



Clinical Correlates of Parenting Stress in Children with Tourette Syndrome and in Typically Developing Children

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Objective To determine the impact of tic severity in children with Tourette syndrome on parenting stress and the impact of comorbid attention-deficit hyperactivity disorder (ADHD) and obsessive-compulsive disorder (OCD) symptomatology on parenting stress in both children with Tourette syndrome and typically developing children.

Study design Children with diagnosed Tourette syndrome (n = 74) and tic-free typically developing control subjects (n = 48) were enrolled in a cross-sectional study.

Results Parenting stress was greater in the group with Tourette syndrome than the typically developing group. Increased levels of parenting stress were related to increased ADHD symptomatology in both children with Tourette syndrome and typically developing children. Symptomatology of OCD was correlated with parenting stress in Tourette syndrome. Parenting stress was independent of tic severity in patients with Tourette syndrome.

Conclusions For parents of children with Tourette syndrome, parenting stress appears to be related to the child's ADHD and OCD comorbidity and not to the severity of the child's tic. Subthreshold ADHD symptomatology also appears to be related to parenting stress in parents of typically developing children. These findings demonstrate that ADHD symptomatology impacts parental stress both in children with and without a chronic tic disorder. (*J Pediatr* 2015;166:1297-302).

Tourette syndrome, a neurodevelopmental disorder characterized by unwanted motor and vocal tics, is relatively common in the pediatric population, affecting 1-10 in 1000 children.^{1,2} An overwhelming majority of children with Tourette syndrome (~90%) have at least 1 comorbid neuropsychiatric diagnosis,³ most commonly attention-deficit hyperactivity disorder (ADHD) and obsessive-compulsive disorder (OCD). Specifically, 50%-62% of children with Tourette syndrome also have ADHD and 20%-60% also have OCD.^{2,3} It has been proposed that the comorbidities associated with Tourette syndrome, rather than tic burden itself, may have a greater impact on quality of life for patients. Indeed, studies have shown that ADHD and OCD symptoms are highly negatively correlated with pediatric quality of life, but severity of the tic is not.^{4,5}

Beyond their impact on the child, neurodevelopmental disorders are known to have a negative impact on parents as well. Parents of children with neurodevelopmental disorders other than Tourette syndrome, such as neurofibromatosis type 1,⁶ fragile X syndrome,⁷ autism,⁸ and Down syndrome,⁹ report increased levels of stress as compared with parents of typically developing children. Parents of children with ADHD report greater levels of stress than parents of children with HIV infections or with asthma,¹⁰ suggesting either that neurodevelopmental disorders contribute to parental stress to a greater extent than other significant medical conditions or that having a child with a neurodevelopmental disorder is correlated with the tendency for increased stress.

Although there is increased caregiver burden in parents of children with Tourette syndrome,¹¹ to our knowledge, in no previous studies have authors investigated the impact of Tourette syndrome and its comorbid conditions on parents' level of stress. The authors of 1 study used mail-in parental report and found that parents of patients with Tourette syndrome and comorbid ADHD and/or OCD reported greater impact on the family (eg, family relationships, finances) than parents of patients with Tourette syndrome alone.¹² Parental stress and dimensional characterizations of comorbid symptomatology, however, were not measured. The goal of the present study was to examine the relationships between severity of tic in childhood Tourette syndrome, comorbid conditions, and parent stress. Our a priori hypotheses were that parent stress levels are: (1) increased in parents of children with Tourette syndrome compared

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ADHD	Attention-deficit hyperactivity disorder
CY-BOCS	Children's Yale-Brown Obsessive Compulsive Scale
OCD	Obsessive-compulsive disorder
PSI	Parenting Stress Index
SF	Short Form
WUMDC	Washington University School of Medicine Movement Disorder Center
YGTSS	Yale Global Tic Severity Scale

with parents of typically developing control subjects; (2) correlated with comorbid ADHD and OCD symptoms; and (3) independent of the severity of the tic, similar to previous results on patient quality of life.^{4,5} We also collected ADHD and OCD inventories in a tic-free population with no clinical diagnoses, allowing us to generalize the relationship between parent stress and ADHD/OCD symptom burden to typically developing children.

Methods

Seventy-four children with a clinical diagnosis of Tourette syndrome (mean age 11.8 years, range 6.7-15.8 years; 13 female) or chronic tic disorder (which involves either chronic motor or vocal tics but not both), together referred to as the Tourette syndrome group, and 48 tic-free unaffected children (mean age 11.6, range 8.1-15.3 years, 17 female) were included in the study. Diagnoses were determined by a pediatric neurology movement disorder specialist, pediatric neurologist, or pediatrician. A convenience sample of participants with Tourette syndrome was recruited through the Washington University School of Medicine Movement Disorder Center (WUMDC) and the local chapter of the Tourette Syndrome Association; some subjects were recruited via flyers posted in the community. All patients seen in the WUMDC who met inclusion criteria were offered participation, and those who were interested and consented participated in the study. Subjects were recruited for a neuroimaging study (neu-

roimaging data not presented here), and the present sample includes all subjects with available clinical data who met inclusion criteria. The presence of diagnosed comorbidities (primarily ADHD and/or OCD) and/or psychoactive medications were not considered exclusion criteria in the participants with Tourette syndrome but were considered exclusion criteria for the unaffected participants. All participants were additionally excluded for: (1) any medical, neurological, or psychiatric diagnosis (other than those commonly comorbid with Tourette syndrome in the participants with Tourette syndrome); (2) IQ less than 80; (3) abnormal findings on a neurological examination; (4) language dysfunction of any kind; (5) non-native English speaker or multilanguage fluency; (6) subnormal visual acuity not corrected by lenses; and (7) contraindications to magnetic resonance imaging (because of primary recruitment for neuroimaging). **Table I** shows participant demographic and clinical information. All children gave assent with parent or guardian consent. The Institutional Review Board at Washington University School of Medicine approved all studies.

Outcome Variable

Parenting stress was measured using the parent-reported Parenting Stress Index/Short Form (PSI/SF).¹³ The PSI/SF is a 36-item questionnaire, scored on a 5-point Likert scale from “strongly agree” to “strongly disagree.” The PSI/SF can be scored in 3 different domains: parenting distress, parent-child dysfunctional interaction, and difficult child.

Table I. Demographic and clinical characteristics

	Participants with Tourette syndrome (n = 74)	Typically developing participants (n = 48)	P value
Male/female (% male)	61/13 (82.4)	31/17 (64.6)	.008
Average age, y (range) SD	11.8 (6.7-15.8) 2.12	11.6 (8.1-15.3) 1.94	.65
Average IQ (range) SD	108 (89-133) 11.50	114 (86-139) 12.68	.01
Left-handed	5	0	
White/minorities (% white)	69/5 (93.2%)	41/7 (86.1%)	.16
African American	1	7	
Asian	1	0	
Hispanic	3	0	
Average household income, \$ (SD)	60 024 (22 113)	66 379 (30 683)	.19
Number taking medications, n (%)	39 (52.7%)*	0	
Centrally acting adrenergic agents	29	0	
Stimulants	18	0	
SSRI antidepressants	9	0	
Atypical neuroleptics	6	0	
Benzodiazepines	3	0	
Antiseizure medications	3	0	
Beta blockers	1	0	
Tetracyclic antidepressants	1	0	
Atomoxetine	1	0	
Number with comorbid diagnoses of number with known comorbidities (%)	33/57 (57.9%)†	0	
Comorbid ADHD	32	0	
Comorbid OCD	4	0	
Comorbid depression/anxiety	6	0	
ADHD, Conners' raw score (range) SD	14.61 (0-36) 9.03	4.77 (0-17) 4.55	<.001
OCD, CY-BOCS rating (range) SD	3.89 (0-18) 5.17	0.27 (0-5) 1.01	<.001
Tics, YGTSS rating (range) SD	16.92 (0-34) 7.61	0	.004
Parenting stress, Total PSI raw score (range) SD	69.77 (37-137) 22.27	58.44 (36-116) 17.50	
Number with PSI percentile ≥90th percentile	13 (17.6%)	2 (4.6%)	.03

NA, not available; SSRI, selective serotonin reuptake inhibitors.

*Twenty participants with Tourette syndrome were taking more than 1 medication.

†Eight participants with Tourette syndrome had more than 1 comorbid diagnosis, and comorbid diagnoses were missing for 17 participants with Tourette syndrome recruited from the community.

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