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Original Article

Social Behavior and Comorbidity in Children With Tics

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ABSTRACT

OBJECTIVES: To examine the characteristics of children with coexisting tics and autism spectrum disorder and determine if children with tics have deficits in social behavior. **METHODS:** Descriptive study of children referred for tics over 18 months. Parents completed the Social Responsiveness Scale and the Social Communications Questionnaire; children screening positive on these measures were evaluated for autism spectrum disorder. Characteristics of children who were diagnosed with both disorders are described. Subscales scores on the Social Responsiveness Scale for children with tics without a comorbid autism spectrum disorder were compared. The relationship between a comorbid diagnosis of attention deficit hyperactivity disorder and autism spectrum disorder symptoms was explored using logistic and linear regression. **RESULTS:** One hundred and fourteen children were evaluated. Children with a tic disorder and autism spectrum disorder had significantly higher rates of comorbid attention deficit hyperactivity disorder ($P = 0.005$), rage attacks ($P = 0.006$), and oppositional defiant disorder ($P = 0.007$) than children without autism spectrum disorder. Mean tic severity and treatment rates did not differ between groups. Mean subscale scores on the Social Responsiveness Scale for children without autism spectrum disorders fell into the clinically significant range for autistic mannerisms only. All Social Responsiveness Scale scores were significantly increased by an attention deficit hyperactivity disorder diagnosis ($P < 0.0001$). **CONCLUSION:** Children referred for assessment of tics should be screened for autism spectrum disorders. There is a subgroup of children with multiple neuropsychiatric comorbidities who suffer from social dysfunction and autistic mannerisms outside of an autism spectrum disorder diagnosis.

Keywords: autism spectrum disorder, tic disorders, comorbidity, attention deficit hyperactivity disorder

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Introduction

Tourette syndrome (TS) is a childhood onset disorder that affects approximately 1%[1] of children. TS consists of multiple motor and one or more vocal tics that persist for more than 1 year.[2] The autism spectrum disorders (ASDs)—including autism, Asperger disorder, and pervasive developmental disorder not otherwise specified—are a group of disorders characterized by perturbed social reciprocity, communication, and stereotyped behaviours.[2] Recent epidemiological studies have shown that ASDs are much more common than previously believed, with a prevalence of approximately 1% in children and adolescents.[3,4]

ASDs and TS are distinct, complex, behaviorally defined disorders of childhood that have almost as many similarities as differences. Their etiologies are largely genetic. They have in common a predominance for the male gender, prominent movement disorders with repetitive motor behaviors, and a number of behavioral traits. A considerable number of individuals with each disorder have other lifelong developmental disorders as well, notably obsessive compulsive disorder, attention deficit hyperactivity disorder (ADHD), and learning disabilities.[5,6]

The underlying pathophysiology of TS and the ASDs is still unknown, but common to both disorders is an alteration of normal brain development and structure.

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Radiologic and pathological studies of patients with TS have confirmed subtle abnormalities in brain structure in these patients, particularly within the basal ganglia.[7,8] More widespread neuroanatomic abnormalities have been reported in patients with ASDs.[9]

The cooccurrence of ASDs and TS in the same individual has been reported at a frequency greater than expected by chance. Simonoff and colleagues[6] identified rates of comorbid psychiatric disorders associated with ASD in a subgroup of 112 10 to 14-year-old children in a population derived cohort. In this group, the 3-month point prevalence of TS was 4.8% and of chronic tic disorder was 9.0%. Currently, only one study has reported on the frequency of ASDs in children with TS. Using the Tourette Syndrome International Database Consortium Registry, Burd[10] identified 334 of 7288 participants (4.6%) in the registry whose clinicians reported had both TS and an ASD. Individuals in the registry, however, were not systematically assessed for both disorders.

Because of the difficulty in confirming the diagnosis of an ASD, several screening instruments have been created to aid clinicians. These measures are meant to help clinicians decide when enough symptoms of an ASD are present to warrant referral for further diagnostic investigation. Previous studies have shown that autistic traits are present in pediatric psychiatric outpatients. Reiersen[11] measured autistic traits in children with ADHD using the Social Responsiveness Scale (SRS). They found that mean SRS scores for children with ADHD were significantly higher than for children without ADHD ($P < 0.001$), with 32% of participants with ADHD scoring in the clinically significant range. Pine[12] compared scores on ASD symptom scales in healthy youths and youths with mood and anxiety disorders using the SRS, the Social Communication Questionnaire (SCQ) and the Children's Communication Checklist. Relative to healthy youths, youths with mood or anxiety disorders exhibited higher scores on each ASD symptom scale. Neither of these studies included follow-up clinical evaluation for ASDs of children scoring above cutoff points on screening questionnaires, so it is uncertain whether the high ASD symptom scale scores represent overlap between psychiatric and ASD symptoms or a true positive screening test result for an ASD.

This study had several objectives. We sought to identify if children with comorbid tic disorders and ASDs had more severe tics or a greater rate of treatment of tics and if there was a relationship between severity of ASD symptoms and tic severity. In children with tic disorders without comorbid ASDs, we examined if any specific deficits in social behaviors as measured using the Social Responsiveness Survey could be identified. Finally, we examined the relationship between comorbid ADHD symptom severity and the severity of ASD symptoms in children with tic disorders.

Methods

Approval for the research study was given by the University of Calgary Regional Ethics Board. This was a cross-sectional, descriptive study of all children referred for the assessment of tics during an 18-month period at a single center. To be eligible for the study, participants were ages 5-17 years and had a legal guardian able to provide informed consent and historical information for the child. Participants had a diagnosis of TS, chronic motor tic disorder, chronic vocal tic disorder, or tic disorder not otherwise specified. Patients of families who could not read or speak in English were excluded. All study participants were grouped into one of two groups: (1) children with a preexisting diagnosis of an autistic spectrum disorder or (2) children without a preexisting diagnosis of an ASD. Children in the first group were asked to supply confirmation of their diagnosis and subtype (by review of diagnostic consultation reports from psychologist and referring physician) and did not have further screening assessments for ASDs performed. They were included for the measurement of the rate of comorbid ASDs in children with TS.

Participants then had a semistructured interview developed by the study physician. The purpose of this interview was to confirm the diagnosis of a tic disorder, assess for common comorbidities such as ADHD and obsessive compulsive disorder, evaluate the child clinically for symptoms of ASD, and to rate symptom severity. This interview was performed by the study physician and included a diagnostic assessment for TS, ADHD, obsessive compulsive disorder, and ASD. The semistructured interview was based on Diagnostic and Statistical Manual-IV-text revision criteria for these disorders. Current diagnoses (e.g., TS only, TS plus ADHD) as determined by the diagnostic assessment were listed. Measurements of symptom severity were performed using the Yale Global Tic Severity Scale (YGTSS) as a measure of current tic severity, the Yale Brown Obsessive Compulsive Symptom Scale as a measure of obsessive compulsive disorder severity, and the Conner's Parent Short as a measure of ADHD symptom severity. Tics were differentiated from motor stereotypies based on core clinical features, including age of onset, phenomenology, and clinical course.

Parents of children who had not previously been diagnosed with an ASD were asked to complete the SRS and the SCQ, which are screening questionnaires for autistic behaviors. The SRS is a questionnaire covering the various dimensions of interpersonal behavior, communication, and repetitive/stereotypic behavior that are characteristic of ASD. The SRS yields five subscale scores: social awareness, social cognition, social communication, social motivation and autistic mannerisms, and a total score. T scores corresponding to raw scores are provided for each subscale score and the total score. Total T scores <59 are considered in the normal range. T scores between 60 and 75 indicate mild to moderate deficiencies in reciprocal social behavior, which are clinically significant and are typical for children with "high functioning" ASDs. T scores >76 represent severe deficiencies and are strongly associated with a clinical diagnosis of ASD. Validation of the SRS has been performed by comparison of SRS results with the Autism Diagnostic Interview-Revised (ADI-R).[13] The SCQ is a measure that taps the symptomatology associated with ASDs. The measure provides a total score that is interpreted with reference to a cutoff score that may indicate a possible ASD. The SCQ shows strong discrimination between ASD and non-ASD patients (sensitivity 0.88, specificity 0.72) and between autism and non-autism patients (sensitivity 0.90, specificity 0.86).[14]

Participants with T scores >76 on the SRS and above the cutoff score on the SCQ or for whom their was clinical suspicion of an ASD based on the clinical consultation were then seen by a psychologist who performed the ADI-R, a standardized diagnostic interview for autistic spectrum disorders for confirmation or refutation of an ASD diagnosis. Children with a positive ADI-R then had the Autism Diagnostic Observation Schedule (ADOS) performed by the same psychologist. The psychologist performing the ADI-R and ADOS holds a doctorate in psychology and is a registered psychologist with more than a decade of experience in the diagnosis of ASD. Based on the results of these examinations and clinical judgement of the psychologist and physician, an ASD diagnosis was determined.

Children with a confirmed diagnosis of an ASD were then grouped with those children previously diagnosed with an ASD for the measurement of comorbid ASDs in children with tic disorders. We compared the rate of comorbidity of ASDs in our tic disorder population to North American population data.[4] Clinical characteristics of children who were ultimately diagnosed with both a tic disorder and an ASD were described and summarized. Children who screened positive on the SRS and SCQ but did not have a confirmed ASD on the ADI-R and/or ADOS were also evaluated as a distinct subgroup to identify any unique clinical characteristics.

Subscales scores on the five domains of the SRS for the study population without a comorbid ASD are described using simple statistics. We