CrossMark

ORIGINAL ARTICLE

The Impact of Tic Severity, Comorbidity and Peer Attachment on Quality of Life Outcomes and Functioning in Tourette's Syndrome: Parental Perspectives

Deirdre O'Hare[1] · Edward Helmes[1] · Valsamma Eapen[2,4] ·
Rachel Grove[2] · Kerry McBain[1] · John Reece[3]

Abstract The aim of this controlled, community-based study based on data from parents of youth (aged 7–16 years) with Tourette's syndrome (TS; n = 86) and parents of age and gender matched peers (n = 108) was to test several hypotheses involving a range of variables salient to the TS population, including peer attachment, quality of life, severity of tics, comorbidity, and psychological, behavioural and social dysfunction. Multivariate between-group analyses confirmed that TS group youth experienced lower quality of life, increased emotional, behavioural and social difficulties, and elevated rates of insecure peer attachment relative to controls, as reported by their primary caregiver. Results also confirmed the main hypothesis that security of peer attachment would be associated with individual variability in outcomes for youth with TS. As predicted, multivariate within-TS group analyses determined strong relationships among adverse quality of life outcomes and insecure attachment to peers, increased tic severity, and the presence of comorbid disorder. Findings suggest that youth with TS are at increased risk for insecure peer attachment and that this might be an important variable impacting the quality of life outcomes for those diagnosed.

Keywords Tourette syndrome · Quality of life · Peer attachment · Comorbidity · Tic severity

✉ Valsamma Eapen
v.eapen@unsw.edu.au

[1] School of Arts and Social Sciences, James Cook University, Townsville, QLD, Australia

[2] School of Psychiatry, University of New South Wales & Ingham Institute, Sydney, NSW, Australia

[3] School of Psychological Sciences, Australian College of Applied Psychology, Melbourne, Australia

[4] Academic Unit of Child Psychiatry, South Western Sydney Local Health District (AUCS), Liverpool Hospital, Mental Health Centre (Level 1: ICAMHS),
Locked Bag 7103, Liverpool, Sydney, NSW 2170, Australia

Introduction

Tourette syndrome (TS) is a childhood onset neurodevelopmental disorder characterised by the presence of two or more motor tics and at least one vocal tic lasting for more than a year [1, 2]. The high rates (80–90 %) of comorbidity found in clinic and community-based TS populations, in particular attention deficit hyperactivity disorder and obsessive compulsive disorder (OCD), contribute to its heterogeneous presentation [3, 4]. Although largely unknown, evidence is emerging for aetiological links between these comorbid disorders and TS occurring at the level of the neural substrate, suggesting that comorbidity is a central feature of the syndrome [5]. Other commonly identified psychopathologies include anxiety, mood and conduct disorders, impulsivity, aggression, learning (LD) and autistic spectrum disorders (ASD) [4, 6, 7].

When compared with results for healthy peers and normative data, TS has been consistently associated with impaired quality of life (QoL) and increased symptoms of maladaptive psychological, behavioural and social functioning [5, 8–10]. Findings from paediatric TS QoL studies universally demonstrate impaired global QoL and reduced psychosocial functioning, with the strongest adverse impact in academic [11], social [9, 12] and emotional domains [13]. Individual differences in outcomes for individuals diagnosed with TS have been demonstrated.

Traditionally, these have been explained by increased tic severity and the presence of comorbidity. Most recent QoL research has demonstrated elevations in adverse outcomes associated with increased tic severity [5, 12] and comorbidity [3, 8, 11, 12, 14–16], particularly when seen in combination [5, 8, 12]. However, a substantial proportion of variance in QoL and functional outcomes remained unexplained [8].

Differential diagnosis in the context of TS is a challenging and protracted process, even for highly experienced clinicians [17, 18]. The majority of TS research has been conducted on clinic-based samples that have better access to comprehensive psychiatric evaluation. Specialist care cannot be guaranteed for community-drawn TS samples. Many TS researchers, therefore, also include psychometric measures to screen for symptoms of psychopathology (e.g., the Child Behaviour Checklist—CBCL [19]), along with various instruments to assess tic severity (e.g. the Parent Tic Questionnaire [20]).

A small number of TS studies point toward another potentially important variable that may affect outcomes; the quality of the close relationships experienced by youth with TS. As demonstrated in prior QoL studies, TS has pronounced negative impacts on social functioning. Others have found that those diagnosed are frequently stigmatised and socially isolated [8, 18]. Peer relationships and family functioning have been identified as key factors in determining the wellbeing and functioning of children and teens with TS [21–25]. Relationship factors have also been shown to moderate the impact of tic severity and comorbidity on outcomes. For example, Storch et al. [26] found that peer victimisation mediated the relationship between tic severity and loneliness. Carter and colleagues [27] also found that positive family functioning was associated with improved outcomes, even for those with more severe tics and comorbid diagnoses.

Within the broader TS population, young people have been found to experience the highest levels of psychosocial stress in the context of their friendships with peers [28]. Problems encountered in peer relationships include experiencing difficulty making and maintaining friendships [29–31], being subjected to negative attitudes and behaviours from peers [8, 18, 26] and having reduced social skills and fewer opportunities to socialise [32–34]. Clinical characteristics such as coprolalia, impulsiveness, obsessive behaviours and aggression also contribute to interpersonal difficulties, and can result in the child's voluntary social withdrawal [35]. Based on these findings it appears that the ability to form close relationships with peers may be an important factor in determining QoL and functioning of children and adolescents with TS.

Attachment theory has been increasingly employed as a conceptual framework within which all close relationships are examined. Attachment theory proposes that children develop a style of attachment during interactions with their primary caregiver that remains stable across time and future relationships [36–40]. Based upon the observed behaviour of children, Ainsworth [38] provided a system for classifying secure and insecure attachment styles.

An extensive body of research has subsequently demonstrated the importance of secure attachment to the healthy psychological, behavioural, social and physical development of the child [40–44]. There is also growing evidence of the importance of child's ability to successfully transfer attachment from parents to peers, with secure peer attachment playing a pivotal role in optimising the child's psychosocial functioning, particularly by adolescence [42–44]. Secure attachment has been most powerfully related to the increased social competence and ability of the child to regulate their emotions, both of which are key factors in determining adjustment and socioemotional wellbeing [40, 41]. The quality of close relationships in youth with TS may, therefore, be an important factor in understanding QoL outcomes for these individuals.

The main aim of the current study was to examine the parent's perspective of the impact of peer attachment security on the quality of life and psychological, behavioural and social functioning of a community-based sample of youth with TS.

The following hypotheses were investigated:

Hypothesis One That youth with TS would experience lower quality of life and increased symptoms of psychological, behavioural and social dysfunction relative to controls.

Hypothesis Two That insecure peer attachment would be associated with lower quality of life and increased symptoms of psychological, behavioural and social dysfunction, and that youth with TS would experience higher rates of insecure peer attachment relative to controls.

Hypothesis Three That increased tic severity and symptoms of comorbid disorder would be associated with lower quality of life and increased psychological, behavioural and social dysfunction for individuals with TS.

Hypothesis Four That secure peer attachment would moderate the relationships between tic severity and comorbidity on quality of life outcomes for youth with TS.

Understanding the impact of security of peer attachment, tic severity and comorbidity on measured outcomes has the potential to substantially improve therapeutic interventions for individuals with TS.

Method

A survey-based methodology was adopted for the current controlled study, with responses restricted to parental reports due to the ethical and practical issues associated with surveying children as young as seven. To minimise ascertainment bias inherent in TS studies employing clinic-based samples, the current study recruited a nation-wide community sample. As undertaken in prior TS research, recruitment was facilitated by the support of national and state TS associations.

Response rate could not be calculated for the TS group due to restricted information maintained on member databases. Inability to accurately record survey distribution for controls over multiple Australia-wide locations precluded response rate calculation. Written informed consent was required, participation was voluntary, confidentiality was guaranteed, and the study was conducted with the approval of the James Cook University Human Research Ethics Committee.

Participants

A total sample ($n = 194$) consisting of two groups was recruited for the current study. The TS group ($n = 86$) comprised parents of youth (7–16 years) formally diagnosed with TS by a medical practitioner. The control group ($n = 108$) contained parents of age and gender matched peers with no known psychiatric or medical diagnosis. Between group differences in demographics were assessed with ANOVAs or χ^2 with results revealing increased variability in racial diversity for the TS group relative to controls, $\chi^2(3, N = 193) = 12.02$, $p = .007$. Demographics are provided in Table 1.

Procedure

Following email and Internet advertising, survey packs were mailed to all members on the databases of Tourette Syndrome Association of Australia (TSAA) and the Tourette Syndrome Association of Victoria (TSAV). Following recruitment of the TS group, control group participants were recruited from multiple Australia-wide sites by localised advertising, Internet advertising, and emails via the TSAA. Hard copy invitations and survey packs were also distributed by research assistants in various locales including the TSAA, TSAV, and among James Cook University faculty and staff. No incentives for participation were offered.

Measures

Parents were administered the "Australian Tourette Survey", a pen and paper instrument assembled for the purposes of the study. All participants were invited to complete the four psychometric measures included in the survey to address demographic questions. Measures were selected on the basis of their employment in similar prior published research, suitability for use in a survey-based study and proven psychometric strength.

Pediatric Quality of Life Inventory (PedsQL)

The widely employed 15 item Pediatric Quality of Life Inventory (PedsQL) [45] was selected to assess health-related quality of life. Parents were asked to rate how often a particular item had been a problem during the past month using a 5-point Likert scale ranging from ''never a problem'' to ''almost always a problem''. Total scores are linearly transformed to a scale of 0–100, with higher scores indicating better quality of life. Four subscales are also calculated to measure physical, emotional, social and school functioning. A psychosocial summary scale score is calculated by summing the emotional, behavioural and social subscales. The reliability and validity of the PedsQL has been assessed, indicating good internal consistency and construct validity [45–47] and demonstrated good to excellent internal consistency for all summary and subscales in the current study (Cronbach's $\alpha = .86–.92$).

The Parent Tic Questionnaire (PTQ)

The Parent Tic Questionnaire (PTQ) [20] was selected to provide a measure of tic severity. This parent-report measures the presence, frequency and intensity of 14 motor and 14 vocal tics during the previous week. To provide consistency with the one-month period required by the PedsQL and to minimise potential for participant error, the observation period for the PTQ was extended to 1 month. Presence of a tic is reported (yes or no), and frequency and intensity are rated on Likert scales ranging from 1 to 4, with greater scores indicating greater frequency and stronger intensity. A score for each tic was calculated by combining the frequency and intensity ratings. Motor and vocal tics were calculated separately as well as a total score computed. The PTQ has demonstrated acceptable internal consistency, temporal stability and convergent and discriminant validity [20] in clinical samples, adequate internal consistency in a community sample, indicating adequate results [8] and good internal consistency for all summary and subscales in the current study (Cronbach's $\alpha = .82–.88$).

Table 1 Demographic characteristics and between group differences

	Control (n = 108)			TS (n = 86)		
	f	%	M (SD)	f	%	M (SD)
Age (years) child/adolescent			11.30 (2.58)			11.44 (2.78)
Parent's relationship to child/adolescent						
Biological mother	97	88.8		77	90.6	
Adoptive mother	2	1.9		1	1.2	
Biological father	9	8.3		7	8.2	
Place of family residence						
City	67	62		50	50	
Regional	33	30.6		20	23.8	
Rural-remote	8	7.4		14	16.7	
Parent's marital status						
Never married	6	5.6		1	1.2	
Married	88	82.2		72	83.7	
Separated/divorced	12	11.2		10	11.6	
Widowed	1	0.9		3	3.5	
Family income						
Low	6	5.6		8	9.8	
Low-middle	26	24.1		19	23.2	
Middle-above	76	70.4		55	76.1	
Gender of child/adolescent						
Male	79	73.1		71	85.4	
Female	29	26.9		12	2.4	
Ethnicity of child/adolescent**						
Caucasian	107	99.1		74	87.1	
A/TSI	0	0		2	2.4	
Asian	0	0.9		3	3.5	
Other	1			6	7.1	
Child has a sibling?						
Yes	101	93.5		76	88.4	
No	7	6.5		10	11.6	

A/TSI = Aboriginal/Torres Strait Islander. Frequencies for TS group do not always sum to 86 because of missing data

** $p < .01$. Between group differences analyzed using χ^2 or ANOVA

The Strengths and Difficulties Questionnaire (SDQ)

The study screened for symptoms of major paediatric psychiatric disorders, behavioural and social difficulties using the Strengths and Difficulties Questionnaire (SDQ) [48]. This is a widely utilised questionnaire (suitable for children aged 4–16 years), which consists of 25 items across five subscales that assesses emotional symptoms, conduct problems, hyperactivity/inattention, peer problems, and prosocial behaviour. A total summary score is calculated, combining the 20 items assessing emotional, conduct, and inattention and peer problems. The SDQ has demonstrated robust psychometric properties and strong internal consistency [49], which was good to excellent for the total difficulties score and all subscales employed in the current study (Cronbach's α = .75–.87), with the exception of the prosocial scale, which was poor (α = .41).

Whilst not a diagnostic tool, the SDQ has been extensively employed to screen for the presence of subclinical and clinical level symptoms of paediatric psychopathology. It correlates strongly with longer form measures such as the Child Behaviour Checklist (CBCL) [50], which has been used to identify symptoms of psychopathology in prior published TS studies, including recent quality of life research by Storch et al. [15].

Attachment Questionnaire for Children (AQC)

Security of peer attachment was assessed via the Attachment Questionnaire for Children (AQC) [51]. As no

continuous measure of attachment suitable for use in middle childhood was able to be located at the time of the current research design, the categorical AQC was employed. This single item measure is a recent adaptation of Hazan and Shaver's [39] self-reported adult attachment typology. Participants were given a choice of three narrative descriptions of feelings and perceptions about their child's relationship with other children, mapping onto either secure, insecure-avoidant or insecure-ambivalent attachment styles. Parents were asked to determine which narrative best matched their child's peer attachment style. To test the main hypothesis and maintain statistical power in the current study, the three categories of the AQC were dichotomised by collapsing "insecure avoidant" and "insecure ambivalent" into a single "insecure" category. Both Hazan and Shaver's typology and the AQC have been successfully employed in multiple published studies and have been found to demonstrate acceptable reliability and validity in the context of categorical measurement [42, 51, 52]. While establishing the reliability of any single item measure can be challenging, inter-rater reliability for the AQC has been shown to be acceptable [53].

Results

Data Screening and Assumption Testing

Data were entered into a single SPSS 20 spreadsheet for analysis. Imputation of missing values was unnecessary due to the absence of missing data. Levene's test revealed adequate homogeneity for all variables. Distance measures did not identify any outliers demanding deletion. Normality was satisfactory for all variables apart from the PTQ, which was successfully corrected by applying a Log 10 transformation. A per comparison critical significance level of $\alpha = .01$ was applied to protect against family-wise error.

Hypothesis One Differences between the TS and control groups in quality of life (PedsQL) and psychological, behavioural, and social dysfunction (SDQ).

Differences in PedsQL and SDQ outcomes for the TS and control groups were examined using two single-factor between-subjects multivariate analyses of variance (MANOVAs). The single between-subjects factor was group (TS vs. Control), with sub-scales of the PedsQL and SDQ forming the combined dependent outcomes for the two analyses. Where significant differences were determined, follow-up univariate ANOVAs were conducted for each of the subscales. The total scores for each scale were analysed separately by univariate ANOVAs.

Results confirmed the hypotheses that youth with TS would experience lower quality of life and higher levels of

psychological, behavioural and social dysfunction than control group peers. Results of the MANOVA indicated significant between group differences in the combined PedsQL subscales, $\Lambda = .57$, $F(5, 188) = 27.87$, $p < .001$, $\eta_p^2 = .43$. Subsequent univariate ANOVAs found that the TS group reported significantly lower quality of life on all sub-scales and the total score of the PedsQL (see Table 2). Results for the SDQ MANOVA indicated significant between group differences on the combined SDQ sub-scales, $\Lambda = .42$, $F(5, 188) = 52.42$, $p < .001$, $\eta_p^2 = .58$ with ANOVAs revealing significantly higher dysfunction for the TS group on all sub-scales and the total score (see Table 2).

Hypothesis Two The impact of peer attachment on quality of life (PedsQL) and psychological, behavioural and social difficulties (SDQ), and between-group differences in rates of insecure peer attachment.

Youth with TS were found to experience a higher rate of insecure peer attachment than controls. Within the TS group, 57 % reported secure peer attachment, compared with 94 % of controls, $\chi^2(1, N = 194) = 36.46$, $p < .001$. Moreover, 38 % of TS children reported difficulty when making friends, compared with 4 % of the control sample $\chi^2(1, N = 194) = 37.34$, $p < .001$.

The very small number of participants in the control group demonstrating insecure attachment ($n = 7$) meant that the analyses comparing participants with secure versus insecure attachment styles needed to focus on the TS group only, given the better balance of participants across the two attachment style groups. Two single-factor between-subjects MANOVAs were conducted with secure versus insecure peer attachment as the between-subjects factor and the subscales of the PedsQL and SDQ as the combined dependent outcomes. Single-factor between-subjects ANOVAs were also conducted to determine significant differences in total PedsQL and SDQ scores. These results are presented in Table 3.

Both MANOVAs were found to be significant: PedsQL, $\Lambda = .62$, $F(4, 81) = 12.70$, $p < .001$, $\eta_p^2 = .39$, and SDQ, $\Lambda = .53$, $F(5, 80) = 14.00$, $p < .001$, $\eta_p^2 = .47$. Only three of the univariate outcomes were not significant at $\alpha = .01$: the physical functioning sub-scale of the PedsQL, and the conduct problems and prosocial subscales of the SDQ.

Hypothesis Three Quality of life (PedsQL) and psychological, behavioural and social difficulties (SDQ) relate to increased tic severity and comorbidity.

Tic Severity

Table 4 provides the correlations between tic severity (motor, vocal, and total) and the sub-scales and total scores

Table 2 Group differences in quality of life outcomes and emotional and behavioural functioning

Outcome measure	Groups				ANOVA		
	TS (n = 86)		Control (n = 108)				
	M	SD	M	SD	F	p	η_p^2
Pediatric quality of life inventory							
Physical functioning	79.91	23.57	91.85	12.63	22.93	<.001	.11
Emotional functioning	49.56	21.62	74.67	16.23	85.29	<.001	.31
Social functioning	60.76	25.46	87.73	14.92	84.74	<.001	.31
School functioning	40.41	28.34	74.07	19.87	94.26	<.001	.33
Psychosocial summary	50.17	19.50	78.43	13.53	141.37	<.001	.42
Total score	59.58	18.06	82.84	11.37	116.98	<.001	.38
Strengths and difficulties questionnaire							
Emotional symptoms	4.27	2.52	0.87	1.28	148.77	<.001	.44
Conduct problems	2.73	2.20	0.68	1.06	72.14	<.001	.27
Hyperactivity/inattention	6.74	2.64	2.25	1.98	183.35	<.001	.49
Peer problems	3.05	2.41	0.87	1.37	62.91	<.001	.25
Prosocial skills	7.52	2.39	8.47	1.73	10.29	.02	.05
Total difficulties	16.80	6.98	4.68	3.94	232.69	<.001	.55

df (1, 192) for all analyses

Table 3 Impact of peer attachment on quality of life and emotional difficulties for TS group (n = 86)

Outcome measure	Attachment style				ANOVA		
	Secure (n = 49)		Insecure (n = 37)				
	M	SD	M	SD	F	p	η_p^2
Pediatric quality of life inventory							
Physical functioning	84.28	20.33	72.43	26.05	5.6	.02	.06
Emotional functioning	56.12	19.28	40.87	21.73	11.8	.001	.12
Social functioning	74.32	19.53	42.79	20.98	51.5	<.001	.38
School functioning	47.62	31.50	30.86	20.21	8.0	.006	.09
Psychosocial summary	59.03	17.40	38.45	15.69	32.1	<.001	.28
Total score	67.44	15.21	49.77	18.69	26.2	<.001	.28
Strengths and difficulties questionnaire							
Emotional symptoms	3.38	2.21	5.43	2.42	16.4	<.001	.16
Conduct problems	2.49	2.36	3.05	1.96	1.4	.24	.02
Hyperactivity/inattention	5.98	2.78	7.75	2.07	16.6	.002	.11
Peer problems	1.75	1.79	4.48	2.03	52.9	<.001	.39
Prosocial skills	7.93	2.03	6.97	2.72	3.54	.06	.04
Total difficulties	13.61	6.21	21.00	5.62	32.3	<.001	.28

df (1, 84) for all analyses

for both the PedsQL and the SDQ. At the multivariate level, a significant relationship was found between the combined three measures of tic severity and the combined sub-scales both the PedsQL, $\Lambda = .60$, $F(12, 209.31) = 3.73$, $p < .001$, $\eta_p^2 = .16$, and the SDQ, $\Lambda = .41$, $F(18, 192.82) = 3.99$, $p < .001$, $\eta_p^2 = .26$.

Thirty of the 36 bivariate correlations were found to be significant at $\alpha < .01$. All correlations were in the expected direction, in that higher tic severity was associated with lower quality of life and higher levels of psychological, behavioural and social difficulties. Notably, the overall pattern of relationships was stronger for vocal tics than motor tics. All correlations involving motor tics were significant, with nine of 12 significant at $p < .001$, whereas five of the 12 correlations involving motor tics were not significant.

Table 4 Correlation between tic severity, quality of life and emotional functioning for the TS group (n = 86)

	Motor tics		Vocal tics		Total tics	
	r	p	r	p	r	p
Pediatric quality of life inventory						
Physical functioning	−.25	.02	−.29	.006	−.31	.004
Emotional functioning	−.36	.001	−.43	<.001	−.45	<.001
Social functioning	−.27	.01	−.50	<.001	−.43	<.001
School functioning	−.37	<.001	−.39	<.001	−.43	<.001
Psychosocial summary	−.43	<.001	−.55	<.001	−.56	<.001
Total score	−.41	<.001	−.53	<.001	−.54	<.001
Strengths and difficulties questionnaire						
Emotional symptoms	.34	.001	.33	.002	.38	<.001
Conduct problems	.16	.15	.50	<.001	.37	.001
Hyperactivity/inattention	.27	.01	.35	.001	.35	.001
Peer Problems	.31	.003	.51	<.001	.47	<.001
Prosocial skills	.07	.52	.39	<.001	−.17	.12
Total difficulties	.38	<.001	.59	<.001	.55	<.001

Comorbidity

Table 5 provides the descriptive and inferential results for the PedsQL and the SDQ comparing participants in the TS group who reported a comorbid disorder versus those with no reported comorbid disorder. MANOVAs found significant differences between those participants with TS who reported a comorbid disorder and those who didn't on both the combined PedsQL sub-scales, $\Lambda = .81$, $F(4, 81) = 4.72$, $p = .002$, $\eta_p^2 = .19$, and the combined SDQ sub-scales, $\Lambda = .86$, $F(5, 80) = 2.58$, $p = .03$, $\eta_p^2 = .14$.

Univariate results are reported in Table 5. Two of the four PedsQL sub-scales plus the total score showed a significant result in the expected direction (i.e., participants with a comorbid disorder demonstrated poorer functioning). No significant differences were found for the physical functioning or social functioning sub-scales.

A significant result at $\alpha = .01$ was found for only one of the SDQ sub-scales, emotional symptoms, and the total SDQ was significant. In both cases, participants with a comorbid disorder were found to have a higher level of problems.

Table 5 Impact of comorbidity on quality of life and emotional difficulties for TS group (n = 86)

Outcome measure	Comorbid disorder				ANOVA		
	Present (n = 63)		Absent (n = 23)				
	M	SD	M	SD	F	p	η_p^2
Pediatric quality of life inventory							
Physical functioning	78.41	25.52	81.30	24.13	0.25	.62	<.01
Emotional functioning	44.94	21.44	62.23	16.80	12.19	.001	.13
Social functioning	58.20	26.50	67.75	21.37	2.41	.12	.03
School functioning	35.05	25.67	55.07	30.64	9.22	.003	.10
Psychosocial summary	45.95	18.34	61.74	18.21	12.54	.001	.13
Total Score	56.77	17.59	68.26	16.95	7.33	.008	.08
Strengths and difficulties questionnaire							
Emotional symptoms	4.73	2.43	3.00	2.35	8.67	.004	.09
Conduct problems	2.92	2.28	2.22	1.93	1.73	.19	.02
Hyperactivity/inattention	7.16	2.42	5.61	2.93	6.16	.02	.07
Peer problems	3.25	2.39	2.48	2.41	1.77	.19	.02
Prosocial skills	7.46	2.40	7.70	2.42	0.16	.69	<.01
Total difficulties	18.06	6.50	13.30	7.21	8.53	.004	.09

df (1, 84) for all analyses

Hypothesis Four Attachment mediates or moderates the relationship between tic severity and both quality of life (PedsQL) and psychological, behavioural and social difficulties (SDQ).

A series of analyses [54] were conducted to test the hypothesis that quality of peer attachment might mediate or moderate the relationship between tic severity and QOL and SDQ outcomes for TS group children and teens. No evidence of mediation was found for any relationship.

The moderation analyses revealed no effect for QoL outcomes and only one significant moderating effect for SDQ outcomes. Attachment style was found to be a significant moderator of the relationship between Peer Problems and the Physical Functioning QoL dimension, $F(3, 82) = 9.04$, $p = .004$, $\eta^2 = .10$. Follow-up testing of this significant effect involved examining separate scatterplots for the two attachment styles for the relationship between peer problems and the physical functioning QoL domain. For secure attachment no significant relationship between peer problems and physical functioning was evident, $R^2 < .01$, but for insecure attachment a negative relationship was evident, $R^2 = .30$.

Interactions Between Tic Severity, SDQ and Attachment Style on Overall Quality of Life for TS Group Children/ Adolescents

In order to establish whether three important variables, tic severity, SDQ, and attachment style, interacted in any meaningful way in their relationship with quality of life, one final analysis explored all possible two and three-way interactions between the three predictors (total tic severity score, the SDQ total difficulties score, and peer attachment security) on the PedsQL total score. In this fully saturated multiple regression model, no significant interactions were found, leading to the conclusion that these variables each had a largely independent relationship with quality of life.

Discussion

The results of the current study supported the main hypothesis that secure peer attachment would be associated with improved quality of life and psychological, behavioural and social functioning, from the perspective of parents of youth with TS. The majority of the remaining hypotheses were also supported. This research represents the largest controlled study of the Australian TS paediatric population and is one of few to employ a community-based TS sample. Limitations must however be considered when reviewing the findings, in particular the need the restrict

participation to the primary caregiver and the inability to control for formal psychiatric evaluation.

The study began by confirming the hypotheses that parents of youth with TS would report lower QoL for those diagnosed relative to controls [12, 15, 27, 55] with effects extending to all functional domains. Mirroring recent findings [8, 15], youth with TS were reported to exhibit greatly reduced psychosocial functioning, with the highest level of impairment demonstrated in the school domain. Multiple factors may contribute to decreased school functioning including the negative effects of tics on academic functioning and performance in the classroom, the presence of comorbid disorders (e.g. ADHD, OCD and LD) and increased psychosocial difficulties of those diagnosed [5, 8, 22, 23, 30, 34], all of which were observed for youth with TS in the current study. TS was also associated with impairment in the emotional and social functional domains and whilst the difference was significant, the effect size for physical functioning was the lowest effect size for all the sub-domains (see Table 2).

Also, as hypothesised and as noted in the TS literature [5, 17], results of the SDQ indicated that parents of youth with TS identified increased symptoms of psychological, behavioural and social dysfunction for their child compared with parents of undiagnosed peers (see Table 2). TS was most strongly associated with elevated rates of hyperactivity and inattention, a finding consistent with the high rate of comorbid ADHD reported in prior studies [56, 57]. However, this result may have been inflated by measurement error associated with some items on the subscale (e.g. distraction, fidgeting), possibly reflecting phenomena associated with both TS and ADHD.

The main aim of the present study was to examine the previously unexplored role of peer attachment in QOL outcomes and functioning of youth with TS. As hypothesised, those diagnosed were reported as experiencing a highly significant increased rate of insecure peer attachment compared with control group peers. The demographic similarity between the two groups (see Table 1) suggests that differences in peer attachment were not related to structural variables often implicated in the development of insecure attachment [58]. Parents also perceived that youth with TS encountered increased difficulty in forming friendships with peers relative to controls. These findings are consistent with prior research identifying difficulties establishing and maintaining friendships, the lower quality and number of friends [29–31] and the multiple documented barriers to friendship youth with TS encounter [18, 29–34].

Also as predicted, secure peer attachment was positively associated with improved QOL outcomes and decreased difficulties (as measured by the SDQ) for youth with TS, with inverse results demonstrated for insecure peer

attachment (see Table 3). Security of peer attachment did not, however, impact the youths' physical functioning or rate of conduct problems. The later finding highlights the highly adverse effects of antisocial and aggressive behaviours of youth with TS, which have been identified in prior studies as having the most detrimental effects on their relationships [59].

The hypothesis that being securely attached to peers would moderate or mediate the adverse impact of TS on QOL outcomes for youth with TS was not supported. Only one significant result was determined; the physical functioning of youth with TS was moderated by increased peer problems relative to controls. This may reflect barriers to participation in actives, such as sport, associated with the increased social anxiety and the negative peer behaviours that youth with TS frequently experience.

Although causality could not be determined in the study, results are consistent with the strong relationship found between secure attachment and optimal child development, wellbeing and functioning [41–44]. Furthermore, the strong relationship between secure attachment and the development of social competence and emotion regulation skills is reflected in the current findings. Insecure attachment was most closely associated with increased psychosocial dysfunction, emotional symptoms and peer relationships problems as assessed by the SDQ [36–38, 40, 41]. On the basis of these findings it may be concluded that having TS places youth at increased risk of insecure peer attachment and its well-documented adverse psychosocial consequences.

The study also included hypotheses regarding the impact of tic severity and comorbidity on outcomes for youth with TS. As demonstrated in prior research [8, 26], increased tic severity predicted adverse outcomes within the TS group. All measured outcomes were significantly negatively associated with increased tic severity with the strongest predictive effects reported for emotional functioning and increased rates of peer difficulties. Although the group experienced a wider range and greater frequency of motor tics, detailed analyses revealed the disproportionately negative impact of vocal tics. This may be partially explained by the relatively high rate of coprolalia (20 %) revealed by the PTQ. Vocal tics, and coprolalia, in particular, have been associated with increased distress, and have highly unfavourable social and behavioural consequences [25, 29, 35].

The third variable to be examined was comorbidity and, consistent with prior studies, having a formally diagnosed co-occurring disorder was associated with lower global QoL [3, 8, 11, 12, 14–16]. Whilst the present findings also identified the significant relationship between comorbidity and impaired emotional and school functioning, comorbidity was not significantly related to physical or social functioning. Similarly, the results of the SDQ indicated that youth with a co-occurring diagnosis experienced a higher rate of overall difficulties than those with "TS only"; however, comorbidity only resulted in elevated levels of emotional problems. When considered alongside the findings for peer attachment, the present results suggest that TS may exert uniquely adverse effects on the social and relational functioning of those diagnosed.

Given previous findings that determined increased impairment in QoL attributable to the combined effect of tic severity and comorbidity [5, 8, 12], one final analysis was conducted. This examined the possibility that increased tic severity, symptoms of psychopathology and behavioural difficulties, as screened by the SDQ, and insecure peer attachment may interact to decrease QoL. No significant interactions were determined, suggesting a model with each variable independently predicting global quality of life in an addition fashion. Finally, a comparison of the effect sizes demonstrated in the study indicated that insecure peer attachment and increased tic severity had equally strong adverse impacts on global QoL, and these exceeded the impact of having a comorbid diagnosis. The relative contribution of these three variables could be more accurately determined in future studies that improve upon the current methodology by controlling for formal psychiatric evaluation. Due to the prevalence of comorbidity, such research may also benefit by examining the variable impact of individual comorbid disorders rather than collapsing them into a single 'comorbidity' variable. These goals were beyond the scope of the current research.

Limitations and Future Research

As previously noted, the current study was limited by the reliance on parental reports. Although parental proxy measures and reports of comorbid diagnoses are evident in more recent TS studies, slight discrepancies between youth and parental outcomes have been noted [8, 15], and the reliability of diagnostic status in this study cannot be assured. Whilst not a diagnostic tool, the inclusion of the psychometrically robust SDQ partially compensated for the lack of control over clinical assessment and results were largely consistent with the high rate of parent reported comorbidity for TS group youth.

Additional limitations include the possibility that recruitment from the TSAA continues to bias the sample towards those with more severe pathology, although the lower than usual rate of comorbidity reported in this study is consistent with the rate expected in a community sample [4, 7]. Due to the inability to randomise sampling, results may also reflect unmeasured characteristics of voluntary participants. Further, the predominantly Caucasian, middle-class backgrounds of participants may restrict the generalisation of findings to more diverse populations. The

study was also limited to comparisons with healthy controls and future studies may benefit by including psychiatric control groups.

Future attachment oriented studies could address the current limitations associated with the need to employ a unidimensional measure of attachment, should a multidimensional measure suitable for use in younger children become available. Additional categories of insecure attachment could also be incorporated though this may require a larger sample, and research extended to include other important attachment relationships.

Summary

TS has been associated with reduced quality of life, impaired functioning across multiple domains and increased psychological, behavioural and social difficulties for diagnosed youth; however outcomes vary widely among individuals. Whilst increased tic severity and the presence of a comorbid disorder has been found to predict adverse QoL outcomes, a significant proportion of variance remains unexplained. The relative lack of controlled, community-based TS studies was addressed by the present research, which tested the hypothesis that a third variable—the security of peer attachment—would account for variability in QoL outcomes. Multivariate analyses revealed that insecure peer attachment, increased tic severity, having a comorbid diagnosis, and elevated symptoms of psychological, social and behavioural difficulty (SDQ) were each independently and strongly associated with adverse outcomes for youth with TS. Also as hypothesised, those diagnosed were found to be at significantly increased risk of insecure peer attachment than undiagnosed peers. Although limited to parental responses, these preliminary findings suggest that secure peer attachment might be an important determinate of quality of life outcomes for youth with TS warranting further investigation. Results also indicate the importance of clinical interventions to improve the social functioning and peer relationships of those diagnosed.

References

1. American Psychiatric Association (2013) Diagnostic and statistical manual of mental disorders—fifth edition (DSM-5). American Psychiatric Publishing, Washington

2. Robertson MM, Eapen V (2014) Tourette's: syndrome, disorder or spectrum? Classificatory challenges and an appraisal of the DSM criteria. Asian J Psychiatr 11:106–113

3. Cavanna A, Servo S, Monaco F, Robertson M (2009) The behavioral spectrum of Gilles de la Tourette syndrome. J Neuropsychiatry Clin Neurosci 21:13–23

4. Robertson MM, Cavanna AE, Eapen V (2015) Gilles de la Tourette syndrome and disruptive behavior disorders: prevalence, associations and explanation of the relationships. J Neuropsychiatry Clin Neurosci 27:33–41

5. Robertson MM (2012) The Gilles de la Tourette syndrome: the current status. Arch Dis Child Educ Pract Ed 97:166–175

6. Termine C, Balottin U, Rossi G, Maisano F, Salini S, Di Nardo R et al (2006) Psychopathology in children and adolescents with Tourette's syndrome: a controlled study. Brain Dev 28:69–75

7. Freeman RD, Fast DK, Burd DL, Kerbashian J, Robertson MM, Sandor P (2000) An international perspective on Tourette syndrome: selected findings from 3500 individuals in 22 countries. Dev Med Child Neurol 42:436–447

8. Conelea CA, Woods DW, Zinner SH, Budman C, Murphy T, Scahill LD et al (2011) Exploring the impact of chronic tic disorders on youth: results from the Tourette syndrome impact survey. Child Psychiatry Hum Dev 42:219–242

9. Eddy CM, Cavanna AE, Gulisano M, Agodi A, Barchitta M, Calì P et al (2011) Clinical correlates of quality of life in Tourette syndrome. Mov Disord 26:735–738

10. Leckman JF, Bloch MH, Scahill L, King RA (2006) Tourette syndrome: the self under Siege. J Child Neurol 21:642–649

11. Storch EA, Lack CW, Simons LE, Goodman WK, Murphy TK, Geffken GR (2007) A measure of functional impairment in youth with Tourette's syndrome. J Pediatr Psychol 32:950–959

12. Eddy CM, Rizzo R, Gulisano M, Agodi A, Barchitta M, Calì P et al (2011) Quality of life in young people with Tourette syndrome: a controlled study. J Neurol 258:291–301

13. Cutler D, Murphy T, Gilmour J, Heyman I (2009) The quality of life of young people with Tourette syndrome. Child Care Health Dev 35:496–504

14. Muller-Vahl K, Dodel I, Müller N, Münchau A, Reese JP, Balzer-Geldsetzer M et al (2010) Health-related quality of life in patients with Gilles de la Tourette's syndrome. Mov Disord 25:309–314

15. Storch EA, Merlo LJ, Lack C, Milsom VA, Geffken GR, Goodman WK et al (2007) Quality of life in youth with Tourette's syndrome and chronic tic disorder. J Clin Child Adolesc Psychol 36:217–227

16. Jalenques I, Galland F, Malet L, Morand D, Legrand G, Auclair C et al (2012) Quality of life in adults with Giles de la Tourette syndrome. BMC Psychiatry 13:109

17. Eapen V, Črnčec R (2009) Tourette syndrome in children and adolescents: special considerations. J Psychosom Res 67:525–532

18. Boudjouk PJ, Woods DW, Miltenberger RG, Long ES (2000) Negative peer evaluation in adolescents: effects of tic disorders and trichotillomania. Child Fam Behav Ther 22:17–28

19. Achenbach TM, Rescorla LA (2000) Manual for the ASEBA preschool forms and profiles. University of Vermont, Research Center for Children, Youth, & Families, Burlington

20. Chang S, Himle MB, Tucker BTP, Woods DW, Douglas W, Piacentini JC (2009) Initial psychometric properties of a brief parent-report instrument for assessing tic severity in children with chronic tic disorders. Child Fam Behav Ther 31:181–191

21. Cooper C, Robertson MM, Livingston G (2003) Psychological morbidity and caregiver burden in parents of children with Tourette's disorder and psychiatric comorbidity. J Am Acad Child Adolesc Psychiatry 42:1370–1375

22. Packer LE (1997) Social and educational resources for patients with Tourette syndrome. Neurol Clin 15:457–473

23. Packer LE (2005) Tic-related school problems: Impact on functioning, accommodations, and interventions. Behav Modif 29:876–899

24. Wilkinson BJ, Marshall RM, Curtwright B (2008) Impact of Tourette's disorder on parent reported stress. J Child Fam Stud 17:582–598

25. Woods DW (2005) Introduction to the special issue on the clinical management of Tourette's syndrome: a behavioral perspective. Behav Modif 29:711–715

26. Storch EA, Murphy TK, Chase RM, Keeley M, Goodman WK, Murray M et al (2007) Peer victimization in youth with Tourette's syndrome and chronic tic disorder: relations with tic severity and internalizing symptoms. J Psychopathol Behav 29:211–219

27. Carter AS, O'Donnell DA, Schultz RT, Scahill L, Leckman JF, Pauls DL (2000) Social and emotional adjustment in children affected with Gilles de la Tourette's syndrome: associations with ADHD and family functioning. J Child Psychol Psychiatry 41:215–223

28. Silva RR, Munoz DM, Barickman J, Friedhoff AJ (1995) Environmental factors and related fluctuation of symptoms in children and adolescents with Tourette's disorder. J Child Psychol Psychiatry 36:305–312

29. Woods DW, Piacentini JC, Scahill L, Peterson AL, Wilhelm S, Chang S et al (2011) Behavior therapy for tics in children: acute and long-term effects on psychiatric and psychosocial functioning. J Child Neurol 26:858–865

30. Woods DW, Koch M, Miltenberger RG (2003) The impact of tic severity on the effects of peer education about Tourette's syndrome. J Dev Phys Disabil 15:67–78

31. Stokes A, Bawden HN, Backman JE, Dooley JM, Camfield PR (1991) Peer problems in Tourette's disorder. Pediatrics 87:936–942

32. Champion LM, Fulton WA, Shady GA (1989) Tourette syndrome and social functioning in a Canadian population. Neurosci Biobehav Rev 12:255–257

33. Cohen DJ, Leckman JF (1994) Developmental psychopathology and neurobiology of Tourette's syndrome. J Am Acad Child Adolesc Psychiatry 33:2–15

34. Dykens E, Leckman J, Riddle M, Hardin M, Schwartz S, Cohen D (1990) Intellectual, academic, and adaptive functioning of Tourette syndrome children with and without attention deficit disorder. J Abnorm Child Psychol 18:607–615

35. Kurlan R, Daragjati C, Como PG, McDermott MP, Trinidad KS, Roddy S et al (1996) Non-obscene complex socially inappropriate behavior in Tourette's syndrome. J Neuropsych Clin Neurosci 8:311–317

36. Bowlby J (1982) Attachment and loss: retrospect and prospect. Am J Orthopsychiatry 52:664–678

37. Ainsworth MD (1989) Attachments beyond infancy. Am Psychol 44:709–716

38. Ainsworth MDS (1982) Attachment: retrospect and prospect. In: Parkes CM, Stevenson-Hinde J (eds) The place of attachment in human behavior. Basic Books, New York, pp 3–30

39. Hazan C, Shaver P (1987) Romantic love conceptualized as an attachment process. J Pers Soc Psychol 52:511–524

40. Sroufe LA (2005) Attachment and development: a prospective, longitudinal study from birth to adulthood. Attach Hum Dev 7:349–367

41. Seibert AC, Kerns KA (2009) Attachment figures in middle childhood. Int J Behav Dev 33:347–355

42. Muris P, Maas A (2004) Strengths and difficulties as correlates of attachment style in institutionalized and non-institutionalized children with below-average intellectual abilities. Child Psychiatry Hum Dev 34:317–328

43. Laible DJ, Carlo G, Roesch SC (2004) Pathways to self-esteem in late adolescence: the role of parent and peer attachment, empathy, and social behaviours. J Adolesc 27:703–716

44. Rubin KH, Dwyer KM, Booth-LaForce C, Kim AH, Burgess KB, Rose-Krasnor L (2004) Attachment, friendship, and psychosocial functioning in early adolescence. J Early Adolesc 24:326–356

45. Varni JW, Limbers CA (2009) The pediatric quality of life inventory: measuring pediatric health-related quality of life from the perspective of children and their parents. Pediatr Clin North Am 56:843–863

46. Varni JW, Seid M, Kurtin PS (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. Med Care 39:800–812

47. Chan KS, Mangione-Smith R, Burwinkle TM, Rosen M, Varni JW (2005) The PedsQL: reliability and validity of the short-form generic core scales and asthma module. Med Care 43:256–265

48. Goodman R (1997) The strengths and difficulties questionnaire: a research note. J Child Psychol Psychiatry 38:581–586

49. Mellor D (2005) Normative data for the strengths and difficulties questionnaire in Australia. Aust Psychol 40:215–222

50. Goodman R, Scott S (1999) Comparing the strengths and difficulties questionnaire and the child behavior checklist: Is small beautiful? J Abnorm Child Psychol 27:17–24

51. Muris P, Meesters C, van Melick M, Zwambag L (2001) Self reported attachment style, attachment quality, and symptoms of anxiety and depression in young adolescents. Pers Indiv Differ 30:809–818

52. Bartholomew K, Shaver P (1998) Methods of assessing adult attachment. Do they converge? In: Simpson JA, Rholes WS (eds) Attachment theory and close relationships. Guilford Press, New York, pp 25–45

53. Muris P, Meesters C (2002) Attachment, behavioral inhibition, and anxiety disorders symptoms in normal adolescents. J Psychopathol Behav 24:97–106

54. Baron RM, Kenny DA (1986) The moderator–mediator variable distinction in social psychological research: Conceptual, strategic and statistical considerations. J Pers Soc Psychol 51:1173–1182

55. Bernard BA, Stebbins GT, Siegel S, Schultz TM, Hays C, Morrissey MJ et al (2009) Determinants of quality of life in children with Gilles de la Tourette syndrome. Mov Disord 24:1070–1073

56. Robertson MM (2008) The prevalence and epidemiology of Gilles de la Tourette syndrome. Part 1: the epidemiological and prevalence studies. J Psychosom Res 65:461–472

57. Robertson MM (2011) Gilles de la Tourette syndrome: the complexities of phenotype and treatment. Br J Hosp Med 72:100–107

58. Main M, Solomon J (1990) Procedures for identifying infants as disorganized–disoriented during the ainsworth strange situation. In: Greenberg M, Cicchetti D, Cummings EM (eds) Attachment in the preschool years: theory, research and intervention. University of Chicago Press, Chicago, pp 121–160

59. De Lange N, Olivier M (2004) Mothers' experiences of aggression in their Tourette's syndrome children. Int J Adv Couns 26:65–77