

Quality of Life in Youth With Tourette's Syndrome and Chronic Tic Disorder

Eric A. Storch

Departments of Psychiatry and Pediatrics, University of Florida

Lisa J. Merlo

Department of Psychiatry, University of Florida

Caleb Lack

Department of Behavioral Sciences, Arkansas Tech University

Vanessa A. Milsom

Department of Clinical and Health Psychology, University of Florida

Gary R. Geffken

*Departments of Psychiatry, Pediatrics, and Clinical and Health Psychology;
University of Florida*

Wayne K. Goodman and Tanya K. Murphy

Department of Psychiatry, University of Florida

This study sought to examine quality of life (QoL) in clinic-referred children and adolescents ($n = 59$, M age = 11.4 ± 2.6 years) with a chronic tic disorder. The QoL scores for tic patients were lower than for healthy controls but higher than for the psychiatric sample on the majority of domains. Children's self-reported QoL scores and a measure of tic severity were moderately and inversely correlated. Parent reports of their child's QoL were weakly related to tic severity. Correlations between parent and child ratings of QoL for children ages 8 to 11 years were generally higher than those for youth ages 12 to 17 years. Finally, externalizing behavior moderated the relations between tic severity and parent-rated QoL, such that tic severity was significantly associated with parent-rated QoL for children with below average externalizing symptoms but not for children with above average externalizing symptoms.

Tourette's Syndrome (TS) is a neuropsychiatric disorder characterized by the presence of multiple motor tics and at least one vocal tic (American Psychiatric Association, 2000). Tics are commonly defined as stereotyped vocalizations or motor movements that are quick, not rhythmic, and occur frequently (cf. Singer, 2005). TS is often comorbid with other psychiatric diagnoses, the most common of which are attention deficit hyperactivity disorder, which co-occurs in 50% to 75% of cases

(King & Scahill, 2001; Robertson, Banerjee, Eapen, & Fox-Hiley, 2002) and obsessive-compulsive disorder, which co-occurs in approximately 50% of cases (Robertson, 1995). In addition, a number of other psychological and behavioral problems occur in higher than average rates among youth with TS, including disruptive behavior (Budman, Bruun, Park, Lesser, & Olson, 2000; Sukhodolsky et al., 2003), mood and anxiety disorders (Carter et al., 2000; Coffey et al., 2000; Coffey & Park, 1997; Robertson et al., 2002), difficulty interacting socially (Carter et al., 2000; Kurlan et al., 1996), and learning disabilities (Comings & Comings, 1985; Robertson, Trimble, & Lees, 1988). In addition, research has

Correspondence should be sent to Eric A. Storch, Department of Psychiatry, Box 100234, University of Florida, Gainesville, FL 32610. E-mail: estorch@psychiatry.ufl.edu

shown negative social perception toward adults (Woods, Fuqua, & Outman, 1999) and children (Boudjouk, Woods, Miltenberger, & Long, 2000) with motor and vocal tics regardless of any comorbid behavioral problems. Both studies also showed that tic severity was inversely related to social acceptableness.

Given the number of problems and difficulties associated with a diagnosis of TS or chronic tic disorder, it is not surprising that quality of life (QoL) may be impacted. Although there is no one agreed-on definition of "quality of life," there are two primary types of information gathered in assessing QoL: functional status of an individual and the individuals' appraisals of how their health impacts their life (Muldoon, Barger, Flory, & Manuck, 1998). For the purposes of this study, QoL was defined "as a patient's perceptions of the impact of disease and treatment functioning in a variety of dimensions including physical, mental, and social domains" (Varni, Seid, & Rode, 1999, p. 126). QoL has been shown to be lowered for children with a variety of medical and behavioral problems, including diabetes (Varni, Burwinkle, Seid et al., 2003), obesity (Williams, Wake, Hesketh, Maher, & Waters, 2005), psychiatric difficulties (Bastiaansen et al., 2003), and asthma (Varni, Burwinkle, Rapoff, Kamps, & Olson, 2004). However, there is a relative lack of research examining QoL in individuals with tics and tic disorders, particularly in pediatric patients.

In an early study examining how QoL is affected in adults with TS, Stefl (1984) found that more than half of the individuals with TS had sought mental health services to assist them in coping with the impact of their tics on daily functioning. In another study, 88% of the sample reported that tics negatively influenced their daily functioning (Ernberg, Cruse, & Rothner, 1987). In both of these samples, a high proportion of participants reported additional non-TS difficulties, including learning problems and psychological disorders, which resulted in additional functional limitations beyond those attributable solely to their tics. Decreased self-esteem and increased social anxiety have also been reported among adults who suffer from tics (Thibert, Day, & Sandor, 1995). In addition, adults with TS report higher unemployment rates (Robertson et al., 1988) and lower income (Brunn, 1988) than those without TS. The QoL of family members is also impacted by TS. Parents of children with TS have reported more marital difficulties, substance abuse, family conflict, lowered quality of parent-child interactions, and higher levels of parenting frustration relative to parents of children without TS (Cohen, Ort, Leckman, Riddle, & Hardin, 1988; Stefl, 1983; Wilkinson et al., 2001).

In the first study to measure QoL with a well-validated quantitative instrument in a sample of adults with tic disorders, results showed a significantly lower QoL for those with tics relative to the general population (Eltner, Selai, Trimble, & Robertson, 2001). No age or gender effects were found, although those with a higher rate of tic severity endorsed greater impairment in social functioning and felt more limited in their capabilities than those with mild or moderate tics. Comparable to results of earlier epidemiological studies (e.g., Robertson et al., 1988), higher rates of depression, anxiety, and obsessive-compulsive disorder were found in the tic sample, with greater tic severity related to higher levels of depression (Eltner et al., 2001).

A formal assessment of the impact of tics on functioning and QoL in a pediatric population has yet to be conducted. The studies that have examined functioning in adolescent populations have used measures such as the Yale Global Tic Severity Scale (YGTSS; Leckman et al., 1989) Impairment rating, which specifically assesses tic-related impairment without measuring general QoL (e.g., Coffey et al., 2004). In a study that examined QoL in a pediatric population referred for general psychiatric difficulties, Bastiaansen, Koot, Bongers, Varni, and Verhulst (2004) found QoL to be significantly lower for children with psychiatric difficulties relative to healthy controls. This finding, combined with significant correlations between QoL and measures of psychopathology, suggests that children with psychiatric problems experience as significant a decrease in QoL as those with chronic medical illnesses.

This study examined QoL in youth with TS and addressed four primary goals. The first goal was to examine child and parent reports of QoL compared to previously reported data on psychiatric (Bastiaansen et al., 2004) and nonclinical (Varni, Burwinkle, Seid & Skarr, 2003) samples. We expected that QoL would be lower in the tic sample than the nonclinical sample but similar to the psychiatric sample. The second goal was to examine the relations among tic severity and QoL scores. We predicted that tic severity would be inversely related to child and parent-proxy reports of overall QoL. Additionally, we were interested in exploring differential relations between tic severity and QoL subdomains. The third goal was to examine parent-child agreement in QoL reports and to determine whether agreement differed by age. Finally, given the high rates of psychiatric comorbidity in pediatric tic disorders, the fourth goal was to examine the extent to which comorbid internalizing and externalizing symptoms were related to QoL. We predicted that tic severity and co-occurring internalizing and externalizing symptoms would interact

in predicting QoL, such that those with additional problems would report lower QoL.

Method

Participants

Participants included 59 children and adolescents (41 boys) with TS or chronic tic disorder consecutive referrals to a university-based clinic for pharmacological or behavioral treatment of tics. Youth ranged in age from 8 to 17 years old ($M = 11.4 \pm 2.6$). The ethnic distribution was White (97%) and Hispanic American (3%). TS or chronic tic disorder and comorbid diagnoses were made in accordance with the *Diagnostic and Statistical Manual of Mental Disorder* (fourth edition, text revision; American Psychiatric Association, 2000) by a board-certified child psychiatrist with 10 years of experience by using all available clinical information (Leckman, Sholomskas, Thompson, Belanger, & Weissman, 1982). Considered the gold standard for diagnosis, this method incorporates information from the YGTSS (Leckman et al., 1989), a semistructured clinical interview, and responses to other measures. Tic and comorbid diagnoses were also confirmed by one of two licensed clinical psychologists with extensive experience based on a discussion of symptoms and viewing relevant quantitative data. Disagreements were resolved through discussion; in the rare instance that this was not possible, the primary psychiatrist (the first author) made the final decision. Comorbid diagnoses, when present, consisted of the following: attention deficit hyperactivity disorder ($n = 28$), obsessive-compulsive disorder ($n = 25$), major depression disorder ($n = 6$), generalized anxiety disorder ($n = 7$), oppositional defiant disorder ($n = 6$), social phobia ($n = 2$), Asperger's disorder ($n = 3$), and panic disorder ($n = 1$). Sixteen youth had one and 27 had multiple comorbidities.

Measures

Pediatric quality of life inventory (PedsQL). The PedsQL (version 4.0; Varni et al., 1999) is a 23-item measure of children's QoL. Items are rated on a 5-point scale with higher scores corresponding to better QoL. The following four domains have been identified: (a) physical functioning (eight items), (b) emotional functioning (five items), (c) social functioning (five items), and (d) school functioning (five items). The individual scales are combined to yield physical (equivalent to the physical functioning domain), psychosocial (sum of emotional, social, and school functioning domains),

and total health scales (all four domains). Excellent and extensive reliability data exist for the PedsQL and PedsQL-Parent Proxy across multiple pediatric illnesses (e.g., Bastiaansen et al., 2004; Varni et al., 2003; Varni, Seid, & Kurtin, 2001). For example, Varni et al. (2001) found the internal consistency for the PedsQL and PedsQL-Parent Proxy among healthy youth to be .88 and .90, respectively. Construct validity was supported by PedsQL differences in the expected directions between youth who were not overweight, at risk for overweight, and overweight (Williams et al., 2005). In addition, the PedsQL has a stable factor structure (Varni et al., 2001, 1999) and differentiates between youth with and without a chronic medical condition (Varni et al., 2004).

YGTSS. The YGTSS (Leckman et al., 1989) is an 11-item clinician-rated interview of motor and phonic tic severity. The clinician initially notes the presence of motor and phonic tics based on child and parent reports over the past week, as well as behavioral observations. Following this, the clinician rates the severity of motor and phonic tics on five separate dimensions each: number, frequency, intensity, complexity, and interference. The YGTSS also includes a separate impairment rating focusing on distress and impairment in interpersonal, academic, and occupational realms. Five index scores are obtained: Total Motor Tic Score, Total Phonic Tic Score, Total Tic Score, Overall Impairment Rating, and Global Severity Score. Excellent reliability data have been reported, including high internal consistency ($\alpha = .92-.93$), interrater reliability (intraclass correlation coefficients for index scores = .62-.85), and 7-week stability (intraclass correlations coefficients = .77-.90; Leckman et al., 1989; Storch et al., 2005). Validity support is derived from a stable factor structure (Storch et al., in press), positive correlations with parent- and clinician-rated tics, and weak nonsignificant correlations with obsessive-compulsive disorder symptoms, depression, and anxiety (Leckman et al., 1989; Storch et al., 2005).

Child Behavior Checklist (CBCL). The CBCL (Achenbach, 1991) is a widely used parent-rated questionnaire assessing the frequency and intensity of behavioral and emotional problems exhibited by the child in the past 6 months. Items are rated on a scale of 0 (*not true*), 1 (*somewhat or sometimes true*), or 2 (*very true or often true*). The CBCL consists of eight syndrome

scales (Withdrawn, Somatic Complaints, Anxious/Depressed, Social Problems, Thought Problems, Attention Problems, Delinquent Behavior, and Aggressive Behavior) and two composite scales (Externalizing and Internalizing Problems). Only the two composite scales were used in this study. Overall, the CBCL has excellent psychometric properties, including 1-week test-retest reliability, adequate internal consistency ($\alpha = .62-.92$), and construct validity (e.g., strong associations with subscales of other measures that assess similar constructs; Achenbach, 1991).

Procedures

The University of Florida Institutional Review Board approved all study procedures. Potentially eligible participants were approached at their clinic visit by a research assistant about participating in a study on QoL and impairment related to having a tic disorder. The consent rate was 91% (59/65) with the most common reasons for declining being time constraints and child refusal to participate. After describing the study and obtaining appropriate written consent and assent, the nature of motor and phonic tics were defined for families. Master's or doctoral-level clinical psychology trainees then administered the YGTSS to participants. All clinicians had extensive experience working with children with tics. YGTSS training included an instructional meeting, four mock practice interviews, and four interviews observed by the first or final author. The intraclass correlation coefficient for the four mock and four observed interviews had to be $\gg .80$ to achieve criterion agreement. The YGTSS was administered to parent(s) and children jointly, with the final ratings based on their responses, clinician judgment, and behavioral observation of the child. Interrater reliability for the YGTSS was calculated on 10 randomly selected participants ($\kappa = .98$). Instructions were given to the parent (primary caregiver when both were present) and child on completing the PedsQL, CBCL, and PedsQL-Parent Proxy. For approximately half of the participants, the parent and child independently completed the measures after administration of the YGTSS. For the other half, child and parent-report measures were completed prior to the YGTSS. Families were compensated \$5 for their participation.

Results

Age and gender differences in the PedsQL scores of tic patients were examined. Using a 2 (Gender) \times 2 (Age: 8–11 and 12–17 years) analysis

of variance, QoL scores for male tic patients ($M = 68.91$, $SD = 15.0$) were significantly lower than those of female tic patients ($M = 78.84$, $SD = 17.08$) on the child-rated PedsQL total score, $F(1, 58) = 5.04$, $p = .03$, but not the PedsQL-Parent Proxy total score, $F(1, 58) = 2.85$, *ns*. There were no significant differences in parent- or child-rated PedsQL total scores for children ages 8 to 11 or 12 to 17 years, nor any Gender \times Age interaction. Using a 2 (Gender) \times 2 (Age: 8–11 and 12–17 years) multivariate analysis of variance, no significant age or gender differences on PedsQL subscale scores were found.

QoL Scores in Youth with Tics Compared to Youth with Psychiatric Disorders and Health Controls

The PedsQL scores of youth with tics were compared to previously reported scores of youth with psychiatric disorders seen at an outpatient clinic (Bastiaansen et al., 2004) and healthy controls (Varni et al., 2003). The psychiatric sample (Bastiaansen et al., 2004) consisted of 310 children, ranging in age from 6.0 to 18.2 years ($M = 11.3$, $SD = 3.18$) who had been referred to an outpatient mental health clinic. The sample was predominantly male (59.7%) and included families of low (33.2%), medium (30.0%), and high (36.8%) socioeconomic status. No description of specific psychiatric conditions was included. The control sample (Varni et al., 2003) consisted of healthy children (ages 2–18 years) who were assessed either in physicians' offices during routine visits or via telephone and whose parents did not report the presence of a significant chronic health condition. To control for Type I error, a Bonferroni correction was used and the alpha level for significance was set at $p < .004$ (.05/12).

Table 1 displays the means and standard deviations of the QoL scores for the tic sample, psychiatric sample, and healthy controls. Using one-sample *t* tests, QoL scores for tic patients were significantly lower than those for healthy controls on the PedsQL total score, $t(58) = -5.25$, $p < .001$, and domains of psychosocial health, $t(58) = -5.89$, $p < .001$; emotional functioning, $t(58) = -4.54$, $p < .001$; social functioning, $t(58) = -3.63$, $p < .001$; and school functioning, $t(58) = -6.06$, $p < .001$. Further, QoL scores for tic patients were significantly lower than those of healthy controls in the PedsQL-Parent Proxy total score, $t(58) = -7.47$, $p < .001$, and the domains of psychosocial health, $t(58) = -8.27$, $p < .001$; emotional functioning, $t(58) = -5.40$, $p < .001$; social functioning, $t(58) = -6.66$, $p < .001$; and school functioning, $t(58) = -7.84$, $p < .001$. There were no differences between the tic and psychiatric

Table 1. *PedsQL Scores for Youth With Tics Compared to a Psychiatric Sample and Healthy Controls*

	Tic Sample ^a		Psychiatric Sample ^b		Healthy Sample ^c		Group Differences
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	
Child Self-Report							
Total score	71.94	16.19	72.20	12.7	83.00	14.79	a < c
Physical functioning	81.73	16.74	81.20	14.2	84.41	17.26	a = b = c
Psychosocial health	68.67	17.89	67.40	14.7	82.38	15.51	a < c
Emotional functioning	67.88	21.96	61.30	19.5	80.86	19.64	a < c
Social functioning	75.08	26.09	73.00	20.4	87.42	17.18	a < c
School functioning	63.05	19.76	67.90	16.7	78.63	20.53	a < c
Parent Proxy Report							
Total score	71.78	16.29	66.90	14.00	87.61	12.33	a < c
Physical functioning	85.09	17.06	80.00	17.70	89.32	16.35	a = b = c
Psychosocial health	67.34	17.87	59.90	15.30	86.58	12.79	b < a < c
Emotional functioning	67.63	21.34	54.40	18.70	82.64	17.54	b < a < c
Social functioning	72.63	21.82	63.30	22.90	91.56	14.20	b < a < c
School functioning	61.78	23.23	62.40	18.20	85.47	17.61	a < c

Note: PedsQL = Pediatric Quality of Life Inventory. One-sample *t* tests were used with Bonferroni correction to make pairwise comparisons.

^a Child self-report *n* = 59; parent proxy report *n* = 59.

^b Child self-report *n* = 296; parent proxy report *n* = 307.

^c Child self-report *n* = 386–401; parent proxy report *n* = 611–717.

p < .004.

sample across PedsQL child report scores. The QoL scores of tic patients were significantly higher than those for the psychiatric sample in the PedsQL–Parent Proxy domains of psychosocial health, *t*(58) = 3.20, *p* < .004; emotional functioning; *t*(58) = 4.76, *p* < .001; and social functioning; *t*(58) = 3.28, *p* < .004. Scores for the two groups were not significantly different in the other domains.

Parent–Child Agreement in QoL Scores

Pearson product–moment correlations between parent and child QoL ratings were computed separately for children ages 8 to 11 and 12 to 17 years (see Table 2). Using the Bonferroni correction, alpha was set at .004 (.05/12). For children ages 8 to 11, strong, positive correlations were found between parent and child reports on each domain

of the PedsQL: total score (*r* = .78, *p* < .001), physical health (*r* = .54, *p* < .001), psychosocial health (*r* = .79, *p* < .001), emotional functioning (*r* = .63, *p* < .001), social functioning (*r* = .66, *p* < .001), and school functioning (*r* = .53, *p* < .002). For children ages 12 to 17, no significant correlations were found.

Relations between QoL, Symptom Severity and Internalizing–Externalizing Problems

To examine the relation between QoL, tic symptom severity, and internalizing and externalizing problems among youth with tics, Pearson product–moment correlations were calculated among PedsQL scores and the YGTSS and CBCL (see Tables 3 and 4). Given the modest sample size and preliminary nature of this study, corrections

Table 2. *Correlations Between PedsQL Child Self-Report and Parent Report*

	Ages 8–11 ^a			Ages 12–17 ^b		
	<i>r</i>	Child Rating <i>M</i> ± <i>SD</i>	Parent Rating <i>M</i> ± <i>SD</i>	<i>r</i>	Adolescent Rating <i>M</i> ± <i>SD</i>	Parent Rating <i>M</i> ± <i>SD</i>
Total Score	.78**	69.39 ± 18.81	71.00 ± 17.76	.34	75.17 ± 11.65	72.76 ± 14.48
Physical Health	.54**	80.59 ± 17.64	84.33 ± 18.58	.28	83.17 ± 15.74	86.06 ± 15.21
Psychosocial Health	.79**	65.66 ± 20.70	66.57 ± 19.40	.42	72.50 ± 12.90	68.33 ± 16.06
Emotional Functioning	.63**	62.58 ± 24.43	66.06 ± 22.56	.38	74.62 ± 16.42	69.61 ± 19.95
Social Functioning	.66**	70.61 ± 30.20	71.97 ± 23.81	.35	80.77 ± 18.75	73.46 ± 19.43
School Functioning	.53**	63.79 ± 18.92	61.67 ± 21.38	.38	62.11 ± 21.13	61.92 ± 25.81

Note: PedsQL = Pediatric Quality of Life Inventory.

^a *n* = 33. ^b *n* = 26.

***p* < .004.

Table 3. Pearson Correlation Coefficients of the PedsQL With YGTSS Scores

	YGTSS Motor	YGTSS Phonic	YGTSS Total Tic	YGTSS Impairment	YGTSS Total
Child self-report					
Total score	-.19	-.33*	-.30*	-.39**	-.37**
Physical health	-.33*	-.38**	-.40**	-.36**	-.40**
Psychosocial health	-.13	-.28*	-.23	-.36**	-.32*
Emotional functioning	-.20	-.29*	-.28*	-.32*	-.32*
Social functioning	-.05	-.22	-.16	-.38**	-.29*
School functioning	-.05	-.14	-.11	-.12	-.12
Parent proxy report					
Total score	.05	-.18	-.09	-.25	-.18
Physical health	.11	-.02	.05	-.07	-.01
Psychosocial health	.02	-.22	-.12	-.28*	-.22
Emotional functioning	.14	-.04	.05	-.09	-.03
Social functioning	.09	-.27*	-.11	-.29*	-.22
School functioning	-.16	-.22	-.22	-.29*	-.27*

Note: PedsQL = Pediatric Quality of Life Inventory; YGTSS Motor = Yale Global Tic Severity Scale Motor Tic score; YGTSS Phonic = Yale Global Tic Severity Scale Phonic Tic score; YGTSS Total Tic = Yale Global Tic Severity Scale Total Tic score; YGTSS Impairment = Yale Global Tic Severity Scale Impairment Rating; YGTSS Total = Yale Global Tic Severity Scale total score. Corrections for Type I error were not made for these analyses.

* $p < .05$. ** $p < .01$.

for Type I error were not made for these analyses. In general, there were moderate, negative correlations among clinician-rated tic severity scores

and children’s self-reported QoL ($r_s = -0.12$ to -0.40). However, parent reports of their child’s QoL were generally not related to tic severity ($r_s = -0.01$ to -0.27). Additionally, there was a moderate, inverse relation between parent-rated internalizing and externalizing behaviors and both parents’ ($r_s = 0.46$ to -0.71) and children’s reports of child QoL ($r_s = -0.22$ to -0.47).

Table 4. Correlations Between PedsQL Scores With CBCL Scores

	CBCL Externalizing	CBCL Internalizing	CBCL Total
Child self-report			
Total score	-.31*	-.49**	-.44**
Physical health	-.10	-.29*	-.22
Psychosocial health	-.34**	-.50**	-.47**
Emotional functioning	-.20	-.38**	-.32*
Social functioning	-.36**	-.50**	-.47**
School functioning	-.23	-.28*	-.28*
Parent proxy report			
Total score	-.64**	-.67**	-.71**
Physical health	-.40**	-.57**	-.53**
Psychosocial health	-.65**	-.63**	-.70**
Emotional functioning	-.56**	-.71**	-.70**
Social functioning	-.50**	-.50**	-.54**
School functioning	-.51**	-.34**	-.46**

Note: PedsQL = Pediatric Quality of Life Inventory; CBCL Externalizing = Child Behavior Checklist Externalizing Scale score; CBCL Internalizing = Child Behavior Checklist Internalizing Scale score; CBCL Total = Child Behavior Checklist total score. Corrections for Type I error were not made for these analyses.

* $p < .05$. ** $p < .01$.

Interaction of Internalizing and Externalizing Symptoms with Tic Severity as a Predictor of QoL

A final goal of this study was to investigate the interaction of internalizing and externalizing symptoms with tic severity as predictors of QoL. Four separate hierarchical linear regressions were computed. The child-rated PedsQL or PedsQL–Parent Proxy total scores were the criterion variables. Per guidelines by Baron and Kenny (1986), the YGTSS Total Score and either CBCL internalizing or externalizing symptoms were entered together in Step 1, and the interaction of the YGTSS Total Score and respective symptom index (internalizing or externalizing) was entered in Step 2. Power analyses showed that a sample of 59 participants gave power greater than 0.8 to detect a medium effect size.

Child PedsQL Scores. In the analysis to determine whether internalizing symptoms interacted with tic severity, Step 1 analyses revealed that both tic severity and internalizing behaviors significantly predicted child-rated QoL

($\beta = -.34, p = .004$ and $\beta = -.45, p < .001$, respectively). There was no significant effect for the interaction of tic severity by internalizing symptoms, $R^2\text{change} = 0.003, F(3, 52) = .25, ns$. In the analysis to determine whether externalizing symptoms interacted with tic severity, both symptom severity and parent-rated externalizing behaviors significantly predicted child-rated QoL ($\beta = -.33, p = .011; \beta = -.25, p < .05$). There was no significant effect for the interaction of tic severity by externalizing symptoms, $R^2\text{change} = 0.001, F(3, 52) = .05, ns$.

Parent PedsQL scores. In the analysis to determine whether internalizing symptoms interacted with tic severity, parent-rated internalizing symptoms significantly predicted parent-rated QoL ($\beta = -.65, p < .001$), but tic severity did not ($\beta = -.14, ns$), see Table 5. There was no significant effect for the interaction of tic severity by internalizing symptoms, $R^2\text{change} = 0.011, F(3, 52) = 1.08, ns$. Similarly, in the analysis to determine whether externalizing symptoms interacted with tic severity, parent-rated externalizing symptoms significantly predicted parent-rated QoL ($\beta = -.61, p < .001$), but clinician-rated tic severity did not ($\beta = -.08, ns$), see Table 6. Of note, there was a significant contribution for the interaction of parent-rated externalizing scores by tic severity for parent-rated QoL, $R^2\text{change} = 0.059, F(3, 52) = 5.64, p < .05$.

Additional analyses were computed to further explore this finding. Following the statistical strategy described by Holmbeck (2002), the slopes for

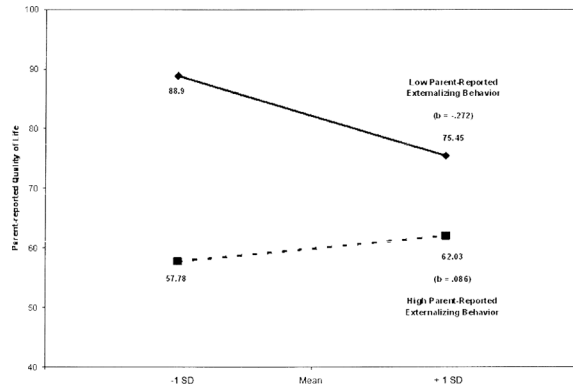


Figure 1. Regression lines for relations between clinician-rated tic severity and parent-reported quality of life as moderated by parent-reported externalizing behavior (a 2-way interaction). *b* = unstandardized regression; SD = standard deviation.

the association between parent-reported QoL and tic severity at high and low levels of parent-reported externalizing behavior were tested for significance using the regression equation from the full sample. As illustrated in Figure 1, at high externalizing behaviors, the association between QoL and tic severity was negative and nonsignificant ($b = -.272, ns$). In contrast, the association was positive and significant ($b = .086, p < .05$) at low levels of externalizing behaviors.

Impact of Comorbidity on QoL

Two one-way analyses of variance were conducted to examine the impact of comorbidity (none, externalizing comorbidity, internalizing comorbidity)

Table 5. Hierarchical Regression Analyses Predicting Parent and Child PedsQL Scores From CBCL Internalizing Scores

Order of Entry	β	r_p	R^2	ΔR^2	$\Delta F (3, 52)$
Child report of quality of life					
Step 1			.339	.339	13.57***
YGTSS Total Score	-.341**	-.39**			
CBCL Internalizing	-.451***	-.48***			
Step 2			.342	.003	0.25
Interaction of YGTSS Total Score and CBCL Internalizing	-.152	-.07			
Parent report of quality of life					
Step 1			.456	.456	22.17***
YGTSS Total Score	-.140	-.19			
CBCL Internalizing	-.651***	-.66***			
Step 2			.467	.011	1.08
Interaction of YGTSS Total Score and CBCL Internalizing	.283	.14			

Note: PedsQL = Pediatric Quality of Life Inventory; CBCL = Child Behavior Checklist; YGTSS = Yale Global Tic Severity Scale. In Step 2, "Interaction" refers to the interaction of YGTSS total score and CBCL Internalizing scores. *** $p < .001$. ** $p < .01$. * $p < .05$.

Table 6. Hierarchical Regression Analyses Predicting Parent and Child PedsQL Scores From CBCL Externalizing Scores

Order of Entry	β	r_p	R^2	ΔR^2	$\Delta F(3, 52)$
Child report of quality of life					
Step 1			.197	.197	6.49**
YGTSS Total Score	-.329*	-.34**			
CBCL Externalizing	-.249*	-.27*			
Step 2			.198	.001	0.05
Interaction of YGTSS Total Score and CBCL Externalizing	-.086	-.03			
Parent report of quality of life					
Step 1			.399	.399	17.60***
YGTSS Total Score	-.082	-.10			
CBCL Externalizing	-.613***	-.63***			
Step 2			.458	.059	5.64*
Interaction of YGTSS Total Score and CBCL Externalizing	.737*	.31*			

Note: PedsQL = Pediatric Quality of Life Inventory; CBCL = Child Behavior Checklist; YGTSS = Yale Global Tic Severity Scale. In Step 2, “Interaction” refers to the interaction of YGTSS total score and CBCL Externalizing scores.
 *** $p < .001$. ** $p < .01$. * $p < .05$.

on the PedsQL and PedsQL–Parent Proxy total scores, see Table 7. Comorbidity was defined as follows: Those with a tic disorder only were classified as “none” ($n = 13$) whereas those with either a comorbid externalizing (e.g., oppositional defiant disorder, attention deficit hyperactivity disorder) or internalizing disorder (e.g., obsessive–compulsive disorder, major depression, generalized anxiety disorder, panic disorder, social phobia) were classified as “externalizing comorbidity” ($n = 10$) and “internalizing comorbidity” ($n = 14$), respectively. Those with both internalizing and externalizing comorbidities were excluded

from this set of analyses. No significant group differences were found for child-rated, $F(2, 34) = 1.33$, *ns*, or parent-rated, $F(2, 34) = 2.69$, *ns*, QoL. Two multivariate analyses of variance were conducted to examine the impact of comorbidity (none, externalizing comorbidity, internalizing comorbidity) on QoL. Separate multivariate analyses of variance were conducted for the PedsQL and PedsQL–Parent Proxy subscales. Although no multivariate main effect of comorbidity was found for the PedsQL subscales, Wilks’s lambda = 0.73, $F(2, 34) = 1.45$, *ns*, there was a significant multivariate main effect of

Table 7. Quality of Life Differences Between Youth Diagnosed With Comorbid Externalizing Disorders and Internalizing Disorders

	Internalizing ^a		Externalizing ^b		No Comorbidity ^c		F Value (3, 55)	p Value
	M	SD	M	SD	M	SD		
Child self-report								
Total score	75.63	12.05	65.95	19.31	76.63	19.23	1.24	0.30
Physical health	82.14	18.07	75.31	14.54	82.69	15.76	0.61	0.61
Psychosocial health	73.45	12.88	62.83	23.16	74.61	21.27	1.56	0.21
Emotional functioning	68.21	22.58	65.00	25.28	75.77	20.50	0.80	0.49
Social functioning	86.43	11.17	58.00	37.95	79.23	25.48	2.67	0.06
School functioning	65.71	18.17	65.50	17.07	68.85	23.64	1.25	0.30
Parent proxy report								
Total score	71.42	15.07	69.19	13.15	81.42	13.06	2.25	0.09
Physical health	79.24	17.23	88.75	15.53	89.90	12.11	1.07	0.37
Psychosocial health	68.81	15.60	62.67	15.24	78.59	14.83	2.92	0.04*
Emotional functioning	62.14	16.49	69.50	25.87	75.00	19.47	0.89	0.45
Social functioning	77.14	16.14	62.50	22.27	80.77	17.78	1.73	0.17
School functioning	67.14	21.81	56.00	12.87	80.00	17.56	6.29	0.001***

Note: Internalizing = comorbid internalizing disorder; externalizing = comorbid externalizing disorder.
^a $n = 14$. ^b $n = 10$. ^c $n = 13$.
 * $p < .05$. ** $p < .01$. *** $p < .001$.

comorbidity for the PedsQL–Parent Proxy subscales, Wilks's $\lambda = 0.56$, $F(2, 34) = 2.79$, $p = .002$. Follow-up analyses of variance revealed significant differences between groups for the PedsQL–Parent Proxy school functioning subscale, $F(2, 34) = 6.29$, $p = .001$. Post hoc testing using a Bonferroni correction revealed significantly lower parent-rated school functioning for children with comorbid externalizing disorders ($M = 56.00$, $SD = 12.87$) as compared to children without any comorbidity ($M = 80.0$, $SD = 17.56$; $p < .05$). Although no other significant differences emerged, children with comorbid externalizing disorders generally had lower QoL scores relative to youth without any comorbidity.

Discussion

This study examined QoL in pediatric tic disorder patients. Generally, results indicated that children with tics experience lower QoL than healthy children in all areas except physical functioning, in which no differences emerged. These results indicate that the presence of tics does have a negative impact on children's lives. However, parents of children with tics generally rated their children's psychosocial functioning higher than did parents of children with other psychiatric disorders. Thus, despite recognizing some psychosocial impairment relative to healthy children, parents may view tics as less distressing and impairing to a child's psychosocial functioning than other emotional and behavioral symptoms (e.g., anxiety, depressive symptoms, externalizing behaviors). The children in this study did not report the same views, however, as children with tics rated their QoL similarly to children with other psychiatric disorders. These findings suggest that children with tics may experience psychosocial distress and functional impairment of which their parents are not aware and are consistent with previous research demonstrating that parents often have difficulty accurately estimating children's internalizing symptoms (Engel, Rodrigue, & Geffken, 1994). Thus, future research should examine the features of tics disorders that are associated with decreased QoL for these patients.

Of note, there were several discrepancies observed among parent and child reports of children's QoL in this study. Results demonstrated strong positive correlations among child and parent reports of the child's QoL in the younger children (i.e., 8–11-year-olds); however, parent and adolescent (i.e., 12–17 year olds) reports were generally not related to one another. These findings may be better understood when developmental considerations are taken

into account. For example, as children mature, their independence increases and they begin to spend less time with parents. In addition, younger children typically volunteer more information to parents about their daily well-being than do adolescents. Thus, parents of younger children may spend more time with their child and communicate more regarding his or her QoL. Parents of teenagers, in contrast, may be rating the adolescent's QoL based solely on their more limited observations of the teen. It is also noteworthy that parents generally rated the younger children's QoL as higher than their child's ratings, whereas parents generally rated the adolescents' QoL as being more negatively affected by their tic disorder than the youth endorsed. These findings have important clinical implications, as they suggest that parents may underestimate or overestimate the impact of a chronic tic disorder on their child's functioning, depending on the child's developmental level. This could lead to undertreatment of children's distress or adolescent's frustration due to parents' unwarranted concerns regarding their symptoms. Taken together, these results suggest that both parent and child reports of QoL in tic patients should be obtained whenever possible, as the two provide relatively different perspectives and both are likely needed to obtain a complete picture of the child's functioning.

In general, tic severity was inversely associated with QoL, though clinician ratings of tic severity were more strongly related to children's self-reported QoL than parent reports of child QoL. It is reasonable to speculate that tic presentations that specifically impact physical well-being (e.g., jerking body parts, tightening muscles) or draw negative attention by others (e.g., loud phonic tics or complex motor tics) may negatively impact QoL. A perceived lack of tic control may also negatively affect QoL. In addition, parent-rated internalizing and externalizing symptoms were significantly related to both child- and parent-rated QoL. These data suggest that children and parents positively link psychiatric symptoms and QoL or, stated alternatively, that they recognize that QoL is impaired by psychiatric symptomatology. Thus, we examined whether there was an interaction between tic severity and internalizing or externalizing symptom severity in predicting QoL. When predicting child-reported QoL, the results were not significant. However, tic severity was significantly related to parent-rated QoL in children with low externalizing symptoms and unrelated to parent-rated QoL in children with high externalizing symptoms. Parent-rated QoL scores were generally lower in children with high externalizing symptoms than children with low externalizing symptoms. Thus, it is possible that parents view

externalizing symptoms as more problematic and disruptive than tics. These results may help to explain why previous research has failed to demonstrate that tics contribute significantly to parent- or teacher-rated functional impairment when controlling for comorbid attention deficit hyperactivity disorder (Sukhodolsky et al., 2003), as parents may be more sensitive to the negative effects of disruptive behaviors than tics. Future research should examine whether child ratings of disruptive behaviors demonstrate divergent results. These findings also have clinical implications. Given the greater negative outcomes reportedly associated with externalizing symptoms, it is likely that children with comorbid tics and externalizing disorders would benefit from undergoing treatment for the externalizing disorder before focusing on the tics. In contrast, children without comorbid difficulties would likely benefit most from interventions primarily targeting the tics.

Some limitations of this study should be noted. First, the sample size was relatively small, predominantly boys, and consisted primarily of White youth. As a result, it is unclear whether the results would generalize to a more diverse population. In addition, given the modest sample size, statistical power was not adequate to detect small effect sizes and we did not correct for Type I error on several sets of analyses. Second, the comparison groups (psychiatric controls and healthy controls) came from previously reported research studies. It is possible that outside factors (e.g., demographic differences, recruitment methods, treatment status, satisfaction with patient care, and so on) may have influenced the QoL ratings, beyond the influence of group differences (e.g., tic patients, psychiatric controls, or healthy controls). Third, analyses examining the contribution of comorbid internalizing or externalizing symptoms were based on a sample of youth with tics who primarily fell below the clinical cutoff for these conditions. It is possible that results may not generalize to youth who meet clinical criteria for a tic disorder and an internalizing or externalizing disorder. Fourth, our rates of oppositional defiant disorder (6/59) were lower than other reports in pediatric tic samples (e.g., Budman et al., 2000; Sukhodolsky et al., 2003). It is possible that children with comorbid oppositional defiant disorder were not presenting for treatment and thus are not fully represented in this study. It is conceivable that those youth with comorbid oppositional defiant disorder may have lower QoL relative to those without this comorbidity. Fifth, diagnoses were based on an unvalidated semistructured clinical interview and interrater agreement was not assessed. On balance, diagnoses were confirmed by a second clinician and all clinicians had considerable experience in childhood psy-

chiatric conditions. Finally, regression analyses that included parent-only indexes may be inflated by shared method variance.

In conclusion, this study presents preliminary findings related to QoL in youth with tic disorders. In general, children and adolescents with tics report more distress and impairment across domains of functioning than do healthy children. However, the impact of tics on QoL, particularly in young children, may not be adequately appreciated by parents. Rather, it appears that parents are more likely to recognize the influence of externalizing symptoms in their children. Results have implications for clinicians working with pediatric tic disorder patients. For example, children with comorbid externalizing symptoms may benefit from treatment targeting those symptoms first. In addition, children with tics may benefit from talking to their parents or mental health care providers about the ways in which the tic disorder interferes with their functioning. Then, the children would be more likely to obtain interventions (e.g., habit reversal therapy, social skills training) that could help them develop skills to minimize the negative impact of the disorder.

References

- Achenbach, T. M. (1991). *Manual for the child behavior checklist/4-18 and 1991 Profile*. Burlington: University of Vermont, Department of Psychiatry.
- American Psychiatric Association. (2000). *Diagnostic and statistical manual of mental disorders* (4th ed., text rev.). Washington, DC: Author.
- Baron, R. M. & Kenny, D. A. (1986). The moderator-mediator variable distinction in social psychological research: Conceptual, strategic, and statistical considerations. *Journal of Personality & Social Psychology*, *52*, 1173-1182.
- Bastiaansen, D., Koot, H. M., Bongers, I. L., Varni, J. W., & Verhulst, F. C. (2004). Measuring quality of life in children referred for psychiatric problems: Psychometric properties of the PedsQL 4.0 generic core scales. *Quality of Life Research*, *13*, 489-495.
- Boudjouk, P. J., Woods, D. W., Miltenberger, R. G., & Long, E. S. (2000). Negative peer evaluation in adolescents: Effects of tic disorders and trichotillomania. *Child & Family Behavior Therapy*, *22*, 17-28.
- Bruun, R. D. (1988). The natural history of Gilles de la Tourette's Syndrome. In D. J. Cohen, R. D. Bruun, & J. F. Leckman (Eds.), *Gilles de la Tourette's Syndrome and related tic disorders* (pp. 22-38). New York: Wiley InterScience.
- Budman, C. L., Bruun, R. D., Park, K. S., Lesser, M., & Olson, M. (2000). Explosive outbursts in children with Tourette's disorder. *Journal of the American Academy of Child & Adolescent Psychiatry*, *39*, 1270-1276.
- Carter, A. S., O'Donnell, D. A., Schultz, R. T., Scchill, L., Leckman, J. F., & Pauls, D. L. (2000). Social and emotional adjustment in children affected with Gilles de la Tourette's Syndrome: Associations with ADHD and family functioning. *Journal of Child Psychology & Psychiatry*, *41*, 215-223.

- Coffey, B. J., Biederman, J., Geller, D., Frazier, J., Spencer, T., Doyle, R., et al. (2004). Reexamining tic persistence and tic-associated impairment in Tourette's disorder. *Journal of Nervous & Mental Disease, 192*, 776–780.
- Coffey, B. J., Biederman, J., Smoller, J. W., Geller, D. A., Sarin, P., Schwartz, S., et al. (2000). Anxiety disorders and tic severity in juveniles with Tourette's disorder. *Journal of the American Academy of Child & Adolescent Psychiatry, 39*, 562–568.
- Coffey, B. J. & Park, K. S. (1997). Behavioral and emotional aspects of Tourette syndrome. *Neurology Clinics, 15*, 277–289.
- Cohen, D. J., Ort, S. I., Leckman, J. F., Riddle, M. A., & Hardin, M. T. (1988). Family functioning and Tourette's syndrome. In D. Cohen, R. Brunn, & J. Leckman (Eds.), *Tourette's syndrome and tic disorders* (pp. 170–196). New York: Wiley.
- Comings, D. E. & Comings, B. G. (1985). Tourette syndrome: Clinical and psychological aspects of 250 cases. *American Journal of Human Genetics, 35*, 435–450.
- Elstner, K., Selai, C. E., Trimble, M. R., & Robertson, M. M. (2001). Quality of life (QOL) of patients with Gilles de la Tourette's Syndrome. *Acta Psychiatrica Scandinavica, 103*, 52–59.
- Engel, N. A., Rodrigue, J. R., & Geffken, G. R. (1994). Parent-child agreement on ratings of anxiety in children. *Psychological Reports, 75*, 1251–1260.
- Ernberg, G., Cruse, R. P., & Rothner, D. A. (1987). The natural history of Tourette's syndrome: A follow-up study. *Annals of Neurology, 22*, 383–385.
- Holmbeck, G. N. (2002). Post-hoc probing of significant moderational and mediational effects in studies of pediatric populations. *Journal of Pediatric Psychology, 7*, 87–96.
- King, R. A. & Scahill, L. (2001). Emotional and behavioral difficulties associated with Tourette syndrome. In D. J. Cohen, C. G. Goetz, & J. Jankovic (Eds.), *Tourette syndrome* (pp. 79–88). Philadelphia, PA: Lippincott, Williams, & Wilkins.
- Kurlan, R., Daragjati, C., Como, P. G., McDermott, M. P., Trinidad, K. S., Roddy, S., et al. (1996). Non-obscene complex socially inappropriate behavior in Tourette's syndrome. *Journal of Neuropsychiatry and Clinical Neurosciences, 8*, 311–317.
- Leckman, J. F., Riddle, M. A., Hardin, M. T., Ort, S. I., Swartz, K. L., Stevenson, J., et al. (1989). The Yale Global Tic Severity Scale: Initial testing of a clinician-rated scale of tic severity. *Journal of the American Academy of Child & Adolescent Psychiatry, 28*, 566–573.
- Leckman, J. F., Sholomskas, D., Thompson, W. D., Belanger, A., & Weissman, M. M. (1982). Best estimate of lifetime psychiatric diagnosis: A methodological study. *Archives of General Psychiatry, 39*, 879–883.
- Muldoon, M. F., Barger, S. D., Flory, J. D., & Manuck, S. B. (1998). What are quality of life measurements measuring? *British Medical Journal, 316*, 542–545.
- Robertson, M. M. (1995). The relationship between Gilles de la Tourette's Syndrome and obsessive compulsive disorder. *Journal of Serotonin Research, 1*(Suppl. 1), 49–62.
- Robertson, M. M., Banerjee, S., Eapen, V., & Fox-Hiley, P. (2002). Obsessive compulsive behaviour and depressive symptoms in young people with Tourette syndrome: A controlled study. *European Child and Adolescent Psychiatry, 11*, 261–265.
- Robertson, M. M., Trimble, M. R., & Lees, A. J. (1988). The psychopathology of the Gilles de la Tourette: A phenomenological analysis. *British Journal of Psychiatry, 152*, 383–390.
- Singer, H. S. (2005). Tourette's syndrome: From behaviour to biology. *Lancet Neurology, 4*, 149–159.
- Stefl, M. E. (1983). *The Ohio Tourette study*. Cincinnati: School of Planning, University of Cincinnati.
- Stefl, M. E. (1984). Mental health needs associated with Gilles de la Tourette's Syndrome. *American Journal of Public Health, 74*, 1310–1313.
- Storch, E. A., Murphy, T. K., Fernandez, M., Krishnan, M., Geffken, G. R., Kellgren, A. R., et al. (2007). Factor analytic study of the Yale Global Tic Severity Scale. *Psychiatry Research, 149*, 231–237.
- Storch, E. A., Murphy, T. K., Geffken, G. R., Sajid, M., Allen, P., Roberti, J. W., et al. (2005). Reliability and validity of the Yale Global Tic Severity Scale. *Psychological Assessment, 17*, 486–491.
- Sukhodolsky, D. G., Scahill, L., Zhang, H., Peterson, B., Kind, R. A., Lombroso, P. J., et al. (2003). Disruptive behavior in children with Tourette's syndrome: Association with ADHD comorbidity, tic severity, and functional impairment. *Journal of the American Academy of Child & Adolescent Psychiatry, 42*, 98–105.
- Thibert, A. L., Day, H. I., & Sandor, P. (1995). Self-concepts and self-consciousness in adults with Tourette's syndrome. *Canadian Journal of Psychiatry, 40*, 35–39.
- Varni, J. W., Burwinkle, T. M., Jacobs, J. R., Gottschalk, M., Kaufman, F., & Jones, K. L. (2003). The PedsQL in Type 1 and Type 2 diabetes. *Diabetes Care, 26*, 631–637.
- Varni, J. W., Burwinkle, T. M., Rapoff, M. A., Kamps, J. L., & Olson, N. (2004). The PedsQL in pediatric asthma: Reliability and validity of the Pediatric Quality of Life Inventory generic core scales and asthma module. *Journal of Behavioral Medicine, 27*, 297–318.
- Varni, J. W., Burwinkle, T. M., Seid, M., & Skarr, D. (2003). The PedsQL 4.0 as a pediatric population measure: Feasibility, reliability, and validity. *Ambulatory Pediatrics, 3*, 329–331.
- Varni, J. W., Seid, M., & Kurtin, P. S. (2001). PedsQL 4.0: Reliability and validity of the Pediatric Quality of Life Inventory version 4.0 generic core scales in healthy and patient populations. *Medical Care, 39*, 800–812.
- Varni, J. W., Seid, M., & Rode, C. A. (1999). The PedsQL: Measurement model for the Pediatric Quality of Life Inventory. *Medical Care, 37*, 126–139.
- Wilkinson, B. J., Newman, M. B., Shytle, R. D., Silver, A. A., Sanberg, P. R., & Sheehan, D. (2001). Family impact of Tourette's syndrome. *Journal of Child and Family Studies, 10*, 477–483.
- Williams, J., Wake, M., Hesketh, K., Maher, E., & Waters, E. (2005). Health-related quality of life of overweight and obese children. *Journal of the American Medical Association, 293*, 70–76.
- Woods, D. W., Fuqua, R. W., & Outman, R. C. (1999). Evaluating the social acceptability of persons with habit disorders: The effects of topography, frequency, and gender manipulation. *Journal of Psychopathology and Behavioral Assessment, 22*, 1–18.

Received June 7, 2006

Accepted October 31, 2006