeletal problems” (Bax et al., 2005; Rosenbaum et al., 2007). A number of important aspects are emphasized in the definition of CP. Firstly, it encompasses CP as a group of

disorders manifesting itself in different phenotypes, which will be further elaborated in the following paragraphs describing classifications of CP. Secondly, it emphasizes the permanent nature of the CP condition as opposed to a progressive disorder, as well as indicating a diverse aetiology of the condition. The condition is influenced by disturbances affecting a vulnerable developing brain, with a time frame stretching from foetal life until the age of 2-3 years. Finally, a number of co-existing conditions such as disturbances of sensation, cognition, communication, behaviour, and seizure disorders may accompany CP, suggesting that a broader, multidisciplinary approach is needed for children with CP.

### *1.2.2 Prevalence*

The prevalence of CP is estimated to be 1.5-3 per 1000 live births, with variations possibly reflecting true differences, or differences in ascertainment and classification (Andersen et al., 2008; Blair, 2010). Numbers are stable worldwide despite differences in types and presentations, with few cases of CP after very pre term birth in low income countries compared to high and middle income countries. On the other hand, birth asphyxia and maternal rhesus auto immunizations are reported more frequently in low income countries (Blair & Watson, 2006).

### *1.2.3 Pathophysiology*

To a large degree, the different phenotypes of CP can be linked to the developmental stage at which the disturbance may have occurred. During the neurogenesis, which takes place in the first and second trimester of foetal life, neuronal cells undergo proliferation, migration and organization. Lesions occurring during the period of neurogenesis therefore often lead to structural malformations of the brain (Krageloh-Mann & Horber, 2007). From the end of second trimester and throughout third trimester, hypoxic-ischemic lesions are more common. These may affect white matter, causing lesions described as periventricular leucomalacia. Further, germinal matrix haemorrhages may occur, causing intra ventricular extensions. Likewise, grey

matter injuries to the cortex or basal ganglia, thalamus, cerebellum or brain stem are seen during this period (Folkerth, 2005). The clinical manifestations largely depend on type of injury, location and severity of the injury (Koman, Smith, & Shilt, 2004).

#### *1.2.4 Aetiology*

The aetiology of CP has been the focus of interest both for obstetricians and paediatricians for centuries. Recent research suggests causative pathways where the interaction of more than one assault may lead to irreversible brain injury (Nelson & Grether, 1999), similar to conclusions from a Norwegian study (Stoknes et al., 2012). Despite much research and new insight into factors predisposing for CP, much is still unknown. However, among the known risk factors, some are listed under the following categories 1) *pre-pregnancy*, 2) *in pregnancy*, 3) *at term*, and 4) *post-neonatal* (i.e factors occurring after 28 days post- partum).

*Risk factors pre-pregnancy* cover factors such as maternal age (both low and high maternal age), as well as high parity (i.e. more than three pregnancies) and short pregnancy intervals. Chronic illnesses for example diabetes mellitus, is a known risk factor. Smoking is also a risk factor (Thorngren-Jerneck & Herbst, 2006).

*Risk factors in pregnancy* cover malformations of the brain, some of which are likely to be related to genetic disturbances (Costeff, 2004), as well as genetic factors related to certain coagulation disorders that may predispose for foetal stroke (Nelson & Grether, 1999). Further, male babies are more vulnerable for CP than girls (Chounti, Hagglund, Wagner, & Westbom, 2013). Other pregnancy related risk factors are low birth weight due to restricted growth, such as small for gestational age (SGA), or on the contrary, high birth weight (>4500g). Twins/triplets, preeclampsia and maternal infections as well as placental abnormalities (Nelson, 2008), and antenatal haemorrhages are risk factors (Palmer, Blair, Petterson, & Burton, 1995). The strongest predictor for CP however, is preterm birth, i.e. birth before 37 weeks of gestation, with significantly increased risk in babies born before 27 weeks gestational age (Himmelmann & Uvebrant, 2014). A recent Norwegian national cohort study of

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risk for CP in relation to pregnancy disorders and preterm birth, found children born after placental abruption, chorioamnionitis, intrauterine growth restriction and congenital malformations at increased risk of developing CP in most gestational groups (Tronnes, Wilcox, Lie, Markestad, & Moster, 2014). They emphasize the assumption that most preterm births are preceded by known or unknown pathological processes, of which some pathological processes leading to preterm birth may be more disruptive than others. One such example is finding the absolute risk of CP much higher in pregnancies with chorioamnionitis than in pregnancies with pre-eclampsia (Tronnes et al., 2014).

*Risk factors at term* cover perinatal stroke (Nelson, 2008), neonatal infections (Ahlin et al., 2013) or induction of labour, breech presentation, placental abruption, birth asphyxia and low Apgar scores (Thorngren-Jerneck & Herbst, 2006).

*Risk factors during the post-neonatal period* cover encephalitis of infectious origin, vascular episodes and trauma to the brain (Germany et al., 2013).

### ***1.3 Classification***

#### *1.3.1 Classification of subtypes*

Subtypes of CP are classified according to the part of the body most affected, e.g. quadriplegia or tetraplegia affecting all four limbs, diplegia affecting the lower limbs, hemiplegia affecting upper and lower limbs on one side. Additionally, one distinguishes between spastic, dyskinetic or atactic types of CP (Gainsborough, Surman, Maestri, Colver, & Cans, 2008; Platt et al., 2009). The subtypes are described in Appendix 2, and the diagnostic decision tree according to ICD-10 criteria developed by the Surveillance of Cerebral Palsy in Europe (SCPE) is described in Appendix 3.



### *1.3.2 Motor functional levels*

Cerebral palsy, being a movement disorder, affects both gross- and fine motor function. In addition to the classification systems describing sub types of CP, functional levels are used to give a more comprehensive picture of the assistive needs of the individual child with CP.

*Gross motor function:* Among the most used motor classification systems, is the *Gross Motor Function Classification System (GMFCS)* (Appendix 1) describing five motor functional levels according to need for assistive aid, such as wheelchair or crutches. Classification of CP according to these criteria has been valid over time, and the GMFCS is being broadly used worldwide (Palisano, Cameron, Rosenbaum, Walter, & Russell, 2006; Palisano, Rosenbaum, Bartlett, & Livingston, 2008).

*Fine motor function:* *The Manual Ability Classification System (MACS)* describing fine motor function is a five level classification system based on the child's self-initiated ability to handle objects, and the need for assistance or adaptation to perform manual activities in everyday life (Eliasson et al., 2006).

*1.3.3 The prevalence of CP in Norway regarding types and gross motor function level is presented in the Table 1.*

**Table1.** Classification of subtypes (types) and gross motor function according to Gross Motor Function Classification System (GMFCS) levels of cerebral palsy in a Norwegian population (N=289) (Andersen, G et al\*).

	Subtypes	Unilateral N (%)	Bilateral N (%)	Dys <sup>a</sup> /Atac <sup>b</sup> N (%)	Not classified N (%)	Total N
GMFCS	I-II	90(56)	51(32)	12( 8)	7( 4)	160
	III	4( 8)	36(75)	5(10)	3( 6)	48
	IV	1( 2)	38(66)	11(19)	8(14)	58
	V	0	15(65)	5(22)	3(13)	23

\* Andersen, G.L.,Irgens,LM.,Haagaas,I.,Skranes,J.S.,Meberg,A.E.,&Vik,T.(2008) Cerebral palsy in Norway: prevalence, subtypes and severity. Eur. J Paediatr Neurol, 12 (1), 4-13.doi:10.1016/j.ejpn.2007.05.00.1

<sup>a</sup>Dyskinetic, <sup>b</sup>Atactic

#### ***1.4 Co-existing conditions***

A number of co-existing conditions are described in table 2(Novak et al., 2012).

Some of these conditions may cause considerable negative impact on the well-being of children with CP, such as pain occurring in three in four children with CP, and behavioural problems occurring in one in four children with CP(Novak et al., 2012).

**Table2.** Prevalence of co-existing conditions in children with Cerebral Palsy\*

Severe hearing impairment/deafness**	4%
Functional blindness**	11 %
Cognitive functioning < 70	49 %
Epilepsy present currently	24 %
Bladder control problems	24 %
Severe feeding problems	6 %
Extensive dribbling	22 %
Displaced hips, migration>30%	28 %
Pathologic sleep pattern	23 %
Pain	75 %
Non-verbal	23 %
Behavioural problems***	26 %

\* Novak.I, Hines.M, Goldsmith.S & Barclay.R (2012).

\*\* Children with less severe auditory or visual impairments are not included.

\*\*\* Mental health problems are described as behavioural- or emotional problems.

## ***1.5 Mental health in children with CP***

### *1.5.1 Mental health problems*

Mental health is defined as a state of well-being in which every individual realizes his or her own potential, can cope with the normal stresses of life, can work productively and fruitfully, and is able to make a contribution to her or his community (WHO, 2014). Contrary to the abovementioned definition of mental health, the term “mental health problems” does not have a similarly distinct definition. Further, the term mental health problems, is an umbrella term encompassing a continuum of symptoms

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of poor mental health, of which some will meet criteria for a well- defined psychiatric disorder.

### *1.5.2 Mental health problems and cerebral palsy*

Mental health problems co-existed in one in four children with CP (Novak et al., 2012; Parkes et al., 2008), similar to findings in a large European multicentre study (Parkes et al., 2008). This was supported by finding children with chronic illnesses generally more prone to mental health problems than their healthy peers (Hysing, Elgen, Gillberg, Lie, & Lundervold, 2007; Lavigne & Faier-Routman, 1992). In a Norwegian epidemiological study including children with chronic illnesses, the subgroup having neurological disorders had a higher prevalence of mental health problems than controls. Children with neurological disorders yielded large effect sizes when comparing to controls, as opposed to small effect sizes when comparing children with other chronic illnesses to controls (Hysing, Elgen, Gillberg, & Lundervold, 2009).

Studies assessing mental health problems in children with CP have often used the Strengths and Difficulties Questionnaire (SDQ), or the Child Behaviour Checklist (CBCL). Relevant papers addressing the issue of CP and mental health problems published prior to the onset of this study in 2009 are presented in appendix 4, demonstrating a high rate of mental health problems in this population. In an Icelandic study using the Child Behaviour Check List (CBCL), preschool children with CP were compared to a randomly selected sample of pre-schoolers. In this study, almost half of children with CP were found to suffer substantially from behavioural and emotional problems, with attention and hyperactivity problems being most common (Sigurdardottir et al., 2010). In another study, one in four children with CP scored within abnormal range on the SDQ in children aged 8-12 years (Parkes et al., 2008), with even higher numbers in a similar study which only included children with CP hemiplegia (Parkes, McCullough, Madden, & McCahey, 2009). Peer problems

were highly prevalent in this study, and the negative impact of peer problems on participation in children with CP has been described previously (Yude, Goodman, & McConachie, 1998). Hyperactivity and attention problems, as well as peer problems, were the most prevalent problem areas, co-occurring extensively with other problem scales (J. Parkes et al., 2009; Parkes et al., 2008; Sigurdardottir et al., 2010).

### *1.5.3 Psychiatric disorders*

Psychiatric disorders are well defined mental- or behavioural patterns. They are distinct diagnostic entities requiring a certain minimum of specified symptom criteria, and a specified level of disrupted function to ascertain a disorder (DSM-IV-TR, 2000; ICD-10). Despite knowing that an increased prevalence of mental health problems had been found when using mental health screening tools (Parkes et al., 2008; J Parkes et al., 2009; Sigurdardottir et al.), less was known regarding the prevalence of psychiatric disorders in children with CP. To our knowledge, only two studies had been conducted previously, attempting to ascertain the prevalence of psychiatric disorders in children with CP (Goodman & Graham, 1996; Graham & Rutter, 1968). A large population study was conducted on the Isle of Wight, describing psychiatric disorders in children with disabilities (Graham & Rutter, 1968). They found an increased prevalence of psychiatric disorders in children with a condition originating from above the brain stem, such as CP and epilepsy (Graham & Rutter, 1968), as opposed to lesions originating from below the brain stem, such as traumatic lesions to the extremities or poliomyelitis. Further, a large prevalence study was conducted in London, assessing psychiatric disorders in a childhood population with unilateral CP (Goodman & Graham, 1996). In this study, psychiatric disorders were confirmed through interviews with parents and children, as well as through questionnaires completed by teachers (Goodman & Graham, 1996). These were both important studies indicating an association between conditions affecting the brain and psychiatric disorders. Although the interviews in the previous two studies were conducted by very experienced doctors, a standardized interview was to our knowledge not used, limiting the comparability to other studies. Further, one of the

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studies included children with hemiplegia only (Goodman & Graham, 1996), limiting our knowledge regarding the prevalence of psychiatric disorders for the whole group of children with CP.

#### *1.5.4 Medical conditions and mental health*

A number of medical conditions are known to co-exist in children CP, as outlined in Table 2. Intellectual disability (ID) was found for half of the children with CP (Novak et al., 2012), and an independent risk for developing psychiatric disorders in persons with ID is known (Emerson, 2003). Likewise, previous studies had found psychiatric disorders prevalent in one in three of children with epilepsy (Davies, Heyman, & Goodman, 2003), similar to findings in a Norwegian study (Alfstad et al., 2011). For children with CP however, little was known regarding a possible association between psychiatric disorders and the co-existing conditions ID and epilepsy. Another highly prevalent co-occurring medical condition is pain, affecting 75% of children with CP (Novak et al., 2012). Pain has been associated with mental health problems (Ramstad et al., 2012). Further, almost one in four children with CP were non-verbal (Novak et al., 2012), and communication problems have been associated with mental health problems (Voorman, Dallmeijer, Van Eck, Schuengel, & Becher, 2010).

In summary, previous questionnaire based studies had found mental health problems in up to every second child with CP, however the prevalence of psychiatric disorders assessed using stringent diagnostic criteria was unknown. Further, the impact of co-existing medical conditions on psychiatric disorders in children with CP was unknown. Assessing a screening instrument against diagnostic criteria, and asking if screening for mental health problems was appropriate in children with CP was considered to be of importance.

## ***1.6 Screening***

### *1.6.1 Screening mental health in general*

Screening for a certain disorder is often used when early detection and treatment is likely to improve the outcome of the condition. The World Health Organization (WHO) has recommended a set of criteria when screening a specific condition (Wilson, 1968):

1. The condition is an important health problem that needs to be addressed.
2. Testing for the condition is simple, safe, precise and validated, ie the sensitivity and specificity of the screening instrument is satisfactory.
3. Treatment is available for the condition.
4. Cost of the screening program is balanced in relation to the expenditure on medical care.

Screening and follow-up programs are established for several conditions with the objective of reducing the rate of adverse development, and to screen for co-existing conditions for which early detection can avoid functional deterioration and/or enhance development (Wilson, 1968). A number of mental health screening tools have been developed for mental health problems in the general population (Goodman, 2001; Goodman, Ford, Simmons, Gatward, & Meltzer, 2000; Novik, 1999, 2000), meeting the WHO guidelines for simple, safe, precise and validated screening instruments with satisfactory psychometric properties. Among the most used mental health screening instruments is the Strength and Difficulties Questionnaire (SDQ) (Goodman et al., 2000). The SDQ has shown good psychometric properties, both in the general childhood population, and in subpopulations of children with chronic illnesses (Goodman, 2001; Goodman et al., 2000; Hysing et al., 2007). It is short, easy to administer, and is developed for multi informants (Goodman, 2001). Further, the SDQ has been validated in large populations cross-culturally (Goodman et al., 2012; Stone, Otten, Engels, Vermulst, & Janssens, 2010).

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### 1.6.2 Screening mental health problems in children with CP

Among children with CP, systematic follow-up and screening for orthopaedic complications has been useful in reducing the rate of hip dislocations (Hagglund et al., 2005). Regular screening for mental health problems for all children with CP has to our knowledge not yet been implemented. By following WHO guidelines for implementing screening for a specific population, the four above mentioned guidelines should be considered. *Firstly*, one needs to decide whether mental health problems in children with CP are of a dimension requiring screening. From questionnaire based studies, we know that mental health problems and peer problems are highly prevalent in children with CP (J. Parkes et al., 2009; Parkes et al., 2008; Sigurdardottir et al.). However, these are not diagnostic instruments, and the prevalence of psychiatric disorders confirmed as a specific diagnosis in children with CP was not known. Confirming the prevalence of psychiatric disorders in children with CP was therefore seen as a prerequisite to decide if the prevalence was of a dimension requiring screening. *Secondly*, if screening for mental health problems in children with CP was appropriate, the screening instrument should have satisfactory psychometric properties, being simple, safe, precise and validated. Although mental health questionnaires were available for the general childhood population, these had not yet been validated for children with CP (Goodman, 2001; Goodman et al., 2000; Novik, 1999, 2000). *Thirdly*, treatment for psychiatric disorders should be available for children with CP. We know that medication for conditions such as AD/HD is effective for children from the general population (MTA-study, 1999), and for children with intellectual disability as well as for children with CP (Aman et al., 2014; Gross-Tsur, Shalev, Badihi, & Manor, 2002; Lipkin, 2013). *Fourthly*, the cost of screening for these disorders should balance the medical expenditure for the conditions in question. To our knowledge, there are no studies regarding the long term expenditure for these conditions in children with CP. However, there is evidence that psychiatric disorders in children and adolescents in the general population entail large expenses, especially related to the educational system (Snell et al., 2013). For AD/HD more specifically, studies have suggested reduced long term expenses in the general population by early interventions (Halmoy, Fasmer, Gillberg, & Haavik,



2009). Further, previous studies have found symptoms of AD/HD leading to later peer problems in children with CP (Yude & Goodman, 1999), indicating benefit from early detection and intervention. Another study has concluded that psychiatric disorders are among the most common reasons for hospitalization in young adults with CP (Young et al., 2011), suggesting a possible benefit of early detection and intervention.

## **2. Aims of the study**

The overall aims of the study were to estimate the prevalence of psychiatric disorders in children with CP at school starting age, and to evaluate the extent to which a short mental health screening instrument could be useful for detecting mental health problems. The following research aims were presented:

- To estimate the prevalence of psychiatric disorders in children with CP.
- To explore the association between psychiatric disorders and type of CP, as well as co-existing conditions.
- To estimate the prevalence of autism spectrum symptoms in children with CP using the ASSQ, and comparing these results to controls.
- To evaluate mental health problems in children with CP compared to controls.
- To evaluate the SDQ as a mental health screening instrument in children with CP.

### **3. Methods**

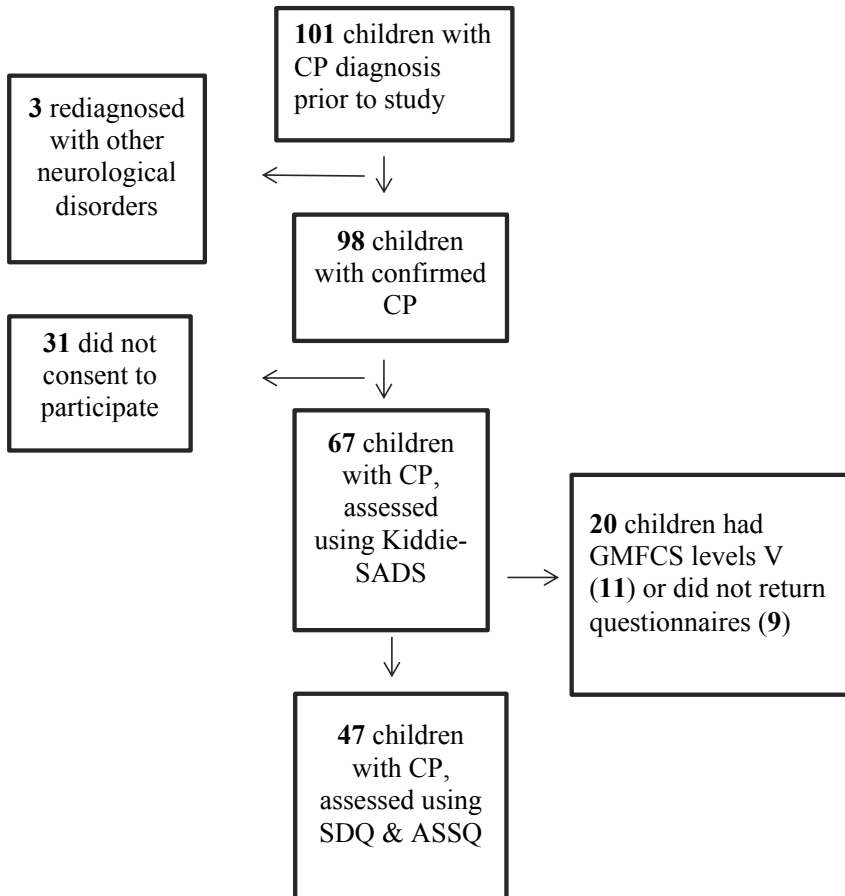
The study was conducted in the Western Health Region of Norway, and all children with cerebral palsy born 2001-2003 and living in the Western Health Region of Norway were invited to take part at school starting age. The Western Health Region of Norway has a stable, mixed urban and rural population of approximately 1 million. All children in Norway are entitled to free medical, educational and social services according to their condition when needed, ensuring the likelihood of including all children with a CP diagnosis in the follow-up programs. Children with CP in the study population are entitled to follow-up provided by the four child habilitation units in the region. From these units, lists of all children being followed-up with a CP diagnosis according to chapters G80.0-G80.9 in the ICD-10 diagnostic manual (ICD-10), born 2001-2003 and living in the Western Health Region of Norway were provided.

#### ***3.1 Participants***

##### *3.1.1 Study population*

The population of 98 children eligible for the study were invited to participate, of whom 67 gave written consent (Figure 1). Paper I, describing prevalence of psychiatric disorders in children with CP, included all 67 children. For papers II and III, using the two screening instruments Strengths and Difficulties Questionnaire (SDQ) and Autism Spectrum Screening Questionnaire (ASSQ), the population sample was reduced to 47, due to exclusion of children with GMFCS levels V (N=11), and those who did not return the questionnaires (N=9).

**Figure1** Population of children with cerebral palsy (CP) in the Western Health Region of Norway born 2001-2003.



### 3.1.2 Controls

Children from the Bergen Child Study (BCS) served as controls, and the BCS population has been described earlier (Heiervang et al., 2007). This is a large population based study involving all children (9155) in the two Norwegian municipalities Bergen and Sund with matching parent SDQ obtained from 6297 children. Children ages 7-9 were used as controls for the present study.

### ***3.2 Classification and medical information***

#### *3.2.1 Types of Cerebral palsy*

Cerebral Palsy was classified according to ICD-10 criteria with the following subtypes: spastic subtypes, dyskinetic, atactic or not further classified (ICD-10, 2014). The spastic subtypes were subdivided into spastic unilateral or spastic bilateral. These were further subdivided into spastic right sided or left sided unilateral, spastic bilateral diplegia or spastic bilateral quadriplegia/ tetraplegia (Appendix 2).

The CP diagnosis was based on the subtype diagnosed prior to the study. Children having been given a diagnosis of CP obtained from the four habilitation units included 101 children. During the verification process, we discovered that three children had been re-diagnosed, and were classified with other neurological disorders. They were therefore no longer eligible for the study and excluded. The CP diagnosis, including type of CP and functional level was verified through information provided in the medical records. Finally, the diagnosis was verified through the medical examination done by the first author as part of the study. The diagnostic process for classifying CP is described in detail in the SCPE decision tree (Appendix 3).

#### *3.2.2 Gross motor function*

For gross motor function, the level of Gross Motor Function Classification System (GMFCS) stated in the medical record prior to the study was used. When classification was not available prior to the study, this was done by the first author during the clinical examination, which also included assessment of motor function. *The Gross Motor Function Classification System (GMFCS)* is a classification system of five levels, emphasizing the child's general motor performance and the need for assistive equipment, described earlier (Palisano et al., 2006) (Appendix 1).

### *3.2.3 Fine motor function*

For fine motor function, the Manual Ability Classification System (MACS) was used. Unless classification was available prior to the study, this was done by the first author. *The Manual Ability Classification System (MACS)* describing fine motor function is a five level classification system similar to the GMFCS, and has been described earlier (Eliasson et al., 2006).

### *3.2.4 Cognitive function*

The cognitive level, ascertained through standardized intelligence tests, was obtained through the child's medical records. It was then classified as normal intellectual ability with the two categories IQ 85 and above and IQ 70-84, or as intellectual disability with the categories mild intellectual disability IQ 50-74, and moderate to severe disability IQ < 50. If standardized tests were not available, cognitive level was recorded through information in the medical record, or through the educational system, and verified during the interview. In these cases, cognitive level was classified in the two categories normal intellectual ability or intellectual disability.

### *3.2.5 Communication skills*

Communication problems were defined as no speech, difficulties with speaking, or needing communication aid. Information about communication problems was obtained through the medical records as well as the medical examination.

### *3.2.6 Medical and social information*

Available information regarding visual and hearing impairment and the need for adaptive measures for such, as well as information regarding epilepsy was obtained from the child's medical record. Data regarding perinatal history as well as socio-demographic factors were obtained through medical records and questionnaires.

### **3.3 Assessment**

#### *3.3.1 Psychiatric disorders - The Kiddie-SADS*

The Schedule for Affective Disorders and Schizophrenia for School-Age Children: Present and Lifetime Version (6-18) 10.04.00 (Kiddie-SADS) (Kaufman et al., 1997) is a semi structured child and adolescent psychiatric diagnostic instrument used to ascertain psychiatric disorders according to DSM IV criteria. Parents of children with CP were interviewed using the Kiddie-SADS to unveil psychiatric symptoms within the following groups of disorders: affective-, anxiety-, psychotic-, eating-, attention/hyperactivity-, oppositional defiant-, conduct-, tics-, substance abuse- and posttraumatic stress disorders, as well as encopresis and enuresis. Diagnostic conclusions were drawn according to the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV-TR, 2000). The Kiddie-SADS has shown excellent test-retest abilities for the following disorders; major depression, any bipolar depression, generalized anxiety, conduct, and oppositional defiant disorder. For the following disorders, test-retest ability was good; posttraumatic stress disorder and attention-deficit hyperactivity disorder. Results from a validation study suggest that the Kiddie-SADS is a valid and reliable child psychiatric diagnostic instrument (Kaufman et al., 1997).

Parents were interviewed, and we recorded present symptoms. All interviews were conducted by the first author, a child-and adolescent psychiatrist. A *psychiatric disorder* was ascertained if criteria listed in the DSM-IV for each specific diagnosis were fulfilled, including severity and duration of specific symptoms. When using the Kiddie-SADS interview, all symptom constellations meeting diagnostic criteria were identified as distinct diagnostic entities. For the whole group of attention and hyperactivity disorders, the abbreviation AD/HD has been used. These disorders were further sub-divided in Attention Deficit Disorders (ADD), Attention Deficit Hyperactivity Disorders (ADHD) and Primarily Hyperactive or Impulsive type according to criteria listed in the Kiddie-SADS.

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*Sub-threshold mental health symptom* is defined in the present study as meeting at least 75% of criteria for a psychiatric disorder according to DSM IV using Kiddie-SADS, without meeting the full criteria for a disorder. It serves the purpose of providing information regarding children at risk of developing a certain psychiatric disorder.

#### *Kiddie-SADS interviews*

All interviews were conducted by H.M. Bjorgaas, first author and child psychiatrist, who had undergone training in diagnosing child- and adolescent psychiatric disorders according to the Kiddie-SADS, and with substantial experience working with children with CP. Six interviews were video-recorded and subsequently viewed together with I. Elgen, Professor of child psychiatry. For the subsequent interviews, whenever in doubt, the interpretation and scorings were discussed for consensus.

Due to long travelling distances for some of the families, interviews were conducted close to the child's home, according to the preference of the parents. Interviews were therefore conducted by the first author at schools, at the nearby health clinics or medical units, or in the home of the child. Questionnaires were given to the parents, and they were offered to fill in the questionnaires after the interviews, returning them the same day. Alternatively, according to their preference, they were left with a pre-stamped envelope, and were asked to return the questionnaires after filling in at home.

#### *3.3.2 Strengths and Difficulties Questionnaire (SDQ)*

The Strengths and Difficulties Questionnaire (SDQ) is a well-documented mental health screening instrument, which has been validated in large study populations (Goodman et al., 2012; Stone et al., 2010). It is translated and validated in many languages, as well as for several subgroups of children (Goodman, 2001; Goodman et al., 2000). It consists of four problem domains, each including five items, and one pro-social domain (scale) including five items. Each item can be answered with "not true", "somewhat true" or "certainly true" rated 0-2 for negatively worded items, and



inversely 2-0 for positively worded items. The problem domains are *Hyperactivity problems* including items such as inattentiveness and distractibility, *Conduct problems* including items such as disobedience and temper tantrums, *Emotional problems* including items such as anxiety and worry, *Peer problems* including items such as loneliness and preference for adult company. *Pro-social behaviour* consists of items such as being helpful and kind. Combining the four problem subscales (0-10) computes the *Total difficulties score (TDS) (0-40)*.

For each of the subscales, a score at or above the 90<sup>th</sup> percentile of the controls (Bergen Child Study) was defined as screen positive (Heiervang et al., 2007; Hysing et al., 2007), and a Total Difficulties Score (TDS) at or above the 90<sup>th</sup> percentile as risk of having a psychiatric disorder as described previously (Bjorgaas, Elgen, Boe, & Hysing, 2013).

The SDQ also includes an *Impact Score (IS)* which measures the impact of mental health problems and the duration of these problems in terms of distress for the child itself. Further, it measures the degree to which mental health problems interfere with the child's everyday life at home, at school, with friends, and during leisure activities, as well as measuring the impact on the family as a whole.

### 3.3.3 *Autism Spectrum Screening Questionnaire (ASSQ)*

The Autism Spectrum Screening Questionnaire (ASSQ) is a 27-item screening instrument designed to detect symptoms within the autism spectrum (Ehlers, Gillberg, & Wing, 1999). ASSQ is rated on a 3-point scale with total symptom scores ranging from 0-54, with highest scores indicating highest symptom load. It has been validated in a Norwegian childhood population, and for the present study, cut-off scores derived from the validation study defined at the 98<sup>th</sup> percentile and corresponding to cut-off scores at  $\geq 17$  were used (Posserud, Lundervold, & Gillberg, 2009). Psychometric properties of the ASSQ against the Diagnostic Interview for Social and

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Communication Disorders (DISCO) are good (Wing, Leekam, Libby, Gould, & Locombe, 2002), with a sensitivity of 0.90 when using parents as informants (Posserud et al., 2009).

### **3.4 Statistics**

Pearsons Chi Square tests were used to examine demographics, clinical characteristics and functional level, and to compare children with CP meeting criteria for a psychiatric disorder, and children not meeting criteria for such in the present study. Results were presented as odds ratios (OR) with 95% confidence intervals and percentages were determined with descriptive analysis. *Sensitivity* was defined as the proportion of actual positives which were correctly identified as such. *Specificity* was defined as the proportion of negatives which were correctly identified as such. *Sensitivity and specificity* above 80% were regarded as high.

Independent t-tests were used to compare mean scores from children in the study population to those from the general population (Bergen Child Study). Effect size was defined using Cohens d, measuring the standardized mean difference between the study population and controls; a large effect size was defined  $\geq 0.8$ , a medium effect size  $\geq 0.5$ , and a small effect size  $\geq 0.3$ . A linear regression analysis was used to explore the influence of each of the five symptom items in the peer problem score, the latter being the dependent variable, and the five items were independent variables. Analyses were performed using The Statistical Package for the Social Sciences (SPSS) for Windows, version 18.0 for paper I and II, and version 20.0 for paper III.

### **3.5 Ethics**

The study was approved by the Regional Committee for Medical Research Ethics in Western Norway. Informed written consent was obtained from all participating caretakers.

## 4. Results

### 4.1 Summary of papers

#### 4.1.1 Paper I “Psychiatric disorders in children with cerebral palsy at school starting age”

Children with CP living in the Western Health Region of Norway were assessed using the child psychiatric diagnostic instrument Kiddie-SADS. This instrument was found to be suitable for children with GMFCS levels I-IV. It was however inappropriate for 11 children in the study population with the combination GMFCS levels V and Intellectual disability (ID), and these were therefore not included in the following analysis. We found 32 (57%) children meeting diagnostic criteria for at least one child psychiatric disorder. AD/HD was the most common disorder found in 28/32 children, and there was a considerable co-occurrence between disorders. Communication problems was the only significant factor associated with a psychiatric disorder in the group as a whole, OR 3.4 (95% CI 1.1-10.8), whereas communication problems, OR 4.0 (95% CI 1.3-12.5) and intellectual disability, OR 5.4 (95% CI 1.3-22.3) were associated with a diagnosis of AD/HD. When including children meeting at least 75% of criteria for a child psychiatric disorder (sub-threshold mental health symptoms), the prevalence rate rose to 75% with extensive co-occurrence of disorders.

#### 4.1.2 Paper II “Mental health problems in children with cerebral palsy - Does screening capture the complexity”?

To assess the prevalence of mental health problems in a Norwegian population of children with CP and to compare the prevalence in children with CP to controls (the Bergen Child Study), the Strengths and Difficulties Questionnaire (SDQ) was used.

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The SDQ was validated against results from the Kiddie-SADS as the gold standard. Two in three children scored above the 90<sup>th</sup> percentile of population controls on the Total Difficulties Score (TDS) of the SDQ. Large effect sizes were found when comparing TDS of children with CP to controls, whereas effect sizes were small when comparing to children with a neurological disorder. Peer problems were found to be highly prevalent. Sensitivity was 0.85 when comparing SDQ-TDS in children with CP to the Kiddie-SADS, whereas specificity was 0.55. Positive predictive Value PPV and Negative Predictive Value NPV were 0.71 and 0.73 respectively. For the problems scales, sensitivity varied from 0.13 to 1.0.

#### *4.1.3 Paper III “Autism spectrum symptoms in children with cerebral palsy-prevalence and co-occurring conditions”*

Finding peer problems prevalent in nine in ten children with CP when using the Strengths and Difficulties Questionnaire, we hypothesized that these findings could be related to autism spectrum symptoms. In paper III, the Autism Spectrum Screening Questionnaire (ASSQ) was used to assess autism symptoms and to relate these to co-existing psychiatric disorders in children with CP. One in five children with CP was found to score above the 98<sup>th</sup> percentile on the ASSQ, indicating a possible autism spectrum disorder. The prevalence rate was unchanged even when omitting the single item “clumsy” which was thought to be a possible confounder. Eight out of nine children scoring above the 98<sup>th</sup> percentile were GMFCS level I-II. A large effect size was found when comparing mean ASSQ total scores in children with CP to those of healthy peers. Following the single ASSQ item “clumsy”, the items ”prefers adult company”, ”uneven abilities” and “lonely” were the most prevalent ASSQ items. Eight in nine children scoring above cut-off met criteria for an AD/HD disorder.

## 4.2 Additional results

### 4.2.1 Non-participating population

Of a total population of 98 children with CP in the Western Health Region of Norway born 2001-2003, 31 children did not participate in the study. Information about type of CP was available for 28, and GMFCS level was available for 22 (Table 3).

**Table 3.** Distribution of classification and gross motor functional levels (GMFCS) for the non-participants.

<i>GMFCS level</i>	I	II	III	IV	V	Missing	Total
N	12	4	2	1	3	6	28
Spastic bilateral CP							
Quadriplegia					1	2	3
Diplegia	2	4	2	1	1	3	13
Spastic unilateral CP							
Hemiplegia	10						10
Atactic/Dyskinetic CP							
					1	1	2

No significant differences were found comparing participants and non-participants for the prevalence of the different types of CP and gross motor functional levels.

### 4.2.2 Subtypes of ADHD

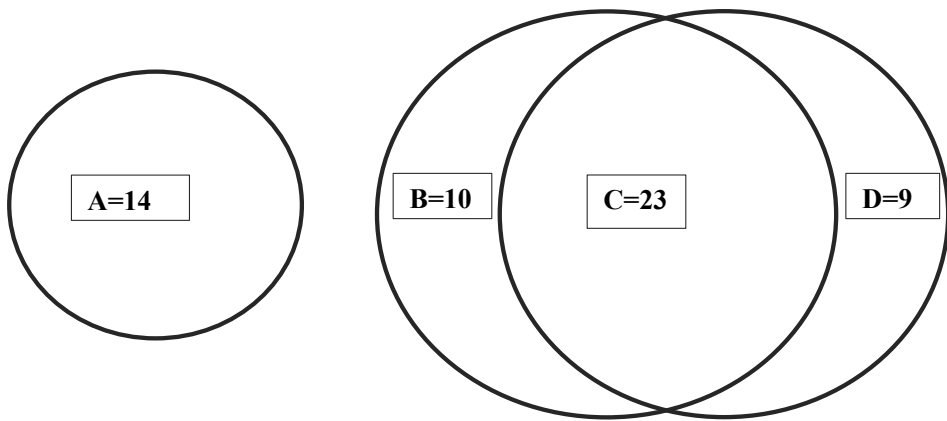
The most prevalent psychiatric disorders in the study population were AD/HD. For the total population of children with GMFCS levels I-IV (N=56), 28 (50%) of the children met criteria for either ADHD combined type N=11, ADD primarily inattentive type N=14, or primarily Hyperactive/Impulsive type N=3, according to DSM IV criteria.

#### 4.2.3 Psychiatric disorders and sub-threshold symptoms

Combining disorders and sub-threshold symptoms, we found 23(41%) children met both the full criteria for a psychiatric disorder according to Kiddie-SADS, and sub-threshold symptoms for another condition (at least 75% of criteria for a psychiatric disorder, but not the full criteria) (Figure 2).

Fourteen of 56 (25%) were reported as “healthy” regarding psychiatric symptoms at school starting age, according to Kiddie-SADS (Figure 2).

**Figure 2.** Prevalence of psychiatric disorders and subthreshold symptoms according to Kiddie-SADS in 56 children with CP, GMFCS I-IV.



**A** No psychiatric symptoms n=14

**B** Sub-threshold psychiatric symptoms only n=10

**C** Psychiatric disorders and subthreshold symptoms n=23

**D** Psychiatric disorders only n=9

#### 4.2.4 Autism spectrum screening questionnaire (ASSQ)

ASSQ high scorers was found in 9/47 children (19%), even when omitting the single, possibly confounding item “clumsy”, as described in paper III. When omitting the following six possibly confounding items from the ASSQ; “expresses sounds involuntarily”, “deviant style of gaze”, “clumsy”, “involuntary movements”, “unusual

facial expressions” and “unusual posture”, we found 8/47 children (17%) scored above the 98<sup>th</sup> percentile on the ASSQ.

## 5. Discussion

### *5.1 Main findings*

In the present thesis, psychiatric disorder was present in more than half of the children with CP, with AD/HD being the most common. Co-existing medical conditions, as well as co-occurring mental health problems were frequent. Communication problems were significantly associated with a psychiatric disorder. Using the screening instrument SDQ to assess mental health problems, two in three children with CP scored above cut-off on the SDQ-total difficulties score. Means on all SDQ subscales were significantly higher compared to controls. Sensitivity for the SDQ was adequate when using the child psychiatric diagnostic interview (Kiddie-SADS) as gold standard, however specificity was sub-optimal. Further, peer problems affected nine in ten children, and one in five scored above cut-off on the Autism Spectrum Screening Questionnaire (ASSQ). A large prevalence of children with CP scored above cut-off on ASSQ compared to healthy controls. Likewise, there was a considerable co-occurrence between high ASSQ score and psychiatric disorder.

### *5.2 Psychiatric disorders and CP*

The Kiddie-SADS interview was used to ascertain a psychiatric disorder. Two previous studies had assessed psychiatric disorders in children with CP (Goodman & Graham, 1996; Graham & Rutter, 1968). However to our knowledge, using a semi-structured diagnostic interview in children with CP had not been done previously. Despite challenges encountered when using a semi structured interview in children with co-existing conditions such as ID, epilepsy, communication problems and pain, the instrument allowed detection of nuances seen as important for diagnostic differentiations. Co-occurrence of disorders was frequent, and more than half of the children with CP met criteria for one or more psychiatric disorders.



### *5.2.1 Behavioural disorders*

By far the most prevalent psychiatric disorders were attention deficit disorders with or without hyperactivity (AD/HD), of whom half of the children met criteria for the inattentive type. During the Kiddie-SADS interviews, parents were asked to evaluate the attention span of their children with CP compared to typically developing children, or healthy siblings. Many of the parents were surprised to find their children having attention problems to a degree qualifying for an ADD diagnosis. For many of the parents, attention problems had previously not been recognized as such. Many parents were used to guiding their child with CP step by step without reflecting over the fact that they were easily distracted, or often forgot parts of the instructions given. Many parents described attention- and shift problems, as well as problems with working memory and distractibility among the most disabling symptoms for their child. Few of the children displayed problems in organizing belongings and schedules. This could be a bias, due to a high level of assistance in activities of daily living required for many children with CP, thereby covering up symptoms of inattention and problems with working memory.

Co-occurrence of psychiatric disorders may also have complicated AD/HD diagnostics. In the present thesis, six children met criteria for ODD, of whom five had co-occurring AD/HD. This is supported by a Swedish study finding at least one co-existing disorder in almost nine in ten children with an AD/HD diagnosis in the general population, of which Oppositional Defiant Disorder (ODD) was the most frequent (Kadesjo & Gillberg, 2001).

### *5.2.2 Emotional disorders*

Among emotional disorders, anxiety was the most common. In the present study, six children met criteria for one or more anxiety disorders of which compulsive disorder was the most common, followed by general anxiety disorder. Many parents described exhausting repetitions of questions, and having to reassure the child time and time

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again, a symptom previously described in children with CP (Bottcher, 2010; Bottcher, Flachs, & Uldall, 2010). The persistence could be interpreted as general anxiety due to inability to understand time and order of actions. Likewise, it could also be interpreted as an expression of Obsessive Compulsive Disorders (OCD), Autism Spectrum Disorders (ASD), Oppositional Defiant Disorders (ODD), or even hyperactivity as part of an AD/HD disorder. We may therefore have misinterpreted symptoms, leading to over or under diagnosing anxiety disorders.

Although only one child in the present study met the full criteria for a depressive disorder, four children met sub-threshold criteria for depression. Difficulties in emotional adjustment have been described as part of the CP conditions (Chang et al., 2014), and carefully evaluating symptoms of emotional instability therefore seems of importance. During interviews with parents, we have learnt that many children with CP seem to become increasingly aware of their shortcomings, and of being “different”, leading to symptoms of depression. We have also learnt that several children had shown grief by the loss of an assistant on whom they were depend for ADL activities, as well as for interpreting social situations. We have not found literature addressing these issues, and we recommend these issues to be included in future mental health studies.

### *5.2.3 Sub-threshold mental health symptoms and psychiatric disorders*

The construct “sub-threshold mental health symptoms”, which consists of mental health symptoms meeting at least 75% of criteria for a psychiatric disorder without meeting the full criteria for such, has also been presented in a previous study (Indredavik et al., 2004). Sub-threshold mental health symptoms were found in more than one in three children, possibly representing burdensome psychiatric symptoms, however not recognized as psychiatric disorders. A considerable overlap between psychiatric disorders and sub-threshold symptoms was found, and half of the children met criteria both for a psychiatric disorder and for sub-threshold symptoms as illustrated in Figure 2.

We found sub-threshold *emotional symptoms* more common than *emotional disorders* meeting the full criteria, indicating a high level of anxiety/ depression symptoms, although not to a degree qualifying for a diagnosis. Sub-threshold *behavioural symptoms* on the other hand, were found in one in three children, whereas criteria for a *behavioural disorder* were met in one in two children. Behavioural symptoms therefore seem to be more apparent at school starting age, with a higher prevalence of children displaying the full criteria for such a disorder, compared to emotional symptoms. Whether the sub-threshold symptoms will subside, or develop into psychiatric disorders later in adolescence, could be ascertained through follow-up studies in the future.

#### *5.2.4 Psychiatric disorders and type and severity of CP*

Psychiatric disorders were not significantly associated with type and severity of CP in the present study. Other studies have found less severely affected children more prone to developing mental health problems (Brossard-Racine et al., 2012; Goodman & Graham, 1996). Perhaps this is due to the nature of the semi structured interview, allowing for more elaborate answers which include a description of symptoms recognized by parents as mental distress in children more severely affected by CP. These could be subtle symptoms such as changes in respiratory rate, or increased spasticity as a response to anxiety provoking instances.

#### *5.3 Psychiatric disorders and co-existing conditions*

When diagnosing psychiatric disorders according to stringent diagnostic criteria, co-existing medical conditions may represent a challenge. Some of the co-existing conditions may be similar to psychiatric symptoms, or even increase psychiatric symptoms. We examined the impact of conditions such as ID, epilepsy, type of CP, visual impairment, and communication problems. In the present study, only communication problems were statistically significant risk factors for psychiatric

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disorders in general. For AD/HD more specifically, both communication problems and ID were associated with psychiatric disorders.

### *5.3.1 Psychiatric disorders and intellectual disability*

Psychiatric disorders being highly prevalent in persons with ID is well-known, and have been found in one in three children with ID as opposed to eight percent among children without ID (Turygin, Matson, Adams, & Williams, 2014). Similarly, children with ID accounted for 14% of all children with a psychiatric disorder in Britain (Emerson, 2003). Evaluating attention problems in children ID could be difficult, as inattention could be due to several factors, such as difficulties understanding instructions given, epileptic seizures or due to AD/HD. We did however try to compensate by asking clarifying questions to differentiate attention problems due to AD/HD, from those caused by being given tasks inappropriate for their cognitive level.

Few psychiatric diagnostic instruments are suitable for persons with ID, however some questionnaires have been developed attempting to compensate for the lack of diagnostic instruments (Dekker, Nunn, Einfeld, Tonge, & Koot, 2002). Other studies have used the Kiddie-SADS in populations including children with ID, attempting to compare psychiatric disorders head to head as in the present study. One of these studies is Norwegian, including children with autism and ID (Gjevik, Eldevik, Fjaeran-Granum, & Sponheim, 2011). In the latter study, the dilemma of using a child psychiatric diagnostic instrument in a population with autism and ID was addressed. The authors concluded that a standard child psychiatric interview provides valuable data on child psychiatric comorbidity. Further, that highlighting diagnostic dilemmas such as using the instrument in children with ID, is of importance in the development of more appropriate instruments, similar to conclusions from another study using the Kiddie-SADS in children with ASD (Simonoff et al., 2008).

### *5.3.2 Psychiatric disorders and epilepsy*

Epilepsy is another highly relevant condition in children with CP, occurring in one in three in the study population. During the interviews, differential diagnoses for AD/HD were thoroughly discussed, and for children with a known epilepsy diagnosis, we asked specifically to assure that recorded attention deficit symptoms were distinguished from epileptic symptoms. To avoid misinterpretation of symptoms, parents were asked about additional symptoms such as postictal tiredness, enuresis, confusion, or any muscle activity differing from their “normal” spastic symptoms linked to periods of inattentiveness. Likewise, to detect nocturnal epileptic seizures, we asked about motor activity during sleep, followed by enuresis, vomiting or waking up with bites and injuries. Still, we are aware of possibly having over- or under diagnosed conditions of AD/HD without having been able to distinguish properly between AD/HD and epilepsy. Connecting many of the children in the study population to long-term-EEG monitoring would have been useful in distinguishing epileptic symptoms, and should perhaps be included in future studies.

### *5.3.3 Psychiatric disorders and pain*

Pain is a frequent symptom occurring in 75% of children with CP (Engel, Jensen, Hoffman, & Kartin, 2003; Novak et al., 2012), and has been significantly associated with mental health problems both in a European multicentre study, and in a Norwegian study (J Parkes et al., 2009; Ramstad et al., 2012). Musculoskeletal pain is well-described in children with CP, and many children may have undergone painful surgery (Parkinson, Gibson, Dickinson, & Colver, 2010; Ramstad, Jahnsen, Skjeldal, & Diseth, 2011). Gastric reflux, tooth ache or constipation could also be sources of pain which many children with CP may be unable to express. Pain was not addressed specifically in the present study, which is a weakness to the study. Assessing pain in children with CP may not always be straight forward, as many of these children may not be able to recognize pain and report to their surroundings. Further, pain in non-verbal children may often be recognized by parents as facial expressions and behavioural changes (Breau, 2014), or as bodily symptoms such as tachycardia and

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sweating, as well as motor unrest and inattention, and could be confused with symptoms of AD/HD. Oral motor dysfunction and feeding problems, as well as energy consuming spastic activity, could lead to malnourishment, adding to attention problems.

#### *5.3.4 Psychiatric symptoms and traumatic experiences*

An association between attention problems and visual problems have been documented (Atkinson & Braddick, 2007). Perhaps visual disorders, found in half of the study population, could also be associated with anxiety disorders. Visual impairment leading to traumatic experiences such as stumbling in stairways or losing sight of persons on whom they rely for assistance, could be related to the high prevalence of traumatic events found in the study population. Considering that one in three children in the present study had experienced traumatic events such as falling out of the wheel chair, stumbling in front of their peers, or losing the relationship to employed assistants without being prepared, attention should be given to the aspect of previous traumatic experiences, which may have led to a need for more reassurance than for other children.

#### **5.4 Mental health problems**

In the present study, a higher prevalence of mental health problems was found when comparing the SDQ in children with CP to controls, supported by previous findings (Brossard-Racine et al., 2012; Parkes et al., 2008; J Parkes et al., 2009). Likewise, mean scores of SDQ were significantly higher across the four problem scales, and highest for peer problems compared to controls, in line with previous questionnaire based studies (Brossard-Racine et al., 2012; Parkes et al., 2008). A previous study even found increasing peer problems in children with CP as they approached their teens (Brossard-Racine et al., 2013), suggesting more focus on the developmental trajectory of peer problems in future studies.

Peer problems and aberrant social functioning seem to be one of the first manifestations of an impact to the vulnerable, developing brain affecting complex cognitive functioning, and leading to the co-occurrence of several mental health problems (Bottcher, 2010; Bottcher et al., 2010; Healy et al., 2013; Kadesjo & Gillberg, 2001; Karmiloff-Smith, 1998). It seems likely to suggest that the early brain lesion such as in CP, could lead both to motor problems and co-occurring mental health problems, as described in a model using the acronym ESSENCE (Early Symptomatic Syndromes Eliciting Neurodevelopmental Examinations), and supported by findings in the present study (Gillberg, 2010).

### ***5.5 Prevalence of autism spectrum symptoms***

The Kiddie-SADS version available at the start of the study did not include a diagnostic section regarding Autism Spectrum Disorders (ASD). Neither did we use other diagnostic instruments to ascertain an autism diagnosis. This is a weakness to the study design, knowing that peer problems are highly prevalent in children with CP (Brossard-Racine et al., 2012; J. Parkes et al., 2009; Parkes et al., 2008). However, attempting to compensate for the lack of information regarding ASD, the Autism Spectrum Screening Questionnaire (ASSQ) was used. ASSQ high scorers were found in 19% of children with CP, indicating autism spectrum problems, or even an ASD. This seems to be a robust finding, as the high prevalence was not altered even when omitting the following six potentially confounding ASSQ items; “expresses sounds involuntarily”, “deviant style of gaze”, “clumsy”, “involuntary movements”, “unusual facial expressions” and “unusual posture”. Further, when comparing ASSQ results in the present study to controls, large differences were found, as opposed to small differences when comparing to children with neurological disorders in general. This is in line with previous studies finding ASD highly prevalent in children with neurological disorders, and ID was present in at least one in three children with ASD (Fernell et al., 2010; Gillberg, 2010; Gillberg & Billstedt, 2000; Gillberg & Coleman, 1996; Kadesjo & Gillberg, 2001; Ryland, Hysing,

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Posserud, Gillberg, & Lundervold, 2012). In a previous study, pervasive developmental disorder was found in almost one in six children with CP (Kilincaslan & Mukaddes, 2009), similar to one in five children with CP being ASSQ high scorers in the present study. The high ASSQ score is a strong indicator of social impairment that should be further addressed through child psychiatric assessment to ascertain a diagnosis within the autism spectrum.

### ***5.6 Challenges diagnosing mental health problems***

Mental health diagnostics is a challenge due to the complexity of co-occurring mental health problems as well as co-existing medical conditions such as epilepsy, ID, pain and communication problems in children with CP (Alfstad et al., 2011; Emerson, 2003; Ramstad et al., 2012; Voorman et al., 2010). Perhaps the combination of biological vulnerability, co-existing medical conditions, as well as the impact of social demands such as trying to keep up with peers, may create a mismatch between social expectations and the child's ability to meet these expectations, leading to mental health problems (Bottcher & Dammeyer, 2013).

Much is still unknown regarding the origins of mental health problems affecting children with CP, let alone the expected developmental trajectory of mental health problems. Despite the complexity of CP and mental health problems, attempting to diagnose psychiatric disorders in children with CP seems to be an important challenge. In the new diagnostic manual DSM-5, the concept of co-occurring psychiatric conditions has been incorporated, allowing for more diagnoses being given concurrently than in the previous manual (Kupfer & Regier, 2011). This focus supports an understanding of the complexity of psychiatric disorders in children with CP and other neurodevelopmental conditions. Considerable co-occurrence of mental health problems as well as co-existing medical conditions, indicate that precise diagnostics could be demanding. Despite these challenges, it seems important to



make an effort to detect symptoms of mental health problems early, to avoid the development of psychiatric disorders.

## **5.7 Mental health screening**

### *5.7.1 SDQ screening properties in children with CP*

Knowing that psychiatric disorders were highly prevalent in children with CP, the important question of how to detect mental health problems within a busy paediatric clinic was raised. In the present study, we therefore wanted to explore the validity of SDQ for screening purposes in children with CP, knowing that the SDQ had good psychometric properties (Goodman, Ford, Corbin, & Meltzer, 2004; Goodman et al., 2000), and was short, concise and user friendly in other populations (Goodman, 2001; Muris, Meesters, & van den Berg, 2003).

Using the Kiddie-SADS as a gold standard (Kaufman et al., 1997), we validated the psychometric properties of the SDQ. The SDQ-total difficulties score was compared against all psychiatric disorders for which diagnostic criteria were met. In the present study, sensitivity for SDQ-total difficulties score was 0.85, indicating that the SDQ is likely to detect most children with CP having a mental health problem. However, a specificity of 0.55 indicates the presence of many false positive cases. The specificity level found in the present study was lower than in previous studies in the general childhood population, where specificity levels varied between 80% and 94% (Goodman, 2001; Goodman et al., 2004; Goodman et al., 2000). On the other hand, the same studies have described sensitivity levels between 63% and 85%, which for some of the studies is lower than in the present one. Sub-optimal specificity could be accounted for by the use of single informants, as poorer sensitivity has been found in previous studies when using multi-informants, whereas specificity was improved by using multi-informants (Goodman, 2001; Goodman et al., 2004; Stone et al., 2010). However, in a study including children with chronic illnesses, both sensitivity and

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specificity levels were almost unaltered for single and multi- informants (Hysing et al., 2007), indicating that the impact of using single vs. multi- informants in children with chronic illnesses seems to be inconclusive.

Psychometric properties varied across the SDQ-subcales in the present study. Low sensitivity for the hyperactivity scale was found, indicating that symptoms of AD/HD largely would go undetected. Low sensitivity may reflect the complexity of a CP condition, where signs of attention problems without hyperactivity could be subtle, requiring additional information through a diagnostic interview to ascertain a diagnosis.

In the presence of highly prevalent co-existing medical conditions as well as co-occurring mental health problems in children with CP, finding a mental health screening instrument with good specificity is perhaps unlikely. Sub optimal specificity, such as in the present study, would imply a higher referral rate than the rate of diagnosed psychiatric disorders. On the other hand, undetected mental health problems are likely to carry much suffering for children with CP and their families. Perhaps the benefit of detecting and intervening against mental health problems early exceeds the costs involved in using a screening instrument with sub-optimal specificity.

#### *5.7.2 WHO guidelines for screening mental health in children with CP*

Following the WHO guidelines for population screening could be a useful approach when considering the implementation of a mental health screening program. The magnitude of mental health problems in children with CP is the *first criterion* to be assessed when considering whether or not to implement screening. Results from the present study, with psychiatric disorders prevalent in more than half of children with CP, should be sufficient to recommend mental health screening. In comparison, psychiatric disorders were estimated to be present in 6% of the general childhood

population in a large Norwegian epidemiological study using the development and well-being assessment (DAWBA) (Heiervang et al., 2007). In support of screening for mental health problems, a recent commentary argued for the use of routine screening for such problems in high prevalence populations (Goldberg, 2014). However, the value of screening programs would be dependent on the ability to assess and diagnose mental health disorders. This brings to *the second WHO criterion* for population screening, which focuses on the validity of the screening instrument. In the present study, sensitivity was at a satisfactory level, however specificity was sub-optimal. Using the SDQ as a screening instrument, few children in need of mental health follow-up services would be missed, however at the expense of a high diagnostic burden on the mental health services. Good psychometric properties of a screening instrument is of little value if treatment is not available for the conditions in question (Goldberg, 2014). Therefore, *the third WHO criterion* for population screening emphasizes the availability of beneficial treatment for the conditions in question. Despite lack of extensive evidence for CP- specific treatment for psychiatric disorders, there is reason to believe that treatment found to be useful for the general population also would benefit children with CP. For emotional disorders, both psycho-therapeutic, psycho-educative as well as pharmacological approaches are being used in children with neurological conditions with positive clinical results (Maguire, Weston, Singh, & Marson, 2014). Research in this area is however still scarce. For children with AD/HD, large studies have found positive outcome by early interventions in the general childhood population (MTA-study, 1999). For children with CP and AD/HD more specifically, medication such as Methylphenidate has shown effect in enhancing learning and social interaction (Gross-Tsur et al., 2002; Symons, Tervo, Kim, & Hoch, 2007). One study has in fact, found untreated AD/HD leading to later peer problems, suggesting early interventions to enhance positive peer relations for children with CP (Yude & Goodman, 1999). Further, pain being highly prevalent in CP, and associated with mental health problems (Engel et al., 2003; Ramstad et al., 2012), suggests a more proactive approach towards health related problems in order to improve mental health. *The fourth WHO criterion* for population screening points to balancing screening costs with expenditure for

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treatment, a criterion for which there is lacking evidence regarding children with CP and mental health problems. There is little knowledge regarding the developmental trajectories of mental health in children with CP. For young adults with CP however, mental illness was found to be the third most common reason for hospitalization preceded only by epilepsy and pneumonia (Young et al., 2011). These studies give reason to believe that early interventions could benefit children with CP on the long run in terms of reduced suffering.

Following the WHO guidelines, screening for mental health problems in children with CP seems indicated due to the high prevalence of psychiatric disorders. Whether psychometric properties are sufficient to recommend using SDQ as a screening tool is not clear-cut. Even if sensitivity is sufficient, specificity is sub-optimal, and a rather large number of false positives must be expected. Despite sub-optimal psychometric properties, one could perhaps still argue for using the SDQ as a screening tool in children with CP due to the availability of treatment for psychiatric disorders, and with early interventions likely to reduce suffering for children with CP.

## ***5.8 Methodological issues***

### *5.8.1 Study population*

The whole population of children with CP born 2001-2003 were invited, of whom 67 consented to participate. Non-participants did not differ significantly from participants regarding type and severity of CP as described in table 3, indicating that the study population is representative. Children were recruited from the child habilitation units where they receive free, regular follow-up, and selection bias is therefore unlikely.

For paper I, describing the prevalence of psychiatric disorders in children with CP, participation rate was 67/98 and considered to be adequate. However for paper II and

III, comparing results from the questionnaires SDQ and ASSQ in children with GMFCS levels I-IV to those from the Kiddie-SADS interviews, the participation rate dropped to 47/98 which is a limitation. A better participation rate could possibly have yielded different validation results for the SDQ. On the other hand, participants were representative of the whole population, and results are therefore likely to be generalizable.

### *5.8.2 Diagnostic procedures*

Child psychiatric diagnostic interviews are usually developed for otherwise healthy children, and we did not know if the use of stringent diagnostics instruments were appropriate. Despite this, we chose to use the Kiddie-SADS as the gold standard, an instrument widely used within the mental health services for normally developing children (Kaufman et al., 1997). A semi-structured interview is designed such that questions can be asked to elaborate answers, and this was of importance in order to distinguish between somatic symptoms such as those encountered in children with CP and mental health symptoms.

All interviews were conducted by the first author, a child and adolescent psychiatrist, which could have resulted in systematic collection bias. All interviews should therefore ideally have been recorded for validation purposes. Due to their high burden of care, and long travelling distances, parents were largely offered to choose the location of the interviews, and video recording was not found to be appropriate in these locations. This is a limitation to the study, and misinterpretations of symptoms may therefore have occurred, despite having discussed challenging diagnostic differentiations. Not having done the Kiddie-SADS interviews under optimal conditions, such as in an office suitable for conducting video-recorded interviews, could perhaps partly be outweighed by a higher participation rate as most parents were offered to choose a location for the interview close to their home.

We chose not to include the diagnoses enuresis and encopresis when using the Kiddie-SADS interview, as we did not have enough information to ensure proper assessment of the origins of these problems. Incontinence in children with CP is common, with bladder problems occurring in one in four children with CP (Novak et al., 2012). Incontinence could be neurogenic, or related to the child not being able to reach the toilet in time due to motor problems. Other factors could be attention problems and distractibility, as well as epileptic seizures. Encopresis could cause discomfort and pain, as well as being socially disabling, and both enuresis and encopresis should have been taken into account as factors that could be associated with mental health problems.

Kiddie-SADS was found to be inappropriate for 11 children with GMFCS V and ID. In these children, it was impossible to distinguish between mental health symptoms related to discomfort due to emotional problems, and symptoms of physical discomfort. Further, some of the questions were inappropriate to ask, due to the severity of the CP condition. This is a limitation to the study, as a high level of psychiatric disorders has been found in other studies, including children severely affected by developmental disability and intellectual disability (Steffenburg, Gillberg, & Steffenburg, 1996). On the other hand, even if many of the children were unable to communicate other than by facial expressions and sounds, many parents showed an impressive ability to interpret, critically differentiate, and contextualize behaviours in these children. We chose to include these mental health symptoms and seemingly appropriate interpretations of subtle signals of anxiety and mood changes as sub-threshold symptoms. Additional analysis performed on these children separately showed sub-threshold mental health symptoms in 36%. This information was therefore collected, however not included when analysing data on the prevalence of psychiatric disorders. Using more appropriate instruments for evaluating mental health in children severely affected by cerebral palsy would have been very interesting, and should be included in future studies.

### *5.8.3 Measuring cognitive level*

A weakness in the present study was not having a standardized measure of cognitive ability in all children with CP in the study protocol. When standardized tests, such as the Wechsler intelligence tests were not available in the medical records, we relied on estimates from the educational system or observations. We were surprised to find that about half of the study population had not been tested with standardized cognitive measures prior to the study. These children were often either severely affected by CP, or had seemingly normal cognitive functioning. Children severely affected by CP could be difficult to assess with standardized tools, as medical conditions such as motor impairment, epilepsy, communication problems, sensory impairments (visual perceptive deficiencies, and auditory problems), and pain could interfere, causing the child not to perform optimally during testing. Likewise, variability in the cognitive profile is prevalent, perhaps causing misinterpretations of the actual cognitive level (Bottcher, 2010; Bottcher et al., 2010). Other children were not tested prior to the study as they were assumed to have a normal cognitive level with no need to undergo testing. We therefore cannot fully exclude the possibility of having misclassified some of the children. For future studies, results from standardized cognitive test should be available for all participants.

### *5.8.4 Methodological issues related to the use of questionnaires*

Using single- informants only, as in the present study, may have resulted in having missed important information from teachers. They are in a position to observe the child interacting with peers and other adults, and they may describe mental health problems differently from parents (Goodman et al., 2004). Further, the AD/HD diagnosis requires information about AD/HD symptoms occurring in different contexts. Despite having asked parents for information regarding the child's behaviour in school, we are aware that we may have missed important details by using parents as the only informants. Likewise, information from the children was lacking, as we did not use self-report questionnaires due to the children's young age. Multi- informants should also have been used for the ASSQ questionnaires, where

information from both teachers and parents probably would have been more nuanced enhancing the ability to differentiate disorders (Posserud et al., 2009). For further studies, information from more than one source should be obtained.

## **5.9 Clinical implications**

When following the WHO guidelines for population screening, the high prevalence of psychiatric disorders found in children with CP is likely to represent enough evidence to recommend mental health screening for all children with CP. Despite sub-optimal specificity, sensitivity was good, and the SDQ could perhaps be a satisfactory screening tool. It is short, concise and easy to administer in a busy paediatric clinic, which adds to the value of using the SDQ in children with CP. Most children and adolescents with CP in Norway are offered to participate in the nationwide follow-up program CPOP (CP oppfølgings program). One of the objectives of CPOP is to prevent orthopaedic complications (CPOP, 2015). Perhaps mental health screening at school starting age, using the SDQ, could be implemented similar to the CPOP in paediatric clinics. Knowing that the specificity of the SDQ is sub-optimal in children with CP, parents should be informed that some children would be false positives, entailing an extra burden of mental health diagnostics for these children and their families.

## **5.10 Conclusion**

In the present thesis, every second child with CP met criteria for a psychiatric disorder at school starting age, and AD/HD was most common.

We found one in five children at risk of having a possible Autism Spectrum Disorder, co-occurring psychiatric disorder was common.



Using the SDQ to assess mental health problems, we found two in three children with CP scored above cut-off, and peer problems were most common. Compared to controls, mental health problems were much more common in children with CP.

Mental health screening is recommended at school starting age in children with CP. When validating the SDQ against the diagnostic instrument Kiddie-SADS, we found sensitivity of the SDQ sufficient for screening purposes, however specificity was sub-optimal.

### **5.11 Future challenges**

A high prevalence of mental health problems at school starting age is known, however little is known regarding the developmental trajectory of these problems into adolescence and adulthood in children with CP. Further, knowledge regarding the child and adolescent's own perception of mental health problems is still fragmented. Follow-up studies at intervals during childhood and adolescence are therefore recommended, and should include self- reported and teacher- reported information as well as information from parents.

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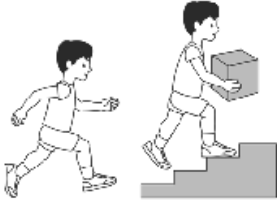
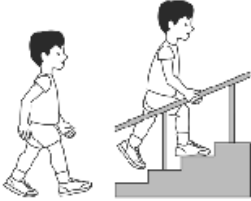
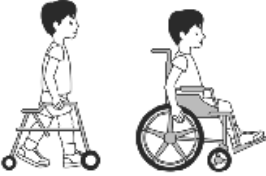
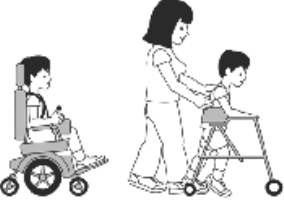
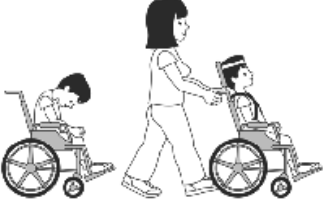


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**Appendix 1.** Description and illustration of gross motor function according to Gross Motor Function Classification System (GMFCS) levels (Palisano et al., 1997).

Figure is included with the courtesy of the authors.

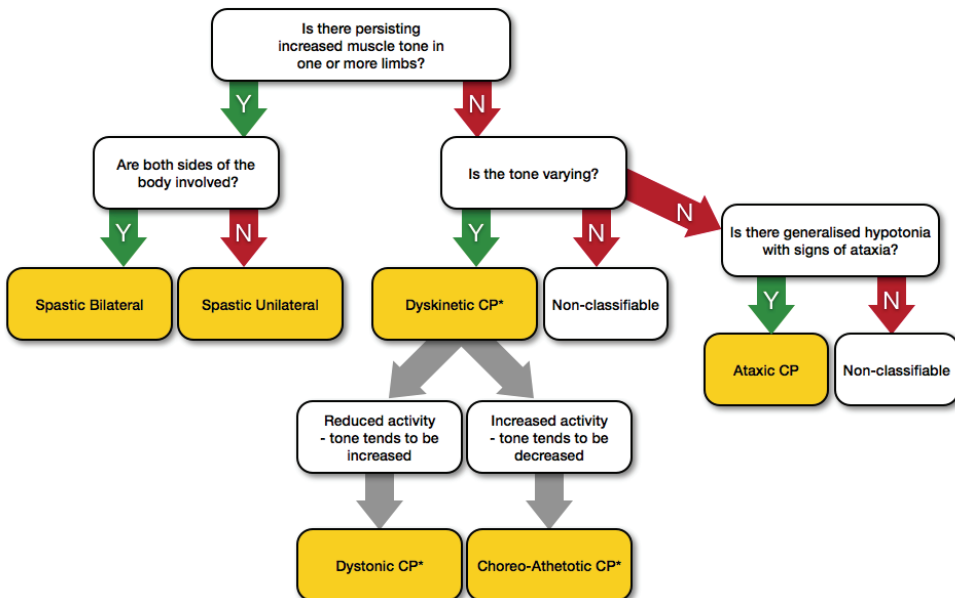
## GMFCS E & R Descriptors and Illustrations for Children between their 6<sup>th</sup> and 12<sup>th</sup> birthday

	<p><b>GMFCS Level I</b></p> <p>Children walk at home, school, outdoors and in the community. They can climb stairs without the use of a railing. Children perform gross motor skills such as running and jumping, but speed, balance and coordination are limited.</p>
	<p><b>GMFCS Level II</b></p> <p>Children walk in most settings and climb stairs holding onto a railing. They may experience difficulty walking long distances and balancing on uneven terrain, inclines, in crowded areas or confined spaces. Children may walk with physical assistance, a hand-held mobility device or used wheeled mobility over long distances. Children have only minimal ability to perform gross motor skills such as running and jumping.</p>
	<p><b>GMFCS Level III</b></p> <p>Children walk using a hand-held mobility device in most indoor settings. They may climb stairs holding onto a railing with supervision or assistance. Children use wheeled mobility when traveling long distances and may self-propel for shorter distances.</p>
	<p><b>GMFCS Level IV</b></p> <p>Children use methods of mobility that require physical assistance or powered mobility in most settings. They may walk for short distances at home with physical assistance or use powered mobility or a body support walker when positioned. At school, outdoors and in the community children are transported in a manual wheelchair or use powered mobility.</p>
	<p><b>GMFCS Level V</b></p> <p>Children are transported in a manual wheelchair in all settings. Children are limited in their ability to maintain antigravity head and trunk postures and control leg and arm movements.</p>

**Appendix 2.** Classification of cerebral palsy according to the Surveillance of Cerebral Palsy Europe (SCPE). Figure is included with the courtesy of the authors.

<b>SCPE Classification of CP Subtypes</b> based on the predominant neurological findings		All CP subtypes have in common an abnormal pattern of movement and posture. <b>Additional features by subtype:</b>
<b>SPASTIC CP</b>	<b>Bilateral Spastic (BS-CP)</b>	Increased tone Pathological reflexes - increased reflexes, e.g. hyperreflexia - pyramidal signs, e.g. Babinski response resulting in abnormal pattern of movement and posture
	<b>Unilateral Spastic (hemiplegia)</b>	
<b>DYSKINETIC CP</b>	<b>Dystonic</b>	Involuntary, uncontrolled, recurring, occasionally stereotyped movements, primitive reflexe patterns predominate, muscle tone is varying
	<b>Choreo-athetotic</b>	
<b>ATAXIC CP</b>		Loss of orderly muscular coordination, so that movements are performed with abnormal force, rhythm and accuracy

**Appendix 3.** Decision tree as developed by the Surveillance of Cerebral Palsy Europe (SCPE). Figure is included with the courtesy of the authors.



**Appendix4.** Papers regarding mental health in children with cerebral palsy published prior to onset of the present study.

Title	Author/Year	Ages	Main findings
Organic brain dysfunction and child psychiatry	Graham, P & Rutter, M 1968	4-14	Psychiatric disorders in 34% of children with organic brain dysfunction, five times more common than general population
A population based analysis of behaviour problems in children with CP	McDermott,S et al. 1996	4-7	One in four had behavior problems, mostly hyperactive, headstrong and dependent.
Psychiatric problems in children with hemiplegia	Goodman, R & Graham, P 1996	6-10	Psychiatric disorders in 61%, few had been in contact with mental health services prior to survey, IQ was strongest predictor.
Psychosocial problems in children with CP; a cross sectional European study	Parkes, J 2008	8-12	Questionnaire based study using SDQ, one in four scored above cut-off, peer problems most common, strongest predictors; better motor function, lower IQ, and pain.
Psychosocial problems in children with hemiplegia, European multicentre study	Parkes, J 2009	8-12	Questionnaire based study using SDQ, every second child scored above cut-off, strongest predictors were; IQ, communication problems, severe pain, single parenthood
Behavioural and emotional symptoms of pre-school children with CP	Sigurdardottir, S 2010	4-6	Questionnaire based study using CBCL, 48% scored above cut-off compared to 18% in general population.







## Psychiatric disorders among children with cerebral palsy at school starting age

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### ABSTRACT

The aim of the present population study was to estimate the prevalence of psychiatric disorders in children with cerebral palsy (CP), as well as the impact of comorbid conditions. A cohort of children with CP born 2001–2003, and living in the Western Health Region of Norway were evaluated at school starting age. Parents were interviewed with the diagnostic instrument Kiddie-SADS, to find the prevalence of psychiatric disorders. Sixty-seven children participated, 43 boys, with mean age 88 months (SD 6.8 months). Most children had spastic CP, Gross Motor Function Classification System (GMFCS) levels I and II were found in 2/3 of the group. We found the diagnostic instrument appropriate for GMFCS levels I–IV. Child psychiatric disorders were found in 57% of the children, including 28 children meeting criteria for an attention deficit disorder, which was the most common. Communication problem was significantly associated with having a psychiatric disorder, whereas intellectual disability, type of CP and functional level did not account for significant differences. Subthreshold symptoms were found in 33 children, and 42 children (75%) met criteria for either psychiatric disorder, or mental health symptoms. One in four (14 children) were considered well-functioning from a mental health point of view. We conclude with a recommendation for early psychiatric evaluation of all children with CP.

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### 1. Introduction

Cerebral palsy (CP) is one of the most frequent neurological conditions in childhood and has shown an increased risk for associated mental health problems according to questionnaire based studies (Carlsson, Olsson, Hagberg, & Beckung, 2008; Parkes et al., 2008; Sigurdardottir et al., 2010). A population study from Iceland, where preschool children with CP were included, found a three to four fold increased risk for emotional and behavioural problems for these children compared to controls (Sigurdardottir et al., 2010). A similar European multicenter study using a mental health screening instrument, Strengths and Difficulties Questionnaire (SDQ), found one in four children aged 8–12 years with CP had scores indicating mental health problems. They concluded with a need for in depth interviews to learn more about the extent of mental health problems in children with CP (Parkes et al., 2008). To our knowledge only one previous study has done this, more specifically, Goodman et al. verified psychiatric disorders in a childhood population with unilateral CP where a sample of parents were interviewed, finding psychiatric disorders present in more than half of the children (Goodman & Graham, 1996). A semistructured diagnostic interview was to our knowledge not yet available at that time, and comparability is therefore

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limited. The sample was restricted to unilateral CP, and little is known about psychiatric disorders in the CP group as a whole, and even more so for the children with multiple disabilities, taking into account the impact of functional level and comorbid conditions. A European questionnaire based multicentre study has shown an apparent lower risk for mental health problems among children with a lower functional level (Parkes et al., 2008). The reliability of these findings has however been discussed, as the more severely affected children will have difficulties expressing their emotional status, and questionnaire based studies may not capture the subtle symptoms of emotional distress.

The aims of the present population based study were to assess the rate of psychiatric disorders using a diagnostic interview, and to evaluate the impact of type of CP, functional level and comorbid conditions on mental health in a cohort of children with CP.

## 2. Methods

### 2.1. Study population

A cohort of children with Cerebral Palsy (CP) in the three western counties of Norway (Western Health Region of Norway) born 2001–2003 were invited to participate in the study. The population is approximately 1 million, it is stable with little migration, and equally distributed between urban and rural areas. All children with CP in Norway are referred to paediatric habilitation units for diagnostic confirmation and follow up, and are offered free services. The four local habilitation units in the region provided in 2009 lists of all children diagnosed with CP according to International Classification of Diseases (ICD-10) criteria under G80.0–G80.9. All children with a diagnosis of CP with an onset of injury up to the age of three years were included in the study.

### 2.2. Classification, functional levels and medical information

#### 2.2.1. Classification

Cerebral Palsy was classified according to ICD-10 criteria with the following subgroups: spastic bilateral and unilateral, dyskinetic, a tactic or not further classified. We recorded status of classification given prior to the study, and stated in the medical record. Functional level was classified by the Gross Motor Function Classification System (GMFCS) and Manual Ability Classification System (MACS) which distinguishes five groups (Eliasson et al., 2006; Palisano et al., 1997). Classification for gross motor function was based on self-initiated movement, functional limitations, and the use of mobility devices in everyday life. Fine motor function was classified according to bimanual ability in daily activities, and five groups were distinguished according to functional level. Functional classification given in the medical record was obtained, or was given during the medical examination if information was not available prior to the study. Classification was grouped as follows: light disability (GMFCS I and II, MACS I and II), moderate disability (GMFCS III and IV, MACS III–IV), and severe disability (GMFCS V, MACS V). Cognitive level was recorded by standardized psychological tests and classified as normal intellectual ability with the two categories IQ 85 and above and IQ 70–84, or as intellectual disability with the categories mild intellectual disability IQ 50–74, and moderate to severe disability IQ < 50. When this information was not available, the children were assessed clinically with information drawn from the medical records and the educational system.

#### 2.2.2. Medical information

From the child's medical record, available information on visual and hearing impairment and the need for adaptive measures for such was recorded, as well as information about epilepsy and other diagnoses. A medical examination was performed, and communication problems were based on this information, as well as information from the medical records. It was then defined as communication problem when having no speech, difficulties with speaking, or having the need for a communication aid.

#### 2.2.3. Mental health assessment using the Kiddie-SADS interview

All children were assessed using the Schedule for Affective Disorders and Schizophrenia for school-age children: present and lifetime version (6–18) 10.04.00 (Kiddie-SADS). It is a semi-structured child psychiatric diagnostic interview, designed to unveil psychiatric symptoms within the following groups of disorders: affective-, anxiety-, psychotic-, eating-, attention/hyperactivity-, oppositional defiant-, conduct-, tics-, substance abuse- and post-traumatic stress disorders, as well as encopresis and enuresis. Diagnostic conclusions were drawn according to the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV). Parents were interviewed, and we recorded present symptoms. All interviews were conducted by the first author, a child- and adolescent psychiatrist. A *psychiatric disorder* was ascertained if criteria listed in the DSM-IV for each specific diagnosis were fulfilled, including severity and duration of specific symptoms. Subthreshold symptoms were defined as *mental health symptoms* present to a degree above 75% of that required to qualify for a distinct disorder according to DSM IV criteria. Specific incidents considered by the parents to have made considerable negative impact on the child's mental health, were recorded as traumatic events.

### 2.3. Statistical analysis

Pearsons Chi Square Tests were used to examine demographics, clinical characteristics and functional level when comparing children meeting criteria for a psychiatric disorder and children not meeting criteria for such, as well as



**Table 1**  
Demographic and clinical characteristics of a cohort of 67 children with cerebral palsy in the Western Health Region of Norway born 2001–2003.

n = 67	n	%
Gender		
Male	43	(64)
CP subtype		
Bilateral	37	(55)
Unilateral	22	(33)
Ataxia/dyskinesia	8	(12)
Gestational length		
<32 weeks	14	(22)
GMFCS <sup>a</sup> level		
I + II	44	(67)
III – V	23	(33)
MACS <sup>b</sup> level		
I + II	34	(51)
III – V	33	(49)
Intellectual disability <sup>c</sup>	25	(39)
Communication problems <sup>d</sup>	34	(51)
Epilepsy	22	(33)
Visual impairment <sup>e</sup>	35	(53)
Auditive impairment <sup>f</sup>	6	(9)

<sup>a</sup> GMFCS **Gross Motor Function Classification System**.

<sup>b</sup> MACS **Manual Ability Classification System**.

<sup>c</sup> Intellectual ability <70.

<sup>d</sup> Speech problems, need for a communication aid or no speech.

<sup>e</sup> Blindness or need for visual aid.

<sup>f</sup> Deafness or need for auditory aid.

comparing children meeting criteria for ADHD/ADD and children not meeting criteria for such. Results are presented as odds ratios (OR) with 95% confidence intervals. Analyses were performed using The Statistical Package for the Social Sciences (SPSS) for Windows, version 18.0.

#### 2.4. Ethics

The study was approved by the Regional Ethical Committee for the Western region of Norway, and by the Norwegian Data Inspectorate. Informed written consent was obtained from all participating parents.

### 3. Results

#### 3.1. Clinical characteristics of the population and classification of cerebral palsy (CP)

Cerebral palsy (CP) was stated for 101 children prior to the study. Three of the children were excluded due to later evaluation, and had been diagnosed with other neurological disorders. This information was provided in the medical journals or by the parents as they were approached during the initial phase of the study. In the present study, 98 children were invited, and 67 participated (68%), of whom 43 were boys (64%) (Table 1). The mean age of the participants was 88 months (SD 6,8 months) at the time of study. 59 (88%) had spastic CP, and two third had GMFCS levels I and II (Table 1). Two third had a cognitive level within normal range (Table 1). Cognitive level had been tested by standardized psychological tests for 32 children. For the remaining 35 children, cognitive level was drawn clinically from medical records and information from the educational system (Table 2).

#### 3.2. Psychiatric disorders and comorbidity

##### 3.2.1. Children severely affected by cerebral palsy and mental health

All the 67 children were assessed according to Kiddie-SADS, however 11 (16%) children with GMFCS level V and intellectual disability could not be diagnosed with a distinct psychiatric disorder due to the severity of their physical and mental condition. Symptoms of mental health problems were nevertheless assessed for this group, as the parents were able to give relevant descriptions of symptoms. Four of these children expressed mental health symptoms to a debilitating degree, one had emotional symptoms, two behavioural symptoms, and one had both. For the following analysis of the results however, these children were omitted.

##### 3.2.2. Prevalence of psychiatric disorders and comorbid conditions

Among the 56 children with GMFCS level I–IV, 32 (57%) met criteria for a child psychiatric disorder according to Kiddie-SADS (Table 3). Behavioural disorders were most frequent, and found in 29/32 children including 28 fulfilling criteria for

**Table 2**Cognitive evaluation of children with Cerebral Palsy born 2001–2003 living in Western Health Region of Norway. *N* = 67.

Assessment method	Normal intellectual ability		Intellectual disability	
	IQ > 85	IQ 70–85	IQ 50–69	IQ < 50
Standardized psychological <i>N</i> = 32	13	5	5	9
Clinical <i>N</i> = 35		24		11

**Table 3**Children with cerebral palsy (56): number of psychiatric disorders and number of subthreshold mental health symptoms according to Kiddie-SADS.<sup>a</sup> A total of 32 children met criteria for a psychiatric disorder, and 33 children met criteria for subthreshold mental health symptoms.<sup>b</sup>

	Psychiatric disorders	Subthreshold mental health symptoms
<b>Behavioural disorders (29)</b>		
ADHD/ADD <sup>c</sup>	28	7
ADHD	12	
ADD	13	
Hyperactivity disorder	3	
ODD <sup>d</sup>	6	12
Conduct disorder		3
<b>Emotional disorders (6)</b>		
Affective disorder	1	4
Anxiety disorder		
Compulsive disorder	5	6
Phobic disorder	1	7
General anxiety disorder	4	1
Separation anxiety disorder	3	6
Panic disorder	1	8

<sup>a</sup> Schedule for affective disorders and schizophrenia for school-age children: present and lifetime version (6–18) 10.04.00 (Kiddie-SADS), a child psychiatric diagnostic interview.<sup>b</sup> Subthreshold mental health symptoms defined as 75% of criteria for a psychiatric disorder fulfilled.<sup>c</sup> Attention deficit hyperactivity disorder/attention deficit disorder.<sup>d</sup> Oppositional defiant disorder.**Table 4**Children with cerebral palsy CP (*n* = 56) fulfilling criteria for any child psychiatric disorder, and children fulfilling criteria for ADHD<sup>a</sup> according to DSM IV, using the Kiddie-SADS<sup>b</sup> interview.

	Children with CP fulfilling diagnostic criteria for any psychiatric disorder <sup>c</sup> <i>N</i> = 32			Children with CP fulfilling diagnostic criteria for ADHD <sup>a</sup> <i>N</i> = 28		
	<i>N</i>	OR	95% CI	<i>N</i>	OR	95% CI
Boys	18/32	0.4	0.1–1.4	16/28	0.5	0.2–1.6
Intellectual disability <sup>d</sup>	11/32	3.7	0.9–15.5	11/28	5.4	1.3–22.3
Epilepsy	9/32	1.6	0.4–5.5	9/28	2.3	0.7–8.1
Communication problems <sup>e</sup>	17/32	3.4	1.1–10.8	16/28	4.0	1.3–12.5
Gest age < 32w <sup>f</sup>	5/32	0.4	0.1–1.6	3/28	0.2	0.1–1.0
GMFCS <sup>g</sup> III–IV	7/32	1.1	0.3–3.9	7/28	1.5	0.4–5.6
MACS <sup>h</sup> III–V	13/32	1.1	0.4–3.4	13/28	1.8	0.6–5.4
Visual impairment	12/31	0.5	0.2–1.6	11/28	0.7	0.2–2.0
Bilateral spastic	17/32	1.1	0.4–3.3	14/28	0.9	0.3–2.5
Unilateral spastic	13/32	1.1	0.4–3.4	12/28	1.3	0.4–3.9
Dystonia/ataxia	2/32	0.5	0.1–3.0	2/28	0.6	0.1–4.2

<sup>a</sup> Attention deficit hyperactivity disorder and attention deficit disorder compared to children who did not meet criteria for such.<sup>b</sup> Schedule for affective disorders and schizophrenia for school-age children: present and lifetime version (6–18) 10.04.00 (Kiddie-SADS), a child psychiatric diagnostic interview.<sup>c</sup> Compared to children who did not meet criteria for any psychiatric disorder.<sup>d</sup> Intellectual quotient < 70.<sup>e</sup> Speech problems, need for a communication aid, or no speech.<sup>f</sup> Gestational age less than 32 weeks.<sup>g</sup> Gross Motor Function Classification System.<sup>h</sup> Manual Ability Classification System.

ADHD/ADD (attention deficit hyperactivity disorder/attention deficit disorder). 10 (15%) children had been diagnosed with ADHD/ADD, or were undergoing diagnostic procedures prior to the study. Comorbidity occurred in six children with ADHD/ADD, five had Oppositional Defiant Disorder (ODD), two had emotional disorder, one had both ODD and emotional disorder. Emotional disorders were found in six children, of whom three had only emotional disorder, one had comorbid ODD and two ADHD (Table 3). Criteria for more than one diagnosis of affective- or anxiety disorder was met in four children (Table 3). A diagnosis of encopresis, enuresis or eating disorder was not given to any of the children, as it was not possible to entangle the origin of these problems within the scope of the study as these symptoms for many children were part of the CP diagnosis. None of the children met criteria for psychotic disorder, substance abuse disorder or Post Traumatic Stress Disorder (PTSD), however 16 (29%) had been exposed to traumatic events.

### 3.2.3. *Mental health symptoms*

Mental health symptoms defined as meeting 75% of criteria for a distinct child psychiatric disorder, were found in more than half of the children (33/56), of whom 23 additionally met criteria for a child psychiatric disorder. Of these 23 children, 21 met criteria for ADHD/ADD, two met criteria for emotional disorder, and two met criteria for both disorders.

One in four were considered mentally healthy, and did not meet criteria for a psychiatric disorder, nor subthreshold mental health symptoms.

### 3.3. *Psychiatric disorders and functional level*

Functional level, measured by level of GMFCS and MACS, as well as subtype of CP, did not impact the risk for psychiatric disorders in children with CP (Table 4). Increased risk for psychiatric disorder was only found for children with communication problems (OR: 3.4; 95% CI 1.1–10.8) (Table 4). Among 56 children with GMFCS level I–IV, 14 were intellectually disabled (ID), of whom 11 met criteria for ADHD/ADD (OR: 5.4; 95%CI 1.3–22.3) (Table 4). Among the children with ID, none met criteria for emotional disorders. When the 14 children with ID were excluded from this group (56), half (21/42) of the children with cerebral palsy met criteria for a psychiatric disorder.

## 4. Discussion

### 4.1. *Psychiatric disorders and the use of Kiddie-SADS among children with CP*

More than half of children with cerebral palsy in the present population based study met diagnostic criteria for a child psychiatric disorder, of which ADHD/ADD was the most common. In addition, one in four met criteria for one or more comorbid psychiatric disorders, and three in four children either met criteria for a psychiatric disorder or had subclinical symptoms of mental health problems. The cohort was representative regarding types of CP and comorbid conditions, and the prevalence of mental health problems is comparable to other populations (Andersen et al., 2008). Kiddie-SADS was found to be an appropriate instrument for detecting psychiatric disorders for the majority of the participants, however for the children with a severe degree of CP (Intellectual Disability (ID) and GMFCS level V), it was impossible to make sufficient distinctions between symptoms of discomfort due to emotional problems and symptoms of physical discomfort. Parents of severely affected children reported relevant mental health symptoms and seemingly appropriate interpretations of subtle signals of anxiety and mood changes. This information was therefore included as mental health symptoms, a separate entity as opposed to psychiatric disorders, and considered to be of importance.

### 4.2. *Prevalence of psychiatric disorders and mental health problems*

More than half of the children met criteria for a psychiatric disorder, ADHD/ADD being the most frequent. This corresponds to a high prevalence found in previous studies using screening instruments (Parkes et al., 2008; Sigurdardottir et al., 2010). The Icelandic study obtained information from teachers as well as parents, and found even higher prevalence when information was provided by teachers, indicating that parents might tend to underreport, especially peer related issues. Studies of general childhood populations have found mental health disorders in one in ten British children, and one in fifteen Norwegian children (Heiervang et al., 2007; Meltzer, Gatward, Goodman, & Ford, 2000). The prevalence in the present study was considerably higher, which correlates to the prevalence of psychiatric disorders found in unilateral CP by Goodman and Graham (1996). One in seven children met criteria for Oppositional Defiant Disorder (ODD), and several parents suggested during the interview, that this contributed to peer problems, loss of friends, and reduced social interaction.

Subthreshold (mental health) symptoms were recorded as a separate entity, and for children meeting criteria for ADHD/ADD, additional emotional symptoms were common, indicating an interaction between behavioural and emotional symptoms which adds to the complexity of the condition. Many of the symptoms described as ODD symptoms, occurred during change of activity, or in incomprehensible situations, and might represent precursor symptoms of compulsive behaviour. Anxiety and compulsive behaviour were prevalent in the present study. Parents gave elaborate descriptions during the interview indicating that compulsive behaviour strongly interfered with social interaction and caused considerable strain to many families.

#### 4.3. *ADHD/ADD and complicating factors*

The complexity of the CP condition constitutes a challenge when diagnosing ADHD/ADD, and differentiating between other causes for inattention such as epileptic disorders, intellectual disability and chronic pain, in addition to communication difficulties, and sleep disorders (Newman, O'Regan, & Hensey, 2006). A study including children with CP with and without epilepsy, found increased risk for mental health problems in children with CP and epilepsy (Carlsson et al., 2008). Epilepsy was not a significant factor in the present study, and we found almost one in four children met criteria for a psychiatric disorder even when children with intellectual disability (ID) and epilepsy were excluded. In addition, half of the CP population with mild or moderate motor function problems without ID met criteria for ADHD/ADD, indicating that the high level of psychiatric disorders in this group is a robust finding. One could suggest that the brain lesion in itself causes an increased prevalence of psychiatric disorders, as did Graham and Rutter in the Isle of Wight study, pointing to a direct brain behaviour link. They found psychiatric disorders five times more common in children with conditions originating from the brain than in the general population, and three times more common than in children with chronic physical disabilities originating from below the brainstem (Graham & Rutter, 1968). Goodman found an association between IQ and psychiatric problems, but suggested low IQ a predictor of a neurobiological condition rather than an etiological factor for psychiatric disorders, as he found a high prevalence of psychiatric disorders even among the least disabled children with CP (Goodman & Graham, 1996).

#### 4.4. *Psychiatric disorders and communication problems*

Communication problem was a significant predictor of psychiatric disorder in the present study. Voorman found an association between communication problems and behaviour problems worsening over time in children with CP in a three year follow-up study (Voorman, Dallmeijer, Van Eck, Schuengel, & Becher, 2010). It remains to be further investigated whether the psychiatric disorders originate from the brain lesion itself, or are related to frustrations due to communicative impairment. This may indicate a need for stronger emphasis on early interventions to enhance communication skills among children with CP.

#### 4.5. *Psychiatric disorders and intellectual- and physical disability*

Parkes found among children with all types of CP, a higher risk for psychological impairment in children with intellectual disabilities (ID), but an apparently lower risk in children with severe physical impairment (Parkes et al., 2008). We did not find differences in rate of psychiatric disorders between GMFCS levels I–IV, types of CP and level of ID, which is contradictory to previous findings. This could be explained by the semistructured interview allowing a broader description of the child's symptoms when identifying psychiatric disorders.

#### 4.6. *Awareness of mental health problems in the follow up of children with CP.*

A substantial number of children were reported by parents to have had experienced one or more traumatic incidences, such as falling out of wheelchairs, sexual insults and loss of friends and helpers, and parents had noticed the negative impact of these incidences on their children. Information of this kind does not necessarily reach the habilitation team, despite its potential importance for the child. With as many as three in four children meeting criteria either for a psychiatric disorder or a mental health symptom, there seems to be a need for clinical tools to detect these symptoms at an early stage. In two third of the children meeting criteria for ADHD/ADD in the present study, the attention problems had not yet been addressed prior to the study, suggesting a need for mental health screening programs for children at preschool age.

#### 4.7. *Limitations*

The whole cohort was included regardless functional level, however small numbers with lack of control group implies limitations to the study.

The clinical population based study design resulted in non-homogenous assessment methods of the cognitive level for half of the population, which reduces the strength of the study. Information was collected from parents only, limiting us to their understanding of their child's mental health status, however we consider parents to be the best informants, understanding the child's often limited communication. Information about ADHD/ADD symptoms was provided by parents only, knowing that a diagnosis is based on symptoms occurring in different settings. Trying to compensate, parents were asked specifically about symptoms of inattention both at home and at school. For the purpose of this study, we diagnosed each disorder individually without taking into account the interaction between disorders, which would have been done in a clinical situation.

Symptoms of Pervasive Developmental Disorder were not addressed in the present version of the Kiddie-SADS interview, and this should be done in further studies. All the Kiddie-SADS interviews were conducted by the same person, which provides interrater stability, but carries a risk for systematic classification bias.

#### 4.8. Conclusions and clinical implications

In a cohort of school age children suffering from cerebral palsy, more than half met criteria for a child psychiatric disorder. The findings indicate that mental health should become part of follow-up services for children with CP. The complexity of the condition asks for a comprehensive approach with a need for establishing shared knowledge about mental health issues in children with CP between child habilitation units, and child psychiatric services.

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## Research Article

# Mental Health in Children with Cerebral Palsy: Does Screening Capture the Complexity?

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**Introduction.** Children with cerebral palsy (CP), one of the most common childhood neurological disorders, often have associated medical and psychological symptoms. This study assesses mental health problems compared to population controls and the ability of a mental health screening tool to predict psychiatric disorders and to capture the complexity of coexisting symptoms. **Methods.** Children with CP ( $N = 47$ ) were assessed according to DSM-IV criteria using a psychiatric diagnostic instrument (Kiddie-SADS) and a mental health screening questionnaire (SDQ). Participants from the Bergen Child Study, a large epidemiological study, served as controls. **Results.** Children with CP had significantly higher means on all problem scores including impact scores. Two in three children scored above 90th percentile cutoff on Total Difficulties Score (TDS), and 57% met criteria for a psychiatric disorder, yielding a sensitivity of 0.85 and a specificity of 0.55. Mental health problems coexisted across symptom scales, and peer problems were highly prevalent in all groups of psychiatric disorders. **Conclusion.** A high prevalence of mental health problems and cooccurrence of symptoms were found in children with CP compared to controls. Screening with SDQ detects mental health problems, but does not predict specific disorders in children with CP. ADHD is common, but difficult to diagnose due to complexity of symptoms. Mental health services integrated in regular followup of children with CP are recommended due to high prevalence and considerable overlap of mental health symptoms.

## 1. Introduction

**1.1. Cerebral Palsy (CP).** CP is one of the most common neurodevelopmental conditions in childhood, affecting 2-3/1000 [1, 2]. While motor impairment is the diagnostic basis, the disorder often presents with associated symptoms and a wide range of related impairments such as epilepsy, pain, and cognitive and communicative impairments. Mental health problems as another main associated symptom, have recently gained awareness, as assessed by screening questionnaires [3-6] and by diagnostic interviews [7]. Using a diagnostic interview, one in two children with CP met criteria for a psychiatric disorder, of which attention deficit hyperactivity disorder (ADHD/ADD) was the most common. Additionally, one in five children had more than one diagnosis, and

the presence of psychiatric disorders was not significantly determined by type and severity of the CP condition [7]. The complex symptom presentation is in accordance with the Early Symptomatic Syndromes Eliciting Neurodevelopmental Clinical Examination (ESSENCE) model that emphasizes the coexistence of symptoms in multiple domains in neurodevelopmental disorders [8]. Further, there is growing evidence that many children with neurodevelopmental disorders are more prone to psychiatric disorders in adulthood, some of which can be screened for and treated in childhood [9, 10]. While there is growing awareness about the complexity of the CP condition, most of the health services are provided in specialized paediatric clinics with motor impairments as the main focus. To detect mental health problems in children with CP, screening questionnaires may be useful. However,

little is known regarding the ability of these questionnaires to disentangle the complexity of mental health problems in children with CP.

**1.2. Mental Health Screening.** Questionnaires such as the Child Behaviour Checklist (CBCL) [11] and the Strengths and Difficulties Questionnaire (SDQ) [12, 13] have both been used to describe the prevalence of mental health symptoms in children with CP [3, 4]. An Icelandic study using CBCL and comparing 36 preschool children having CP with a randomly selected sample of preschool children found that 40–50% of children with CP suffered substantially from behavioural and emotional problems as reported by parents, demonstrating the early coexistence of these symptoms as suggested by the ESSENCE model [4, 8]. A high prevalence of diverse mental health problems have also been found in other studies including school age children when using the SDQ [3, 14, 15]. Children with CP scoring above abnormal range were found in one of four children in a European multicentre study, and in a recent Canadian study [3, 15], and an even higher number in a study including children with CP hemiplegia only, a subtype generally less severely affected [14]. All three studies has used British comparison norms. Peer problems were found to affect more than one third of children with CP in these studies. Likewise, hyperactivity problems were found in one of four children, conduct problems in one of six children, and emotional problems ranging from 17% to 32%. These findings indicate a high prevalence of mental health problems in children with CP and a considerable coexistence of problems.

**1.3. The Strengths and Difficulties Questionnaire.** The SDQ is a short 25-item screening tool to detect mental health problems and prosocial behaviour in children. In combination with the impact scores, which depict the impairment from mental health problems in response to family, social, and school situations, it has shown accuracy in detecting psychiatric disorders, and has been thoroughly validated both in general populations, and in populations of children suffering from chronic illnesses [12, 13]. Psychometric properties of the Total Difficulties Score (TSD) of the SDQ compared to the Development and Well-being Assessment (DAWBA) has shown a sensitivity level of 0.63 in the general British childhood population, up to 0.87 in at-risk populations, likewise specificity has shown values from 0.47 in an at-risk populations, to 0.95 in a general British childhood population [12, 16, 17]. To our knowledge, none of the previously used questionnaires have yet been validated against a diagnostic interview in a population of young children with CP, despite knowledge that there is a considerable overlap of mental health symptoms in addition to a complexity of somatic symptoms which may affect the psychometric properties of the screening tool.

**1.4. Using the ESSENCE Model as a Framework.** We wanted to assess mental health problems in children with CP compared to population-based controls and to assess frequency and coexistence of symptoms. Secondly, we wanted to assess the ability of a mental health screening instrument (SDQ) to sufficiently detect prevalence and coexistence of mental health problems in children with CP, comparing SDQ findings to

results from a diagnostic psychiatric interview (the Kiddie-SADS).

## 2. Methods

**2.1. Population.** A cohort of children with cerebral palsy (CP) in the three western counties of Norway born 2001–2003 were invited to participate in the study. Cerebral palsy (CP) was stated for 101 children prior to the study. Three of the children had however subsequently been re-diagnosed and given a different neurological diagnosis and were therefore excluded from the study. Of the 98 children invited, 67 (68%) participated and were examined regarding psychiatric disorder. For 11 children however, diagnosing a psychiatric disorder using a diagnostic interview was inappropriate due to severe disability, and these children were omitted from the study. The remaining 56 children were included in the present study. The population has been described in detail in an earlier study [7].

The Bergen Child Study (BCS) served as control group [18]. This study is a large longitudinal population-based study involving all children (9155) in the two Norwegian municipalities Bergen and Sund with matching parent SDQ obtained from 6297 children. It has been described in detail elsewhere [19], and we will only give a brief description here. The whole population was assessed for mental health problems using the Strengths and Difficulties Questionnaire (SDQ) as well as answering a question about chronic illness or disability. Data collected when the children were 7–9 years old were used as comparisons for the present study.

**2.2. Classification, Functional Levels, and Medical Information.** Cerebral palsy was classified according to ICD-10 criteria with the following subgroups: spastic bilateral and unilateral, dyskinetic, atactic, or not further classified. Functional level was classified by the Gross Motor Function Classification System (GMFCS) and Manual Ability Classification System (MACS) which distinguishes five levels (I–V), with level V being the most severe [20, 21]. The cognitive level was recorded through information in the medical record, or through the educational system, and was verified by parents during the interview if results from updated cognitive tests were not available. The population is described in detail in a previous study [7].

**2.3. Mental Health: SDQ.** The Strengths and Difficulties Questionnaire (SDQ) consists of 25 items, of which four record problem domains, each including five items, and one prosocial domain (scale) including five items. Each item can be answered with “not true,” “somewhat true,” or “certainly true” rated 0–2 for negatively worded items, and inversely 2–0 for positively worded items. The problem domains are *hyperactivity problems* including items such as inattentiveness and distractibility, *conduct problems* including items such as disobedience and temper tantrums, *emotional problems* including items such as anxiety and worry, *peer problems* including items such as loneliness and preferring adult company. *Prosocial behaviour* consists of items such as being helpful and kind.

Combining the four problem subscales (0–10) computes the Total Difficulties Score (TDS) (0–40). The SDQ also includes an impact score (IS) which measures the impact of mental health problems. For each of the subscales, a score at or above the 90th percentile of the controls was defined as screen positive and a Total Difficulties Score at or above the 90th percentile as risk of having a psychiatric disorder.

**2.4. Psychiatric Disorders: Kiddie-SADS.** Parents of children with CP were interviewed using the Schedule for Affective Disorders and Schizophrenia for School-Age Children: Present and Lifetime Version (6-18) 10.04.00 (Kiddie-SADS), a semistructured child psychiatric diagnostic interview designed to unveil psychiatric symptoms within the following groups of disorders: affective, anxiety, psychotic, eating, attention/hyperactivity, oppositional defiant, conduct, tics, substance abuse, and posttraumatic stress disorders, as well as encopresis and enuresis. Diagnostic conclusions were drawn according to the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV). Parents were interviewed, and we recorded present symptoms. All interviews were conducted by the first author, a child and adolescent psychiatrist. A *psychiatric disorder* was ascertained if criteria listed in the DSM-IV for each specific diagnosis were fulfilled, including severity and duration of specific symptoms.

**2.5. Mental Health Screening and Psychiatric Disorder.** Mental health problems recorded using the SDQ were compared to psychiatric disorders (DSM-IV criteria) for the following symptom-disorder pairs: SDQ-emotional problems compared to emotional disorders, SDQ-hyperactivity problems compared to ADHD/ADD, and SDQ-conduct problems were compared to ODD and conduct disorders. Finally, the SDQ-Total Difficulties, SDQ-peer problems, and SDQ-impact scores were compared to any psychiatric disorder.

**2.6. Statistical Analyses.** Independent *t*-tests were used to compare mean scores from children in the CP group to those from the general population. *Sensitivity* was defined as the proportion of actual positives which were identified as such. *Specificity* was defined as the proportion of actual negatives which were correctly identified as such. *Positive predictive value (PPV)* was defined as the proportion of positive tests that were true positives, and *negative predictive value* was defined as the proportion of negative tests that were true negatives. *Sensitivity, specificity, PPV, and NPV above 80% were regarded as high.* Cross-tabulations and 90th percentile cutoff were used to calculate these parameters.

Effect size was defined using Cohen's *d* measuring the standardized mean difference between the study population and controls; a large effect size was defined as  $\geq 0.8$ , a medium effect size as  $\geq 0.5$ , and a small effect size as  $\geq 0.3$ . Cross-tabulations were done to evaluate possibly significance between high scorers on SDQ-TDS and types and severity of CP.

A linear regression analysis was used to explore the influence of each of the five symptom items in the peer

TABLE 1: Demographics and functional data for a population of children with cerebral palsy living in the Western Health Region of Norway at school starting age.

	N (%)
N = 47	
Boys	30 (64)
Cerebral palsy subtype	
Bilateral	25 (53)
Unilateral	18 (38)
Ataxia/dyskinesia	4 (9)
GMFCS level <sup>i</sup>	
I-II	38 (81)
III-IV	9 (19)
Intellectual disability <sup>ii</sup>	10 (21)
Epilepsy	9 (19)
Visual impairment <sup>iii</sup>	19 (40)

<sup>i</sup>Gross Motor Function Classification System.

<sup>ii</sup>Defined as IQ < 70.

<sup>iii</sup>Defined as any visual impairment.

problem score, the latter being the dependent variable, and the five items were independent variables.

### 3. Results

**3.1. Population.** Of the 56 children in the present study, 47 completed the SDQ. The mean age was 7 years and 3 months (87.6 months, SD 6.5). Thirty (64%) were boys, more than half had CP diplegia, and four of five children had Gross Motor Function Classification System (GMFCS) levels I and II. One of five had an intellectual disability, and approximately the same number suffered from epilepsy (Table 1). Type and severity of CP conditions were not significantly associated with high scorers on SDQ-Total Difficulties Score.

**3.2. Mental Health Problems in Children with Cerebral Palsy Compared to a Population-Based Control.** Children with CP had significantly higher mean scores compared to controls, affecting all problem scales as well as impact score (Table 2). A large effect size was found for the Total Difficulties Score, hyperactivity problems, conduct problems, peer problems and impact score, and moderate effect size for emotional problems, and a small effect size for prosocial behaviour (Table 2).

**3.3. Coexisting Symptoms.** When comparing all SDQ screen positives across the problem scales, including TDS and impact score, with the following four groups of psychiatric disorders, emotional disorders, conduct disorders, ADHD/ADD, and any psychiatric disorders, we found a high prevalence of coexisting screen positives across groups of psychiatric disorders in children with CP (Table 3). There was a discrepancy between screen positives and children meeting criteria for a psychiatric disorder, most prevalent for SDQ-hyperactivity problems (3/24) and SDQ-conduct problems (2/4). For the 24 children meeting criteria for ADHD/ADD,

TABLE 2: Mental health for children with cerebral palsy (CP) using mean scores of the Strengths and Difficulties Questionnaire compared with controls.

	CP group (47)		Controls <sup>a</sup> (7007)		<i>t</i> (df)	<i>P</i>	95%	CI	Cohen's <i>d</i>
	Mean	SD	Mean	SD					
Emotional problems	2.6	2.2	1.3	1.7	4.0 (46)	<.001	0.6	1.9	0.66
Conduct problems	2.5	1.5	1.0	1.3	6.7 (45)	<.001	1.1	2.0	1.07
Hyperactivity problems	4.3	1.5	2.7	2.1	7.8 (47)	<.001	1.2	2.1	0.88
Peer problems	4.5	1.5	1.0	1.5	16.0 (46)	<.001	3.1	4.0	2.33
Total difficulties scores	13.9	5.3	5.6	5.0	10.7 (46)	<.001	6.8	9.9	1.61
Prosocial behaviour <sup>b</sup>	7.8	2.1	8.5	1.5	-2.2 (45)	0.03	1.3	-0.1	0.39
Impact score	2.9	3.8	0.4	1.5	4.6 (46)	<.001	1.4	3.7	0.94

<sup>a</sup>Bergen Child Study, <sup>b</sup>higher scores indicate more prosocial behaviour.

TABLE 3: Coexisting mental health symptoms in children with cerebral palsy meeting criteria for a psychiatric disorder according to DSM-IV criteria assessed by Kiddie-SADS.

Psychiatric disorder	Children scoring above 90th percentile cutoff on the Strengths and Difficulties Questionnaire					
	Emotional problems	Conduct problems	Hyperactivity problems	Peer problems	TDS <sup>a</sup>	Impact score
Emotional ( <i>N</i> = 5)	5/5	3/5	1/5	5/5	5/5	3/5
Conduct/ODD <sup>b</sup> ( <i>N</i> = 4)	2/4	2/4	2/4	4/4	4/4	4/4
ADHD/ADD <sup>c</sup> ( <i>N</i> = 24)	9/24	10/24	3/24	23/24	19/24	19/24
Any psych <sup>d</sup> ( <i>N</i> = 27)	12/27	12/26	4/27	26/26	22/26	20/27

<sup>a</sup>TDS: Total Difficulties Score; <sup>b</sup>ODD: oppositional/defiant disorder; <sup>c</sup>ADHD: attention-deficit/hyperactivity disorder; ADD: attention deficit disorder; <sup>d</sup>Any psych: any psychiatric disorder.

23 were screen positives for peer problems. To gain understanding of the large proportion experiencing peer problems in the present study, a regression analysis was done to identify the impact of each of the five items in the peer problem scale. We found preference for adult company, the most weighted item, followed by lack of close friendships, loneliness, being bullied, and not being liked in diminishing order.

**3.4. SDQ and Psychiatric Disorder.** Screening efficiency of the SDQ-TDS in children with cerebral palsy was assessed by comparing SDQ screen positives (i.e., scores above 90th percentile) with children meeting criteria for a psychiatric disorder according to Kiddie-SADS (Table 4). Sensitivity for all symptom groups varied between 0.13 and 1.0, and specificity varied from 0.55 to 0.87. For TDS, sensitivity was 0.85 and specificity 0.55 (Table 4).

## 4. Discussion

**4.1. Main Findings.** Children with CP in the present population based study had significantly higher means on all SDQ subscales compared to the general childhood population. There was considerable mental health symptom overlap across all categories, and peer problems were highly prevalent in all four groups of psychiatric disorders. Sensitivity for TDS was adequate for SDQ compared to any psychiatric disorders using a semistructured child psychiatric diagnostic interview (Kiddie-SADS).

**4.2. Mental Health Problems in Children with Cerebral Palsy.** Compared to controls, mean scores in children with cerebral palsy were significantly higher on all scales, which is consistent with previous findings. Previous studies have found high TDS mean scores 11.0–12.4, which are similar to those found in the present study [3, 6, 14, 15], even when children with GMFCS level V and intellectual disability (ID) were excluded and therefore as expected had less medical complications than a population consisting of all five GMFCS levels [1, 7].

Mean scores for emotional problems in the present study were similar to the study by Parkes et al. since 2008 [3] and Brossard-Racine et al. where all CP subgroups were included [15]. In the study by Parkes et al. including children with CP hemiplegia only however [14], a fourfold higher mean score for emotional problems was found. This may indicate that anxiety and depressive disorders are more prevalent in children having CP with a functional level closer to their healthy peers, as they perhaps more often are met with the same expectations in all areas as children without CP, leading to a sense of shortcoming.

For the conduct and hyperactivity subscales, mean scores in the present study were similar (2.5 and 4.3, resp.) to those found in comparable studies, 1.7–2.0 for conduct problems and 4.3–4.8 for hyperactivity problems [3, 14, 15]. This finding underlines a consistently high prevalence of behavioural problems in children with CP, which was also confirmed in a recent meta-analysis where one of four children with CP were found to have behaviour disorder [22]. Mean scores

TABLE 4: Mental health screening compared to having a psychiatric disorder (according to DSM-IV criteria assessed by Kiddie-SADS) for children with cerebral palsy at school starting age.

SDQ-symptoms versus psychiatric disorders	Above 90th percentile on SDQ <sup>a</sup> N (%)	Psychiatric disorder present N (%)	Sensitivity	Specificity	PPV	NPV
Emotional symptoms versus emotional disorders <sup>b</sup>	14 (29.8)	5 (10.6)	1.00	0.79	0.36	1.00
Conduct problems versus conduct disorder/ODD <sup>c</sup>	16 (34.8)	4 (8.5)	0.50	0.67	0.13	0.93
Hyperactivity problems versus ADHD/ADD <sup>b</sup>	6 (12.8)	24 (51.1)	0.13	0.87	0.50	0.49
Total Difficulties Score versus any psychiatric disorder <sup>c</sup>	31 (67.4)	26 (56.5)	0.85	0.55	0.71	0.73
Peer problems versus any psychiatric disorder	41 (89.1)	26 (56.5)	1.0	0.25	0.63	1.0
Impact score versus any psychiatric disorder	27 (57.4)	27 (57.4)	0.74	0.65	0.74	0.65

PPV: positive predictive value; NPV: negative predictive value; <sup>a</sup>SDQ: strengths and difficulties questionnaire <sup>b</sup>N = 47; <sup>c</sup>N = 46.

for peer problems however were higher in the present study compared to previous studies of children with cerebral palsy (2.7–3.0) [3, 14, 15]. Peer problems were found in nine of ten children with CP in the present study, whereas Parkes et al. found peer problems in one of three children in their studies and two of five children in the Canadian study by Brossard-Racine et al. Peer problems were highly prevalent across all diagnostic groups, that is, emotional disorders, conduct disorders/ODD, as well as hyperactivity disorders, and could be related symptomatically to autism spectrum disorders (ASD) which have been found to be prevalent in children with CP [23]. Similarly, ASD is often part of neurodevelopmental conditions in general with a considerable overlap of conditions as described in the ESSENCE model [8]. When analysing the peer problems construct in detail, preference for adult company and loneliness were highly prevalent in the present study. This finding is coherent with information given by parents during the Kiddie-SADS interview, where many parents pointed to their children taking part in organized school and leisure activities, although they often had few mutual own-aged friendships. This could indicate that some might be passively watching rather than being active participants in peer activities. Another possible hypothesis for peer problems is slow cognitive processing and speech, which for many children with CP with normal IQ may result in difficulties matching their peers in spontaneous play activities, perhaps resulting in a preference for adults to accompany during activities [24].

**4.3. Behaviour Problems.** For ADHD/ADD which was the most prevalent psychiatric disorder in the present study, the screening efficacy using SDQ in the present study was 13%, about half of that found in the studies by Parkes et al. and Brossard-Racine et al. [3, 14, 15], despite a high prevalence of ADHD/ADD (52%) when parents of the same children were interviewed with the Kiddie-SADS and diagnosed according to DSM-IV criteria. On the contrary, symptoms of conduct disorders in the present study were almost twice as high as those found in the studies by Parkes et al. and Brossard-Racine et al. [3, 14, 15], despite a much lower prevalence

(8.5%) when diagnosing according to Kiddie-SADS. Other studies have pointed to cooccurrence of symptoms between ODD (oppositional defiant disorders) and ADHD/ADD, emotional problems and peer problems [25]. Although the studies by Parkes et al. and Brossard-Racine et al. include children with all CP subtypes, similar to the present one, mental health symptoms may be recognized as conduct or peer problems rather than hyperactivity problems in the study population. Attention problems may not be recognized as such, as these children in Norway to a large degree have individual support during most of their time at school, and parents are often primed to assist their children with all practical issues at home and may not recognize their children's challenges in memory or attention span. During the semistructured Kiddie-SADS interview, these issues were elaborated, and many children met criteria for attention problems when parents were asked to judge their child's attention span in a situation where they were left to do their school work or organize their belongings as would their peers or siblings. Perhaps the screening tools are more sensitive to symptoms such as peer and conduct problems in a population of children with CP than to hyperactivity problems and therefore may not fully capture the attention and/or hyperactivity problems these children suffer, as motor symptoms due to the CP condition may disguise restlessness and distractibility which may be interpreted differently.

**4.4. Coexisting Conditions.** In accordance with the ESSENCE model, we found an extensive overlap of mental health symptoms in children with CP meeting criteria for a psychiatric disorder. Gillberg et al. found a high prevalence of other psychiatric disorders such as emotional problems and ODD in children with ADHD [26], which has also been found in a previous study including children in the present study [7]. Similarly, ADHD was found in half of all children with mild mental retardation MMR in association with other developmental problems [27]. ADHD seems to be a common associated factor in many different developmental disorders or neurological conditions stemming from the brain and may be associated with the immature nervous system being

vulnerable to brain injury in the neonatal period. Perhaps this is one of the reasons for a high level of diversity and overlap of mental health symptoms in children with disorders such as CP which occurs in the neonatal period and why mental health symptoms in children with CP seem to present differently from the general childhood population. It may also explain why the SDQ seems to be a less accurate screening tool for ADHD/ADD in children with CP than one would expect from studies conducted in a general childhood population. The extensive overlap of symptoms constitutes a considerable challenge both in providing diagnostic tools and in gaining competence in recognizing psychiatric disorders in children with a complexity of interacting symptoms.

**4.5. Screening Efficiency of the SDQ.** The child psychiatric diagnostic interview Kiddie-SADS and the Strengths and Difficulties Questionnaire were both used to assess the same children which enabled us to evaluate the psychometric properties of the SDQ when using the Kiddie-SADS as the gold standard. When comparing specific mental health symptoms and psychiatric disorders, we found a large range in sensitivity and specificity between the least and the most sensitive symptom-disorder pairs in this study. Sensitivity for TDS compared to any psychiatric disorder according to Kiddie-SADS in the present study was 0.85 and specificity 0.55, which differ from those found in the general childhood population yielding sensitivity of 0.63 and specificity of 0.95 [12]. For a population of children referred to mental health services in Norway, sensitivity was 0.85 and specificity 0.52 for TDS [16], and for children with chronic illnesses in a Norwegian study, sensitivity was 0.81 and specificity 0.69 for TDS [17] which is comparable to the present study. For emotional, conduct, and hyperactivity disorders, however, we found that the positive predictive value was low, indicating that there may be a need for caution when using the SDQ to predict these disorders in children with CP in early childhood.

**4.6. Clinical Implications.** In the present study, 57% met criteria for a psychiatric disorder, and 67% were screen positive on Total Difficulties Score (TDS). Sensitivity of the SDQ for TDS may be considered adequate however predictive value for specific psychiatric disorders is found to be inadequate in the present study. Knowing that mental health problems seem to affect two in three children with CP, with a considerable overlap of mental health symptoms and psychiatric disorders, we are faced with challenges in disentangling and diagnosing a conglomerate of symptoms. Establishing mental health services as part of the regular follow-up program for children with CP seems relevant. In addition, using the SDQ as a screening instrument to identify children with CP without mental health problems, not in need for mental health services, may be legitimate, bearing in mind, however, the high rate of psychiatric diagnosis and the low diagnostic specificity especially for AD/HD which is one of the most frequent diagnosis.

**4.7. Limitations.** The version of Kiddie-SADS used in the present study did not contain a section on autism spectrum

Disorders (ASD), which is a weakness, since all children diagnosed with a psychiatric disorder were screen positive for peer problems. Further, one of three had peer problems without having a psychiatric disorder, and these might represent children with an ASD. Likewise, we did not use the SDQ algorithm for predicting psychiatric disorders as we had a single informant.

A small study population was also a limitation to the study. Similarly, omitting children with GMFCS V and ID from the study may represent a weakness as information on mental health among severely affected children is lacking. Previous studies have attempted to include the most severely affected children; however, it has not been possible to draw diagnostic conclusions regarding psychiatric disorders due to the complexity of motor, cognitive, somatic, and epileptic disorders affecting these children [7, 28]. The current study was however representative of children with CP with GMFCS levels I–IV.

**4.8. Conclusions.** The present study supports the previous literature indicating a high prevalence of mental health problems in children with CP compared to controls. The SDQ disclosed problems relating to peers in more than three in four children, and further studies regarding ASD are recommended. We recommend establishing mental health services for children with CP as part of the regular follow-up program; however screening children with CP for mental health problems in the paediatric clinics using the SDQ seems relevant when established mental health service programs are not available.

## Ethical Approval

The study was approved by the Regional Committee for Medical Research Ethics in Western Norway.

## Conflict of Interests

The authors declare no conflict of interests.

## Authors' Contribution

H. M. Bjorgaas has contributed to the study design, collection of data, and writing the paper. I. Elgen has contributed to the study design, draft, and revision of the paper. T. Boe has contributed to the statistical analysis, draft, and revision of the paper. M. Hysing has contributed to data analysis, draft, and revision of the paper.

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## Research in Autism Spectrum Disorders

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# Autism spectrum symptoms in children with cerebral palsy: Prevalence and co-occurring conditions

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## ABSTRACT

**Purpose:** To explore autism spectrum symptoms in children with cerebral palsy (CP), and the association between autism spectrum symptoms and medical and psychiatric comorbidity.

**Methodology:** Parents of children with CP in a Norwegian population were interviewed with a child psychiatric diagnostic instrument, and completed the Autism Spectrum Screening Questionnaire (ASSQ). Medical and socio-demographic data were obtained. ASSQ mean scores were compared to the Bergen Child Study (BCS), both to healthy controls and to subgroups of children with chronic illness in general, and neurological disorders specifically.

**Results:** Interviews and data collection were completed for 47 children, of whom 30 were boys, most had spastic CP, and were less severely affected by CP. Large effect sizes were found when comparing ASSQ mean scores in children with CP to children with chronic illnesses and normal controls. One in five children was ASSQ high scorers defined as a score above the 98th percentile of normal controls. A high rate of co-occurring psychiatric disorders, mainly AD/HD, was found in ASSQ high scorers.

**Conclusions:** More attention should be given to autism spectrum symptoms in the regular follow-up of children with CP in an attempt to enhance social functioning.

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## 1. Introduction

Cerebral palsy (CP) is primarily a motor disorder, caused by a cerebral lesion occurring in the developing brain. It is one of the most prevalent neurological disorders in childhood, affecting 2–3/1000 children (Andersen et al., 2008). Motor impairment varies from mild to serious, and is often classified according to GMFCS (Gross Motor Function Classification System) levels I–V, where level V is the most disabling. Conditions co-occurring with CP, such as intellectual disability (ID), communication problems, epilepsy, pain, and psychiatric disorders, are commonly described (Bjorgaas, Hysing, & Elgen, 2012; Parkes et al., 2008; Parkes, White-Koning, McCullough, & Colver, 2009; Sigurdardottir et al., 2010).

The social functioning of children with CP has gained increasing attention, with parents reporting consistently more peer problems in children with CP than in controls (Brossard-Racine et al., 2012; Parkes et al., 2008, 2009). Most studies assessing

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social functioning have used short screening questionnaires covering peer problems, such as the Strengths and Difficulties Questionnaire (SDQ) (Brossard-Racine et al., 2012; Parkes et al., 2008, 2009). This questionnaire was also used in a recent Norwegian study, where parents reported peer problems in 90% of children with CP (Bjorgaas, Elgen, Boe, & Hysing, 2013). Why problems with social functioning are highly prevalent in children with CP, is an interesting question.

Intellectual disability is a well-known risk factor for both mental health problems and problems with social functioning (Parkes et al., 2008), and may account for some of the increased rates of peer problems in children with CP. Studies with focus on peer problems in children with CP have however been conflicting. Some studies have found ID significantly associated with peer problems (Parkes et al., 2009), or predictive of later peer problems (Yude & Goodman, 1999). Others however, have not found ID related to peer problems, but rather peer problems related to less severe motor impairment (Brossard-Racine et al., 2013).

Problems with social functioning have been described in children with other neurological conditions occurring in the developing brain, such as associations between developmental disorders, seizures in early life, intellectual disability (ID) and autism spectrum disorder (ASD) (Matson, Dempsey, Lovullo, & Wilkins, 2008; Saemundsen, Ludvigsson, & Rafnsson, 2007). This is further supported by results from a large scale epidemiological study using the Autism Spectrum Screening Questionnaire (ASSQ) (Ehlers, Gillberg, & Wing, 1999; Posserud, Lundervold, & Gillberg, 2009), where children with neurological disorders were found to have an increased rate of autism spectrum symptoms compared to their peers (Ryland, Hysing, Posserud, Gillberg, & Lundervold, 2012). Supporting the notion that peer problems in children with CP could be related to ASD, a diagnosis of Autism was found in eight percent of children with CP in a register study (Kirby et al., 2011). In a clinical study however, 14% of children with more severe CP met criteria for autism or pervasive developmental disorder (PDD) (Kilincaslan & Mukaddes, 2009). While shared etiology between CP and autism spectrum symptoms might be possible, the exact factors accounting for the overlap are still unclear.

Problems in social functioning may also be intrinsically related to other mental health problems, with considerable overlap between mental health- and peer problems demonstrated in children with CP (Parkes et al., 2008, 2009). A similar pattern was found between psychiatric disorders extensively overlapping with peer problems (Bjorgaas et al., 2012, 2013). Similarly, longitudinal studies have found AD/HD symptoms leading to later peer problems in children with CP (Yude, Goodman, & McConachie, 1998). The demonstrated problems related to social functioning, has spurred the interest in exploring autism spectrum symptoms, and the interplay between these symptoms and co-occurring medical conditions and psychiatric disorders in children with CP.

### *1.1. Aims of the present study*

The aims of the present study were threefold; firstly, to explore the prevalence of autism spectrum symptoms using the ASSQ in children with CP. The second aim was to compare the rate of autism spectrum symptoms in children with CP to children with other chronic illnesses as well as to healthy peers. Thirdly, we wanted to explore if autism spectrum symptoms were related to medical- and psychiatric co-morbidity in children with CP.

## **2. Materials and methods**

### *2.1. Population*

#### *2.1.1. Study population*

All children with a diagnosis of cerebral palsy (CP) living in the Western Health Region of Norway, born in 2001–2003, were invited to take part in the study. The population has earlier been described in detail (Bjorgaas et al., 2012). In short, 67 of a total population of 98 participated. Children with the combination GMFCS level V and ID were excluded from the study as child psychiatric diagnostic conclusions were not possible (Bjorgaas et al., 2012).

#### *2.1.2. Controls*

The Autism Spectrum Screening Questionnaire (ASSQ) data obtained from children with CP were compared to those from the Bergen Child Study (BCS), a large longitudinal population study involving all children in the two Norwegian municipalities Bergen and Sund (Heiervang et al., 2007). Data collected when the children were 11–13 years old ( $N = 5781$ ) were used as comparisons for the present study. Data from children with CP were compared to the general BCS population, and to a subgroup of children having chronic illnesses ( $N = 496$ ), as well as a subgroup within the chronic illness group suffering from neurological disorders ( $N = 99$ ). The BCS and the studies involving subgroups of children with chronic illness are described extensively elsewhere (Heiervang et al., 2007; Hysing, Elgen, Gillberg, Lie, & Lundervold, 2007; Hysing, Elgen, Gillberg, & Lundervold, 2009; Ryland et al., 2012).

### *2.2. Classification*

Cerebral palsy was classified according to ICD-10 criteria with the following subgroups: spastic bilateral and unilateral, dyskinetic, atactic or not further classified.

Functional level was classified by the Gross Motor Function Classification System (GMFCS) which distinguishes five groups, GMFCS levels V being the most severely affected.

The cognitive level ascertained through standardized intelligence tests were obtained through the child's medical record. If these were not available, cognitive level was recorded through information in the medical record, or through the educational system, and verified by parents during the interview. The following variables were recorded through the medical examination, parent interview, and information in the child's medical record; gestational age, epilepsy, intellectual disability (ID), type and severity of CP, as well as communication problems.

### 2.3. Mental health outcomes

#### 2.3.1. Autism Spectrum Screening Questionnaire (ASSQ)

The ASSQ is a 27 item screening questionnaire designed to identify school aged children displaying autism spectrum symptoms, where a comprehensive evaluation of suspected ASD may be indicated. The ASSQ covers a range of symptoms predictive of a diagnosis within the autism spectrum. It is rated on a 3-point scale with total symptom scores ranging from 0 to 54, with highest scores indicating highest symptom load. The ASSQ has been validated in a Norwegian childhood population. For the present study, cut-off scores derived from the validation study defined at the 98th percentile and corresponding to cut-off scores at  $\geq 17$  were used (Posserud et al., 2009). Psychometric properties of the ASSQ as a population screening instrument were found to be good, with a sensitivity of 0.90 when parents were informants (Posserud et al., 2009). Means of ASSQ total scores including all items were compared to children with neurological disorders (Ryland et al., 2012), children with chronic illnesses in general (Hysing et al., 2007), and children from the general population (Posserud, Lundervold, & Gillberg, 2006).

#### 2.3.2. Psychiatric disorders – Kiddie-SADS

Parents of children with CP were interviewed using the Schedule for Affective Disorders and Schizophrenia for School-Age Children: Present and Lifetime Version (6-18) 10.04.00 (Kiddie-SADS), a semi-structured child psychiatric diagnostic interview. It is designed to unveil psychiatric symptoms within the following groups of disorders: affective-, anxiety-, psychotic-, eating-, attention/hyperactivity-, oppositional defiant-, conduct-, tics-, substance abuse- and posttraumatic stress disorders, as well as encopresis and enuresis. Diagnostic conclusions were drawn according to the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV). Parents were interviewed, and we recorded present symptoms. All interviews were conducted by the first author, a child-and adolescent psychiatrist. A psychiatric disorder was ascertained if criteria listed in the DSM-IV for each specific diagnosis were fulfilled, including severity and duration of specific symptoms.

### 2.4. Statistics

All analyses were performed using the PASW SPSS 20.0. For the ASSQ data, population cut-off norms at  $\geq 17$  were used to determine the 98th percentile. Numbers and percentages as well as means of the ASSQ total scores were analyzed for the following items; gender, gestational age, type and severity of CP, communication problem, epilepsy as well as intellectual disability. Means of ASSQ total scores were analyzed with all ASSQ single items, and without the potentially confounding item "clumsy". These were compared to those of children with neurological disorders, children with chronic illnesses, and healthy controls. Effect sizes (Cohen's *d*) were calculated using the ASSQ total scores, including all items, and defined as follows; large effect size at or above 0.8, medium at or above 0.5, small at or above 0.2. Percentages of single ASSQ symptom items were determined with descriptive analysis, and results were compared with and without the confounding item "clumsy".

## 3. Results

### 3.1. Population

For the 56 consenting participants, 9 children did not return the ASSQ, resulting in 47 children taking part in the present study. For these children, results from both the Kiddie-SADS interview and the ASSQ's were available. Mean age was 7 years and 3 months (87.6 months, SD 6.5). Almost two in three were boys, more than half had bilateral CP, and four in five children had GMFCS level I or II. One in five had intellectual disability (ID), and the approximate same number suffered from epilepsy (Table 1).

### 3.2. ASSQ high scorers, percentage of ASSQ single items and mean scores in children with CP

Nine of the 47 children (19%) had scores at or above 17 (98th percentile) on the ASSQ. Omitting "clumsy", which was the most prevalent single item, made no difference in the rate of children scoring above the 98th percentile. Mean total ASSQ scores with and without the item "clumsy" are listed in Table 2. Numbers and percentage of ASSQ single items, presented in

**Table 1**

Characteristics of children with cerebral palsy at school starting age. Means of ASSQ<sup>a</sup> total scores according to functional and medical variables with *p*-values derived for each variable paired. *N* = 47.

	<i>N</i> (%)	ASSQ means (SD)	<i>p</i> -Value
Gender			
Boys	30 (64)	10.1 (9.9)	0.07
Girls	17 (36)	5.3 (5.8)	
Gestational age			
<32weeks	11 (24)	6.6 (9.3)	0.45
>32weeks	35 (76)	8.9 (8.9)	
Type of CP <sup>b</sup>			
Spas. unilat <sup>c</sup>	18 (38)	6.7 (8.5)	0.30
Spas. bilat <sup>d</sup>	25 (53)	9.4 (9.0)	0.39
Atactic/dyskinetic	4 (9)	9.5(11.1)	0.80
GMFCS <sup>e</sup> level			
III–IV	9 (19)	11.3 (8.5)	0.27
I–II	38 (81)	7.7 (8.9)	
Intellectual disability			
Yes	10 (21)	14.4 (6.7)	0.01
No	37 (79)	6.8 (8.7)	
Epilepsy			
Yes	9 (19)	13.2 (7.3)	0.07
No	38 (81)	7.2 (8.9)	
Communication problem			
Yes	18 (38)	11.1 (9.1)	0.10
No	29 (62)	6.7 (8.4)	

<sup>a</sup> Autism Spectrum Screening Questionnaire.

<sup>b</sup> *p*-Values are analyzed for each type of CP comparing against all other types of CP.

<sup>c</sup> Spastic unilateral.

<sup>d</sup> Spastic bilateral.

<sup>e</sup> Gross Motor Function Classification System.

**Table 2**

Mean ASSQ<sup>a</sup> total score for children with cerebral palsy at school starting age compared to groups of children derived from the Bergen Child Study, effect sizes (cohen's *d*) for the magnitude of the mean difference are measured between children with CP (all items) and comparison groups. *N* = 47.

	Cerebral palsy all items	Omitting "clumsy"	Neurological disorders, <i>N</i> = 99	<i>d</i>	Other chronic illnesses, <i>N</i> = 496	<i>d</i>	Healthy controls, <i>N</i> = 5781	<i>d</i>
Mean total score (SD)	8.4 (8.9)	7.6 (8.6)	10.4 (9.7)	−0.2	3.3 (4.3)	0.7	2.9 (4.1)	0.8

<sup>a</sup> Autism Spectrum Screening Questionnaire.

diminishing order, revealed the following items most frequent; "clumsy", "uneven abilities", and "fails to make friends" (Table 3).

### 3.3. Autism spectrum symptoms in children with cerebral palsy compared to children with neurological disorders, other chronic illnesses and healthy peers

Effect sizes were large when comparing mean ASSQ total score of children with CP to children from the general population, medium when comparing to children with a chronic illness, and small when comparing to those of children with a general neurological disorder (Table 2).

### 3.4. Autism spectrum symptoms and co-occurring medical conditions and psychiatric disorders in children with CP

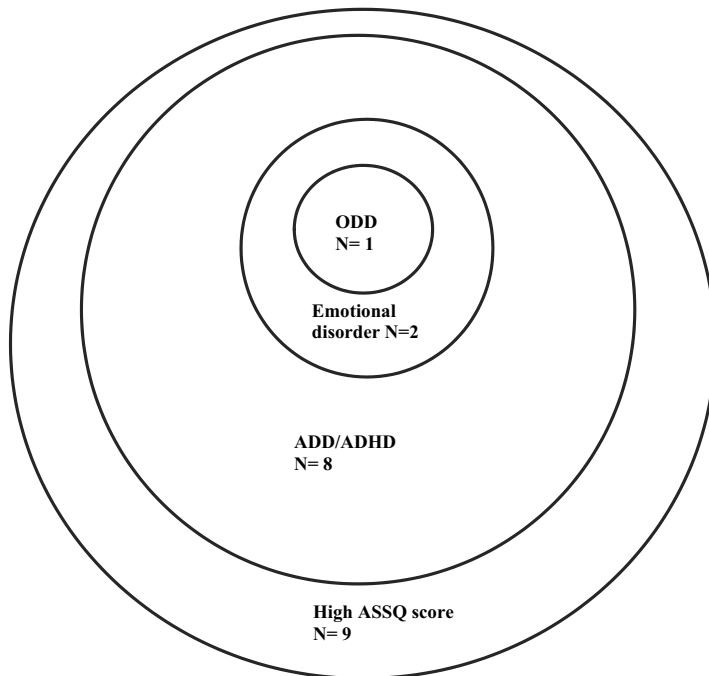
Mean ASSQ scores according to types and severity of CP as well as co-occurring factors are presented (Table 1). Intellectual disability was the only factor significantly associated with a high ASSQ total score  $p = 0.01$ . Nine children scored above the 98th percentile on the ASSQ, of whom eight were boys, one was gestational age <32 weeks, one was GMFCS level III–IV, three had ID, two had epilepsy and four had communication problems. The overlap between ASSQ high scorers and those meeting criteria for psychiatric disorders according to the Kiddie-SADS interview are described in Fig. 1. Eight of the nine children with ASSQ high score met criteria for a child psychiatric disorder. Of these children, all eight met criteria for AD/HD, further two met criteria for an emotional disorder, and one met criteria for a third diagnosis, oppositional defiant disorder (ODD).

**Table 3**

Numbers and percentage of Autism Spectrum Screening Questionnaire (ASSQ) single items in children with cerebral palsy presented in diminishing order of percentage. *N* = 47.

ASSQ symptoms	<i>N</i> (%) <sup>a</sup>
Clumsy ( <i>N</i> = 45)	27 (60)
Uneven abilities ( <i>N</i> = 45)	18 (40)
Fails to make friends	18 (38)
Old-fashioned or precocious	18 (38)
No social fit in language ( <i>N</i> = 46)	17 (37)
Idiosyncratic words	17 (36)
Lives in own world	16 (34)
Accumulate facts ( <i>N</i> = 46)	15 (33)
Poor at games, own rules ( <i>N</i> = 46)	15 (33)
Sociable on own terms only ( <i>N</i> = 46)	14 (30)
Lacks best friend	13 (28)
Different voice/speech	13 (28)
Literal understanding ( <i>N</i> = 46)	11 (24)
Lacks common sense ( <i>N</i> = 46)	11 (24)
Insists on no change ( <i>N</i> = 46)	10 (22)
Idiosyncratic attachment ( <i>N</i> = 46)	10 (22)
Naïve remarks ( <i>N</i> = 46)	10 (22)
Involuntary movements ( <i>N</i> = 46)	9 (20)
Bullied by other children ( <i>N</i> = 46)	9 (20)
Lacks empathy	9 (19)
Unusual posture ( <i>N</i> = 46)	8 (17)
Involuntary sounds	8 (17)
Deviant style of gaze	8 (17)
Robot like language	6 (13)
Compulsory repetitive ( <i>N</i> = 45)	6 (13)
Eccentric professor	3 (6)
Unusual facial expression ( <i>N</i> = 46)	1 (2)

<sup>a</sup> Number and percentage of children with CP scoring answer alternatives "sometimes true" and "always true".



**Fig. 1.** Children with cerebral palsy at school starting age scoring above 98th percentile on ASSQ (Autism Spectrum Screening Questionnaire) with overlapping psychiatric disorders.

#### 4. Discussion

Children with cerebral palsy (CP) had a high rate of autism spectrum symptoms in the present population based study. One in five children with CP scored above the 98th percentile on the Autism Spectrum Screening Questionnaire (ASSQ). The mean level of autism spectrum symptoms was higher than in healthy peers, and in children with other chronic illnesses. A high ASSQ score was associated with intellectual disability (ID). There was a considerable overlap between ASSQ high scorers and children meeting criteria for AD/HD.

An increased prevalence of autism spectrum symptoms was found in the present study, which is in accordance with other studies (Kilincaslan & Mukaddes, 2009; Kirby et al., 2011). Having excluded children with both GMFCS level V and ID, this finding may indicate that ASD is more common than previously known, even among children with GMFCS levels I–II. In fact, peer problems were more common in those least affected by motor impairment in the present study, similar to previous findings (Brossard-Racine et al., 2013). Perhaps children with less severe CP more often take part in activities alongside healthy peers, experiencing shortcomings both in social functioning and in motor activities, as also discussed in other studies (Yude et al., 1998). Some of the autism spectrum symptoms may be overlapping for both cerebral palsy and autism, impairing the conclusions drawn. However, the same ASSQ high score prevalence was found, even when omitting the item “clumsy”. This perhaps indicates a certain robustness in finding a high prevalence of autism spectrum symptoms in children with CP. Mean ASSQ total score from the study population was higher than those found in children with other chronic illnesses and in healthy controls, supporting previous studies finding high ASSQ scores in children with neurological disorders (Ryland et al., 2012).

“Uneven abilities” ranked as the second most frequent ASSQ single item in the present study. Difficulties in cognitive functioning, such as ID or uneven cognitive profiles, may be factors accounting for an increased rate of autism spectrum symptoms found in children with CP. Studies of children with ASD, have found that discrepancies between performance IQ (PIQ) and verbal IQ (VIQ) were related to autism symptom presentation, although the direction of the discrepancy was inconsistent across studies (Black, Wallace, Sokoloff, & Kenworthy, 2009; Joseph, Tager-Flusberg, & Lord, 2002). Although no specific cognitive profiles were obtained in the present study, ID was the only factor significantly associated with ASSQ high score. This is in accordance with other studies involving children with CP, where ID was significantly associated with autism spectrum symptoms or peer problems (Kilincaslan & Mukaddes, 2009; Parkes et al., 2008; Yude & Goodman, 1999). One study however, analyzing ID and peer problems did not come to the same conclusion (Brossard-Racine et al., 2012). Other studies have described hampered cognitive functioning resulting in reduced social functioning and fragile social relationships as a self-reinforcing process in children with CP (Bottcher, Flachs, & Uldall, 2010). An increased prevalence of peer problems is perhaps mirrored in finding the single ASSQ item “fails to make friends” the third most common item in the present study. Other studies have found children with CP twice as likely to be rejected and have no friends, and three times as likely to be victimized as their healthy peers (Yude & Goodman, 1999), similar to results in a study by Nadeau & Tessier (2006). Knowing that many children with CP face motor, cognitive and social limitations, efforts to nurture social skills from an early age seem important (Yude et al., 1998).

The study population was representative for children with CP having GMFCS levels I–IV, regarding type of CP, severity and comorbid medical conditions (Andersen et al., 2008; Dolk, Parkes, & Hill, 2006). Medical conditions related to CP such as epilepsy, severity of CP, and communication problems were not significantly related to autism symptoms, similar to other studies (Brossard-Racine et al., 2012; Parkes et al., 2008). We did however find a large mean difference for the variables epilepsy and communication problems for ASSQ high scorers. A larger population sample however, might have shown a significant association, supporting findings in the Turkish study where presence of epilepsy and speech problems was related to autism (Kilincaslan & Mukaddes, 2009). The etiology leading to autism spectrum symptoms in children with early lesions affecting the developing brain is still largely unknown, despite evidence of an overlap between developmental disorders, autism spectrum disorders (ASD), motor difficulties and epilepsy (Suren et al., 2012). Previous studies have found ASD and autistic traits genetically related (Lundstrom et al., 2012), and recent studies suggest some of the CP conditions may be caused by genetic disturbances (Lin et al., 2013). The notion of common etiology leading to a high prevalence of autistic symptoms, AD/HD and other psychiatric disorders in a developing brain affected by early adverse incidences, has been described previously (Kadesjo & Gillberg, 2001). Likewise, a high prevalence of comorbid psychiatric disorders, developmental coordination disorders (DCD) and ASSQ high scores, similar to the present study, was found in in 87% of children with ADHD (Kadesjo & Gillberg, 2001). A high number of overlapping psychiatric disorders is a challenge to precise diagnostics.

##### 4.1. Limitations

There are some limitations to the present study, such as parents being the only informants, thus reducing the validity of the ASSQ (Mattila et al., 2009). Parents were however all interviewed with the Kiddie-SADS, which is a semi structured interview allowing for elaborate answers. The ASSQ is a screening instrument for ASD, and is inappropriate for ascertaining an autism diagnosis. It is therefore a drawback that the version of the diagnostic instrument used in the present study did not include a section on ASD. Validation studies have however shown a good correlation between ASSQ high scores, and an ASD diagnosis (Posserud et al., 2009). The most severely affected children were omitted as diagnostic conclusions regarding child



psychiatric disorders were not possible (Bjorgaas et al., 2012). Despite the study being population based, numbers were small, thus interpretations drawn from the study should be done with caution.

## 5. Conclusions

Finding one in five children with CP being ASSQ high scorers suggests a need for more comprehensive ASD diagnostics in a large number of children with CP. The ASSQ may be used with caution as part of a mental health screening package to detect autism spectrum symptoms. However, diagnostic instruments should be used to ascertain an autism diagnosis, taking into account the considerable overlap of co-occurring psychiatric disorders.

## Ethical approval

The study was approved by the Regional Committee for Medical Research Ethics in Western Norway.

## Conflict of interest

The authors declare no conflict of interests.

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