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Autism traits: The importance of “co-morbid” problems for impairment and contact with services. Data from the Bergen Child Study

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ABSTRACT

Background: Co-occurring problems are common in individuals with clinical autism spectrum disorder (ASD) but their relevance for impairment and contact with health services in ASD is largely unexplored.

Aims: We investigated the extent of co-occurring problems in children with high ASD traits from a total population sample. We explored the contribution of co-occurring problems to impairment and service contact, and whether there were children without co-occurring problems in this group; as proxy for “ASD only”.

Methods and procedures: Children screening positive on the Autism Spectrum Screening Questionnaire (ASSQ) were used as proxy for ASD. Attention Deficit/Hyperactivity Disorder (ADHD) and Oppositional Defiant Disorder (ODD) were operationalised using symptom counts. A parent or teacher report above the 95th percentile counted as “problem” present for other symptom domains.

Outcomes and results: 92% of ASSQ high-scorers had a minimum of two other problems. Emotional problems, ADHD symptoms and learning problems were the most commonly reported problems, also predicting impairment and contact with services.

Conclusions and implications: Co-occurring problems were common in ASD screen positive children and contributed strongly to both impairment and to contact with services. Gender differences indicated that female symptoms were perceived as less impairing by parents and teachers.

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What this paper adds?

Autism spectrum disorder (ASD) has historically been associated to poor outcome and lack of independent life in adulthood, but very few studies have examined the extent to which this poor outcome may be due to the extensive comorbid problems also suffered by individuals with ASD. This study contributes with unique information showing that also children from a population-based sample who screen positive for ASD have considerable and clinically relevant impairment and

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psychiatric comorbid problems as reported by parents and teachers. Furthermore, their reported impairment is to a large degree explained by their comorbid problems. The present findings indicate that very few children with ASD traits have autism traits alone, and that they most likely also suffer from other cognitive and mental health problems, e.g. learning problems, tics, emotional problems and ADHD symptoms. This indicates that “autism plus” is the most common autism problem also in population based samples and not only a result of a referral bias. It also indicates that the “plus” part accounts for a large part of the impairment children with ASD traits have. A broad assessment of mental health, learning and cognition is thus crucial in the management and treatment of children with suspected ASD. There were differences between female and male symptomatology. Eating problems and obsessive–compulsive symptoms were more common in girls, but were not rated as impairing and had no bearing towards service use. The implications of this gender difference are unknown.

1. Introduction

Adults with autism spectrum disorders (ASD) comprise a heterogeneous group (Happé, Ronald, & Plomin, 2006) many of whom grew up undiagnosed (Brugha et al., 2011; Nylander, Holmqvist, Gustafson, & Gillberg, 2013). Over the past 10 years, we have seen a surge in diagnoses, due to a combination of increased awareness, changes in the perception of the ASD phenotype, and revisions to the diagnostic criteria (Fombonne, Quirke, & Hagen, 2009). Functional ability in individuals with ASD may vary broadly and some individuals are able to lead independent and successful lives (Grandin, 2011). Studies to date however indicate that a large majority of adults with ASD have poor outcome (Billstedt, Gillberg, & Gillberg, 2005) and have indicated that autism symptoms themselves are predictive of dysfunction in adulthood (Howlin, Moss, Savage, & Rutter, 2013; Magiati, Tay, & Howlin, 2014). The factors influencing functional outcome have been difficult to tease apart. There is a dearth of studies assessing the extent of impairment arising from ASD traits vs. co-occurring conditions although co-occurring conditions are common (Gillberg & Fernell, 2014).

The Early Symptomatic Syndromes Eliciting Neurodevelopmental Clinical Examinations – ESSENCE – framework, suggests that a wide range of neurodevelopmental difficulties tend to appear and persist together (Gillberg, 2010). In children with ASD, the ESSENCE framework is supported by a large body of research showing that ASD is commonly accompanied with different kinds of neurodevelopmental disabilities (Carlsson et al., 2013; Levy et al., 2010), including epilepsy (Canitano, 2007), intellectual disability (Matson & Shoemaker, 2009) and a broad range of other psychiatric disorders, notably ADHD (Ghaziuddin, Weidmer-Mikhail, & Ghaziuddin, 1998; Joshi et al., 2010), Tourette's syndrome (TS) and tics (Baron-Cohen, Mortimore, Moriarty, Izaguirre, & Robertson, 1999; Baron-Cohen, Scahill, Izaguirre, Hornsey, & Robertson, 1999; Canitano & Vivanti, 2007; Simonoff et al., 2008), and anxiety disorders (Muris, Steerneman, Merckelbach, Holdrinet, & Meesters, 1998; White, Oswald, Ollendick, & Scahill, 2009). Depression and obsessive–compulsive disorders (OCD) become more common in older children (Ghaziuddin, Ghaziuddin, & Greden, 2002; Gjevik, Eldevik, Fjaeran-Granum, & Sponheim, 2011). Furthermore, learning disability, language and learning problems are intricately linked to autism (O'Brien & Pearson, 2004). Exact prevalence estimates vary greatly depending on definition, assessment and patient group (O'Brien & Pearson, 2004), but both language problems and intellectual problems have been reported to occur in around 80% of children with ASD (Carlsson et al., 2013; Fombonne, 1999). These co-occurring problems are expected to contribute to poor functional outcome, although to our knowledge, no study has examined their respective contribution to impairment and outcome in children with ASD.

Both the ESSENCE model and findings from the Dunedin study suggest that the *amount* of co-occurring problems may be interpreted as indexing an overall factor of severity of functional impairment rather than specific psychopathology alone (Caspi et al., 2014). Diagnosing an increasing number of higher functioning children with ASD could thus lead to less functional disability due to low prevalence of co-occurring problems. The cited studies above have been performed in mainly clinical/special school populations, raising the question of whether the high rates of comorbidity could be related to a referral bias. If ASD by itself is not clinically impairing, children with only autism would not be found in clinical populations. Park et al. (2014) however found that children diagnosed as part of a population screen had *higher* scores on several of the comorbidity scales than children already clinically diagnosed with ASD (Park, Kim, Koh, Song, & Leventhal, 2014). The authors suggested that externalising symptoms combined with better adaptive functioning were masking the ASD problems in these children, hindering their access to ASD services. Similarly, Levy et al. found that children with other diagnoses and ASD symptoms were diagnosed with their ASD at a later age, indicating that the ASD was masked by other problems (Levy et al., 2010). Gillberg and Fernell point out that the reverse is often true; that children are increasingly diagnosed with autism only, while the functional impairment often stem from the important but overlooked co-occurring problems (Gillberg & Fernell, 2014). As co-occurring symptoms/problems in ASD thus impact presentation, help-seeking behaviour, assessment and the final diagnosis of ASD, population-based studies of ASD symptoms, co-occurring problems and associated impairment are warranted.

Gender differences characterise the abovementioned disorders, with male:female ratios varying depending on disorder studied, level of ability, age, and a range of other factors. In ASD, the male predominance is considerably higher in individuals with an intellectual function in the normal range than in samples including mainly intellectually disabled children (Van Wijngaarden-Cremers et al., 2014). For ADHD, the male predominance is lower in adult samples than in children (Haavik, Halmøy, Lundervold, & Fasmer, 2010). The underpinnings of the male predominance in childhood neuropsychiatric problems are poorly understood, but some of the differences seem to relate to ascertainment bias, cultural norms and expectations rather than biological differences (Bussing, Zima, Gary, & Garvan, 2003; Ohan & Visser, 2009; Van Wijngaarden-Cremers et al., 2014). Gender expectations have also been shown to be a consistent barrier for girls in access to mental health services (Alegria et al., 2004; Bussing, Zima, Perwien, Belin, & Widawski, 1998; Derks, Hudziak, & Boomsma, 2007).

The present study was thus launched with a view to examining the prevalence of co-occurring problems, impairment and service use in a population-based sample of children defined as screen positive on the Autism Spectrum Screening Questionnaire (ASSQ). We asked if the high level of impairment and the high rate of co-occurring problems reported previously in clinical studies of children with ASD could be replicated in a total population based sample of autism screen positive children. We further explored the contribution of co-occurring mental health problems on the children's functional impairment, contact with health and school services and the effect of gender.

2. Method

2.1. Sample

The first wave of the longitudinal Bergen Child Study (BCS) assessed a broad range of mental health problems in a total population of school children aged 7–9 years ($n = 9430$) through teacher and parent questionnaires to all schools, including special schools and private schools. Teacher questionnaires ($n = 9152$) covered 97%, whereas parent questionnaires ($n = 6295$) covered 67% of the population. The present study included all children who had both a parent and a teacher ASSQ, excluding questionnaires missing more than four items ($n = 58$), leading to a final sample consisting of 6237 children (50.1% girls).

2.2. ASSQ

The Autism Spectrum Screening Questionnaire (ASSQ) was used to screen for ASD (Ehlers, Gillberg, & Wing, 1999). An ASSQ score ≥ 17 points on either parent and teacher ASSQ was used to define ASD high-scorers, corresponding to a sensitivity of 0.91 and specificity of 0.86 for ASD when defined as screen positive by either informant (Posserud, Lundervold, & Gillberg, 2009). ASD screen positive children are referred to as the ASD group in the present study. A significant number of these children may have received a clinical diagnosis of ASD, but as we did not have access to clinical data in this population-based sample, we do not know to whom a clinical diagnosis of ASD applies.

2.3. Co-occurring problems

The BCS questionnaire was designed to screen for a range of mental health problems and included in addition to the ASSQ, the Strength and Difficulties Questionnaire (SDQ) (Goodman, 2001), the DSM-IV criteria for ADHD and ODD, five items targeting OCD (Thomsen, 1998), 5 items targeting tics, and 6 items targeting learning problems (4 items on language, one item on reading/writing and one item on arithmetic). In addition, parent questionnaires included the Eating Disorders Scale (EDS-5) developed for screening (Rosenvinge et al., 2001), while the teacher version included two items on hypoactivity and one item targeting selective mutism from the FTF questionnaire (Kadesjo et al., 2004). All items were scored on a three-point Likert scale (not true, somewhat true, definitely true). An ASD co-occurring problem was defined as follows: ADHD and ODD were conceptualised as in previous publications from the BCS; ADHD items scored as “somewhat true” and “definitely true” counted as “symptom” but requiring both informants to endorse minimum 6 symptoms in at least one of the two ADHD dimensions. ODD required an item to be scored as “definitely true” to be counted as “symptom”, but here it sufficed for only one informant (parent or teacher) scoring 4 or more symptoms for a child to be categorised as having ODD (Munkvold, Lundervold, Lie, & Manger, 2009; Ullebo, Posserud, Heiervang, Obel, & Gillberg, 2012).

All other symptoms scales were dichotomised and counted as present if endorsed above the 95th population percentile on that scale. Both teacher and parent information was included, so that a symptom was counted as present if either parents or teachers endorsed it above the 95th percentile. *Learning difficulties (LD)* included all 6 items on learning problems. *Selective mutism (SM)* and *Hypoactivity* items were only addressed to teachers, and *eating disorder symptoms (ED)* only to parents, thus the 95th percentile for these domains were based on one informant only. For *Tics* the 95th percentile coincided with the 90th percentile for both parents and teachers. As ADHD and ODD were already defined, only the emotional subscale (“*Emotional problems*”) and the impairment items were used from the SDQ.

2.4. Impairment

Parent and teacher impact scores from the SDQ were calculated according to the scoring algorithm recommended on the SDQ scoring site (www.sdqinfo.com), but included all impairment items except duration of problem. The impairment scale thus consisted of: *overall impact* (parent (P) + teacher (T)), *overall burden for the child* (P + T), *overall burden to family* (P), *overall burden to class room/teacher* (T), *impact on learning* (P + T), *impact on friends* (P + T), *impact on family life* (P) and *impact on leisure activities* (P). Scores from parents and teachers were then combined to a joint *Impairment score* with a scoring range of 0–24.

2.5. Service use

Both informants were asked whether the child, to their knowledge, had been referred to child and adolescent mental health services (CAMHS) or to school psychology services (SPS) for any of the problems reported in the questionnaire. Response options were “yes,” “no,” or “I don't know.” A child was defined as referred to a service if the parent and/or the teacher reported “yes” regarding that service.

2.6. Statistical analyses

Significant overlap between symptom domains was tested with Pearson Chi-Square or Fisher's exact test (two-tailed) with $p < 0.05$ as the required level of significance. To understand the contribution of symptoms to impairment, the joint impairment scale was used as dependent variable in a multiple linear regression analysis including all the symptom domains and gender as predictor variables. For this analysis, symptoms were not dichotomised, but the scores from teachers and parents added to produce a dimensional measure. Differences in mean scores were calculated using independent samples mean test. To analyse the relative contribution of symptoms to contact with health services, logistic regression with CAMHS and SPS as dependent variables and problem domains and gender as predictor variables was performed. The variables were entered hierarchically, and then removed stepwise with decreasing significance until only retaining significant predictors in the model. Effects of gender and number of problems on impairment were analysed using ANCOVA with gender as covariate. SPSS version 21 was used for all analyses.

3. Results

3.1. Co-occurring problems

A total of 226 children were defined as ASD screen positive (3.6%), 66 girls (2.1% of all girls) and 160 boys (5.1% of all boys). The average number of co-occurring problems in this ASD group was 3.8 (vs. 0.5 in the screen negative children, $p < 0.001$), with an average impairment score of 8.0 (vs. 0.5 in the screen negative children, $p < 0.001$). Of the total ASD group, 163/226 children (72.1%) had a caseness score of ≥ 2 for either informant as defined by Goodman (Goodman, 1999), 35/66 girls (53.0%) and 128/160 boys (80.0%). The gender difference was statistically significant ($p < 0.001$).

Five of the 226 children had no other problems reported and another 17 children had one problem, meaning that 90.3% of ASSQ high-scorers had problems within at least two areas in addition to their high ASSQ score, and 57.5% had four or more problem areas. Almost half of the children (43.6%) met the symptom criteria for ADHD. Language/learning difficulties (LD), emotional problems, ODD, tics and OCD were also all very common. Table 1 shows the baseline rate of other problem and the percentages of children having both these problems (in addition to their high ASSQ score). p -Values indicate where the paired problems co-occur more or less frequently than expected. Emotional problems were overrepresented in children with ODD and OCD symptoms, so that 70.8% of the children with ODD had emotional problems, compared to 41.6% of the children without ODD. More than 60% (62.6%) of the children with OCD symptoms were reported to have emotional problems, compared to 40.3% in children with low OCD score. Hypoactivity problems were commonly occurring with ADHD. Both LD and ODD were related to hypoactivity, but this overlap seemed to be accounted for by their common overlap with ADHD, as the overrepresentation between these domains disappeared for both ODD and LD when analysed by ADHD status. It should however be noted that the relationship was almost significant for ODD ($\chi^2 = 3.347$, $p = 0.067$).

3.2. Impairment

Impairment was associated to number of co-occurring problems in children with ASD symptoms with $r = .50$, $p < 0.001$. Looking at specific problem domains, the main predictors of impairment included inattentive and hyperactivity symptoms, ODD symptoms, LD, emotional problems and ED problems, together explaining more than 60% of the variance of the reported impairment (Table 2). Interestingly, ED problems were negatively associated with impairment. The analyses were also repeated omitting impairment regarding friends (as this impairment is intrinsic to ASD problems), but the proportion of explained variance and the strength of the associations all remained the same (data not shown).

Table 1

Prevalence rates of problem domains and the prevalence of each paired overlap in ASSQ high scorers.

Prevalence	ADHD	LD	ODD	Emo	OCD	Tics	SM	Eat	Hypo
	43.8%	62.4%	31.9%	50.9%	47.3%	73.9%	9.7%	22.1%	40.7%
ADHD	–	33.6%**	21.2%**	26.5*	20.4%	36.7*	4.4%	4.9%**	27.0%**
LD		–	19.9%	31.4%	32.3%	49.1*	7.5%	12.8%	29.2%**
ODD			–	22.6%**	15.0%	22.1%	1.8%	5.3%	18.1%**
Emo				–	29.6%**	36.3%	5.8%	9.3%	25.7%**
OCD					–	40.3%**	5.8%	14.6*	17.3%
Tics						–	7.1%	17.7%	31.4%
SM							–	2.2%	5.8%
Eat								–	4.4%**
Hypo									–

* $p < 0.05$.

** $p \leq 0.001$.

Table 2
Multiple regression analysis for symptoms predicting impairment (only showing values of significant predictors).

Variable	Impairment		
	B	SE B	β
Gender	–	–	–
Inattention	0.23	0.05	.31***
Hyperactivity	0.13	0.05	.17**
Learning problems	0.23	0.05	.21***
ODD	0.22	0.06	.25***
Emotional	0.23	0.10	.13**
OCD	–	–	–
Tics	–	–	–
Selective mutism	–	–	–
Eating problems	–0.56	0.23	–.10*
Hypoactivity	–	–	–
R ²		0.63	
F		63.60***	

* $p < .05$.

** $p < .01$.

*** $p < .001$.

3.3. Contact with CAMHS and school psychology services

Two of the 54 children with two or fewer co-occurring problems were in contact with CAMHS and 19/54 (35%) were in contact with SPS, vs. 68/172 (39.5%) and 113/172 (65.7%) of the children with three or more co-occurring problems. Hierarchical logistic regression analyses showed that ADHD predicted contact with both CAMHS (ADHD: OR 4.6, 95%CI: 2.4–8.5, $p < 0.001$) and SPS (ADHD: OR 3.2, 95%CI 1.5–6.8, $p = 0.002$). Emotional problems also predicted contact with CAMHS (OR 2.0, 95%CI: 1.1–3.8, $p = 0.025$), whereas LD was the most important predictor for SPS contact (OR: 6.6, 95%CI 3.2–13.4, $p < 0.001$). Both selective mutism¹ and ED predicted less contact with SPS ((ED:OR:0.22, 95%CI 0.1–0.5, $p < 0.001$), (SM:OR:0.10, 95%CI 0.0–0.3, $p < 0.001$)). Gender did not predict service contact after ADHD and LD were introduced into the models.

3.4. Gender differences

Although the number of co-occurring problems were almost the same for boys and girls (4.0 vs. 3.5, $p = 0.04$), the impairment score was much higher for boys (9.3 vs. 4.9, $p < 0.001$). There was an interaction effect between number of co-occurring problems and gender in predicting impairment ($F_{7,209} = 2.56$, $p = 0.015$), in addition to main effects of number of co-occurring problems ($F_{7,209} = 9.73$, $p < 0.001$) and gender ($F_{1,209} = 10.33$, $p < 0.01$) (Fig. 1). The results remained identical when repeated omitting an outlier with 8 co-occurring problems and no impairment (a boy).

Examining gender differences by specific symptom domains, ADHD showed the most salient difference in frequency with only 20% of the girls reported to have ADHD problems vs. 54% of the boys ($\chi^2 = 22.01$, $p < 0.001$). Girls were less frequently reported to be hypoactive (26% vs. 47% in boys, $\chi^2 = 8.633$, $p = 0.003$) and to show ODD problems (21% vs. 36%, $\chi^2 = 4.87$, $p = 0.03$), but they were significantly more often reported to have OCD and ED problems than boys. OCD symptoms were reported in 61% of the girls vs. 42% of the boys ($\chi^2 = 6.58$, $p = 0.01$). Only 17% of boys had ED problems, whereas this was true for 23/66 girls, i.e. 35% ($\chi^2 = 8.76$, $p = 0.003$). Emotional problems, learning difficulties and tics were equally common in boys and girls. Gender did not account for any difference in CAMHS contact after adding symptoms of ADHD to the statistical model, although, as shown above, there were clear gender differences in the prevalence of ADHD. Furthermore, while ADHD and ODD were more often co-occurring than not in boys, there was no such overlap in girls (Table 3). Girls were more often reported to have ED problems. This symptom domain was however associated with less contact with SPS and less impairment. Girls with OCD and ED problems were significantly less impaired than boys with OCD and ED problems (OCD: girl mean impairment score = 4.4, boys = 9.6, $p < 0.001$, ED: girl mean impairment score = 3.0, boys = 6.9, $p < 0.05$).

¹ All four children with the response “completely true” to the item selective mutism were in contact with SPS and two of them also with CAMHS, so the protective effect vs. contact with SPS seemed to be accounted for by the children with “partly true” responses, where only 5/17 (29%) were in contact with SPS.

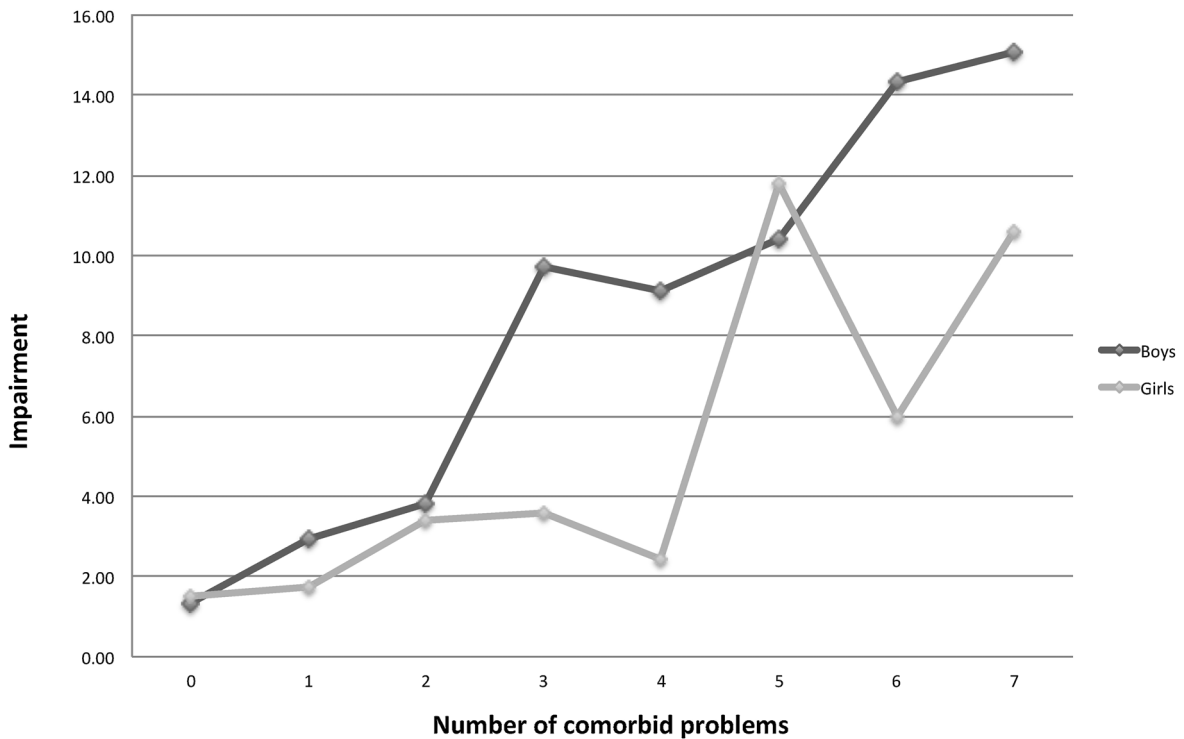


Fig. 1. Level of impairment (SDQ score) and number of co-occurring problems by gender.

Table 3
Interaction between gender, ADHD and ODD.

Gender	ODD	ADHD		Total (%)	Association
		No (%)	Yes (%)		
Female	No	44 (84.6)	8 (15.4)	52 (78.8)	$p = 0.128^a$
	Yes	9 (64.3)	5 (35.7)	13 (21.2)	
Male	No	59 (57.8)	43 (42.2)	102 (63.8)	$p < 0.001$ $\chi^2 (1) = 15.21$
	Yes	15 (25.9)	43 (74.1)	58 (36.3)	
Total		127 (56.2)	99 (43.8)	226 (100)	

^a Using Fisher's exact test, as expected count was below 5.

4. Discussion

The present study showed that ASD screen positive children were characterised by a high symptom load on co-occurring problems and high scores on the impairment scale. More than 70% had an SDQ impairment score indicating caseness as defined by Goodman (1999). Both impairment and contact with health services were to a large extent explained by problems in the domains of ADHD, LD and emotional problems. Only 2% of children could be characterised as having “autism only”, i.e. having no other problems in this population-based sample. Although only examined at symptom level, the findings indicate that children with a high level of ASD traits need a comprehensive assessment: most of these children have symptom profiles suggestive of several other problems contributing to their suffering and need for help. The present study findings thus accord with clinical studies, supporting that the wide array of co-occurring problems demonstrated in clinical studies of children with ASD are also found in a population-based sample of children scoring high on ASD traits.

The high prevalence (62.4%) and substantial impact of LD reported in the present study have important clinical implications. Language and learning ability should be an intrinsic part of the assessment procedure in all children suspected of suffering from an autism spectrum disorder, also in children perceived as “high functioning”, and clinicians should bear in mind that problems understanding language may present as non-compliance or disruptive behaviours (Helland, Lundervold, Heimann, & Posserud, 2014). Identifying language problems is thus crucial to understanding and intervention for children with ASD traits.

The gender differences were another important finding of the present study. Although there was only a slight difference in number of problems reported, there was a strong difference in degree of impairment. There was both an independent main effect of gender and an interaction effect between gender and number of problem areas. Girls were generally scored as less impaired and as less often in contact with services, but this association seemed to be accounted for by their lower rate of ADHD, and for the girls with ADHD, it was less often associated with ODD. It was also noted that ED and OCD problems were significantly more common in girls with a high ASSQ score than in boys. However, ED was found to be associated to less impairment and against contact with SPS, and girls with OCD problems were rated as less impairing than boys with OCD problems. In clinical studies of ASD, females have been shown to have less stereotypical and repetitive behaviour (Van Wijngaarden-Cremers et al., 2014). However, as ASD research is dominated by the male ASD phenotype, female ASD symptoms may be misinterpreted and overlooked (Kirkovski, Enticott, & Fitzgerald, 2013; Van Wijngaarden-Cremers et al., 2014). The present findings suggest that ED and OCD symptoms should be explored in further studies as possible female expressions of repetitive and stereotype behaviour and interests (RRBI) in ASD.

The present study indicates that girls are reported to have different kinds of problems than boys, and that their problems are viewed as less impairing. The question is whether females are truly less impaired or whether their teachers and parents just fail to identify their problems (or both). At least when related to the child's own impairment score, parents have been found to rate girls as less impaired than boys (Wille, Bettge, Wittchen, Ravens-Sieberer, & group, 2008). Wille et al. also found that parents rated impairment mainly for externalising symptoms, whereas the children themselves reported impairment in relation to emotional symptoms (Wille et al., 2008). The strong contribution of disruptive behaviour to impairment and to contact with health services in the present study further supports the conclusion that internalising problems in general are underestimated and overlooked by adults around the child. Although more than 50% of the children in the current study had emotional problems, it had less bearing towards service contact than ADHD problems. This is supported by previous research from the BCS, showing that only 13% of children suffering from anxiety disorders in this age group were in touch with CAMHS compared to 75% of children with ADHD (Heiervang et al., 2007). Unfortunately in the present study, no child rating of impairment was available.

In addition to the general large overlap between problem domains, there were some specific patterns of overlap. The overrepresentation of ODD and emotional problems in children with ASD has been shown previously in a study by Mattila et al. (2010), where most children with ODD (7/8) also had an anxiety disorder (Mattila et al., 2010). ODD and emotional disorders have been shown to be associated in both clinical and population based samples (Boylan, Vaillancourt, Boyle, & Szatmari, 2007). The present findings could be used to support the emotional dysregulation hypothesis that has been postulated as an important vulnerability factor for both emotional problems and ODD (Cavanagh, Quinn, Duncan, Graham, & Balbuena, 2014). Hypoactivity was defined by two items only, and the results should thus be interpreted with caution. However, a previous study on a subsample from the BCS has shown that children who were perceived as hypoactive by teachers had lower processing speed on psychometric tests (Lundervold, Posserud, Ullebo, Sorensen, & Gillberg, 2011). This knowledge together with the high overlap between hypoactivity and other problem domains in the present study highlights hypoactivity as an important symptom that deserves further attention in future studies.

A factor contributing to the large gender difference found regarding ADHD could have been due to the different definitions applied to ADHD vs. other problem areas in the present study, as the ADHD diagnosis required a minimum of six symptoms from *both* informants to comply with previous work. The analyses were therefore recalculated applying the same definition as for the other categories. Apart from almost doubling the number of children defined as ADHD, it did not alter the results related to gender (data not shown).

A large part of the impairment found in individuals with a high ASSQ score was explained by problems within other symptom domains. Howlin et al. found that the strongest childhood predictor of adult dysfunction was the reciprocal social difficulties themselves (Howlin et al., 2013; Magiati et al., 2014), but it has to be noted that these studies did not include detailed scrutiny of other ESSENCE problems. Furthermore, "autism only" may not cause the same degree of impairment in a child, as parents normally cater for the majority of daily necessities such as hygiene, nutrition and care. Family and school provide a mandatory social context that prevents social isolation in youth. However, as these children grow up, and are expected to become independent, their lack of age-appropriate self-help skills and social skills may lead to profound deprivation and complete social isolation as parents withdraw their support (Hendricks & Wehman, 2009; Magiati et al., 2014). Longitudinal studies including the entire spectrum of problems in "autism plus" vs. "autism only" are required to increase our knowledge of the relative contributions to functional outcome.

4.1. Limitations

We have used the ASSQ screen positive score as a proxy for ASD and autism traits. However, ASD is a clinical diagnosis that cannot be derived from questionnaire data alone. Validation studies have shown that many of the children scoring high on ASSQ do not necessarily have autism spectrum disorders (ASD). They may rather fulfil criteria for ADHD and intellectual disability (Posserud, Lundervold, & Gillberg, 2009). One must therefore be cautious in extending the results to clinical populations of ASD. Similarly, high symptom counts on other problem domains should not be understood as clinical disorders, but only as an estimate and indication of the problems that may be present at group level.

4.2. Clinical implications

The impairment and wide range of co-occurring problems found in relation to autism traits already in childhood combined with the bleak outcome shown in previous studies of adults with ASD, indicate that a high score on the ASSQ should lead to a comprehensive clinical assessment including neurodevelopmental, psychiatric and cognitive assessment and follow-up regardless of the final clinical diagnosis. The ESSENCE approach was supported in our study, given that the overlap across a wide range of symptom domains was very high. Furthermore, a large part of the impairment was accounted for by co-occurring problems. Language/learning and ADHD problems are areas meriting special attention, due to their high prevalence in these children, their known relevance for impairment and treatment implications. Future research should include other ESSENCE symptoms/disorders in studies of ASD adult outcome both in clinical and population-based samples to better understand their relative role for long-term prognosis.

Although gender itself did not explain differences in impairment and contact with health services, there were differences in the pattern of reported symptoms and how impairing these problems were perceived by parents and teachers. Further studies are required to understand whether and how these differences are related to less identification of girls with psychiatric problems in childhood and the later age of diagnosis in girls.

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References

- Alegria, M., Canino, G., Lai, S., Ramirez, R. R., Chavez, L., Rusch, D., et al. (2004). Understanding caregivers' help-seeking for Latino children's mental health care use. *Medical Care, 42*(5), 447–455.
- Baron-Cohen, S., Mortimore, C., Moriarty, J., Izaguirre, J., & Robertson, M. (1999). The prevalence of Gilles de la Tourette's syndrome in children and adolescents with autism. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 40*(2), 213–218.
- Baron-Cohen, S., Scahill, V. L., Izaguirre, J., Hornsey, H., & Robertson, M. M. (1999). The prevalence of Gilles de la Tourette syndrome in children and adolescents with autism: A large scale study. *Psychological Medicine, 29*(5), 1151–1159.
- Billstedt, E., Gillberg, I. C., & Gillberg, C. (2005). Autism after adolescence: Population-based 13- to 22-year follow-up study of 120 individuals with autism diagnosed in childhood. *Journal of Autism and Developmental Disorders, 35*(3), 351–360.
- Boylan, K., Vaillancourt, T., Boyle, M., & Szatmari, P. (2007). Comorbidity of internalizing disorders in children with oppositional defiant disorder. *European Child and Adolescent Psychiatry, 16*(8), 484–494. <http://dx.doi.org/10.1007/s00787-007-0624-1>
- Brugha, T. S., McManus, S., Bankart, J., Scott, F., Purdon, S., Smith, J., et al. (2011). Epidemiology of autism spectrum disorders in adults in the community in England. *Archives of General Psychiatry, 68*(5), 459–465. <http://dx.doi.org/10.1001/archgenpsychiatry.2011.38>
- Bussing, R., Zima, B. T., Gary, F. A., & Garvan, C. W. (2003). Barriers to detection, help-seeking, and service use for children with ADHD symptoms. *Journal of Behavioral Health Services and Research, 30*(2), 176–189.
- Bussing, R., Zima, B. T., Perwien, A. R., Belin, T. R., & Widawski, M. (1998). Children in special education programs: Attention deficit hyperactivity disorder, use of services, and unmet needs. *American Journal of Public Health, 88*(6), 880–886.
- Canitano, R. (2007). Epilepsy in autism spectrum disorders. *European Child and Adolescent Psychiatry, 16*(1), 61–66. <http://dx.doi.org/10.1007/s00787-006-0563-2>
- Canitano, R., & Vivanti, G. (2007). Tics and Tourette syndrome in autism spectrum disorders. *Autism, 11*(1), 19–28. <http://dx.doi.org/10.1177/1362361307070992>
- Carlsson, L. H., Norrelgen, F., Kjellmer, L., Westerlund, J., Gillberg, C., & Fernell, E. (2013). Coexisting disorders and problems in preschool children with autism spectrum disorders. *The Scientific World Journal, 2013*, 213979. <http://dx.doi.org/10.1155/2013/213979>
- Caspi, A., Houts, R. M., Belsky, D. W., Goldman-Mellor, S. J., Harrington, H. L., Israel, S., et al. (2014). The p factor one general psychopathology factor in the structure of psychiatric disorders? *Clinical Psychological Science, 2*(2), 119–137.
- Cavanagh, M., Quinn, D., Duncan, D., Graham, T., & Balbuena, L. (2014). Oppositional defiant disorder is better conceptualized as a disorder of emotional regulation. *Journal of Attention Disorders, 18*(10), 1087–1094. <http://dx.doi.org/10.1177/1087054713520221>
- Derks, E. M., Hudziak, J. J., & Boomsma, D. I. (2007). Why more boys than girls with ADHD receive treatment: A study of Dutch twins. *Twin Research and Human Genetics, 10*(5), 765–770. <http://dx.doi.org/10.1375/twin.10.5.765>
- Ehlers, S., Gillberg, C., & Wing, L. (1999). A screening questionnaire for Asperger syndrome and other high-functioning autism spectrum disorders in school age children. *Journal of Autism and Developmental Disorders, 29*(2), 129–141.
- Fombonne, E. (1999). The epidemiology of autism: A review. *Psychological Medicine, 29*(4), 769–786.
- Fombonne, E., Quirke, S., & Hagen, A. (2009). Prevalence and interpretation of recent trends in rates of pervasive developmental disorders. *McGill Journal of Medicine, 12*(2), 73.
- Ghaziuddin, M., Ghaziuddin, N., & Greden, J. (2002). Depression in persons with autism: Implications for research and clinical care. *Journal of Autism and Developmental Disorders, 32*(4), 299–306.
- Ghaziuddin, M., Weidmer-Mikhail, E., & Ghaziuddin, N. (1998). Comorbidity of Asperger syndrome: A preliminary report. *Journal of Intellectual Disability Research, 42*(Pt 4), 279–283.
- Gillberg, C. (2010). The ESSENCE in child psychiatry: Early symptomatic syndromes eliciting neurodevelopmental clinical examinations. *Research in Developmental Disabilities, 31*(6), 1543–1551. <http://dx.doi.org/10.1016/j.ridd.2010.06.002>
- Gillberg, C., & Fernell, E. (2014). Autism plus versus autism pure. *Journal of Autism and Developmental Disorders, 44*(12), 3274–3276. <http://dx.doi.org/10.1007/s10803-014-2163-1>
- Gjevik, E., Eldevik, S., Fjæraen-Granum, T., & Sponheim, E. (2011). Kiddie-SADS reveals high rates of DSM-IV disorders in children and adolescents with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 41*(6), 761–769. <http://dx.doi.org/10.1007/s10803-010-1095-7>
- Goodman, R. (1999). The extended version of the Strengths and Difficulties Questionnaire as a guide to child psychiatric caseness and consequent burden. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 40*(5), 791–799.
- Goodman, R. (2001). Psychometric properties of the strengths and difficulties questionnaire. *Journal of the American Academy for Child and Adolescent Psychiatry, 40*(5), 791–799.
- Grandin, T. (2011). *The way I see it: A personal look at autism & asperger's*. Future Horizons.

- Haavik, J., Halmoy, A., Lundervold, A. J., & Fasmer, O. B. (2010). Clinical assessment and diagnosis of adults with attention-deficit/hyperactivity disorder. *Expert Review of Neurotherapeutics*, 10(10), 1569–1580. <http://dx.doi.org/10.1586/ern.10.149>
- Happe, F., Ronald, A., & Plomin, R. (2006). Time to give up on a single explanation for autism. *Nature Neuroscience*, 9(10), 1218–1220. <http://dx.doi.org/10.1038/nrn1770>
- Heiervang, E., Stormark, K. M., Lundervold, A. J., Heimann, M., Goodman, R., Posserud, M.-B., et al. (2007). Psychiatric disorders in Norwegian 8- to 10-year-olds: An epidemiological survey of prevalence, risk factors, and service use. *Journal of the American Academy of Child & Adolescent Psychiatry*, 46(4), 438–447.
- Helland, W. A., Lundervold, A. J., Heimann, M., & Posserud, M. B. (2014). Stable associations between behavioral problems and language impairments across childhood – The importance of pragmatic language problems. *Research in Developmental Disabilities*, 35(5), 943–951. <http://dx.doi.org/10.1016/j.ridd.2014.02.016>
- Hendricks, D. R., & Wehman, P. (2009). Transition from school to adulthood for youth with autism spectrum disorders review and recommendations. *Focus on Autism and Other Developmental Disabilities*, 24(2), 77–88. <http://dx.doi.org/10.1177/1088357608329827>
- Howlin, P., Moss, P., Savage, S., & Rutter, M. (2013). Social outcomes in mid- to later adulthood among individuals diagnosed with autism and average nonverbal IQ as children. *Journal of the American Academy of Child and Adolescent Psychiatry*, 52(6). <http://dx.doi.org/10.1016/j.jaac.2013.02.017>. 572–581 e571
- Joshi, G., Petty, C., Wozniak, J., Henin, A., Fried, R., Galdo, M., et al. (2010). The heavy burden of psychiatric comorbidity in youth with autism spectrum disorders: A large comparative study of a psychiatrically referred population. *Journal of Autism and Developmental Disorders*, 40(11), 1361–1370. <http://dx.doi.org/10.1007/s10803-010-0996-9>
- Kadesjo, B., Janols, L. O., Korkman, M., Mickelsson, K., Strand, G., Trillingsgaard, A., et al. (2004). The FTF (Five to Fifteen): The development of a parent questionnaire for the assessment of ADHD and comorbid conditions. *European Child and Adolescent Psychiatry*, 13(Suppl. 3), 3–13. <http://dx.doi.org/10.1007/s00787-004-3002-2>
- Kirkovski, M., Enticott, P. G., & Fitzgerald, P. B. (2013). A review of the role of female gender in autism spectrum disorders. *Journal of Autism and Developmental Disorders*, 43(11), 2584–2603. <http://dx.doi.org/10.1007/s10803-013-1811-1>
- Levy, S. E., Giarelli, E., Lee, L. C., Schieve, L. A., Kirby, R. S., Cunniff, C., et al. (2010). Autism spectrum disorder and co-occurring developmental, psychiatric, and medical conditions among children in multiple populations of the United States. *Journal of Developmental and Behavioral Pediatrics*, 31(4), 267–275. <http://dx.doi.org/10.1097/DBP.0b013e3181d5d03b>
- Lundervold, A. J., Posserud, M. B., Ullebo, A. K., Sorensen, L., & Gillberg, C. (2011). Teacher reports of hypoactivity symptoms reflect slow cognitive processing speed in primary school children. *European Child and Adolescent Psychiatry*, 20(3), 121–126. <http://dx.doi.org/10.1007/s00787-010-0153-1>
- Magiati, I., Tay, X. W., & Howlin, P. (2014). Cognitive, language, social and behavioural outcomes in adults with autism spectrum disorders: A systematic review of longitudinal follow-up studies in adulthood. *Clinical Psychology Review*, 34(1), 73–86. <http://dx.doi.org/10.1016/j.cpr.2013.11.002>
- Matson, J. L., & Shoemaker, M. (2009). Intellectual disability and its relationship to autism spectrum disorders. *Research in Developmental Disabilities*, 30(6), 1107–1114. <http://dx.doi.org/10.1016/j.ridd.2009.06.003>
- Mattila, M. L., Hurtig, T., Haapsamo, H., Jussila, K., Kuusikko-Gauffin, S., Kielinen, M., et al. (2010). Comorbid psychiatric disorders associated with Asperger syndrome/high-functioning autism: A community- and clinic-based study. *Journal of Autism and Developmental Disorders*, 40(9), 1080–1093. <http://dx.doi.org/10.1007/s10803-010-0958-2>
- Munkvold, L., Lundervold, A. B., Lie, S. A., & Manger, T. (2009). Should there be separate parent and teacher-based categories of ODD? Evidence from a general population. *Journal of Child Psychology and Psychiatry*, 50(10), 1264–1272. <http://dx.doi.org/10.1111/j.1469-7610.2009.02091.x>
- Muris, P., Steerneman, P., Merckelbach, H., Holdrinet, I., & Meesters, C. (1998). Comorbid anxiety symptoms in children with pervasive developmental disorders. *Journal of Anxiety Disorders*, 12(4), 387–393.
- Nylander, L., Holmqvist, M., Gustafson, L., & Gillberg, C. (2013). Attention-deficit/hyperactivity disorder (ADHD) and autism spectrum disorder (ASD) in adult psychiatry. A 20-year register study. *Nordic Journal of Psychiatry*, 67(5), 344–350. <http://dx.doi.org/10.3109/08039488.2012.748824>
- O'Brien, G., & Pearson, J. (2004). Autism and learning disability. *Autism*, 8(2), 125–140. <http://dx.doi.org/10.1177/1362361304042718>
- Ohan, J. L., & Visser, T. A. (2009). Why is there a gender gap in children presenting for attention deficit/hyperactivity disorder services? *Journal of Clinical Child and Adolescent Psychology*, 38(5), 650–660. <http://dx.doi.org/10.1080/15374410903103627>
- Park, J. H., Kim, Y.-S., Koh, Y.-J., Song, J., & Leventhal, B. L. (2014). A contrast of comorbid condition and adaptive function between children with Autism Spectrum Disorder from clinical and non-clinical populations. *Research in Autism Spectrum Disorders*, 8(11), 1471–1481.
- Posserud, M. B., Lundervold, A. J., & Gillberg, C. (2009). Validation of the autism spectrum screening questionnaire in a total population sample. *Journal of Autism and Developmental Disorders*, 39(1), 126–134. <http://dx.doi.org/10.1007/s10803-008-0609-z>
- Rosenvinge, J. H., Perry, J. A., Bjørgum, L., Bergersen, T. D., Silvera, D. H., & Holte, A. (2001). A new instrument measuring disturbed eating patterns in community populations: Development and initial validation of a five-item scale (EDS-5). *European Eating Disorders Review*, 9(2), 123–132.
- Simonoff, E., Pickles, A., Charman, T., Chandler, S., Loucas, T., & Baird, G. (2008). Psychiatric disorders in children with autism spectrum disorders: Prevalence, comorbidity, and associated factors in a population-derived sample. *Journal of the American Academy of Child and Adolescent Psychiatry*, 47(8), 921–929. <http://dx.doi.org/10.1097/CHI.0b013e318179964f>
- Thomsen, P. H. (1998). Obsessive-compulsive disorder in children and adolescents. Clinical guidelines. *European Child and Adolescent Psychiatry*, 7(1), 1–11.
- Ullebo, A. K., Posserud, M. B., Heiervang, E., Obel, C., & Gillberg, C. (2012). Prevalence of the ADHD phenotype in 7- to 9-year-old children: Effects of informant, gender and non-participation. *Social Psychiatry and Psychiatric Epidemiology*, 47(5), 763–769. <http://dx.doi.org/10.1007/s00127-011-0379-3>
- Van Wijngaarden-Cremers, P. J., van Eeten, E., Groen, W. B., Van Deurzen, P. A., Oosterling, I. J., & Van der Gaag, R. J. (2014). Gender and age differences in the core triad of impairments in autism spectrum disorders: A systematic review and meta-analysis. *Journal of Autism and Developmental Disorders*, 44(3), 627–635. <http://dx.doi.org/10.1007/s10803-013-1913-9>
- White, S. W., Oswald, D., Ollendick, T., & Scahill, L. (2009). Anxiety in children and adolescents with autism spectrum disorders. *Clinical Psychology Review*, 29(3), 216–229. <http://dx.doi.org/10.1016/j.cpr.2009.01.003>
- Wille, N., Bettge, S., Wittchen, H. U., & Ravens-Sieberer, U. BELLA study group. (2008). How impaired are children and adolescents by mental health problems? Results of the BELLA study. *European Child and Adolescent Psychiatry*, 17(Suppl. 1), 42–51. <http://dx.doi.org/10.1007/s00787-008-1005-0>