Pervasive developmental disorder behavior in adolescents with intellectual disability and co-occurring somatic chronic diseases

B. Oeseburg a,b,*, J.W. Groothoff a, G.J. Dijkstra a, S.A. Reijneveld a, D.E.M.C. Jansen a,c

a Department of Health Sciences and Graduate School for Health Research (SHARE), University Medical Center Groningen, University of Groningen, Groningen, The Netherlands
b Wenckebach Institute, School of Nursing and Health, University Medical Center Groningen, Groningen, The Netherlands
c Department of Sociology, Faculty of Behavioral and Social Sciences, University of Groningen, Groningen, The Netherlands

1. Introduction

Studies on the association between somatic chronic diseases in adolescents with intellectual disability (ID-adolescents) and the full range of pervasive developmental disorder behavior (PDD behavior) are hardly available. Mild PDD behavior is a term reserved for those who do not meet the criteria for severe PDD behavior like in autism or Asperger syndrome. Mild PDD behavior is widespread among ID-adolescents and has a profound effect on their daily functioning (de Bildt et al., 2005a; de Bildt, Sytema, Kraijer, & Minderaa, 2005; Hartman, Luteijn, Serra, & Minderaa, 2006; Kraijer, 2000).

Evidence on the association between somatic chronic diseases in ID-adolescents and the full range of pervasive developmental disorder behavior (PDD behavior) is scarce. The aim of the present study is to assess the association between somatic chronic diseases in ID-adolescents and mild PDD behavior. We obtained data on 1044 ID-adolescents, aged 12–18, attending secondary schools in the Netherlands. Parents of the adolescents completed the Dutch version of the Children's Social Behavior Questionnaire (CSBQ) parent version, covering a wide range of PDD behavior, and questions about chronic diseases and background characteristics of their child. ID-adolescents with somatic chronic diseases showed more PDD behavior, in particular milder forms, than their peers without chronic diseases. In addition, ID-adolescents with somatic chronic diseases in combination with pervasive developmental disorders (PDD) and attention deficit hyperactivity disorder (ADHD) also showed more PDD behavior than their peers with only PDD/ADHD. Clinicians should be extra alert on PDD behavior, in particular the milder forms, in ID-adolescents when somatic chronic diseases are present. However, to strengthen our results about the relationship between somatic chronic diseases in ID-adolescents and PDD behavior studies are needed using both the CSBQ and standardized diagnostic instruments.

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greater chance on both pervasive development disorders (PDD) (de Bildt et al., 2005a; Harris et al., 2008; Matson & Shoemaker, 2009; Walker et al., 2004) and attention deficit hyperactivity disorder (ADHD) (Emerson & Hatton, 2007; Matson & Shoemaker, 2009). Although ADHD and PDD have different nosological diagnoses, both diagnostic categories partly include similar symptoms like deficits in social interaction, impulsivity, attention and hyperactivity deficit (Gallagher, Bellgrove, Hawi, Segurado, & Fitzgerald, 2007). These deficits are also associated with mild PDD behavior (de Bildt et al., 2005a; Hartman et al., 2006; Nijmeijer et al., 2009).

Literature also suggests that adolescents with somatic chronic diseases show more PDD behavior than adolescents in the general population (Ekström, Hakenas-Plate, Samuelsson, Tulinius, & Wentz, 2008; Fombonne, 2009; Freeman, Roberts, & Daneman, 2005; Hendriksen & Vles, 2008; Kilincaslan & Mukaddes, 2009; Nordin & Gillberg, 1996; Steffenburg, Gillberg, & Steffenburg, 1996; Thome-Souza et al., 2004), but only two studies have focused explicitly on ID-adolescents with somatic chronic diseases (Nordin & Gillberg, 1996; Steffenburg et al., 1996). None of the aforementioned studies used instruments that were suitable for screening or diagnosing mild PDD behavior (Hartman et al., 2006).

Studies on the association between somatic chronic diseases in ID-adolescents and PDD behavior, in particular milder forms of PDD behavior, are highly needed. Professionals do not always recognize mild PDD behavior in (ID-)adolescents (de Bildt et al., 2005b; Kilincaslan & Mukaddes, 2009). Evidence on an association between somatic chronic diseases in ID-adolescents and mild PDD behavior may thus increase the attentiveness of professionals for mild PDD behavior, enabling earlier diagnosis and treatment. PDD behavior is very disabling in social and interpersonal situations and hinder successful participation in society (de Bildt et al., 2005a; Haccou & Hamond, 2006; Matson, Wilkins, Smith, & Ancona, 2008).

The aim of this study is to assess the association between somatic chronic diseases in ID-adolescents and PDD behavior, in particular the milder forms of PDD behavior.

2. Methods
2.1. Participants and procedure

We collected data on adolescents with a borderline, mild, moderate or severe ID aged 12–18 years in two provinces in the north of the Netherlands: Groningen and Drenthe (total population about 1.1 million people).

Nearly all adolescents of the target population attended secondary schools (schools for practical training) or special secondary schools (regional expertise centers). ID-adolescents attending schools for practical training can be classified as mainly educable and have IQs between 60 and 80. ID-adolescents attending regional expertise centers can be classified as mainly trainable and have IQs between 30 and 60 (Dekker, Koot, Van der Ende, & Verhulst, 2002). ID-adolescents not attending secondary schools, most of them with profound ID, were not included.

In the current school-based cross-sectional research project, 88% of the schools for practical training and regional expertise centers in both provinces participated. Non-participating schools did not differ from participating schools regarding urbanization of the catchment area and number of students. All parents of the 2156 adolescents aged 12–18 years received a questionnaire and a reminder when they did not respond. One thousand forty four parents returned the questionnaire (48.4%). Adolescents in the response and non-response group did not differ regarding age (t-test = 1.751, ns), but the response group had a higher proportion of girls ($\chi^2 = 5.9; p < 0.05$) and a higher proportion of adolescents with borderline or mild ID ($\chi^2 = 9.8; p < 0.05$). However, the effect sizes for both variables were negligible; Cohen’s $W$ were 0.06 and 0.07, respectively (Cohen, 1988).

The study protocol was approved by the Medical Ethics Committee of the University Medical Center Groningen, the Netherlands.

2.2. Measures
2.2.1. Intellectual disability

The target population had been officially classified as having ID by an independent committee established by the Dutch Ministry of Education, Culture and Science (Dutch Eurydice Unit, 2007). The classification of ID is based on a set of objective criteria, with the Dutch version of the Wechsler Intelligence Scale for Children–3rd Edition (Kort et al., 2002; Wechsler, 1991), and the Snijders-Oomen Nonverbal Intelligence Test-Revised (Snijders et al., 2003) as core ones.

2.2.2. Chronic diseases

Chronic diseases in ID-adolescents were measured by the National Permanent Survey on Living Conditions questionnaire (POLS); module health and labor, part chronic diseases in children (Statistics Netherlands, 2003). POLS part chronic diseases in children covers the most prevalent chronic diseases such as: ear, eye, skin diseases, diseases of the nervous, musculoskeletal, blood and circulatory, respiratory, digestive, and endocrine, nutritional and metabolic systems and ADHD. Questions were added about the presence of pervasive developmental disorders. Parents were asked to fill in the presence or absence of each specific chronic disease in the last 12 months for their child. Parents were also offered the possibility to mention the presence of chronic diseases that were not listed in the questionnaire. POLS was developed by Statistics Netherlands and is yearly used in a representative sample ($n \approx 10,000$) of the Dutch population (Otten & Winkels, 1998).
2.2.3. Pervasive development disorder behavior

PDD behavior was measured by the Dutch version of the Children’s Social Behavior Questionnaire (CSBQ) parent version. The CSBQ has 49 items describing a broad range of behavioral features, including milder forms that are typical of PDD. The items can be allocated to six subscales: “not optimally tuned to the social situation” (not tuned behavior; 11 items addressing emotional overreacting and stubbornness/disobedience), “reduced contact and social interest” (social withdrawn; 12 items), “orientation problems in time, place, or activity” (orientation problems; 8 items), “difficulties in understanding of social information” (not understanding; 7 items), “stereotyped behavior” (stereotyped behavior; 8 items), and “fear of and resistance to changes” (fear of changes; 3 items). Each item can be marked as “does not apply” to the child (score 0), “sometimes or somewhat applies” (score 1), or “clearly or often applies” (score 2) (de Bildt et al., 2005a; Hartman, Luteijn, Moorlag, de Bildt, & Minderaa, 2007; Hartman et al., 2006). Subscale scores can be computed by summing scores on the items; range 0–6 (fear of changes) to 0–24 (social withdrawn). Lower scores on the subscales reflect milder forms of PDD behavior, whereas higher scores reflect more severe forms of PDD behavior. A total score can also be calculated by summing the scores of the subscales (range 0–98). Although the CSBQ was originally developed for and investigated in children with normal intelligence, the psychometric qualities of the CSBQ in children with ID were found to be good (de Bildt et al., 2009; Luteijn, Luteijn, Jackson, Volkmar, & Minderaa, 2000).

2.2.4. Background characteristics

The questionnaire comprised questions on age, gender and school type of the adolescent.

2.3. Analysis

The CSBQ item scores were transformed to summative scores on the six subscales and a total CSBQ score, respectively. Subsequently, adolescents were categorized in a group without chronic diseases, with only somatic chronic diseases, with only PDD/ADHD, and with somatic chronic diseases in combination with PDD/ADHD. Adolescents with other psychiatric disorders (n = 102) were excluded from the analyses.

We tested via Univariate Analysis of Variance with Bonferroni post-hoc correction the mean differences between the group with somatic chronic diseases versus without chronic diseases and the mean differences between the group with somatic chronic diseases in combination with PDD/ADHD versus the group with PDD/ADHD. Both analyses were adjusted for level of ID (as measured by school type). For all mean differences, effect sizes (Cohen’s D) were calculated (Cohen, 1988).

3. Results

Table 1 shows the background characteristics of the adolescents. The gender ratio, 58.2% boys and 41.8% girls, was similar to the ratio boys and girls with ID in the Netherlands (van Schrojenstein Lantman-de Valk et al., 2002).

Tables 2 and 3 shows the mean scores and standard deviations of the four subgroups of ID-adolescents on all CSBQ scales. Table 2 shows that, adjusted for level of ID, ID-adolescents with somatic chronic diseases had statistically significant higher mean scores on all CSBQ scales compared to ID-adolescents without chronic diseases. According to Cohen’s criteria, all the effect sizes were small, except for those associated with the Orientation problem scale; medium effect size.

In addition, Table 3 shows that, adjusted for level of ID, ID-adolescents with comorbidity of somatic chronic diseases in combination with PDD/ADHD had statistically significant higher mean scores on five of the six CSBQ subscales and total CSBQ score compared to ID-adolescents with only PDD/ADHD. The effect sizes were small for all of the significant associations.

As shown in both Tables 2 and 3, all level of ID effects were statistically significant. This indicates that ID-adolescents with IQ between 30 and 59 had higher mean scores on CSBQ subscales and total CSBQ score compared to ID-adolescents with IQ between 60 and 80.

<table>
<thead>
<tr>
<th>Characteristics</th>
<th>Years</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (N = 1028)</td>
<td></td>
</tr>
<tr>
<td>Mean</td>
<td>15.4</td>
</tr>
<tr>
<td>SD</td>
<td>1.6</td>
</tr>
<tr>
<td>Range</td>
<td>12–18</td>
</tr>
<tr>
<td>Gender (N = 1035)</td>
<td></td>
</tr>
<tr>
<td>Boys</td>
<td>602 (58.2)</td>
</tr>
<tr>
<td>Girls</td>
<td>433 (41.8)</td>
</tr>
<tr>
<td>Level of ID* (N = 1038)</td>
<td></td>
</tr>
<tr>
<td>IQ 60–80</td>
<td>785 (75.6)</td>
</tr>
<tr>
<td>IQ 30–59</td>
<td>253 (24.4)</td>
</tr>
</tbody>
</table>

* As measured by school type.
4. Discussion

ID-adolescents with somatic chronic diseases have more PDD behavior than those without chronic diseases, in particular milder forms of PDD behavior. This association is independent of having PDD/ADHD or not. Our findings suggest a relationship between somatic chronic diseases in ID-adolescents and mild PDD behavior.

Differences are rather large, whereas effect sizes are mostly relative low. This can be explained by the fact that the differences in mean scores on the CSBQ scales between the groups as well as the deviations of the means within each group are considerably large, indicating considerable variation within each group.

4.1. Fit with other studies

To our knowledge, no previous studies have examined the association between somatic chronic diseases in ID-adolescents and the full range of pervasive developmental disorder behavior (PDD behavior), including milder forms of PDD behavior. However, our findings are mostly in line with two previous studies on ID-adolescents that used instruments suitable for diagnosing severe forms of PDD behavior and reported prevalence rates of PDD based on diagnostic classification (Nordin & Gillberg, 1996; Steffenburg et al., 1996). Both studies used the Autism Behavior Checklist (ABC) and Childhood Autism Rating Scale (CARS) and found higher prevalence rates of autistic spectrum disorder in ID-adolescent with epilepsy (38%) (Steffenburg et al., 1996), cerebral palsy (11%) (Nordin & Gillberg, 1996) and severe visual impairment (50%) (Nordin & Gillberg, 1996). Moreover, our findings are also mostly in line with studies on autism spectrum disorders and somatic chronic diseases among adolescents without ID and among a mixed group of adolescents with and without ID (Ekström et al., 2008; Freeman et al., 2005; Hendriksen & Vles, 2008; Kilincaslan & Mukaddes, 2009; Thome-Souza et al., 2004). These studies showed that adolescents with somatic chronic diseases had a greater chance on autism spectrum disorders compared to adolescents in the general population (Fombonne, 2009).

Our study thus shows that findings on severe cases of PDD can be extended to the full range of PDD behaviors.

4.2. Strength and limitations

The strength of this study is that it examined the association between somatic chronic diseases in adolescents with ID and PDD behavior in a large school-based sample representative for about 95% of the adolescents with ID. Another strength of

Table 2
Means and standard deviations on CSBQ scales and mean differences, adjusted for level of ID, and effect sizes between CSBQ scale scores in ID-adolescents without chronic diseases and ID-adolescents with somatic chronic diseases (SCD).

<table>
<thead>
<tr>
<th>Chronic diseases</th>
<th>None</th>
<th>SCD</th>
<th>MD*</th>
<th>p</th>
<th>95% CI</th>
<th>Cohen's d b</th>
</tr>
</thead>
<tbody>
<tr>
<td>CSBQ scales</td>
<td>n</td>
<td>M</td>
<td>SD</td>
<td>n</td>
<td>M</td>
<td>SD</td>
</tr>
<tr>
<td>Total score</td>
<td>392</td>
<td>15.27</td>
<td>12.89</td>
<td>233</td>
<td>21.95</td>
<td>14.66</td>
</tr>
<tr>
<td>Not tuned behavior</td>
<td>389</td>
<td>4.34</td>
<td>4.33</td>
<td>232</td>
<td>5.48</td>
<td>4.80</td>
</tr>
<tr>
<td>Social withdrawn</td>
<td>392</td>
<td>2.76</td>
<td>3.56</td>
<td>233</td>
<td>4.29</td>
<td>4.23</td>
</tr>
<tr>
<td>Orientation problems</td>
<td>383</td>
<td>2.65</td>
<td>2.81</td>
<td>232</td>
<td>4.18</td>
<td>3.40</td>
</tr>
<tr>
<td>Not understanding</td>
<td>391</td>
<td>3.89</td>
<td>3.23</td>
<td>231</td>
<td>5.22</td>
<td>3.45</td>
</tr>
<tr>
<td>Stereotyped behavior</td>
<td>389</td>
<td>0.88</td>
<td>1.71</td>
<td>231</td>
<td>1.59</td>
<td>2.19</td>
</tr>
<tr>
<td>Fear of changes</td>
<td>388</td>
<td>0.68</td>
<td>1.12</td>
<td>229</td>
<td>1.17</td>
<td>1.59</td>
</tr>
</tbody>
</table>

Note: CI, confidence interval; LL, lower limit; MD, mean differences; SCD, somatic chronic diseases; UL, upper limit.

* All level of ID effects p < 0.001.

b Cohen thresholds: negligible effect (<0.20); small effect (0.20 and <0.50); medium effect (0.50 and <0.80); large effect (≥0.80).

Table 3
Means and standard deviations on CSBQ scales and mean differences, adjusted for level of ID, and effect sizes between CSBQ scale scores in ID-adolescents with only PDD/ADHD and ID-adolescents with somatic chronic diseases (SCD) in combination with PDD/ADHD.

<table>
<thead>
<tr>
<th>Chronic diseases</th>
<th>PDD/ADHD</th>
<th>SCD + PDD/ADHD</th>
<th>MD*</th>
<th>p</th>
<th>95% CI</th>
<th>Cohen's d b</th>
</tr>
</thead>
<tbody>
<tr>
<td>CSBQ scales</td>
<td>n</td>
<td>M</td>
<td>SD</td>
<td>n</td>
<td>M</td>
<td>SD</td>
</tr>
<tr>
<td>Total score</td>
<td>163</td>
<td>33.82</td>
<td>16.60</td>
<td>144</td>
<td>41.03</td>
<td>16.62</td>
</tr>
<tr>
<td>Not tuned behavior</td>
<td>163</td>
<td>9.92</td>
<td>5.71</td>
<td>144</td>
<td>10.38</td>
<td>5.46</td>
</tr>
<tr>
<td>Social withdrawn</td>
<td>163</td>
<td>6.50</td>
<td>5.16</td>
<td>144</td>
<td>8.59</td>
<td>5.35</td>
</tr>
<tr>
<td>Orientation problems</td>
<td>163</td>
<td>5.73</td>
<td>3.47</td>
<td>144</td>
<td>7.32</td>
<td>3.72</td>
</tr>
<tr>
<td>Not understanding</td>
<td>161</td>
<td>7.34</td>
<td>3.70</td>
<td>144</td>
<td>8.44</td>
<td>3.35</td>
</tr>
<tr>
<td>Stereotyped behavior</td>
<td>162</td>
<td>2.45</td>
<td>3.06</td>
<td>143</td>
<td>3.73</td>
<td>3.83</td>
</tr>
<tr>
<td>Fear of changes</td>
<td>163</td>
<td>1.90</td>
<td>1.87</td>
<td>143</td>
<td>2.49</td>
<td>1.97</td>
</tr>
</tbody>
</table>

Note: CI, confidence interval; LL, lower limit; MD, mean differences; SCD, somatic chronic diseases; UL, upper limit

* All level of ID effects p < 0.001.

b Cohen thresholds: negligible effect (<0.20); small effect (0.20 and <0.50); medium effect (0.50 and <0.80); large effect (≥0.80).
this study was the use of a specifically developed screening instrument that covers the full range of PDD behavior, including milder forms. The CSBQ is useful for describing the severity and pattern of social deficits in groups other than PDD. A limitation is that the CSBQ is not intended for purposes of diagnostic classification (Hartman et al., 2006). Another limitation of the study was the relatively low response rate (48%), but non-response analyses revealed that adolescents in the response and non-response group did not differ on age, gender and educational level.

4.3. Implications for clinicians

Clinicians should be extra alert on PDD behavior, in particular the milder forms, in ID-adolescents when somatic chronic diseases are present. Detecting PDD behavior in ID-adolescents and adequate treatment of those with mild PDD behavior is important because these problems are very disabling for ID-adolescents in social and interpersonal situations and hinder successful participation in the society (de Bildt et al., 2005a; Haccou & Hamond, 2006; Matson et al., 2008). The CSBQ may be helpful as a first screening device when there is a suspicion of PDD behavior, even when the problems are associated with diagnoses outside autism spectrum disorders. In these cases, the score profile of the adolescent may help to plan for more specific diagnostic assessment and treatment, as well as their monitoring (Hartman et al., 2006).

4.4. Implications for research

Our study was the first to assess the association between somatic chronic diseases in ID-adolescents and mild PDD behavior. Therefore, they need confirmational studies, which preferable use both the CSBQ and standardized diagnostic instruments.

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