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ODD irritability is associated with obsessive–compulsive behavior and not ADHD in chronic tic disorders

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ABSTRACT

Gilles de la Tourette syndrome (TS) and chronic tic disorder (CT) are often associated with a variety of behavioral comorbidities including attention-deficit hyperactivity disorder (ADHD), obsessive–compulsive behavior (OCB), oppositional-defiant disorder (ODD) and temper outbursts. ODD is often associated with ADHD but its links to other symptoms of TS/CT is not as clear. This study examined whether the various symptoms of ODD were differentially linked to the various comorbidities in TS. A clinical sample of 135 children diagnosed with TS was evaluated through parent questionnaires and semi-structured interviews. Regressions and structural equation modeling confirmed that ODD is multidimensional in a TS/CT sample and showed that OCB was associated with the irritability symptoms of ODD whereas ADHD was associated with the Headstrong symptoms of ODD. Results suggest that increased attention to the different facets of ODD may help improve our understanding of emotional symptoms in TS/CT.

1. Introduction

Gilles de la Tourette syndrome (TS) and chronic tic disorder (CT) are characterized by repetitive and stereotyped tics with wax and wane in severity. In CT, either vocal or motor tics are present whereas in TS both types of tics are present. The prevalence of TS remains unclear partly because of symptom fluctuations but it is estimated that about 1–2% of the school age population is affected by TS and 3–6% if chronic tic disorders (CT) are included (Hornsey et al., 2001; Knight et al., 2012). In a majority of cases, TS is also associated with behavioral symptoms including obsessive–compulsive behavior (OCB), attention deficit hyperactivity disorder (ADHD) and temper outbursts (Spencer et al., 1998; Stephens and Sandor, 1999; Budman et al., 2000; Freeman et al., 2000; Kurlan et al., 2002; Bloch and Leckman, 2009; Cavanna et al., 2009; Grados and Mathews, 2009) while only about 8–25% of children with TS do not show any comorbidity (Freeman et al., 2000; Khalifa and von Knorring, 2005; Roessner et al., 2007; Robertson, 2012). Behavioral symptoms such as ADHD, OCB and temper outbursts are more related to the quality of life and adaptation of TS children than is the severity of tics (Bernard et al., 2003; Bloch and Leckman, 2009; Rizzo et al., 2012).

Oppositional defiant disorder (ODD) is also a frequent comorbidity in TS (Comings and Comings, 1987; Kurlan et al., 2002; Roessner et al., 2007; Robertson, 2012). ODD symptoms often have an impact on family functioning and social adaptation, especially in children with multiple symptoms such as TS children. ODD has often been considered as an ADHD-related comorbidity in TS, as a majority of TS children showing ODD also show ADHD (Spencer et al., 1998; Sukhodolsky et al., 2003; Roessner et al., 2007). However, some ODD symptoms show a clinical overlap with non-ADHD symptoms often present in TS such as irritability which is often observed in OCB.

There is evidence that some ODD symptoms are associated with emotional disorders such as mood disorders, anxiety disorders, OCB, and temper outbursts (Pierre et al., 1999; Stephens and Sandor, 1999; Burke et al., 2002; Greene et al., 2002; Maughan et al., 2004; Burke et al., 2005; Nock et al., 2007; Drabick et al., 2010; Sobanski et al., 2010). ODD symptoms can be an early indicator of risk of behavior problems, anxiety, or mood disorders (Speltz et al., 1999; Lavigne et al., 2001; Kim-Cohen et al., 2003; Burke et al., 2005; Boylan et al., 2007; Nock et al., 2007) and may

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contribute to the comorbidity between internalizing and externalizing symptoms (Burke et al., 2005). In TS, comorbid internalizing and externalizing symptoms are very frequent and thus ODD may also be associated with other symptoms than ADHD in this population.

Even though ODD symptoms are intercorrelated, ODD is increasingly viewed as a multifactorial disorder. Some ODD symptoms show a stronger association to emotional symptoms and others, a stronger association to ADHD (Stringaris and Goodman, 2009b; Burke et al., 2010; Rowe et al., 2010). In a community sample, ADHD was preferentially predicted by a portion of ODD symptoms (Headstrong component) whereas emotional disorders such as anxiety and depression were preferentially predicted by other ODD symptoms (Irritable component) and aggressive symptoms were predicted by a third set of ODD symptoms (Hurtful component) (Stringaris and Goodman, 2009b). In children with ADHD, the Irritable component of ODD was linked to emotional lability whereas the Headstrong component of ODD was not (Aebi et al., 2010). Also, there is evidence that the co-occurrence of ODD and generalized anxiety disorder (GAD) could not be explained by their joint co-occurrence with ADHD (Drabick et al., 2008).

Despite clear evidence for differential prediction of behavioral problems by different ODD symptoms, there have been several suggestions as to the number of interrelated components in ODD and which ODD symptoms are included in each component. For instance, Rowe et al. (2010) obtained a two-component model of ODD symptoms (Irritable and Headstrong), while Aebi et al. (2010) and Stringaris and Goodman (2009b) obtained three components. ODD-Irritable (temper outbursts, angriness, and touchiness) is identical in the three previous models, while the ODD-Headstrong and the ODD-Hurtful components vary. Rowe et al. (2010) include the five remaining ODD symptoms (argumentation, defiance, annoyance, blaming and spitefulness) in the ODD-Headstrong component whereas Stringaris and Goodman (2009b) and Aebi et al. (2010) combined these symptoms differently in ODD-Headstrong and ODD-Hurtful components. For Aebi et al. (2010), the criterion “annoys others” is included in the ODD-Hurtful component along with the spitefulness criteria, while for Stringaris and Goodman (2009b) the annoyance criterion is in the ODD-Headstrong component, leaving the ODD-Hurtful component with spitefulness as its only criterion.

A large majority of TS children in clinical samples show one or more behavioral comorbidities which have previously been linked to ODD. This makes this population particularly well suited to study ODD components and their links to other symptoms. In TS, ODD has traditionally been linked to ADHD only but the links between ODD and other symptoms need further investigation. The aim of the present study was to examine the factor structure of ODD symptoms in a clinical TS/CT sample and to explore the specific links between different ODD symptoms and behavioral comorbidities in TS/CT. We hypothesized that some components of ODD are preferentially linked to ADHD while others are preferentially linked to emotional symptoms like OCB.

2. Method

2.1. Participants

The present sample was composed of 135 children (118 boys) aged between 5 and 17 ($M=10.3$, S.D.$=2.6$) taking part in a large genetic study and diagnosed with TS ($N=129$) or CT ($N=6$) as defined by the Tourette Syndrome Classification Study Group (TSCSG, 1993): multiple motor tics and phonic tics (motor or phonic tics for CT) must be present at some time during the illness, although not necessarily concurrently; tics must occur many times a day, nearly every day, or intermittently throughout a period of more than 1 year; the anatomical location, number, frequency, type, complexity, or severity of tics must change over time; the onset must occur before the age of 21 years; involuntary movements and noises must not be explainable by other medical conditions; and tics must be witnessed directly or from video by a reliable examiner or concluded from clinical history. All affected individuals had four grand-parents of French Canadian descent. Families were recruited at the Tourette clinic of Sainte-Justine hospital. Patients (above 14 years) or their parents gave written and informed consent to the research coordinator which was not involved in their care. The research was approved by the institutional review board. Exclusion criteria were: a) inability to provide consent, b) a history of head injury or other neurological disorder which may cause tics, c) tics linked to drugs, d) a psychotic disorder, or e) a pervasive developmental disorder.

2.2. Measures

Parents first filled out questionnaires including the Parent Rating Scale (CPRS-L; Conners, 2003), as well as questionnaires on medical, obstetric, and developmental history for their children and for themselves. A clinical evaluation followed and consisted in multiple semi-structured interviews targeting tics using the Yale Global Tic Severity Scale (Y-GTSS; Leckman et al., 1989), as well as behavioral symptoms including OCB through the Yale-Brown Obsessive–Compulsive Scale (Y-BOCS; Goodman et al., 1989 and DSM-IV criteria; American Psychiatric Association, 2000), ADHD (DSM-IV criteria including age of onset, duration and interference; American Psychiatric Association, 2000) and ODD (DSM-IV criteria including duration and interference; American Psychiatric Association, 2000). Interviews were conducted by trained professionals including neurologists, neuropsychologists and psychiatrists, and all had previous experience with the Tourette syndrome population and with their specific evaluation. During interviews, professionals reviewed the questionnaires with the parents to ensure clarity and correspondence with the information shared during interviews. In interviews and questionnaires, parents were asked to consider the behavior of the child when no medication was taken.

The number of ODD symptoms was selected as the main measure of ODD because there is evidence that many children with fewer than the four criteria required for a clinical ODD diagnosis show significant functional impairment (Rowe et al., 2005). ODD criteria include: (1) loses temper, (2) argues with adults, (3) actively defies or refuses to comply with adults' requests, (4) deliberately does things that annoy other people, (5) blames others for his/her mistakes or misbehavior, (6) touchy or easily annoyed by others, (7) angry and resentful, (8) spiteful or vindictive. The main measure for ADHD was based on the number of DSM-IV criteria for ADHD met by the child. The criteria included inattention, hyperactivity and impulsivity related symptoms as detailed by the DSM-IV. If the participant showed 6 or more symptoms of either hyperactivity/impulsivity or inattention, a score of 3 was attributed (definite ADHD), for 5 symptoms a score of 2 was attributed (probable ADHD), for 4 symptoms a score of 1 was attributed (possible ADHD) and finally a score of 0 was attributed when less than 4 symptoms were documented (non-ADHD). As for OCB, the measure used was the total Y-BOCS score ranging from 0 to 40 since it is the most widely used measure of OCB severity (Deacon and Abramowitz, 2005). The Y-BOCS measure examines obsessions and compulsions which are presented as internalized symptoms (e.g., fear of harming others) as opposed to externalized or disruptive behaviors as in ODD. We selected this measure because TS/CT patients often have significant symptoms that may not meet the criteria for full-blown OCD on the level of distress and interference. Trained professionals evaluating OCB were aware of the array of tics of each patient before addressing OCB symptoms to ensure that the same symptoms were not counted as both tics and OCB. Compulsions were defined as clearly goal-directed behaviors to distinguish them from tics. The measures for tics, ODD, ADHD and OCB included in the analyses were all evaluations of the worst severity of symptoms in the patient's life.

2.3. Data analysis

The data were analyzed using the Statistical Package for Social Sciences (SPSS) 17.0. Pearson correlations were used to examine the associations between symptoms. Confirmatory factor analyses were used to determine the number of components which best accounted for the pattern of correlations between ODD criteria in our sample. Linear regressions were used to predict TS comorbidities with the two ODD components. The low multicollinearity assumption was respected since the Variance Inflation Factor (VIF) value was 1 for all regressions (less than 10) (Belseley et al., 1980). Some measures were statistically transformed to respect the normality assumption for linear regressions (i.e. OCB measure and ODD-Headstrong component). Structural equation models of the predictions of comorbidities by ODD components were tested using MXGui version 1.7.03. The following standard cut-offs were used as indications of an acceptable model: $RMR \leq 0.05$, $RMSEA \leq 0.06$ and $CFI \geq 0.95$ (Hu and Bentler, 1999).

3. Results

Table 1 shows clinical and demographic data for the sample. The proportion of boys was 87%. The sample reflected patients referred to a specialized TS clinic in that only 6.7% of the sample presented