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Parent and Self-Report Health-Related Quality of Life Measures in Young Patients With Tourette Syndrome

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Abstract

Tourette syndrome is a neurodevelopmental disorder characterized by tics and comorbid behavioral problems. This study compared child- and parent-reported quality of life and everyday functioning. We assessed 75 children with Tourette syndrome, of which 42 (56%) had comorbid conditions (obsessive-compulsive disorder = 25; attention-deficit hyperactivity disorder = 6; both comorbidities = 4). All patients completed psychometric instruments, including the Gilles de la Tourette Syndrome–Quality of Life Scale for Children and Adolescents (child report) and the Child Tourette's Syndrome Impairment Scale (parent report). Data were compared for patients with pure Tourette syndrome, Tourette syndrome + obsessive-compulsive disorder, Tourette syndrome + attention-deficit hyperactivity disorder, and Tourette syndrome + both comorbidities. There were no group differences in quality of life. However, there were differences for total, school, and home activities impairment scores. Children and parents may not share similar views about the impact of Tourette syndrome on functioning. The measurement of health-related quality of life in Tourette syndrome is more complex in children than adults.

Keywords

Tourette syndrome, tics, health-related quality of life, parent report, self-report

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Tourette syndrome is a childhood-onset neurodevelopmental condition characterized by multiple motor tics and at least 1 vocal/phonic tic. Tics are rapid, recurrent, nonrhythmic, and stereotyped movements/vocalizations that can be simple or complex, are usually suggestible, and may be preceded by premonitory urges and suppressed voluntarily. Although prevalence rates show wide variability, most recent studies suggest a prevalence in youngsters aged 5 to 18 of about 1%, with a male-female ratio of 4:1.[1]

Tourette syndrome is a complex disorder. In 90% of cases, additional behavioral problems are present (full-blown Tourette syndrome and Tourette syndrome–plus). The spectrum of behavioral difficulties ranges from complex tic-like symptoms (self-injurious behaviors; non-obscene socially inappropriate behaviors; and coprophenomena, echophenomena, and paliphenomena) to symptoms of attention-deficit hyperactivity disorder (ADHD), obsessive-compulsive disorder, mood disorders, impulse control disorders, and personality disorders.[1]

The presence of these comorbidities has been shown to be associated with social difficulties and impulse dyscontrol in young patients. Both tics and comorbid disorders associated with Tourette syndrome are potentially socially disabling, with the latter sometimes exerting greater detrimental impact.[2] In recent years, health-related quality of life has begun to be established as an essential measure of clinical outcome, as it

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takes into account the patient's own subjective perspective. The first disease-specific health-related quality of life assessment tool for youths with Tourette syndrome was recently developed and validated: the Gilles de la Tourette Syndrome–Quality of Life Scale for Children and Adolescents.[3]

In the clinical assessment of young patients with Tourette syndrome, both self- and proxy report instruments are routinely used to assist the specialist. These instruments allow thorough investigation of the clinical characteristics of Tourette syndrome patients, and in particular, an assessment of comorbid conditions. However, few previous studies have investigated the usefulness of generic parent or self-report health-related quality of life measures in complementing data obtained from neuropsychiatric assessments and clinician-rated scales in young Tourette syndrome patients.[2,4]

The current study explored the functional impact of tics and comorbid conditions among children and adolescents with Tourette syndrome on health-related quality of life, comparing both parent and child perceptions of well-being, using disease-specific questionnaires.

Methods

Over the period 2009-2010, all consecutive outpatients with Tourette syndrome seen at 3 specialist centers in Italy (the Child and Adolescent Neuropsychiatry Unit at the University of Insubria, Varese; the Department of Child Neuropsychiatry at the University of Catania; and the Department of Child Neuropsychiatry at the University of Rome) were invited to participate in the study. The study was approved by the local Ethics Committee and written informed consent was obtained from all subjects prior to enrolment.

All patients underwent neurologic examination, cognitive assessment, and instrumental analysis (electroencephalography, brain magnetic resonance imaging [MRI], standard laboratory test) to rule out secondary tics and tourettism. We also excluded subjects with learning disabilities (Intelligence Quotient <70) or other neurologic conditions.

The 75 young patients recruited met *Diagnostic and Statistical Manual for Mental Disorders* (4th edition, text revised) criteria for Tourette syndrome (Varese: n = 21; Rome: n = 24; Catania: n = 30). All participants were clinically evaluated by a neuropsychiatrist with substantial experience in Tourette syndrome. The clinical interview included the National Hospital Interview Schedule for Gilles de la Tourette syndrome, a detailed semi-structured interview schedule,[5] and the Diagnostic Confidence Index, which rates the lifetime likelihood of having Tourette syndrome.[6] The Schedule for Affective Disorders and Schizophrenia for School-Age Children–Present and Lifetime Version[7] (a semistructured diagnostic interview designed to assess current and past episodes of psychopathology in children and adolescents according to *Diagnostic and Statistical Manual for Mental Disorders* [3rd and 4th edition, text revised] criteria) was used to validate the diagnosis of various Tourette syndrome–associated disorders, such as ADHD and obsessive-compulsive disorder. Tic severity was assessed using the Yale Global Tic Severity Scale,[8] whereas the severity of obsessive-compulsive symptoms was rated using the Children's Yale-Brown Obsessive Compulsive Scale.[9]

All participants completed a standardized psychometric battery, including the following self-report scales:

- The Gilles de la Tourette Syndrome-Quality of Life Scale for Children and Adolescents: a 27-item scale (Italian version) consisting of 4 subscales (psychological, physical, obsessive-compulsive and cognitive) in 2 age-adjusted versions: (1) an interview to be administered by a qualified clinician for children aged 6-12 years and (2) a self-report questionnaire for adolescents aged 13-18 years.[3]
- The Child Depression Inventory: a 27-item self-report instrument that assesses depressive symptoms in 7- to 17-year-olds.[10]
- The Multidimensional Anxiety Scale for Children: a 39-item self-report scale that robustly represents the factor structure of anxiety in children aged 8 to 18 years.[11]
- Child Tourette's Syndrome Impairment Scale, a 37-item parent-rated instrument covering school, home, and social activities that may be impaired by tics or comorbid problems (including obsessive-compulsive symptoms, depression, anxiety, oppositional/disruptive behavior, hyperactivity, and inattention).[12]

We split the sample into 4 groups. These were pure Tourette syndrome without comorbid conditions (n = 33), Tourette syndrome + obsessive-compulsive disorder (n = 25), Tourette syndrome + ADHD (n = 6), and Tourette syndrome + obsessive-compulsive disorder + ADHD (n = 11). These groups were compared using nonparametric Kruskal-Wallis *H* tests for quantitative variables (age; mean ratings on clinical measures; mean scale and subscale scores), and chi-square analysis for qualitative variables (sex; presence of tic-related symptoms; pharmacotherapy). All statistical analyses were performed using SPSS for Macintosh, version 18.0.2.

Results

We recruited 75 patients fulfilling diagnostic criteria for Tourette syndrome (60 males; age range 6.8-18.3; mean $\pm$ SD = 12.4 $\pm$ 3.2 years; mean age of disease onset = 6.7 $\pm$ 2.4 years; mean disease duration = 2.0 $\pm$ 2.0 years). This sample was characterized by marked tic severity (Yale Global Tic Severity Scale total score mean = 44.3 $\pm$ 20.5).

Demographic and clinical characteristics for the 4 groups (pure Tourette syndrome = 44%, Tourette syndrome + obsessive-compulsive disorder = 33.3%, Tourette syndrome + ADHD = 8%, Tourette syndrome + obsessive-compulsive disorder + attention-deficit hyperactivity disorder = 14.7%) are summarized in Table 1. The Tourette syndrome + obsessive-compulsive disorder group was significantly older than the other groups ($P = .014$) and had higher Children's Yale-Brown Obsessive-Compulsive Scale scores ($P < .001$). The Tourette syndrome + ADHD group showed higher total tic severity scores than the other 3 groups ($P = .008$). There were further differences between the groups for disease duration.

Table 2 shows the scores obtained from each group for the quality of life scale and impairment scale. The comparison between the 4 groups did not reveal any significant difference in Gilles de la Tourette syndrome–quality of life, child and adolescent version, total and subscale scores. However, when we compared data obtained from the impairment scale across the 4 groups, we found significant differences for total ($P = .012$), school activities ($P = .001$), and home activities ($P = .004$) scores. No differences were found for the social activities subscale.

Table 1. Demographic and Clinical Characteristics of the Tourette Syndrome Sample.

	Pure TS (n = 33)	TS + OCD (n = 25)	TS + ADHD (n = 6)	TS + OCD + ADHD (n = 11)	*P*
Male, n (%)	26 (78.8)	22 (88.0)	5 (83.3)	7 (63.6)	.405
Age (y), mean (SD)	11.4 (2.8)	13.8 (3.4)	10.6 (2.4)	13.7 (3.1)	**.014**
Age at onset of tics (y), mean (SD)	6.9 (2.2)	6.7 (2.6)	6.3 (3.2)	6.2 (2.3)	.656
Disease duration (y), mean (SD)	1.4 (1.7)	2.7 (2.1)	1.1 (1.8)	2.9 (2.2)	**.011**
DCI total score, mean (SD)	80 (23.4)	88.7 (25.1)	92.2 (19.6)	92.2 (23.5)	.330
YGTSS global severity score, mean (SD)	35.8 (17.9)	48.2 (21.3)	56.3 (13.4)	54.5 (20.7)	**.008**
YGTSS total motor tic score, mean (SD)	11.3 (4.6)	14.0 (6.3)	15.5 (3.5)	13.6 (5.5)	.102
YGTSS total vocal tic score, mean (SD)	7.4 (5.3)	9.0 (5.6)	12.5 (4.6)	11.7 (6.1)	.070
CY-BOCS total score, mean (SD)	4.9 (6.2)	18.3 (9.4)	7.3 (8.2)	17.5 (8.2)	**<.001**
Tic-related symptoms, n (%)	10 (30.3)	12.0 (48.0)	4 (66.7)	7 (63.6)	.130
CDI total score, mean (SD)	7.7 (5.1)	10.1 (7.4)	13.8 (6.7)	11.6 (9.2)	.193
MASC total score, mean (SD)	49.1 (9.4)	49.4 (11.5)	53.3 (11.8)	49.3 (9.7)	.803
Pharmacotherapy, n (%)	10 (30.3)	19 (76.0)	3 (50.0)	8 (72.7)	.003

Abbreviations: ADHD, attention-deficit hyperactivity disorder; CDI, Child Depression Inventory; CY-BOCS, Children's Yale-Brown Obsessive Compulsive Scale; DCI, Diagnostic Confidence Index; MASC, Multidimensional Anxiety Scale for Children; OCD, obsessive-compulsive disorder; TS, Tourette syndrome; YGTSS, Yale Global Tic Severity Scale.
Note: Significant differences in bold.

Table 2. Total and subscales scores of the Gilles de la Tourette Syndrome–Quality of Life Scale for Children and Adolescents and Child Tourette's Syndrome Impairment Scale.

	Pure TS (n = 33)	TS + OCD (n = 25)	TS + ADHD (n = 6)	TS + OCD + ADHD (n = 11)	*P*
Completed by children					
Mean GTS-QOL-C&A total score (SD)	22.56 (15.27)	30.33 (22.35)	31.17 (11.69)	27.82 (16.52)	.443
Mean GTS-QOL-C&A psychological score (SD)	9.88 (8.22)	13.64 (10.79)	15.67 (7.23)	10.73 (6.93)	.346
Mean GTS-QOL-C&A physical/ADL score(SD)	5.66 (4.16)	6.17 (5.68)	6.17 (3.31)	7.00 (4.07)	.719
Mean GTS-QOL-C&A obsessive-compulsive score(SD)	3.52 (3.40)	5.76 (4.25)	4.67 (3.08)	6.27 (3.88)	.077
Mean GTS-QOL-C&A cognitive score (SD)	3.91 (2.49)	4.36 (3.79)	4.67 (2.73)	3.82 (3.68)	.898
Mean GTS-QOL-C&A VAS score (SD)	77.58 (22.40)	67.58 (26.44)	61.67 (34.30)	72.27 (30.77)	.384
Completed by parents					
Mean CTIM-P total score (SD)	8.29 (12.10)	11.60 (15.80)	18.83 (16.49)	34.00 (25.23)	**.012**
Mean CTIM-P school activities score (SD)	3.42 (4.32)	3.40 (4.81)	8.33 (5.85)	12.70 (7.88)	**.001**
Mean CTIM-P home activities score (SD)	1.16 (2.76)	2.44 (4.59)	3.33 (4.50)	6.82 (6.48)	**.004**
Mean CTIM-P social activities score (SD)	3.34 (6.46)	5.76 (9.31)	7.17 (9.70)	13.80 (14.31)	.161

Abbreviations: ADHD, attention-deficit hyperactivity disorder; ADL, activities of daily living; CTIM-P, Child Tourette's Syndrome Impairment Scale; GTS-QOL-C&A, Gilles de la Tourette Syndrome–Quality of Life Scale for Children and Adolescents; OCD, obsessive-compulsive disorder; TS, Tourette syndrome; VAS, Visual Analog Scale.
Note: Significant differences in bold.

Discussion

In this study, we examined the impact of tics and comorbid conditions on the health-related quality of life of children and adolescents with Tourette syndrome, using parent report on the Child Tourette's Syndrome Impairment Scale and child report on the Gilles de la Tourette Syndrome–Quality of Life Scale for Children and Adolescents. The sample formed 4 subgroups, according to the presence (Tourette syndrome + obsessive-compulsive disorder; Tourette syndrome + ADHD; Tourette syndrome + obsessive-compulsive disorder + ADHD) or absence (pure Tourette syndrome) of comorbid conditions. There were differences between the 4 groups for the severity of obsessive-compulsive symptoms, as would be expected. Of greater interest, we found that group with comorbid ADHD exhibited greater tic severity than the pure Tourette syndrome and Tourette syndrome + obsessive-compulsive disorder groups, as reported by previous studies.[13] Despite this, we found no differences between the subgroups for the quality of life physical subscale, which should have been sensitive to the influence of tic severity.[14] However, other studies have shown that tic severity is not always related to the presence of comorbid conditions,[15] though our finding could have been affected by the small number of patients with Tourette syndrome + ADHD.

We found significant differences between the 4 Tourette syndrome subgroups for the Child Tourette Syndrome Impairment Scale, which is consistent with prior research[4] using generic instruments. Eddy et al.[2] found that comorbidity subgroups could be differentiated using a generic health-related quality of life measure. The quality of life scale used in the

current study did not reveal differences in total score across the 4 patient groups. This finding is likely to reflect a strength in the scale in focusing on core symptoms of Tourette syndrome (eg, tics), which were shared by subgroups.

Significant differences were apparent when comparing child self-report versus parent report of health-related quality of life. These results suggest that children and parents may be unlikely to share similar views about the overall functional impact of Tourette syndrome. There are several potential explanations for these findings. First, parents may place most emphasis on comorbid conditions, which they perceive to be worse than tics, leading to differences in the impairment scores across patient subgroups. Children, however, may more readily notice the impact of tics. Second, parent report of quality of life for children with Tourette syndrome may be additionally influenced by personal anxiety and feelings of responsibility that concerned parents are likely to experience.

This study has some methodological limitations. The relatively small number of patients limits the statistical power of the analysis, and the small sample contained few patients with Tourette syndrome + ADHD. Two-thirds of the sample was aged 6 to 11 years, and these young patients may have been less aware of their difficulties. Finally, all patients were recruited from specialist neuropsychiatric settings where more complex/severe cases are seen, resulting in a sample characterized by marked tic severity. Further studies addressing health-related quality of life assessment in Tourette syndrome should be carried out on larger samples and in community settings.

This study has demonstrated the utility of the Child Tourette's Syndrome Impairment Scale in differentiating between subgroups of individuals with Tourette syndrome and comorbid conditions. Perhaps more important, we have shown there can be significant differences between health-related quality of life as reported by young people with Tourette syndrome and their parents. Future studies should therefore avoid relying exclusively on parent report, as this may not constitute an accurate reflection of the well-being of a young patient with Tourette syndrome.

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Author Contributions

AEC and CT were responsible for research project conception and organization.CL, CS, RB, PRS, PVC, EG, UB, FC, and RR undertook research project execution and supervision. CL and CS designed and executed the statistical analysis. CME and AEC prepared the manuscript.

Ethical Approval

The study was approved by the local Ethics Committee and written informed consent was obtained from all subjects prior to enrolment.

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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