Published in final edited form as:

J Pediatr. 2015 May ; 166(5): 1297–1302.e3. doi:10.1016/j.jpeds.2015.01.041.

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

Stephanie B. Stewart, MD,

Department of Neurology, School of Medicine, Washington University in St Louis, St Louis, MO

Deanna J. Greene, PhD,

Department of Psychiatry, School of Medicine, Washington University in St Louis, St Louis, MO

Department of Radiology, School of Medicine, Washington University in St Louis, St Louis, MO

Christina N. Lessov-Schlaggar, PhD,

Department of Psychiatry, School of Medicine, Washington University in St Louis, St Louis, MO

Jessica A. Church, PhD, and

Department of Psychology, The University of Texas at Austin, Austin, TX

Bradley L. Schlaggar, MD PhD

Department of Neurology, School of Medicine, Washington University in St Louis, St Louis, MO

Department of Psychiatry, School of Medicine, Washington University in St Louis, St Louis, MO

Department of Radiology, School of Medicine, Washington University in St Louis, St Louis, MO

Department of Pediatrics, School of Medicine, Washington University in St Louis, St Louis, MO

Abstract

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 48 tic-free typically developing controls were enrolled in a cross-sectional study.

Results—Parenting stress was higher in the group with Tourette syndrome than the typically developing group. Higher levels of parenting stress were related to greater ADHD symptomatology in both children with Tourette syndrome and typically developing children. OCD

© 2015 Published by Elsevier Inc.

Corresponding Address: Stephanie B. Stewart, MD, Department of Neurology, 4525 Scott Ave, Washington University School of Medicine, St Louis, MO 63110, Phone (314) 362-3317, Fax (314) 362-2186.

Publisher's Disclaimer: This is a PDF file of an unedited manuscript that has been accepted for publication. As a service to our customers we are providing this early version of the manuscript. The manuscript will undergo copyediting, typesetting, and review of the resulting proof before it is published in its final citable form. Please note that during the production process errors may be discovered which could affect the content, and all legal disclaimers that apply to the journal pertain.

The authors declare no conflicts of interest.

symptomatology was correlated with parenting stress in Tourett syndrome. Parenting stress was independent of tic severity in patients with Tourette syndrome.

Conclusions—For parents of children with Tourett syndrome, parenting stress appears to be related to the child's ADHD and OCD comorbidity and not to the child's tic severity. 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.

Keywords

Attention-deficit disorder; obsessive compulsive disorder; tics

Tourette syndrome, a neurodevelopmental disorder characterized by unwanted motor and vocal tics, is relatively common in the pediatric population, affecting 1–10 in 1,000 children.^{1,2} An overwhelming majority of children with Tourett syndrome (~90%) have at least one comorbid neuropsychiatric diagnosis,³ most commonly attention deficit hyperactivity disorder (ADHD) and obsessive-compulsive disorder (OCD). Specifically, 50–62% of children with Tourett syndrome also have ADHD and 20–60% also have OCD.^{2,3} It has been proposed that the comorbidities associated with Tourett 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 tic severity is not.^{4,5}

Beyond affecting the child, neurodevelopmental disorders are known to have a negative impact on parents as well. Parents of children with neurodevelopmental disorders other than Tourett syndrome, such as neurofibromatosis type 1,⁶ fragile X syndrome,⁷ autism,⁸ and Down syndrome⁹ report increased levels of stress as compared with parents of typically developing children. Parents of children with ADHD report higher 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 TS,¹¹ to our knowledge no prior studies have investigated the impact of Tourett syndrome and its comorbid conditions on parents' level of stress. One study that used mail-in parental report found that parents of patients with Tourett syndrome and comorbid ADHD and/or OCD reported greater impact on the family (eg, family relationships, finances) than parents of patients with Tourett syndrome alone.¹² However, parental stress and dimensional characterizations of comorbid symptomatology were not measured. The goal of the present study was to examine the relationships between tic severity in childhood Tourett syndrome, comorbid conditions, and parent stress. Our *a priori* hypotheses were that parent stress levels are: (1) elevated for parents of children with Tourett syndrome as compared with parents of typically developing controls; (2) correlated with comorbid ADHD and OCD symptoms; and (3) independent of tic severity, 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 Tourett 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 Tourett 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 meeting inclusion criteria were offered participation, and those who were interested and consented participated in the study. Subjects were recruited for a neuroimaging study (neuroimaging data not presented here), and the present sample includes all subjects with available clinical data who met inclusion criteria. The presence of diagnosed co-morbidities (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 Tourett syndrome in the participants with Tourette syndrome); (2) intelligence quotient (IQ) less than 80; (3) abnormal findings on a neurological exam; (4) language dysfunction of any kind; (5) non-native English speaking or multi-language fluency; (6) subnormal visual acuity not corrected by lenses; and (7) contraindications to magnetic resonance imaging (due to 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 Parenting Stress Index/Short Form¹³ (PSI/SF). 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 three different domains: parenting distress (PD), parent-child dysfunctional interaction (PCDI), and difficult child (DC). The PD subscale measures parents’ level of distress resulting from personal (non-child) factors. The PCDI subscale measures parents’ degree of dissatisfaction with the interactions they have with their child. The DC subscale measures characteristics of the child, such as defiance and demandingness, which make parenting more difficult. Raw PSI/SF scores were used for descriptive statistics and t-scores were used for final analyses.

Covariates of Primary Interest

Investigators administered the Children’s Yale-Brown Obsessive Compulsive Scale¹⁴ (CY-BOCS) to all participants and the Yale Global Tic Severity Scale¹⁵ (YGTSS) to all patients

with Tourette syndrome. The CY-BOCS and YGTSS use both clinician-observation and report from the child and parent of the frequency, intensity, and interference of obsessive-compulsive symptoms and tics, respectively. Raw YGTSS scores were used in all analyses. Raw OCD scores were used in the correlations. Given the preponderance of zeros of CY-BOCS scores, OCD was coded as a three-level variable with recommended clinical cut-off scores of 0–3 corresponding to no/low anxiety, 4–15 corresponding to subclinical anxiety, and scores above 16 corresponding to clinical anxiety to be used for the regressions.^{16, 17}

To assess child ADHD symptoms, all parents completed the Conners' Parent Rating Scale - Revised: Short Form,¹⁸ a validated 27-item questionnaire, scored on a 4-point Likert scale from "not true at all" to "very much true." To allow for distinction between the subtypes of ADHD, it can also be scored in three subdomains: oppositional, cognitive problems/inattention, and hyperactivity, in addition to a composite score. Raw ADHD scores were used for descriptive statistics and t-scores were used for final analyses.

Other Covariates

All participants underwent a two subtests of the Wechsler Abbreviated Scale of Intelligence (Vocabulary and Matrix Reasoning) to assess general intelligence.¹⁹ To estimate household income (as a proxy for socio-economic status) for each participant, their home addresses were geocoded using ArcGIS 10 (ESRI Products, Redlands, CA) and overlaid onto median household income by census tract, obtained from the American Community Survey for 2010 (participants were studied between 2008–2010). Two participants with Tourette syndrome had PO box addresses and could not be geocoded. The total sample mean household income was substituted for the missing data. Because the scale of the household income variable was different from the rest of the variables, for the purpose of analysis, household income was standardized (z-score).

The sample was predominantly Caucasian (Table I) according to parental report. For the purpose of analysis, a single Asian subject was grouped with the Caucasian sample and the African American and Hispanic subjects were grouped together. Grouping was based on similarity in age, IQ, and family income. The result was a dichotomous ethnicity variable.

Medication use in the group with Tourette syndrome was dichotomously coded reflecting any medication use or no medications. Child age (continuous) and sex (dichotomous) were also included as covariates.

Statistical analyses

Descriptive statistics such as mean and prevalence estimates were conducted on raw scale scores. Group differences between participants with Tourette syndrome and typically developing controls were examined using chi-square (for categorical measures) and one-way analysis of variance (for continuous measures). Within the participants with Tourette syndrome, possible differences between subjects receiving treatment at WUMDC and those recruited from the community, as well as differences between medicated and unmedicated subjects were also examined. Pearson correlations for pairwise relationships between tic severity (raw score), OCD symptomatology (raw score), ADHD symptomatology (t-score),

and parent stress (t-score) were conducted separately in the Tourette syndrome and typically developing groups.

Multivariate linear regression models were used for hypothesis testing with the parenting stress variable as the outcome variable. Separate regression models were conducted for the participants with Tourette syndrome and the typically developing controls. All models included child age, sex, IQ, estimated household income, and ethnicity/race as covariates. For the participants with Tourette syndrome, medication use was included as an additional covariate. Possible non-linear effects of age were tested by including age squared as a confounding variable; however, such a non-linear effect of age was not statistically significant and was dropped from the final models. For both Tourette syndrome and control groups, ADHD and OCD symptomatology were the main independent variables of interest; in the Tourette syndrome group, tic severity was an additional covariate of interest.

Four pairs of participants with Tourette syndrome were siblings, and 11 pairs plus one set of three typically developing participants were siblings. Correlations were additionally calculated on a sample that included only one sibling out of each sibling set. Regression models were corrected for the correlated data in family members by using the Huber-White sandwich estimator for standard errors.^{20,21} Descriptive analyses and regression models were also carried out including only one child from each of the sets of siblings. Statistical analyses were performed using STATA (StataCorp LP, College Station, TX) and SPSS version 22 (SPSS, Inc., Chicago IL).

Results

The group with Tourette syndrome, as compared with typically developing controls, had slightly but significantly lower IQ, more males, more left-handed children, and greater severity of ADHD and OCD symptomatology (Table I). The group with Tourette syndrome also had significantly higher levels of parenting stress than the typically developing group (examined as a continuous score as well as at a clinically significant 90th percentile cutoff score, when patients should be referred for psychological services¹³). There were no significant group differences in age or estimated household income.

Among the participants with Tourette syndrome, there was no significant difference in demographic characteristics (sex, age, IQ, and estimated household income), ADHD, OCD, tic severity or parenting stress between the participants treated at WUMDC and those recruited from the community (Table II; available at www.jpeds.com). There was a higher rate of medication use in the participants treated at WUMDC. Comparing the participants with Tourette syndrome receiving psychoactive medication with the unmedicated participants, there were also no significance differences in demographic characteristics or parenting stress (Table III; available at www.jpeds.com). Medicated participants had greater severity of OCD and tics than unmedicated participants, but there was no significant difference in ADHD score. When comparing participants with Tourette syndrome specifically taking stimulants to those not taking stimulants, there was a greater severity of ADHD symptoms in the stimulant group ($p = .05$), as expected.

Bivariate correlations showed significant associations between parenting stress and ADHD measures in both the Tourett syndrome and typically developing groups, and between parenting stress and OCD in the Tourett syndrome group (Table IV). These results suggest that childhood neurodevelopmental problems are generally associated with parenting stress. In the typically developing group, there was also a significant correlation between parenting stress and OCD; however, the predominance of zero scores on the CY-BOCS makes it difficult to assess whether this association is meaningful. In the Tourett syndrome group, higher severity of ADHD was significantly associated with higher severity of OCD, but there was no significant relationship between these two measures in the typically developing group. In the group with Tourette syndrome, greater tic severity was significantly related to greater severity of both ADHD and OCD; the significant pairwise associations between tic, ADHD, and OCD severity in the patients with Tourette syndrome confirm the known comorbidity of these disorders. Results were similar when only one child from each set of siblings was included in the correlation (Table V; available at www.jpeds.com).

Analyses examining ADHD subscales showed that within the Tourett syndrome group, all ADHD subscales were correlated with parenting stress (Table VI; available at www.jpeds.com). In the typically developing group, oppositional and hyperactive subscales were correlated with parenting stress, but the cognitive problems/inattention subscale was not (Table VII; available at www.jpeds.com).

In the Tourett syndrome group, parenting stress was not significantly correlated with tic severity (Table IV). This result, in combination with the significant positive association of parenting stress with higher severity of ADHD and OCD, suggests that in patients with Tourette syndrome, comorbid disorders, but not tic severity *per se*, are associated with parenting stress. This latter point was confirmed in multivariate regression models in the Tourett syndrome group, as higher severity of ADHD and OCD independently contributed to higher levels of parenting stress, but tic severity was not significantly associated with parenting stress (Table VIII). In the typically developing controls, who do not have diagnoses of ADHD or OCD, subthreshold ADHD symptoms were also significantly and independently associated with higher levels of parenting stress (Table VIII), but subthreshold OCD symptoms were not, further suggesting some generalizability of parenting stress with childhood neurodevelopmental symptomatology. Interestingly in the typically developing group, but not the Tourett syndrome group, higher child IQ was significantly associated with higher parenting stress.

The importance of ADHD and OCD in the relationship with parenting stress was underscored by removing these covariates from the regression, which reduced the amount of variance explained. Removing ADHD from the covariate list reduced R-squared from 0.41 to 0.17 in the Tourett syndrome group and from 0.48 to 0.36 in the typically developing group. Removing OCD from the covariate list reduced the R-square from 0.41 to 0.37 in the Tourett syndrome group. OCD was not significant in the regression for the typically developing group.

Correlations with household income demonstrated that lower estimated household income was negatively associated with parenting stress in the Tourett syndrome group, but did not

reach statistical significance ($p = .08$) (Table XI; available at www.jpeds.com). In typically developing participants, household income was not correlated with parenting stress ($p = .29$).

Discussion

Parents of children with Tourett syndrome reported a higher level of parenting stress than parents of typically developing children, consistent with prior studies that found elevated parenting stress in other neurodevelopmental disorders.^{6,7,9,10} In addition, we found that parenting stress was strongly correlated with ADHD and OCD symptoms, independent of tic severity, for children with Tourett syndrome. Although previous studies have found that ADHD and OCD symptoms had a greater impact on the child than tic burden,^{4,5} our study extends that finding to impact on the parents and parent-child relationship. Further, we found that ADHD symptomatology correlated with parenting stress in typically developing children as well, demonstrating that this relationship extends to children without such diagnoses. In both children with Tourette syndrome and typically developing controls, the oppositional and hyperactive subscales were correlated with parenting stress but the inattentive/cognitive problems subscale was only related to parenting stress in the participants with Tourette syndrome. Further studies should investigate whether oppositional or hyperactive traits, independent of ADHD symptomatology, are associated with parental stress.

There are two clinical conclusions that can be drawn from this work. First, treating tics in isolation will likely not address the elevated parenting stress reported by parents of children with Tourett syndrome. Thus, it would be helpful for pediatricians, pediatric neurologists, pediatric psychiatrists, pediatric psychologists and other treating clinicians to address comorbid ADHD and OCD symptoms in addition to tic severity, even in children without formal comorbid diagnoses. Second, the negative impact of ADHD symptoms on parenting stress extends to a typically developing pediatric population. Even though there was an overall lower level of parenting stress in this group compared with the Tourett syndrome group, there was still a notable proportion (4%) with clinically significant parenting stress. Children with a diagnosis of ADHD were specifically excluded from our typically developing population, yet subclinical levels of ADHD symptoms still negatively influenced parenting stress. Thus, general pediatricians may want to inquire about dimensional ADHD symptoms; even when these symptoms do not reach clinical/diagnostic threshold, they still may negatively impact the parent-child relationship. In addition, physicians evaluating and managing pediatric patients may consider assessing parent stress clinically as a screening tool or as a measure of either pharmacological or behavioral treatment efficacy.

It is noteworthy that there was no difference in parenting stress between the medicated and unmedicated participants with Tourett syndrome. This finding has at least two possible explanations: the medications that alleviate Tourett syndrome, ADHD, and OCD symptoms may not alleviate parenting stress or parents of medicated children had even higher levels of stress prior to medication, and the effects of medication brought parenting stress down to the level observed in the parents of unmedicated children. As our data collection did not ascertain which specific symptoms the medications were prescribed to target or the extent to

which they were effective, it is difficult to determine the relationship between medication and parenting stress in the present study. It is also noteworthy that there was a negative relationship between parenting stress and estimated household income in the participants with Tourette syndrome at a trend level. Previous studies have also found that lower family income was significantly associated with increased parenting stress, both in parents of children with neural tube defects²² and in parents of children with a variety of chronic childhood diseases (cancer, asthma, type 1 diabetes or cystic fibrosis).²³

It should be noted that anxiety, OCD, and attention difficulties are highly heritable and that these traits may influence ratings of parenting stress. Parental reporting may not only reflect the impact of their child's behavior, but also the parents' own underlying anxiety and ADHD symptomatology. Further studies may benefit from collecting anxiety, ADHD, and OCD measures in parents as well as in the children under study.

Limitations of this study include a lack of formal comorbid diagnoses for 17 of the 74 children with Tourette syndrome. However, we collected dimensional ratings of ADHD and OCD symptoms for all children, and our findings indicate that this dimensionality is important. Additionally, the participants with Tourette syndrome in this study were largely seen in a tertiary regional referral center by a Tourette syndrome sub-specialist, and hence, may not be fully representative of the Tourette syndrome population, as tertiary referral centers may attract more complex cases with Tourette syndrome as compared with the general population with Tourette syndrome. However, comparing participants with Tourette syndrome receiving treatment at WUMDC with those recruited from the community demonstrated no differences in ADHD, OCD, tic severity, or parenting stress.

Our findings, together with previous studies,^{4,5} demonstrate that comorbid ADHD and OCD symptoms – even subthreshold of diagnostic criteria – affect children with Tourette syndrome and their parents more so than tics themselves. Thus, in many children treating tics alone may not be as effective in improving the well being of the child and caregivers as treating comorbid symptoms in addition to tics. Further, we extended these results to typically developing children, suggesting subclinical ADHD symptoms affect children and their caregivers more broadly.

Acknowledgments

We are grateful to Kelly McVey, Rebecca Lepore, Rebecca Coalson, Josh Siegel, and Mary Downey-Jones for their assistance on this project.

Funded by NINDS (F32NS065649 [to J.C.]), NIMH (R21 MH091512 [to B.S.]), and the Intellectual and Developmental Disabilities Research Center at Washington University (NIH/NICHD P30 HD062171).

List of Abbreviations

(ADHD)	Attention deficit hyperactivity disorder
(CY-BOCS)	Children's Yale-Brown Obsessive Compulsive Scale
(OCD)	Obsessive-compulsive disorder

(PSI/SF)	Parenting Stress Index/Short Form
(WUMDC)	Washington University School of Medicine Movement Disorder Center
(YGTSS)	Yale Global Tic Severity Scale

References

1. Kurlan R, McDermott MP, Deeley C, Como PG, Brower C, Eapen S, et al. Prevalence of tics in schoolchildren and association with placement in special education. *Neurology*. 2001; 57:1383–1388. [PubMed: 11673576]
2. CDC. Prevalence of diagnosed Tourette syndrome in persons aged 6–17 years -United States, 2007. *MMWR Morb Mortal Wkly Rep*. 2009; 58:581–585. [PubMed: 19498335]
3. Freeman RD, Fast DK, Burd L, Kerbeshian J, Robertson MM, Sandor P. An international perspective on Tourette syndrome: selected findings from 3,500 individuals in 22 countries. *Dev Med Child Neurol*. 2000; 42:436–447. [PubMed: 10972415]
4. Eddy CM, Cavanna AE, Gulisano M, Agodi A, Barchitta M, Cali P, et al. Clinical correlates of quality of life in Tourette syndrome. *Movement disorders : official journal of the Movement Disorder Society*. 2011; 26:735–738. [PubMed: 21506153]
5. Bernard BA, Stebbins GT, Siegel S, Schultz TM, Hays C, Morrissey MJ, et al. Determinants of quality of life in children with Gilles de la Tourette syndrome. *Movement disorders : official journal of the Movement Disorder Society*. 2009; 24:1070–1073. [PubMed: 19306279]
6. Esposito M, Marotta R, Roccella M, Gallai B, Parisi L, Lavano SM, et al. Pediatric neurofibromatosis 1 and parental stress: a multicenter study. *Neuropsychiatric disease and treatment*. 2014; 10:141–146. [PubMed: 24489471]
7. Johnston C, Hessel D, Blasey C, Eliez S, Erba H, Dyer-Friedman J, et al. Factors associated with parenting stress in mothers of children with fragile X syndrome. *Journal of developmental and behavioral pediatrics : JDBP*. 2003; 24:267–275. [PubMed: 12915799]
8. Davis NO, Carter AS. Parenting stress in mothers and fathers of toddlers with autism spectrum disorders: associations with child characteristics. *Journal of autism and developmental disorders*. 2008; 38:1278–1291. [PubMed: 18240012]
9. Sanders JL, Morgan SB. Family stress and adjustment as perceived by parents of children with autism or Down syndrome: Implications for intervention. *Child Fam Behav Ther*. 1997; 19:15–32.
10. Gupta VB. Comparison of parenting stress in different developmental disabilities. *J Dev Phys Disabil*. 2007; 19:417–425.
11. Cooper C, Robertson MM, Livingston G. Psychological morbidity and caregiver burden in parents of children with Tourette's disorder and psychiatric comorbidity. *Journal of the American Academy of Child and Adolescent Psychiatry*. 2003; 42:1370–1375. [PubMed: 14566175]
12. Wilkinson BJ, Newman MB, Shytle RD, Silver AA, Sanberg PR, Sheehan D. Family Impact of Tourette's Syndrome. *Journal of Child and Family Studies*. 2002; 10:477–483.
13. Abidin, RR. *The Parenting Stress Index Professional Manual*. Odessa, FL: Psychological Assessment Resources; 1995.
14. Scahill L, Riddle MA, McSwiggin-Hardin M, Ort SI, King RA, Goodman WK, et al. Children's Yale-Brown Obsessive Compulsive Scale: reliability and validity. *J Am Acad Child Adolesc Psychiatry*. 1997; 36:844–852. [PubMed: 9183141]
15. Leckman JF, Riddle MA, Hardin MT, Ort SI, Swartz KL, Stevenson J, et al. The Yale Global Tic Severity Scale: initial testing of a clinician-rated scale of tic severity. *Journal of the American Academy of Child and Adolescent Psychiatry*. 1989; 28:566–573. [PubMed: 2768151]
16. Janardhan Reddy YC, Srinath S, Prakash HM, Girimaji SC, Sheshadri SP, Khanna S, et al. A follow-up study of juvenile obsessive-compulsive disorder from India. *Acta Psychiatrica Scandinavica*. 2003; 107:457–464. [PubMed: 12752023]

17. Team. POTSP. Cognitive-Behavior Therapy, Sertraline, and Their Combination for Children and Adolescents With Obsessive-Compulsive Disorder. *JAMA : the journal of the American Medical Association*. 2004; 292:1969–1976.
18. Conners, CK. Conners' Rating Scales – Revised: Short Form. North Tonawanda, NY: Multi-Health Systems; 1997.
19. Wechsler, D. Wechsler abbreviated scale of intelligence. Third ed.. San Antonio: The Psychological Corporation; 1999.
20. White H. A Heteroskedasticity-Consistent Covariance-Matrix Estimator and a Direct Test for Heteroskedasticity. *Econometrica*. 1980; 48:817–838.
21. Huber P. The behavior of maximum likelihood estimates under nonstandard conditions. *Proceedings of the Fifth Berkeley Symposium on Mathematical Statistics and Probability*. 1967; 1:221–233.
22. Al-Akour NA, Khader YS, Hamlan A. Stress among parents of infants with neural tube defect and its associated factors. *International journal of nursing practice*. 2013; 19:149–155. [PubMed: 23577972]
23. Hullmann SE, Wolfe-Christensen C, Ryan JL, Fedele DA, Rambo PL, Chaney JM, et al. Parental overprotection, perceived child vulnerability, and parenting stress: a cross-illness comparison. *Journal of clinical psychology in medical settings*. 2010; 17:357–365. [PubMed: 21086027]

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 (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	
Caucasian/minorities (% cauc.)	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 (%)	39 (52.7%) ^a	0	
Centrally acting adrenergic agents	29	0	
Stimulants	18	0	
SSRI antidepressants	9	0	
Atypical neuroleptics	6	0	
Benzodiazepines	3	0	
Anti-seizure 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%) ^b	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	NA
Parenting Stress, Total PSI raw score (range) SD	69.77 (37–137) 22.27	58.44 (36–116) 17.50	.004
Number with PSI percentile 90 th percentile	13 (17.6%)	2 (4.6%)	.03

^a Twenty participants with Tourette syndrome were taking more than one medication.

^b Eight participants with Tourette syndrome had more than one comorbid diagnosis and comorbid diagnoses were missing for 17 participants with Tourette syndrome recruited from the community.

Table II

Demographic and Clinical Characteristics of Tourette Syndrome Participants Treated at an Academic Medical Center and in the Community

	Treated at WUMDC (n=57)	Treated in the community (n=17)	P value
Males/females (% Male)	48/9 (84.2%)	15/2 (88.2%)	.17
Age (SD)	11.98 (2.23)	11.24 (1.65)	.20
IQ (SD)	109 (11.95)	107 (9.99)	.50
Income (SD)	60110 (22702)	59743 (20741)	.95
On medication/unmedicated (% on meds)	34/23 (59.6%)	4/13 (23.5%)	.009
ADHD raw score (SD)	14.68 (8.88)	14.35 (9.80)	.90
OCD rating (SD)	3.904 (4.91)	3.824 (6.13)	.96
Tics rating (SD)	17.54 (7.86)	14.82 (6.47)	.20
Parenting Stress raw score (SD)	67.89 (21.31)	76.06 (24.87)	.19

Table III

Demographic and Clinical Characteristics of Tourette syndrome Participants Receiving and Not Receiving Medication

	Receiving medication (n=38)	Unmedicated (n=36)	P value
Males/females (% Male)	31/7	32/4	.78
Age (SD)	12.08 (2.29)	11.53 (1.91)	.26
IQ (SD)	108 (11.99)	108 (11.12)	.91
Income (SD)	62818 (21298)	57229 (22851)	.29
ADHD raw score (SD)	15.71 (8.13)	13.44 (9.88)	.28
OCD rating (SD)	5.237 (5.76)	2.458 (4.06)	.02
Tics rating (SD)	18.82 (8.41)	14.92 (6.17)	.03
Parenting Stress raw score (SD)	69.84 (21.30)	69.69 (23.56)	.98

Table IV

Correlation Coefficients between Parenting Stress and ADHD, OCD, Tic Severities Measures

Correlation between	Participants with Tourette syndrome (n=74)		Typically developing participants (n=48)	
	Pearson <i>r</i>	<i>P</i> value	Pearson <i>r</i>	<i>P</i> value
ADHD-PSI	0.571	<.001	0.489	<.001
OCD-PSI	0.395	<.001	0.401	.005
Tics-PSI	0.176	.13	NA	NA
ADHD-OCD	0.316	.006	0.093	.53
ADHD-Tics	0.324	.005	NA	NA
OCD-Tics	0.356	.002	NA	NA

Table V

Correlation Coefficients between Parenting Stress and ADHD, OCD, Tic Severities Measures with Only One of Each of the Siblings

Correlation between	Participants with Tourette syndrome (n=70)		Typically developing participants (n=36)	
	Pearson <i>r</i>	<i>P</i> value	Pearson <i>r</i>	<i>P</i> value
ADHD-PSI	0.582	<.001	0.677	<.001
OCD-PSI	0.401	.001	0.410	.01
Tics-PSI	0.196	.10	NA	NA
ADHD-OCD	0.293	.01	0.106	.54
ADHD-Tics	0.311	.009	NA	NA
OCD-Tics	0.331	.005	NA	NA

Subscale Correlation Coefficients Between Parenting Stress, ADHD, OCD and Tic Severities Measures in Participants with Tourette Syndrome

Table VI

	ADHD- OPP	ADHD- INATT	ADHD- HYPER	PSI-PD	PSI- PCDI	PSI-DC	OCD	Tics
ADHD- OPP	<i>r</i>	0.566 <.001	0.627 <.001	0.290 .01	0.645 <.001	0.704 <.001	0.362 .002	0.308 .008
	<i>P</i> value							
ADHD- INATT	<i>r</i>	0.566 <.001	0.458 <.001	0.245 0.04	0.486 <.001	0.544 <.001	0.292 .01	0.145 .21
	<i>P</i> value							
ADHD- HYPER	<i>r</i>	0.627 <.001	0.458 <.001	0.308 .008	0.307 .008	0.401 <.001	0.275 .01	0.429 <.001
	<i>P</i> value							
PSI-PD	<i>r</i>	0.290 .01	0.245 .04	0.308 .008	0.435 <.001	0.503 <.001	0.366 .001	0.114 .33
	<i>P</i> value							
PSI- PCDI	<i>r</i>	0.645 <.001	0.486 <.001	0.307 .008	0.435 <.001	0.782 <.001	0.306 .008	0.152 .20
	<i>P</i> value							
PSI-DC	<i>r</i>	0.704 <.001	0.544 <.001	0.410 <.001	0.503 <.001	0.782 <.001	0.380 .001	0.142 .23
	<i>P</i> value							
OCD	<i>r</i>	0.362 .002	0.292 .01	0.275 .01	0.366 .001	0.306 .008	0.380 .001	0.356 .002
	<i>P</i> value							
Tics	<i>r</i>	0.308 .008	0.145 .22	0.429 <.001	0.114 0.33	0.152 0.20	0.142 0.23	0.356 0.002
	<i>P</i> value							

Abbreviations: ADHD, attention deficit disorder; ADHD-HYPER, Conners' Hyperactivity subscale; ADHD-INATT, Conners' Cognitive Problems/Inattention subscale; ADHD-OPP, Conners' Oppositional subscale; OCD, obsessive-compulsive disorder;; PSI-DC, Parenting Stress Index- Difficult Child; PSI-PCDI, Parenting Stress Index- Parent-Child Dysfunctional Interaction; PSI-PD, Parenting Stress Index- Parental Distress+

Table VII

Subscale Correlation Coefficients Between Parenting Stress, ADHD, and OCD Severities Measures in Typically Developing Participants

		ADHD- OPP	ADHD- INATT	ADHD- HYPER	PSI-PD	PSI-PCDI	PSI-DC	OCD
ADHD- OPP	<i>r</i>		0.221	0.620	0.534	0.536	0.600	0.304
	<i>P</i> value		.13	<.001	<.001	<.001	<.001	.04
ADHD- INATT	<i>r</i>	0.221		0.217	0.146	0.196	0.088	0.032
	<i>P</i> value	.13		.14	.32	.18	.55	.83
ADHD- HYPER	<i>r</i>	0.620	0.217		0.331	0.421	0.326	0.292
	<i>P</i> value	<.001	.14		.02	.003	.02	.004
PSI-PD	<i>r</i>	0.534	0.146	0.331		0.852	0.686	0.305
	<i>P</i> value	<.001	.32	.02		<.001	<.001	.04
PSI- PCDI	<i>r</i>	0.536	0.196	0.421	0.852		0.722	0.349
	<i>P</i> value	<.001	.18	.003	<.001		<.001	.02
PSI-DC	<i>r</i>	0.600	0.088	0.326	0.686	0.722		0.289
	<i>P</i> value	<.001	.55	.02	<.001	<.001		.05
OCD	<i>r</i>	0.304	0.032	0.292	0.305	0.349	0.289	
	<i>P</i> value	.04	.83	.004	.04	.02	.05	

Abbreviations: ADHD, attention deficit disorder; ADHD-HYPER, Conners' Hyperactivity subscale; ADHD-INATT, Conners' Cognitive Problems/Inattention subscale; ADHD-OPP, Conners' Oppositional subscale; OCD, obsessive-compulsive disorder;; PSI-DC, Parenting Stress Index- Difficult Child; PSI-PCDI, Parenting Stress Index- Parent-Child Dysfunctional Interaction; PSI-PD, Parenting Stress Index- Parental Distress

Table VIII

Association of Clinical and Demographic Variables with Parenting Stress

Independent Variable	Participants with Tourette syndrome (n=74)			Typically developing participants (n=48)		
	Regression Coefficients	T	P value	Regression Coefficients	T	P value
ADHD	0.718	5.13	<.001	0.958	2.69	.01
OCD	5.595	2.00	.05	15.54	1.18	.25
Tics	0.037	0.15	.88	NA	NA	NA
Medication	-3.615	-1.11	.27	NA	NA	NA
Age	1.057	1.65	.10	-0.339	-0.57	.57
IQ	0.201	1.43	.16	0.263	2.15	.04
Sex	0.684	0.20	.84	0.428	0.14	.89
Ethnicity	-3.196	-0.49	.62	9.254	1.53	.13
Income	-1.252	-0.64	.52	-1.072	-0.86	.40

Table IX

Correlation Coefficients between Income and Parenting Stress, ADHD, OCD, Tic Severities Measures in TS and Typically Developing Participants

Correlation between	Participants with Tourette syndrome (n=74)		Typically developing participants (n=48)	
	Pearson <i>r</i>	<i>P</i> value	Pearson <i>r</i>	<i>P</i> value
ADHD-Income	−0.247	.04	−0.345	.02
OCD-Income	−0.072	.55	0.137	.35
Tics-Income	−0.211	.08	NA	NA
PSI-Income	−0.209	.08	−0.156	.29