Mother-Child Agreement on Behavioral Ratings in Tourette Syndrome: A Controlled Study

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Abstract

In Tourette syndrome, motor and phonic tics are associated with a spectrum of psychiatric disorders. As proxy report instruments are commonly used to assess children with Tourette syndrome, we investigated the relationship between child and mother ratings of behavioral problems. We enrolled 28 children with Tourette syndrome (25 males; mean age, 13.9 years) and 61 gender- and age-matched healthy controls (55 males; mean age, 14.7 years). Clinicians completed measures of tic severity, and all children completed the Youth Self-Report version of the Child Behavior Checklist, while their mothers completed the Child Behavior Checklist. In the clinical group, Youth Self-Report scores were significantly lower than mothers' Child Behavior Checklist scores across the majority of subscales (especially affect and somatization). In contrast, for the control group, mother and child ratings only differed for the externalizing behavior subscales. Clinicians should be aware of these differences between self and mother ratings for specific behavioral problems in Tourette syndrome.

Keywords

Tourette syndrome, tics, behavior, ratings, parent, ADHD, obsessive-compulsive

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Tourette syndrome is a neurodevelopmental condition characterized by multiple motor tics and at least 1 phonic tic.¹ Although prevalence rates show wide variability, most studies suggest a prevalence in youngsters aged 5 to 18 years of approximately 1%, with a male:female ratio of 3:1.² Tourette syndrome is a complex disorder with a wide spectrum of behavioral problems in 90% of cases, ranging from complex ticlike symptoms (self-injurious behaviors; nonobscene, socially inappropriate behaviors; coprophenomena, echophenomena, and paliphenomena) to symptoms of attention-deficit hyperactivity disorder (ADHD), obsessive-compulsive disorder, mood disorders, impulse control disorders, and personality disorders.³⁻⁵ The presence of behavioral comorbidities has been shown to be associated with social difficulties and impulse dyscontrol in young patients and can have a detrimental impact on quality of life.⁶ It is therefore essential to obtain accurate information about both tic symptoms and related behavioral problems as part of the routine clinical assessment in Tourette syndrome.

Expert neuropsychiatric examination is usually complemented by standardized psychometric instruments to evaluate young patients' behavioral and emotional problems. In particular, both self-report and proxy report instruments are routinely used to assist the specialist in the clinical assessment of the Tourette syndrome spectrum.⁷ The Child Behavior Checklist 6-18 is a widely used psychometric instrument for the screening of behavioral problems, which, once identified, need to be investigated in greater depth using more refined clinical instruments, such as structured interviews and projective tests.⁸ There is a self-report version of the Child Behavior Checklist, the Youth Self-Report, which is also used as a

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screening tool for behavioral problems. Both the Child Behavior Checklist and the Youth Self-Report show good reliability and validity, as demonstrated by previous research.^{9,10} However, recent studies suggested that self-report instruments can have limited usefulness in assisting the assessment of behavioral symptoms of young patients with Tourette syndrome¹¹ and that parents' evaluation of behavioral and emotional problems in Tourette syndrome is more objective than young patients'.⁸ In this case control study, we compared parent reports (Child Behavior Checklist) to self-reports (Youth Self-Report) when assessing behavioral problems in both young patients with Tourette syndrome and healthy children.

Methods

All consecutive outpatients seen over the period from 2006 to 2008 at the specialist movement disorder clinics of the Child Neuropsychiatry Units at the University of Insubria in Varese, Italy, and the University of Pavia in Pavia, Italy, were invited to participate. The research protocol was approved by the local ethics committee, and written informed consent was obtained from each participant prior to enrollment.

We recruited 28 young patients meeting the *Diagnostic and Statistical Manual for Mental Disorders, Fourth Edition, Text Revision* criteria for Tourette syndrome. All patients underwent neurological examination, neurophysiology (electroencephalography) and neuroradiology (brain magnetic resonance imaging) investigations, and standard laboratory tests (cupremia, ceruloplasmin, cupriuria, erythrocyte sedimentation rate, anti–streptolysin O antibodies, acanthocytes, antinuclear antibody test) to rule out secondary tics and other neurological disorders. At cognitive assessment, all participants showed an intelligence quotient >70.

All patients were clinically evaluated by a neuropsychiatrist with substantial experience in Tourette syndrome, who collected a comprehensive medical and family history and reviewed comorbid diagnoses, such as ADHD and obsessive-compulsive disorder. Clinical assessment involved the Schedule for Affective Disorders and Schizophrenia for School Age Children, a semistructured diagnostic interview designed to assess current and past psychopathology in children and adolescents according to the *Diagnostic and Statistical Manual of Mental Disorders, Third Edition, Revised* and *Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition* criteria.¹² Tic severity was assessed using the Yale Global Tic Severity Scale.¹³ Severity of obsessive-compulsive symptoms was assessed in patients with comorbid obsessive-compulsive disorder using the Children's Yale-Brown Obsessive Compulsive Scale.¹⁴

Sixty-one age- and sex-matched healthy controls were recruited from among 247 children from local schools. None had a personal or family neuropsychiatric history.

The mothers of all participants completed the Child Behavior Checklist, a parent-reported 118-item questionnaire for young people aged 6 to 18 years, which assesses the child's emotional and behavioral functioning by rating both internalizing (anxiety, depression, withdrawal, and somatic complaints) and externalizing (including thought problems, attention problems, rule breaking, and aggressiveness) behaviors. Separate ratings assess competencies (social and school functioning) and social, thought, and attention problems.¹⁵ All participants completed the Youth Self-Report, a self-reported questionnaire derived from the Child Behavior Checklist and designed for use with adolescents aged 12 to 18 years. The Youth Self-Report comprises 2 subscales: (1) a 20-item scale to measure the child's participation in school and social and leisure activities (eg, sports, chores, etc), and (2) a 112-item scale to assess internalizing and externalizing behaviors as the Child Behavior Checklist.¹⁶

Ratings obtained from patients with Tourette syndrome and mothers were compared with similar data obtained from healthy controls. Further analyses compared Child Behavior Checklist scores with Youth Self-Report scores for both Tourette syndrome and healthy control groups. Statistical analyses were performed using the PASW (SPSS) Statistics version 17.0.3 (IBM, New York, USA) for Macintosh. χ^2 analysis was used for categorical data, whereas continuous variables were assessed with parametric (Student *t*) and nonparametric (Mann-Whitney *U* and Wilcoxon) tests. *P* values <.05 were considered statistically significant.

Results

Demographic and clinical characteristics are summarized in Table 1. Participants' ages ranged between 12 and 16 years for both groups. The mean age at tic onset was 7.0 \pm 2.0 years. First tics were simple motor tics (n = 7), complex motor tics (n = 4), simple phonic tics (n = 1), and mixed motor/phonic tics (n = 16). The mean Yale Global Tic Severity Scale total score was 36.5 ± 20.54 (mean motor score, 11.6 ± 3.7 ; mean phonic score, 6.0 ± 5.2 ; mean impairment score, 18.9 ± 14.2), indicating moderate tic severity. The mean Children's Yale-Brown Obsessive Compulsive Scale score for the subgroup of patients with Tourette syndrome plus obsessive-compulsive disorder was 5.6 ± 7.4 , indicating mild obsessive-compulsive symptoms.

Table 1 shows the comparison between behavioral ratings (Child Behavior Checklist and Youth Self-Report) for the Tourette syndrome and healthy control groups, with significantly higher mothers' ratings (Child Behavior Checklist) for the Tourette syndrome group versus healthy controls for all subscales except withdrawal and somatic complaints and for overall competence, although the competence subscales did not reach significance. In contrast, the only significant differences for Youth Self-Report scores indicated more thought and attention problems in Tourette syndrome and lower competence overall, including the social subscale.

Table 2 shows comparisons between mother and child reports within each of the 2 groups. For the Tourette syndrome group, there were many differences including higher ratings given by mothers across all subscales (including competence), except withdrawal, delinquent, and aggressive behavior subscales. For healthy controls, there were differences for all competence subscales as seen with Tourette syndrome. However, there were fewer differences seen for behavioral scales. Mothers gave lower ratings for externalizing subscales (including delinquent and aggressive behavior).

Discussion

While previous studies have compared parent and child agreement on the Child Behavior Checklist and Youth Self-Report in

Characteristics	Patients with Tourette syndrome (n $=$ 28)	Healthy controls $(n = 61)$	P .898
Male gender, n (%)	25 (89.3)	55 (90.2)	
Age, mean \pm SD, y	13.9 ± 1.9	14.7 ± 1.8	.053
Presence of OCD, n (%)	7 (25)	0 (0)	<.001
Presence of ADHD, n (%)	9 (32.1)	0 (0)	<.001
Presence of ADHD $+$ OCD, n (%)	5 (21.4)	0 (0)	<.001
Presence of depression, n (%)	3 (3.6)	0 (0)	<.001
Presence of anxiety, n (%)	6 (21.4)	0 (0)	<.001
Pregnancy problems, n (%)	4 (15.4)	0 (0)	.025
Prematurity, n (%)	3 (11.1)	2 (5.4)	.211
Dystocic delivery, n (%)	9 (33.3)	0 (0)	<.001
Postnatal problems, n (%)	6 (23.1)	I (2.7)	.017
CBCL problem scale			
Total score, mean \pm SD	61.0 <u>+</u> 8.6	49.1 <u>+</u> 8.9	<.001
Internalizing, mean \pm SD	58.1 <u>+</u> 9.7	51.9 <u>+</u> 9.4	.005
Withdrawn, mean \pm SD	56.7 <u>+</u> 8.7	54.7 <u>+</u> 6.8	.292
Somatic complaints, mean \pm SD	57.7 <u>+</u> 7.6	56.0 <u>+</u> 7.5	.178
Anxious/depressed, mean \pm SD	61.0 <u>+</u> 10.1	54.0 <u>+</u> 5.1	.00 ا
Externalizing, mean \pm SD	56.5 <u>+</u> 9.1	47.2 <u>+</u> 8.1	<.001
Delinquent behavior, mean \pm SD	54.5 ± 5.1	52.0 <u>+</u> 3.6	.006
Aggressive behavior, mean \pm SD	57.0 <u>+</u> 6.1	52.1 <u>+</u> 4.0	<.001
Social problems, mean \pm SD	60.0 ± 7.3	54.3 <u>+</u> 6.5	<.001
Thought problems, mean \pm SD	61.7 ± 9.2	51.9 ± 5.0	<.001
Attention problems, mean \pm SD	67.5 <u>+</u> 9.4	54.1 <u>+</u> 4.8	<.001
CBCL competence scales			
Total, mean \pm SD	34.7 <u>+</u> 8.1	39.5 <u>+</u> 7.8	.011
Activities, mean \pm SD	34.6 ± 8.8	36.3 <u>+</u> 9.4	.423
Social, mean \pm SD	4I.2 ± 8.1	43.1 ± 7.2	.273
YSR problem scales			
Total, mean \pm SD	53.4 ± 11.6	50.4 ± 10.5	.229
Internalizing, mean \pm SD	50.1 ± 12.1	50.1 ± 11.1	.982
Withdrawn, mean \pm SD	54.4 ± 6.4	53.5 ± 6.0	.748
Somatic complaints, mean \pm SD	53.1 ± 4.8	54.5 ± 6.8	.399
Anxious/depressed, mean \pm SD	55.8 ± 9.4	54.3 ± 6.2	.895
Externalizing, mean \pm SD	52.3 ± 9.5	52.2 ± 9.6	.968
Delinquent behavior, mean \pm SD	54.6 ± 5.1	55.7 ± 7.6	.901
Aggressive behavior, mean \pm SD	55.2 ± 6.9	54.9 ± 7.0	.676
Social problems, mean \pm SD	56.4 ± 6.8	53.8 ± 6.3	.055
Thought problems, mean \pm SD	53.0 ± 4.6	52.2 ± 6.2	.027
Attention problems, mean \pm SD	61.2 ± 10.1	54.0 ± 6.2	.00
YSR competence scales	12.0 1 10.2	40.0 1 0.2	000
Total, mean \pm SD	42.9 ± 10.2	49.9 ± 9.3	.002
Activities, mean \pm SD	40.7 ± 11.7	46.1 ± 7.5	.050
Social, mean \pm SD	46.0 ± 7.2	48.8 <u>+</u> 7.2	.033

Abbreviations: CBCL, Child Behavior Checklist 6-18; YSR, Youth Self-Report; SD, standard deviation; OCD, obsessive-compulsive disorder; ADHD, attentiondeficit hyperactivity disorder.

clinical and nonclinical samples,¹⁷⁻²³ this may be the first study to do so in children with Tourette syndrome. Importantly, our findings showed that young patients with Tourette syndrome tend to rate themselves (Youth Self-Report) as significantly less symptomatic than their mother (Child Behavior Checklist) on internalizing, externalizing, and total problem scales. This is consistent with previous literature.^{18,24,25}

Our findings may highlight a tendency for mothers' ratings to overestimate both internalizing and especially externalizing problems, as suggested by previous studies.^{11,18,26} One explanation for this finding is that parents' perception of their children's behavior is different to children's own self-concepts.²⁷ Indeed, previous studies found that the stress experienced by parents of young children with chronic illness appears to relate to their perception of their children's behavior problems.²⁸ Moreover, Hauser-Cram et al²⁹ showed that the children's type of disability also predicted changes in maternal (but not paternal) child-related and parent-related stress. Finally, parents perceived tics and tic-related symptoms (eg, compulsions and anger attacks) as voluntary and defiant, which may affect their identification of these behaviors as symptoms.³⁰

The mothers of both healthy controls and children with Tourette syndrome gave significantly lower competence ratings than their children, perhaps highlighting a consistent

	Tourette syndrome group			Control group		
	CBCL	YSR	Р	CBCL	YSR	Р
Problem scales						
Total	61.0 ± 8.6	53.4 ± 11.2	.001	49.1 ± 8.9	50.4 ± 10.5	.367
Internalizing	58.I <u>+</u> 9.7	50.1 ± 12.1	.001	51.9 ± 9.34	50.1 ± 11.1	.212
Withdrawn	56.7 <u>+</u> 8.7	54.4 <u>+</u> 6.4	.650	54.7 ± 6.9	53.5 ± 6.0	.144
Somatic complaints	57.7 <u>+</u> 7.6	53.I ± 4.8	.002	56.0 <u>+</u> 7.5	54.5 ± 6.8	.099
Anxious/depressed	61.0 ± 10.1	55.8 ± 9.4	.015	54.0 ± 5.1	54.3 ± 6.2	.766
Externalizing	56.5 [—] 56.5 [—]	52.3 [—] 9.5	.046	47.2 [—] 8.1	52.2 [—] 9.6	.001
Delinquent behavior	54.5 ± 5.1	54.6 ± 5.1	.871	52.0 ± 3.6	55.7 <u>+</u> 7.6	<.001
Aggressive behavior	57.0 + 6.I	55.2 + 6.9	.204	52.1 + 4.0	54.9 + 7.0	.004
Social problems	60.0 [—] 7.3	56.4 [—] 6.8	.034	54.3 [—] 6.5	53.8 [—] 6.3	.629
Thought problems	6I.7 [—] 9.2	53.I [—] 4.6	<.001	51.9 [—] 51.9 [—]	52.2 [—] 6.2	.699
Attention problems	67.5 [—] 9.4	61.2 [—] 10.1	.005	54.I [—] 4.8	54.0 [—] 6.2	.923
Competence scales	_	_		—	_	
Total	34.7 ± 8.1	42.9 ± 10.2	<.001	39.5 ± 7.8	49.9 ± 9.3	<.001
Activities	34.6 [—] 8.8	40.7 ± 11.7	.008	36.3 [—] 9.4	46.I <u>+</u> 7.5	<.001
Social	41.2 + 8.1	46.0 + 7.2	.009	43.I [—] 7.2	48.8 + 7.2	<.001

Table 2. Comparison Between Mothers' and Children's Reports in the Tourette Syndrome Group and Healthy Control Group.

Values are expressed as mean ± standard deviation unless otherwise indicated. CBCL, Child Behavior Checklist 6-18; YSR, Youth Self-Report.

difference in the way the parent and the child judge the child's ability. However, children with Tourette syndrome may underestimate the severity of their behavioral problems and the impact of these. It has been suggested that self-report instruments in this age group could possess intrinsic limitations.²⁶ However, some authors argue youths are better equipped to judge their problems than parents/caregivers.¹⁰ Other studies have shown that internalizing difficulties are reported more frequently by young patients themselves than their parents.³¹

The limitations of this study include sample size, which limits statistical power, and possible referral bias, as all patients were recruited from specialist clinics where more complex/ severe cases are seen. Further studies are needed to investigate the use of parent report and self-report measures across larger samples and in community settings.

In conclusion, the Child Behavior Checklist is a sensitive instrument for the screening of behavioral problems in children with Tourette syndrome. While self-report resulted in lower ratings, mothers' ratings for patients often fell within the "borderline" range for behavioral disorders, consistent with the high prevalence of comorbid conditions in the Tourette syndrome group (comorbid ADHD: 50%; comorbid obsessive-compulsive disorder: 42.9%). We caution that comprehensive evaluation of the behavioral spectrum of Tourette syndrome requires the integration of both parents' and patients' perspectives. Indeed, the knowledge of parental perceived intensity of child behavioral problems could account for a significant proportion of unique variance in predicting the quality of family life.

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Author Contributions

AEC and CT were responsible for the conception and organization of the research project; CL, CS, VB, and UB were responsible for the execution and supervision of the research project; CL and CS were responsible for the design and execution of statistical analysis; and CME and AEC were responsible for article preparation.

Declaration of Conflicting Interests

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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Ethical Approval

The study was approved by the local ethics committee, and written informed consent was obtained from all patients prior to enrollment.

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