Neurodevelopmental correlates of behavioural and emotional problems in a neuropaediatric sample

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Abstract

Most research does not address the overlap between neurodevelopmental disorders when investigating concomitant mental health problems. The purpose of the present study was to examine the association of intellectual disability (ID), autism spectrum disorder (ASD), and attention-deficit/hyperactivity disorder (ADHD) with the presence of behavioural and emotional problems after controlling for other well-known correlates and risk factors. The sample included 4- to 18-year-old children who attended neuropaediatric clinics ($N = 331$). After controlling for adversity, age, gender, other developmental/neurological disorders, parental emotional problems, and parenting strategies, the presence of ADHD but not ASD or ID was uniquely associated with behaviour problems. Neither ADHD nor ASD nor ID was significantly associated with emotional problems after controlling for other risk factors. However, ADHD, ASD and behavioural/emotional disorders but not ID were significantly associated with functional impairment in everyday activities after controlling for other risk factors. Because children with neurodevelopmental disorders have complex needs, a holistic approach to diagnosis and interventions is highly warranted, including the assessment and treatment of behavioural and emotional disorders.

Key words: attention-deficit/hyperactivity disorder; autism spectrum disorders; intellectual disability; mental health disorders; neurodevelopmental disorders

1. Introduction

Population-based studies have established that children and adolescents with neurodevelopmental disorders such as intellectual disability (ID), autism spectrum disorder (ASD), and attention-deficit/hyperactivity disorder (ADHD) have a high risk of comorbid mental health problems compared with other children (Einfeld, Ellis, & Emerson, 2011; Ford, Goodman, & Meltzer, 2003; Larson, Russ, Kahn, & Halfon, 2011; Simonoff et al., 2008). Emotional disorders, such as anxiety disorders and depression, as well as behavioural disorders in the form of irritability and oppositionality, are among the most frequent comorbid mental disorders (Costello, Mustillo, Erkanli, Keeler, & Angold 2003; Dekker & Koot, 2003; Emerson, 2003; Larson et al., 2011; Simonoff et al., 2008). Concurrent behavioural and emotional problems typically emerge during the early preschool years, are relatively persistent, and are associated with parental aggravation, parental mental health problems, and family dysfunction (Chadwick, Kusel, Cuddy, & Taylor, 2005; Einfeld et al., 2006; Herring et al., 2006; Larson et al., 2011; Wallander, Dekker, & Koot, 2006). However, the mechanisms for these putative broader effects of ID, ASD, and ADHD are poorly understood because population-based studies have not, with very few exceptions, distinguished between the underlying neurodevelopmental disorders associated with behavioural and emotional problems (Anderson, Maye, & Lord, 2011; Totsika, Hastings, Emerson, Berridge, & Lancaster, 2011a; Totsika, Hastings, Emerson, Lancaster, & Berridge, 2011b). The co-existence of neurodevelopmental disorders and the sharing of symptoms across disorders are highly common (American Psychiatric Association [APA], 2013). For instance, 50% to 70% of children and adolescents with ASD also have ID (Yeargin-Allsopp et al., 2003). Among children with ID, approximately 17% have ASD (De Bildt, Systema, Kraijer, & Minderaa, 2005). In addition, approximately 45% of children with ADHD are reported to have a concurrent learning disability (DuPaul, Gormley, & Laracy, 2013). Furthermore, substantial

comorbidity exists between ASD and ADHD (Simonoff et al., 2008). The vast majority of studies, for instance, compare children with ASD and IDs to children with IDs or typically developing children and have found an elevated prevalence of concurrent mental health problems in the combined ASD/ID group (Bradley, Summers, Wood, & Bryson, 2004; Eisenhower, Baker, & Blacher, 2005; Hastings et al., 2006). Accordingly, the validity of these findings indicating the presence of elevated behavioural problems in children with ASD compared to other children may be restricted because of the lack of separation between ASD and any co-existing ID (Totsika et al., 2011b). In two population-representative cross-sectional samples aimed at disentangling the putative effects of ASD and ID on mental health problems, Totsika et al. (2011a, 2011b) found that ASD and ID significantly and independently contributed to elevated odds of behavioural and emotional problems, even after controlling for the effects of other well-known risk factors such as age, gender, adversity, and maternal mental health. Moreover, ASD was consistently found to be associated with higher odds ratios than ID. On the other hand, in a prospective study of referred children with ASD, Anderson et al. (2011) reported that the high rate of behavioural difficulty was primarily explained by the severity of ID rather than by core ASD features. Likewise, findings from population-based studies among children and adolescents with ADHD have suggested that the significant association between two disorders may be explained by the presence of other disorders (Bauermeister et al., 2007). For example, the high prevalence of emotional disorders among children with ADHD may be specific to that disorder or due to an indirect association between these two and oppositional defiant disorder (ODD) (Costello et al., 2003; Ford et al., 2003). In a large population-based sample, after adjusting for the presence of behavioural disorders, there was no longer a significant association between ADHD and either anxiety or depression (Ford et al., 2003).

Overall, there is a striking lack of studies examining the association between neurodevelopmental disorders and mental health problems, taking into account the effects of other comorbid neurodevelopmental disorders. Comorbidity has most often been examined between pairs of diagnoses without controlling for the indirect associations that may occur between disorders because both are related to a third disorder (Ford, Goodman, & Meltzer, 2003). Accordingly, the common overlap between neurodevelopmental disorders may exacerbate behavioural and emotional findings, as neurodevelopmental factors are involved in the ontogenesis and epigenesis of mental health problems (Moreno-De-Luca et al., 2013). The identification of the neurodevelopmental disorder or aspect of a given disorder that is associated with an increased risk for mental health problems is an important first step towards the development of a causal hypothesis for subsequent testing. To address the lack of separation of the effects of comorbid neurodevelopmental disorders, we examined the independent association of multiple neurodevelopmental disorders with the presence of concurrent mental health problems and functional impairment.

1.1. The current study

The present study introduced a standardized assessment of concurrent mental disorders as part of the ordinary assessment of neurodevelopmental and neurological conditions among children and adolescents attending neuropaediatric outpatient clinics. This approach enabled the simultaneous examination of the independent association of ID, ASD, and ADHD with the presence of behavioural and emotional problems, controlling for a broad range of other well-known correlates and risk factors reported in the general child psychopathology literature (i.e., adversity, age, gender, other developmental and neurological disorders, parental emotional problems, and parenting strategies) (e.g., Ford, Goodman, & Meltzer, 2004; Shanahan, Copeland, Costello, & Angold, 2008). Based on earlier findings (Anderson et al., 2011; Ford

et al., 2003; Totsika et al., 2011a, 2011b), we hypothesized that i) ASD and ID would be independently associated with both behavioural and emotional problems; ii) ADHD would be independently associated with behavioural problems; and, finally, iii) ASD, ID, ADHD, and comorbid behavioural/emotional disorders would all be independently associated with functional impairment in everyday activities.

2. Methods

2.1. Design and participants

A cross-sectional study was conducted with children and adolescents referred for a developmental/neurological assessment to the neuropaediatric outpatient clinics at the University Hospital of North Norway (UNN) and the Finnmark Hospital Trust by a general practitioner, by child protective services, or by a medical specialist in the specialist health services. Those two neuropaediatric outpatient clinics are specialized health service units in the counties of Troms and Finnmark in northern Norway serving a population of 266,000 residents. Both institutions provide services to children and adolescents with neurodevelopmental disorders or early-acquired disabilities, developmental delays, and intellectual and developmental disabilities. Assessments are interdisciplinary, including specialists such as paediatricians specializing in neurology, neuropsychologists, special education therapists and physiotherapists. The inclusion criteria were that the patients were scheduled to attend one of the selected clinics between October 2012 and July 2016 at the UNN or between January 2014 and July 2016 at the Finnmark Hospital Trust and that they were between 4 and 18 years of age. The presence of mental disorders was identified with the Development and Well-Being Assessment (DAWBA) (Goodman, Ford, Richards, Gatward, & Meltzer, 2000). A total of 518 children and adolescents were eligible for the present study.

One hundred and fifty-three patients (30 %) were excluded either because they were not treated at the clinic as a result of time constraints or because of a lack of parental motivation or insufficient knowledge of the Norwegian language. Additionally, thirty-four participants were excluded because their DAWBA ratings were either missing or incomplete, making diagnostic assessment impossible. DAWBA ratings were available for the remaining 331 children and adolescents.

Written informed consent was obtained before inclusion in the study. The study was approved by the appropriate ethics committee.

2.2. Measures

2.2.1. Child mental disorders

The DAWBA (Goodman et al., 2000) was used to establish diagnoses of mental disorders based on DSM-IV diagnostic criteria (www.dawba.info). The DAWBA is a package of diagnostic interviews with modules for parents, adolescents (11–17 years old), and teachers. The module for parents (n = 115) takes approximately 50 min, the youth interview (n = 115) takes approximately 30 min, and the brief teacher questionnaires (n = 249) take approximately 10 min to complete. The DAWBA has shown good properties for discriminating between population-based and clinical samples as well as between different diagnoses (Goodman et al., 2000). In both Norway and Great Britain, the DAWBA generates realistic estimates of the prevalence of mental illness and has a high predictive validity when used in public health services (Heiervang et al., 2007; Meltzer, Gatward, Goodman, & Ford, 2003). Good to excellent interrater reliability has been reported in both British and Norwegian studies, with k = 0.86–0.91 reported for diagnoses in general, k = 0.57–0.93 for emotional diagnoses, and k = 0.93–1.0 for conduct diagnoses (Ford, Goodman, & Meltzer, 1999;

Heiervang, Goodman, & Goodman, 2008). Good to excellent agreement between diagnoses from clinical practice and those based solely on the DAWBA has also been reported, with k = 0.57–0.76 (Foreman & Ford, 2008; Foreman, Morton, & Ford, 2009).

The parent version of the Strengths and Difficulties Questionnaire (SDQ, Goodman, 1999) was used to assess mental health symptoms. The SDQ, administered as part of the DAWBA (Goodman et al., 2000), is a 25-item mental health questionnaire covering four problem areas (emotional, hyperactivity-inattention, conduct and peer problems), one area of strength (prosocial behaviour), and additional questions related to distress and functional impairment. The SDQ impact score is calculated by summing the distress and impairment items relating to the duration of the problem(s), its degree of impact and areas of manifestations, and the burden that the problem(s) places on the respondents' social environment. In the current study, we included the SDQ emotional symptoms (Cronbach's α = 0.75) and conduct problems (Cronbach's α = 0.69) as continuous measures of mental health problems, in addition to the SDQ impact score (Cronbach's α = 0.79) as a measure of functional impairment. The SDQ has been validated in different cultures, with results indicating good psychometric properties (Achenbach et al., 2008).

2.2.3. Intellectual and adaptive function

Children were individually assessed with a standardized Wechsler intelligence test appropriate for their age (Wechsler, 2007, 2008a, 2008b, 2009, 2012). A small number of children (n = 11) were assessed with Raven's Coloured Progressive Matrices (Raven, 2004). Intellectual level was defined by the Full Scale IQ (FSIQ) score. For thirty-five children, the FSIQ scores were missing due to the administration of a test appropriate for chronologically younger children.

To measure the children's adaptive abilities, we administered the Vineland-II (Sparrow, Cicchetti, & Balla, 2011), a semi-structured interview that includes the following four domains with related subdomains: communication (receptive, expressive and written), daily living skills (personal, domestic, and community), socialization (interpersonal relationships, play and leisure time, and coping skills) and motor skills (gross and fine). In the present study, the total score was used.

2.2.4. Parent mental health

The emotional well-being of the children's parents was measured using the self-report version of the 10-item Everyday Feelings Questionnaire (EFQ) (Uher & Goodman, 2010). The EFQ was administered as part of the DAWBA (Goodman et al., 2000). The EFQ measures symptoms related to depression and anxiety during the past four weeks, as well as items reflecting psychological well-being, such as optimism, self-esteem and coping. Higher scores indicate the presence of greater emotional problems. The EFQ has been validated in both epidemiological (Uher & Goodman, 2010) and clinical (Mann, Henley, O´Mahen, & Ford, 2013) populations. Cronbach's α for the total score in the present study was 0.87.

2.2.5. Parental practice

Parental practice was measured using the 13-item Family Life Questionnaire (FaLQ) (Last, Miles, Wills, Brownhill, & Ford, 2012). The FaLQ, administered as part of the DAWBA (Goodman et al., 2000), consists of four scales: affirmation (four items related to the child-parent relationship, e.g., receives love and affection, is praised and rewarded), negative discipline (four items related to punishment, e.g., is physically punished, is blamed unfairly), rules (two items measuring structure and organization within the family, e.g., have clear rules,

rules are applied consistently) and special allowances (three items related to parental over- and underinvolvement; this scale was not used in the current analyses). A higher score indicates greater adherence to a particular parental practice. In the current study, three scales from the FaLQ were used: affirmation (Cronbach's $\alpha = 0.83$), rules (Cronbach's $\alpha = 0.76$), and negative discipline (Cronbach's $\alpha = 0.53$). The lower Cronbach's α for the negative discipline scale was partially due to the relatively low item-total correlation ($r = 0.20$) for the "Physical punishment" item. Last et al. (2012) found that the internal consistency and test-retest reliability for affirmation and rules varied between moderate and very good, whereas the negative discipline subscale had poor internal consistency.

2.2.6. Social factors

A social disadvantage sum score variable was created by including the following variables: unemployment, a home inadequate for the family's needs, and economic difficulties reported on the background section in the DAWBA (Goodman et al., 2000), as well as the lowest reported parental education level and single-parent home status. Higher scores indicate the presence of greater social disadvantage.

A negative life events sum score variable was created based on parents' reports on the total number of stressful life events that were experienced by the family or by the child in the last 12 months. This sum score variable included the following variables from the background section in the DAWBA (Goodman et al., 2000): accident/serious injury, severe disease/hospitalization, death (of a parent or sibling/friend), loss of friendship, separation/divorce and other serious events. Higher scores indicate a greater number of negative life events.

2.3. Procedure

The parents and children who agreed to participate underwent the ordinary interdisciplinary assessment of neurodevelopmental/neurological disorders and an additional assessment of the presence of comorbid behavioural and emotional disorders at the same time, typically over two consecutive days. Paediatricians specializing in neurology examined subjects for the presence of a neurological/neurodevelopmental disorder; the examinations included, for instance, MR Caput, EEG and/or genetic testing if indicated. Children with muscle disease or motor delays were also examined by a physiotherapist. All children were examined by a clinical psychologist/neuropsychologist; these examinations included using a standardized intelligence scale and the Vineland-II. Diagnoses of neurological and neurodevelopmental disorders, including ASD, ADHD, and ID, were obtained from interdisciplinary assessments in the neuropaediatric clinics. The ICD-10 criteria (World Health Organization [WHO], 1993, 2010) were used to code diagnoses. The presence of an ID was operationalized as scoring below 70 on both the standardized intelligence test and the Vineland-II. The parents and children who agreed to participate, in addition to their teachers, further underwent an assessment (using the web-based version of the DAWBA) of the presence of comorbid mental disorders among the children. After completion of the DAWBA interview, two expert raters (X and Y, both of whom are senior clinical specialists in neuropsychology with at least 15 years of experience in the field and are trained in DAWBA rating (Publication removed due to double-blind peer review), generated diagnostic ratings based on the structured (yes/no) and semi-structured (free text) answers provided by parents, teachers and young persons after reviewing the full DAWBA information. The decision rules from the DSM-IV (APA, 2000) diagnostic criteria were used. Comorbidity was registered whenever the diagnostic criteria for more than one diagnosis were met, without attention to the exclusion rules of the DSM-IV. To ensure that there were enough cases for analysis, we

grouped the diagnoses into categories: emotional disorders (diagnoses related to separation anxiety, specific phobias, social phobia, panic attacks and agoraphobia, post-traumatic stress disorder, generalized anxiety, compulsions and obsession, depression, and deliberate self-harm) and conduct disorders (diagnoses related to awkward and troublesome behaviour).

2.4. Statistical analyses

All analyses were conducted using SPSS 24. The relationships between ID, ASD, ADHD, other correlates and risk factors and behavioural or emotional disorder diagnoses, symptoms and functional impairment were examined using Pearson's correlation coefficients. Two hierarchical linear regression analyses were conducted to examine whether ID, ASD and ADHD were associated with behavioural/emotional symptoms after controlling for the other well-known correlates and risk factors. For all analyses, independent variables included in step 1 were social disadvantage, negative life events, age, gender (0 = girl, 1 = boy), and neurological/other developmental disorders, excluding ID, ASD and ADHD (0 = no, 1 = yes). Step 2 included parental mental health problems, affirmation, negative discipline, and rules, and step 3 included ID (0 = no, 1 = yes), ASD (0 = no, 1 = yes), and ADHD (0 = no, 1 = yes). Since research in the general child population has indicated an interaction between age and gender for mental disorders (i.e., greatest risk for younger boys and older girls) (Waddell, Offord, Shepherd, Hua, & McEwan, 2002), we initially included the interaction term in the regression analyses but subsequently excluded it from the analyses as it was non-significant. The dependent variables were SDQ behavioural symptoms and SDQ emotional symptoms. We assessed the significance of change in R^2 attributable to ID, ASD, and ADHD after first entering all the other independent variables. By convention, an R^2 change of 2% is a small effect, a change of 13% is a medium effect, and a change of 26% is a large effect (Cohen, 1988). Individual independent variables were interpreted only if the corresponding step was

significant. In addition, the BIC (Bayesian information criterion) was calculated as an additional criterion for model selection, where the model with the lowest BIC is preferred (Schwartz, 1978). When choosing between models, $\Delta BIC > 2$ is considered positive, 6-10 strong, and >10 very strong evidence against the model with the higher BIC (Rafferty, 1995). In cases where the BIC and the R^2 criteria differed concerning model selection, R^2 criterion was preferred.

Likewise, two hierarchical logistic regression analyses were conducted to examine whether ID, ASD and ADHD were associated with behavioural/emotional disorders beyond that gained from the other well-known correlates and risk factors. Independent variables were the same as in the linear regression analyses. The interaction term between gender and age was non-significant and was thus excluded from the analyses. The dependent variables were dummy variables for behavioural disorder (0 = no, 1 = yes) and emotional disorder (0 = no, 1 = yes). We assessed the significant contributions of ID, ASD and ADHD after first entering all the other independent variables. The overall model was tested using the chi-squared statistic, and differences between the models (step 1 vs 2 and step 2 vs 3) were tested using the difference in model chi squared for each step. In addition, the BIC was calculated as a criterion for model selection. Individual predictor variables were only interpreted if the corresponding step was significant. The following guidelines were used for assessing the effect sizes: an OR of 1.68 is a small effect, an OR of 3.47 is a medium effect, and an OR of 6.71 is a large effect (Chen, Cohen, & Chen, 2010). The equivalent values for OR less than 1.00 would be 0.59 (small effect), 0.29 (medium effect), and 0.15 (large effect). Finally, a hierarchical linear regression analysis was conducted to examine whether ID, ASD, ADHD, and a behavioural/emotional disorder status (0 = no, 1 = yes) were associated with distress and functional impairment after controlling for the other well-known risk factors. The independent variables were the same as in the regression analyses above. The

behavioural/emotional disorder status was included as a separate step (step 4). The dependent variable was the SDQ impact score. Tolerance values were inspected to examine the possibility of multicollinearity. For the present data set, a minimal tolerance problem was not observed for the independent variables. The number of participants included in the different regression analyses ranged from $N = 267$ (logistic regression) to $N = 239$ (SDQ impact score). The reason for this range was the missing items on the EFQ, the FaLQ, items related to the social disadvantage and negative life events sum score variables, and the SDQ impact section. The participants with the missing data did not differ significantly from the sample with the complete data included in the analyses in terms of age, gender, FSIQ or Vineland-II scores (i.e., $N = 267$ vs. $N = 64$: $t_{age}(329) = -1.11, p > .05$; $\chi^2_{gender}(1) = 0.03, p > .05$; $t_{FIQ}(294) = 1.31, p > .05$; $t_{Vineland\ total}(313) = 1.46, p > .05$. $N = 239$ vs. $N = 92$: $t_{age}(329) = -1.49, p > .05$; $\chi^2_{gender}(1) = 0.42, p > .05$; $t_{FIQ}(294) = 0.46, p > .05$; $t_{Vineland\ total}(313) = -1.64, p > .05$).

3. Results

3.1. Participant characteristics

Descriptive statistics were calculated for the total sample of children and adolescents ($N = 331$). The participants had a mean age of 10.12 years ($SD = 3.79$, range 4–18 years), and 64.7% were boys. The mean FSIQ was 76.46 ($SD = 16.99$, range 40–140), and the mean level of adaptive functioning (Vineland-II total score) was 67.15 ($SD = 14.98$, range 20–112) (see Table 1 for additional information). The majority of children lived with both parents (80.6%), with very few children living in child disability care (0.9%). The most frequent neurodevelopmental disorders in the sample were, in descending order, specific developmental disorders (33.5%), ID (20.8%, none with severe IDs), other diseases of the nervous system such as epilepsy and cerebral palsy (17.5%), ASD (14.5%), ADHD (13.6%),

and congenital malformations and chromosomal abnormalities (10.0%). The diagnoses were not mutually exclusive. Accordingly, among children with ASD, 18.8% had a comorbid diagnosis of ID, 8.3% had a comorbid ADHD diagnosis, 2.1% had the comorbid diagnoses of ID and ADHD, and 20.8% had a comorbid diagnosis of other developmental/neurological disorders. Among children with ADHD, 17.8% had a comorbid diagnosis of ID, 8.9% had a comorbid ASD diagnosis, and 48.9% had a comorbid diagnosis of other developmental/neurological disorders (mostly specific developmental disorder). In addition, 14.8% of the sample did not receive a neurodevelopmental and/or neurological disorder diagnosis (69.4% boys, mean age 11.02 (*SD* = 3.87), mean FSIQ 89.33 (*SD* = 19.04), mean Vineland-II total score 76.11 (*SD* = 13.83)).

As shown in Table 2, the DMS-IV mental disorders included anxiety disorders (17.1%), depression (4.5%) and behavioural disorders (14.2%) in the form of irritability and oppositionality. Overall, 25.7% of the children had a behavioural and/or emotional diagnosis. Two or more diagnoses were present in 8.4% of the children. Of the respondents, 40.5% were mothers, 25.7% included both parents, 15.1% were a parent whose gender was not specified, 8.5% were fathers, 6.6% were foster mothers, and the remainder were mainly step-fathers and foster fathers. Half of the respondents had obtained a college or university degree (50.5%), with 35% obtaining a high school diploma alone.

Table 1 about here

Table 2 about here

3.2. Relationship between ID, ASD, ADHD and other risk variables and behavioural and emotional problems

Table 1 shows the correlations among the variables examined, as well as descriptive statistics. Overall, the independent variables were moderately to weakly correlated ($r < 0.40$) with the outcome variables in the expected directions (Table 1). ID, ASD and ADHD diagnostic status all lacked any significant relation to a diagnosis of emotional disorder or related symptoms (SDQ emotional symptoms). The diagnostic status of ADHD was significantly related to a behavioural diagnosis (ODD), symptoms (SDQ conduct problems), and functional impairment (SDQ impact) with small to medium correlations. An ID diagnostic status was significantly related to behavioural symptoms and functional impairment (small correlations). The diagnostic status of ASD was significantly related only with the outcome variable functional impairment with a small correlation

As shown in Table 3, regarding the association with the presence of behavioural symptoms, the overall model was significant ($F(12) = 11.69, p < .001$), accounting for 37% of the variance in behavioural symptoms. Adversity/child factors (step 1) and family factors (step 2) accounted for 10% and 23% of the variance in behavioural symptoms, respectively,

reflecting effects of medium and large magnitudes. It is of particular interest that the diagnostic status of neurodevelopmental disorders (i.e., ID, ASD, and ADHD) accounted for an additional 4% of the variance in behavioural symptoms, reflecting effects of small magnitude. The BIC values were calculated for the three models (step 1 to step 3), and the corresponding numbers were: 359.46, 307.89 and 307.43. The delta BICs were -51.57 (between model 1 and model 2) and -0.46 (between model 2 and model 3) favouring the simpler model in step 2 compared to step 3, whereas and inspection of the ΔR^2 indicated that even step 3 was significant More specifically, when inspecting the significant independent variables, a lack of a neurological diagnosis/other developmental disorder, low use of affirmation, high use of negative discipline, and presence of ADHD were significantly associated with elevated levels of behavioural symptoms.

The overall model for the presence of emotional symptoms was significant ($F(12) = 3.50, p < .001$), accounting for 14% of the variance in emotional symptoms. As shown in Table 3, the diagnostic status of ID, ASD, and ADHD (step 3) did not account for a significant incremental variance in emotional symptom scores. Adversity/child factors (step 1) and family factors (step 2) accounted for 9% and 5% of the variance in emotional symptoms, respectively, reflecting effects of medium and small magnitude. The BIC values were calculated for the three models (step 1 to step 3), and the corresponding numbers were: 519.43, 527.61 and 543.36. The delta BICs were 8.18 (between model 1 and model 2) and 15.75 (between model 2 and model 3) favouring the simpler model in step 1, whereas an inspection of the ΔR^2 indicated that even step 2 was significant. More specifically, female gender and parental emotional problems were significantly associated with higher levels of emotional symptoms.

Table 3 about here

To investigate the correspondence of variables to the presence of mental health problems as symptoms (i.e., continuous variables) versus mental health problems as a diagnosis (i.e., categorical variables), we used the same hierarchical model, but in this case, we modelled the presence of a behavioural or emotional diagnosis in two separate hierarchical logistic regression analyses. As shown in Table 4, the overall model for the presence of a behavioural diagnosis was significant ($\chi^2(12) = 48.99, p < .001$), correctly specifying 85.8% of group memberships. Adversity/child factors (step 1) and family factors (step 2) were significantly associated with the presence of a behavioural diagnosis. The diagnostic status of neurodevelopmental disorders (step 3) did not significantly improve the model. The BIC values were calculated for the three models (step 1 to step 3), and the corresponding numbers were: 245.94, 250.20 and 261.62. The delta BICs were -4.25 (between model 1 and model 2) and -11.42 (between model 2 and model 3) favouring the simpler model in step 1, whereas an inspection of the ΔR^2 indicated that even step 2 was significant. More specifically, compared to children without a behavioural diagnosis, children with such a diagnosis had an increased probability of not having a neurological diagnosis or other developmental disorders, and their parents reported using lower levels of affirmation and more negative discipline strategies than did the parents of children who did not have a behavioural diagnosis (see Table 4). The ORs ranged from 0.19 (neurological/other developmental disorder) to 1.39 (negative discipline), respectively, reflecting effects of large and small magnitude.

Likewise, the overall model for the presence of an emotional diagnosis was significant ($\chi^2(12) = 23.74, p = 0.02$), correctly specifying 85.4 % of group memberships. The BIC values were calculated for the three models (step 1 to step 3), and the corresponding numbers were: 237.87, 254.52 and 269.35. The delta BICs were -16.65 (between model 1 and model 2)

and -14.82 (between model 2 and model 3) which favours the simpler model in step 1. As shown in Table 4, the family factors (step 2) and the diagnostic status of ID, ASD, and ADHD (step 3) did not significantly improve the model fit . On the other hand, adversity and child factors (step 1) were significantly associated with the presence of an emotional diagnosis. Of individual predictors, only gender was significant, where girls were likely to have an emotional diagnosis compared to boys.

Table 4 about here

Finally, to test how the diagnostic status of ID, ASD, ADHD, and behavioural/emotional disorder explained overall child functional impairment, we conducted a separate hierarchical multiple regression analysis, controlling for any associations with other risk variables. The independent variables were entered in the same steps and order as in the previous analyses. However, in this analysis, we also included the presence of a comorbid behavioural/emotional disorder as an independent variable in a separate step 4. As shown in Table 5, the overall model was significant ($F(13) = 8.67, p < 0.001$), accounting for 33% of the variance in functional impairment. Adversity/child factors (step 1) and family factors (step 2) accounted for 6% and 12% of the variance in functional impairment, respectively, reflecting effects of small and medium magnitude. It is of particular interest that the diagnostic status of neurodevelopmental (step 3) and behavioural/emotional disorder (step 4) accounted for an additional 7% and 8% of the variance in functional impairment, reflecting effects of small to medium magnitude. The BIC values were calculated for the four models (step 1 to step 4), and the corresponding numbers were: 498.96, 488.32, 478.43 and 455.76.

The delta BICs were -10.64 (between model 1 and model 2), -9.89 (between model 2 and model 3) and -22.67 (between model 3 and model 4) favouring the more complex model in step 4. More specifically, based on an inspection of individual independent variables, we found that parental emotional problems, greater use of rules, and ASD, ADHD, and behavioural/emotional disorder status (i.e., ODD and anxiety disorder) were significantly associated with higher levels of functional impairment.

Table 5 about here

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4. Discussion

A large number of studies indicate a higher prevalence of mental health problems among children with ASD and ADHD than among other children (Larson et al., 2011; Simonoff et al., 2008). However, the validity of these findings may be restricted due to the lack of separation of the associations of ASD, ADHD and any co-existing ID. Therefore, the aim of the present study was to examine the independent association of ASD, ADHD and ID after controlling for other well-known correlates and risk factors. Although an initial bivariate link between ID and behavioural problems was found, regression analyses did not support our first hypothesis that ID and ASD were independently associated with behavioural and

emotional problems after controlling for a range of other factors (including neurodevelopmental/neurological disorders). Our failure to find the hypothesized relationship between ASD and ID and behavioural and emotional problems is not in accordance with initial findings from two population-based studies (Totsika et al., 2011a, 2011b). One obvious potential explanation for the divergent result of the present study might be methodological differences. The present study recruited participants from clinical referrals to neuropaediatric clinics. Referred cases potentially have different clinical characteristics than children in the general population (for example, a chronic course and high comorbidity) (Biederman et al., 2008). Thus, among the participants with ID and ASD in the present study, a substantial proportion had other comorbid developmental and neurological conditions (e.g., specific learning disorder, epilepsy, and genetic syndromes). The presence of co-existing conditions other than ASD and ID in the population-based studies by Tostika et al. (2011a, 2011b) was unknown, in addition to the lack of clinical ascertainment of ASD and ID cases. Hence, it cannot be ruled out that the individual associations of ASD and ID with behavioural and emotional problems in the studies of Tostika et al. (2011a, 2011b) may be epiphenomena of multiple comorbidities within the neurodevelopmental disorders. The causes of the interrelationship between ASD and mental health problems are not fully understood. Thus, future population-based studies disentangling the putative effects of ASD, ID, and other frequently co-occurring neurodevelopmental disorders on behavioural and emotional problems will be an important first step towards developing causal hypotheses for subsequent testing.

In keeping with the second hypothesis, we found a significant association between ADHD and behavioural problems when using a dimensional approach to behavioural problems mostly ODD symptoms, controlling for the presence of ID, ASD and a range of other factors (including other developmental/neurological disorders such as a specific learning

disability). This finding suggests that the comorbidity between ADHD and ODD symptoms is unique and not solely accounted for by diagnostic overlaps with other neurodevelopmental disorders and well-known risk factors such as specific learning disorders and the use of harsh negative discipline by parents. Indeed, Dick, Viken, Kaprio, Pulkkinen, and Rose (2005), analysing data from a large sample of Finnish twin pairs, found evidence indicating that the comorbidity among ADHD, ODD, and conduct disorder was primarily explained by shared genetic influences; however, each disorder was also under a unique set of genetic influences. Common genetic risk factors may include genes influencing temperament. One suggested interpretation of the ADHD/behavioural problems comorbidity is that ADHD indexes a pattern of neurocognitive impairments or delays, which, in combination with adverse temperamental features, contributes to the risk of developing more global behavioural dysregulation marked by oppositionality and defiance (Maughan, Rowe, Messer, Goodman, & Meltzer, 2004). As expected, a recent longitudinal community-based representative sample suggested that shared temperament factors in the form of high levels of temperamental activity underlie the co-occurrence of ADHD (the hyperactive-impulsive and combined types) and ODD (Stringaris, Maughan, & Goodman, 2010).

With respect to our third hypothesis, we found that ASD, ADHD, and behavioural/emotional disorder but not ID were significantly related to distress and functional impairment in everyday activities after accounting for the effects of other developmental/neurological disorders, adversity, age, gender, and parent variables. Behavioural/emotional disorder explained approximately the same amount of variance (8%) in functional impairment as did the step with ID, ASD and ADHD (7%). This finding highlights the importance of addressing comorbid mental health problems in neuropaediatric assessments, as such comorbidities adversely influence the everyday life of the child and family (e.g., Herring et al., 2006; Larson et al., 2011). The lack of evidence uniquely

associating ID with functional impairment after accounting for the other variables may reflect parents' expectations in their reporting of functional problems. That is, perhaps parents do not view certain behaviours as "abnormal" for a child with a global cognitive disability and instead may evaluate their child's functioning based on expectations adjusted for the child's lower levels of cognitive and adaptive functioning (Gioia, Isquith, Guy, & Kenworthy, 2000).

4.1. Strengths and limitations of the study

The strengths of the present study included the use of a structural diagnostic interview (i.e., the DAWBA) in diagnosing mental health disorders, whereas the majority of population-based studies have employed symptom measures (Dekker, Koot, van der Ende, & Verhulst, 2002; Einfeld et al., 2006; Emerson & Einfeld, 2010; Emerson et al., 2010; Totsika et al., 2011a, 2011b; Wallander et al., 2006). The DAWBA has not been specifically validated for children and adolescents with ID. However, the subgroup with ID in the present study was only mildly impaired, with none of the patients having severe ID. The DAWBA interview has also been used frequently in recent population-based studies on the ID population, making the findings from divergent studies more comparable (Emerson, 2003; Emerson & Hatton, 2007; Hatton & Emerson, 2004). Another strength was the use of a standardized assessment of the children's intellectual and adaptive levels. A restricted range of correlates or predictors have been investigated with any degree of regularity in previous studies, with the significance of age, gender, level of functioning, and socioeconomic status remaining uncertain (Einfeld et al., 2011). The present study simultaneously investigated multiple neurodevelopmental disorders, controlling for a relatively broad range of correlates and risk factors found in the general child population. Nevertheless, some limitations should be noted.

The present sample included only participants who had sufficient knowledge of the Norwegian language and whose parents were motivated to contribute. We employed a clinical sample; accordingly, the results may not generalize to children with ASD and ID in the general population. However, the results are likely to generalize to ADHD children seen in neuropaediatric and psychiatric settings. Additionally, the cross-sectional design precludes any interpretations regarding the causality of the identified correlates.

Notwithstanding these limitations, the main conclusion from the study is that the presence of an ADHD diagnosis is uniquely related to concurrent ODD symptoms even after accounting for the presence of other neurodevelopmental/neurological disorders and risk factors. Accordingly, preventive efforts to address the risk of ODD among children with ADHD are imperative to improve compromised outcomes for ADHD youths. A second conclusion indicates that, as behavioural and emotional disorders are uniquely associated with functional impairment after accounting for the presence of neurodevelopmental and neurological disorders, neuropaediatric clinics should routinely address mental health problems.

Acknowledgements

This study was supported by the Northern Norway Regional Health Authority (PFP1178-14). The study was also supported in part by The Norwegian Directorate of Health and the Regional Centre for Child and Youth Mental Health and Child Welfare North at UiT The Arctic University of Norway, Tromsø, Norway. The authors would like to thank the participants who contributed to the dataset. We would also like to thank Professor Eric

Emerson for commenting on an early draft of the manuscript. Finally, we would like to thank the anonymous reviewers for helpful suggestions that improved this paper.

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A publication is removed due to double-blind peer review.

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Table 1

Descriptive statistics and correlations among main variables

Variable	*M (SD)/ n (%)*	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17
Predictor variables																		
1. Social disadvantage	2.38 (1.24)	-																
2. Neg. life events	0.57 (0.87)	.19**	-															
3. Age	10.12 (3.79)	.01	.07	-														
4. Gender[a]	214 (64.7)	.10	-.07	-.12*	-													
5.Neurological/develop.[b]	189 (57.1)	-.13*	.08	-.01	-.11	-												
6. Parent mental health	11.57 (5.05)	.24***	.13*	.02	-.01	-..04	-											
7. Affirmation	10.55 (1.87)	-.05	-.04	-.20***	-.09	.06	-.25***	-										
8. Neg. discipline	2.90 (1.50	.13*	.01	-.12*	.12*	-.06	.26***	-.08	-									
9. Rules	3.99 (1.26)	-.03	-.05	-.04	-.02	.06	-.17**	.35***	.12*	-								
10. ID[c]	69 (20.8)	.03	.02	-.02	.01	.-.26***	.04	.01	.04	-.02	-							
11. ASD[d]	48 (14.5)	.00	.02	-.05	.04	-.30***	.07	-.01	-.02	.00	-.02	-						
12. ADHD[e]	45 (13.6)	-.01	-.04	.04	.16**	-.07	-.01	-.11	.20***	-.07	-.03	-.06	-					
13. Emo./behav. diag.[f]	85 (25.7)	.16**	.03	.14**	-.06	-.15**	.16**	-.13*	.13*	-.03	.04	.03	.11*	-				
Mental health																		
14. Emotional diag.[g]	48 (14.5)	.10	.12*	.17**	-.14**	.03	.16**	-.00	-.03	-.04	.00	-.02	.04	.70***	-			
15. Behavioural diag.[h]	47 (14.2)	.09	-.03	.01	.05	-.24***	.07	.-.17**	.22***	.02	.05	.10	.17**	.69***	.08	-		
16. SDQ emo. symp.	3.39 (2.60)	.15*	..15**	.15**	-.18***	-.03	.26***	-.05	.01	-.00	.05	.03	-.01	.47***	.56***	.14*	-	
17. SDQ behav. symp.	2.03 (1.93)	.16**	.13*	.03	.07	-.23***	.21***	-.35***	.39***	-.12*	.17**	-.00	.27***	.43***	.13*	..49***	..28***	-
18. SDQ Impact score	3.97 (2.75)	.15*	.06	.14*	-..03	-.11	.30***	-.01	.19**	.10	.12*	.16**	.24***	.38***	.35***	.22***	.42***	.28***

Note. [a]Boy = 1 and 0 = girl. [b] Neurological/other developmental disorders except ID, ASD, and ADHD = 1 and 0 = absence. [c]ID = 1 and 0 = absence. [d]ASD = 1 and 0 = absence. [e]ADHD = 1 and 0 = absence. [f]Emotional or behavioural disorder = 1 and 0 = absence. [g]Emotional disorder = 1 and 0 = absence. [h]Behavioural disorder = 1 and 0 = absence. * $p < .05$, ** $p < .01$, *** $p < .001$ (2-tailed).

Table 2

Prevalence of mental health disorders in a neuropaediatric sample ($N = 331$)

Disorder[a]	*n* (%)
Major depression	15 (4.5 %)
Separation anxiety	13 (3.9 %)
Specific phobia	12 (3.6 %)
Social phobia	13 (3.9 %)
Panic Disorder	4 (1.2 %)
Posttraumatic stress disorder	1 (0.3 %)
Obsessive compulsive disorder	4 (1.2 %)
Generalized anxiety	9 (2.7 %)
Other anxiety	1 (0.3 %)
Undifferentiated Anxiety/Depression	1 (0.3 %)
Oppositional defiant	41 (12.4%)
Conduct disorder	6 (1.8 %)

Note. [a]No children met criteria for Agoraphobia, Selective mutism, Other disruptive disorder NOS, Reactive attachment disorder, Eating disorders, Bipolar disorder, or Psychosis. Diagnoses are not mutually exclusive.

Table 3

Hierarchical regression analyses results for variables associated with behavioural and emotional symptoms

Variable	Behavioural symptoms		Emotional symptoms	
	ΔR^2	β	ΔR^2	β
Step 1	.10***		.09***	
Social disadvantage		.07		.10
Negative life events last year		.10		.10
Age		-.04		.11
Gender[a]		-.07		-.18**
Neurological/other develop.[b]		-.20***		-.01
Step 2	.23**		.05**	
Parental emotional problems		.02		.23***
Affirmation		-.30***		-.02
Negative discipline		.30***		-.05
Rules		-.02		.08
Step 3	.04***		.00	
ID[c]		.06		.02
ASD[d]		-.05		.06
ADHD[e]		.20***		.03
Total R^2	.37***		.14***	
N	253		267	

Note. The beta-coefficients were taken from the last step of the model. All independent variables except neurological/other developmental disorders, ID, ASD and ADHD, were based on DAWBA. The dependent variables were based on SDQ. [a]Boy = 1 and 0 = girl. [b]Neurological/other developmental disorders except ID, ASD, and ADHD = 1 and 0 = absence. [c]ID = 1 and 0 = absence. [d]ASD = 1 and 0 = absence. [e]ADHD = 1 and 0 = absence. *$p < .01$, **$p < .001$.

Table 4

Hierarchical logistic regression results for variables associated with behavioural and emotional diagnoses ($N = 267$)

Variable		Behavioural diagnosis[a]				Emotional diagnosis[b]		
	$\Delta\chi^2$	β	*SE*	*OR*	$\Delta\chi^2$	β	*SE*	*OR*
Step 1	25.55***				16.10**			
Social disadvantage		0.16	0.17	1.17		0.16	0.16	1.18
Negative life events		-0.14	0.23	0.87		0.31	0.19	1.36
Age		0.01	0.06	1.01		0.10	0.06	1.10
Gender[c]		-0.41	0.42	0.67		-0.85*	0.40	0.43
Neurological/other develop.[d]		-1.65***	0.45	0.19		0.17	0.45	1.19
Step 2	18.10***				5.70			
Parental emotional problems		-0.01	0.04	0.99		0.10**	0.04	1.10
Affirmation		-0.23*	0.11	0.79		0.03	0.11	1.03
Negative discipline		0.33**	0.13	1.39		-0.15	0.14	0.86
Rules		0.22	0.17	1.25		0.06	0.16	1.07
Step 3	5.34				1.94			
ID[e]		-0.22	0.50	0.80		-0.19	0.58	0.83
ASD[f]		0.64	0.50	1.90		0.07	0.61	1.08
ADHD[g]		0.94*	0.48	2.55		0.77	0.56	2.16

Note. All independent variables except neurological/other developmental disorders, ID, ASD and ADHD, were based on DAWBA. The dependent variables were based on DAWBA[a]Model $\chi^2(12) = 48.99$, $p < .001$. Cox & Snell $R^2 = .17$, Nagelkerke $R^2 = .29$. [b]Model $\chi^2(12) = 23.74$, $p = .02$. Cox & Snell $R^2 = .09$, Nagelkerke $R^2 = .15$. [c]Boy = 1 and 0 = girl. [d]Neurological/other developmental disorders except ID, ASD, and ADHD = 1 and 0 = absence. [e]ID = 1 and 0 = absence. [f]ASD = 1 and 0 = absence. [g]ADHD = 1 and 0 = absence. * $p < .05$, ** $p < .01$, ***$p < .001$.

Table 5

Hierarchical regression analysis results for variables associated with distress and functional impairment

Variable	Functional impairment	
	ΔR^2	β
Step 1	.06**	
Social disadvantage		.02
Negative life events		.02
Age		.07
Gender[a]		-.03
Neurological/other develop.[b]		-.03
Step 2	.12***	
Parental emotional problems		.26***
Affirmation		.06
Negative discipline		.05
Rules		.18**
Step 3	.07***	
ID[c]		.05
ASD[d]		.18**
ADHD[e]		.19***
Step 4	.08***	
Behavioural/emotional disorder[f]		.31***
Total R^2	.33***	
N	239	

Note. All independent variables except neurological/other developmental disorders, ID, ASD and ADHD, were based on DAWBA. The dependent variable was based on SDQ. The beta-coefficients were taken from the last step of the model. [a]Boy = 1 and 0 = girl. [b]Neurological/other developmental disorder except ID, ASD, and ADHD = 1 and 0 = absence. [c]ID = 1 and 0 = absence. [d]ASD = 1 and 0 = absence. [e]ADHD = 1 and 0 = absence. [f]Behavioural/emotional disorder = 1 and 0 = absence.
*p < .01, **p < 0.01.