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**“The Impact of Tourette’s Syndrome on the Functioning of Young  
Australians: The Roles of Tic Severity, Comorbid Diagnoses and  
Attachment to Peers.”**

Thesis submitted by

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## Statement on the Contribution of Others

I wish to acknowledge the invaluable and ever constant assistance of Prof. Edward Helmes, my primary supervisor; the early assistance of Dr. Todd Jackson during the process of the development of the research proposal for this study, ethics approval and confirmation; Dr. Kerry McBain and Dr. Beryl Buckby as secondary supervisors and for their support and encouragement; Prof. Valsamma Eapen, from the University of New South Wales, as an associate supervisor, and for providing expertise in the area of Tourette's; and Prof. John Reece for ongoing statistical consultation.

The specific contribution of coauthors of the research reports included in this thesis is indicated below.

### Specific Contribution of Co-authors of Publications Included in this Thesis

<b>Publications</b>	<b>Nature and Extent of the intellectual input of each author, including the candidate for all papers</b>
<p>1. "Recognising and treating tourette's in young australians: A need for informed multidisciplinary support" <i>Australian Psychologist</i> (in press)</p> <p>2. "The impact of tic severity, comorbidity and peer attachment on quality of life outcomes and functioning in tourette's syndrome: Parental perspectives" <i>Child Psychiatry &amp; Human Development</i> (october 2015) published doi: 10.1007/s10578-015-0590-</p> <p>3. "Factors impacting the quality of peer relationships of youth with tourette's syndrome" <i>BMC Psychology</i>, (september, 2015) published</p>	<p><b>Deirdre O'Hare<sup>1</sup></b>, principal author and primary researcher, responsible for the development of the research questions, research design, data collection, data analyses, interpretation of the findings and first authorship of all research reports and conference presentations</p> <p><b>Edward Helmes<sup>1</sup></b>, Primary PhD Supervisor. Intellectual input in the capacity as primary supervisor of all aspects of the research; review of findings and reviews of reports</p> <p><b>Kerry McBain<sup>1</sup></b>, Secondary Supervisor. Intellectual Input, particularly regarding qualitative</p>

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## **Declaration of Ethics**

The research presented and reported in this thesis was conducted within the guidelines for research ethics outlined in the National Statement on Ethics Conduct in Research Involving Humans (2007), the Joint NHMRC/AVCC Statement and Guidelines on Research Practice (1997), The James Cook University Policy on Experimentation Ethics, Standard Practices and Guidelines (2001). The proposed research methodology received clearance from the James Cook University Experimentation ethics Review Committee (Approval number H4380).

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Thank you also to Dr Douglas Woods and colleagues for permission use the PTQ and Dr James Varni and associates for permission to the use of the PedsQL. Every reasonable effort has been made to gain permission and acknowledge the owners of copyright material. I would be pleased to hear from any copyright owner who has been omitted or incorrectly acknowledged.

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**Dedication**

For Boy

Mike

To all who live bravely and proudly with Tourette's Syndrome

## **Abstract**

This research was conducted in response to the under-involvement of psychologists in the provision of care and advancement of research regarding Tourette's syndrome (TS). The major goal was to improve current understandings of individual differences in outcomes for youth with this distressing disorder. It represents one of the largest controlled studies of TS, is one of few to employ a community-based sample, and is the first comprehensive study of TS in young Australians. The research was conducted in two parts. Study One was an Australia-wide, controlled, mixed-method survey-based study. Participants were parents of youth with TS (n = 86) and parents of typically developing peers (n = 108). Study Two was a qualitative, interview-based study of the attachment relationships and functioning of youth with TS, as perceived by their biological mothers (n = 22). The ethical and practical constraints associated with including young children in this study, limited participation to primary caregivers.

In a novel application of attachment theory, the study tested the hypothesis that individual differences in outcomes would be predicted by the security of the peer attachments of youth with TS. Tourette's was conceptualised as a stressor with the capacity to disrupt or impair the quality of the youth's close relationships.

Multivariate analyses in Study One study confirmed that youth with TS experienced significantly lower health-related quality of life and functioning across all domains (assessed with the Pediatric Quality of Life Inventory, PedsQL) and higher rates of psychopathology, behavioural and social dysfunction (measured by the



Strengths and Difficulties Questionnaire, SDQ) relative to controls. Also as expected, the clinical presentation of TS group youth mirrored the phenomenology of the disorder reported in the international literature.

The major hypothesis was also supported. Multivariate results indicated highly significant ( $p < .01$  -  $p < .001$ ) associations between insecure peer attachment (classified by the Attachment Questionnaire for Children) and negative outcomes of all measures (PedsQL & SDQ), with converse findings for secure attachment. Also as predicted, youth with TS experienced a highly significant increased rate (threefold) of insecure peer attachment in comparison with controls. The further prediction that secure peer attachment would moderate or mediate the adverse impact of tic severity and comorbid disorder, however, received limited support.

Also as predicted, increased tic severity and having a comorbid diagnoses (72% of the TS group) were both strongly associated with negative outcomes on all measures. No interactions were found between tic severity, comorbid disorder and insecure peer attachment, indicating that each variable independently affected the individual's quality of life. Further analyses of the impact of individual comorbid diagnoses on quality of life and levels of dysfunction, however, revealed restricted and highly disorder specific effects. This analysis also determined the exclusive contribution of TS to impaired social functioning and peer relationships problems. Furthermore, a high rate of undiagnosed or subclinical level mood disorder was evident in TS group youth. As depression has been found to be the strongest predictor of highly adverse outcomes for those with the TS by adulthood, this result is of particular clinical interest.

The two exploratory qualitative studies revealed the impact of TS on the attachment relationships and attachment-related functioning of youth with TS, using methodology designed for the study. Personality (classified into “Big Five” traits) was the most commonly identified factor to impact peer relationships. Extraversion and Agreeableness facilitated the friendships for all youth, whilst higher rates of Neuroticism impaired the peer relationships of TS group youth. The explanatory theories of TS group parents were also more complex, variable and included more “non-personality” related factors than those of controls.

Factors enhancing secure attachments for TS group youth included successful psychological adjustment to diagnosis; low level of self-consciousness and adaptive cognitive appraisal of their symptomatology; the ability to defend themselves against the negative behaviour of others; the ability to manage tics on occasions; and having the acceptance and understanding of peers. The strongest barriers to friendships were the negative behaviour of peers (bullying, teasing and social rejection); the experience of severe tics; the inability to control tics at critical times; non-tic related or comorbid symptoms such as impulsivity, cognitive rigidity and obsessive-compulsive behaviours; social anxiety; maladjustment to diagnosis of TS; effort required to suppress tics when in company of friends; and high levels of self-consciousness.

The findings from Study Two illustrated the powerful influence of TS on the quality of the Mother-Child relationship (MCR), by determining the unique “closeness” of the MCR and directing many maternal roles and functions within this relationship. The study also identified the multiple threats TS posed to the security of the MCR. These included high levels of maternal fear, anxiety, stress, and caregiver burden; relationship ruptures associated with negative aspects of the child’s

symptoms, behaviours and developmental transitions; reduced social support; and maternal over-involvement. The impact of the child's diagnosis of TS, however, appeared to strengthen the mother-child bond. The study also revealed the stress associated with parenting a child or adolescent with TS, with participants' experiences mirroring those of parents of children with other serious chronic disorders.

Finally, the integrated results of the research facilitated the development of an inclusive, predictive quality of life model for youth with TS. Findings also informed the development of guidelines for psychological interventions to improve the quality of the youth's attachment relationships. It was concluded that improving the attachment relationships of youth with TS is an important treatment goal that should be considered alongside the management of tic severity and comorbid disorder. Broader recommendations for policy, services and advocacy were also made in response to current findings of continuing stigma, and the inadequacy of TS services and supports in Australia.

## Table of Contents

<b><u>“THE IMPACT OF TOURETTE’S SYNDROME ON THE FUNCTIONING OF YOUNG AUSTRALIANS: THE ROLES OF TIC SEVERITY, COMORBID DIAGNOSES AND ATTACHMENT TO PEERS.”</u></b> .....	<b>I</b>
<b><u>STATEMENT ON THE CONTRIBUTION OF OTHERS</u></b> .....	<b>II</b>
<b><u>DECLARATION OF ETHICS</u></b> .....	<b>IV</b>
<b><u>ABSTRACT</u></b> .....	<b>VII</b>
<b><u>SECTION 1. INTRODUCTION TO THE STUDY</u></b> .....	<b>5</b>
<b><u>SECTION 2. CHAPTER 1. LITERATURE REVIEW</u></b> .....	<b>16</b>
<b>CURRENT DEFINITION AND DIAGNOSIS OF TS</b> .....	<b>16</b>
<b>HISTORY OF TOURETTE’S SYNDROME</b> .....	<b>17</b>
<b>THE DIAGNOSTIC EXPERIENCE</b> .....	<b>21</b>
<b>AETIOLOGY, EPIDEMIOLOGY AND PHENOMENOLOGY OF TOURETTE’S SYNDROME</b> .....	<b>22</b>
LIMITATIONS OF PREVIOUS TS RESEARCH .....	22
AETIOLOGY.....	23
PREVALENCE.....	26
ONSET AND COURSE OF TS .....	27
TIC PHENOMENOLOGY AND CLASSIFICATION.....	29
PREVALENCE AND TYPE OF COMORBID DISORDERS.....	32
<b>THE IMPACT OF TOURETTE’S SYNDROME</b> .....	<b>36</b>
TS AND QUALITY OF LIFE (QOL).....	36
TS AND SOCIO-EMOTIONAL FUNCTIONING OF CHILDREN AND ADOLESCENTS.....	39
TS AND FAMILY FUNCTIONING .....	42
TS AND THE INTELLECTUAL AND EDUCATIONAL FUNCTIONING OF CHILDREN AND ADOLESCENTS ..	44
<b>VARIABLES AFFECTING THE IMPACT OF TOURETTE’S SYNDROME</b> .....	<b>46</b>
RELATIONSHIPS BETWEEN TIC SEVERITY, COMORBIDITY, FUNCTIONAL OUTCOMES AND QUALITY OF LIFE .....	47
THE IMPACT OF TIC SEVERITY .....	51
<b>THE IMPACT OF PEER ATTACHMENT ON CHILDREN AND ADOLESCENTS WITH TS</b> .....	<b>54</b>
<b>ATTACHMENT THEORY: THE DEVELOPMENT OF ATTACHMENT STYLE</b> .....	<b>57</b>
<b>ATTACHMENT STABILITY AND CONTINUITY ACROSS TIME AND RELATIONSHIPS</b> .....	<b>60</b>
<b>DEVELOPMENTAL STAGES AND THE EXTENSION OF ATTACHMENT</b> .....	<b>64</b>
<b>THE IMPACT OF ATTACHMENT ON CHILD AND ADOLESCENT PSYCHOLOGICAL AND BEHAVIOURAL FUNCTIONING</b> .....	<b>69</b>
<b>ATTACHMENT AND PEER RELATIONS</b> .....	<b>73</b>
<b><u>SECTION 2 CHAPTER 2 OVERVIEW OF THE RESEARCH</u></b> .....	<b>78</b>
<b>PRIMARY RESEARCH OBJECTIVES</b> .....	<b>78</b>
<b>HYPOTHESES FOR STUDY 1</b> .....	<b>80</b>
<b>HYPOTHESES FOR STUDY 2</b> .....	<b>81</b>
<b>OUTLINE OF THE PRESENTATION OF THE RESEARCH</b> .....	<b>82</b>

<b><u>SECTION 3 CHAPTER 1. RECOGNISING AND TREATING TOURETTE'S IN YOUNG AUSTRALIANS: A NEED FOR INFORMED MULTIDISCIPLINARY SUPPORT AUSTRALIAN PSYCHOLOGIST (IN PRESS)</u></b> .....	<b>84</b>
<b><u>SECTION 3 CHAPTER 2. THE IMPACT OF TIC SEVERITY, COMORBIDITY AND PEER ATTACHMENT ON QUALITY OF LIFE OUTCOMES AND FUNCTIONING IN TOURETTE'S SYNDROME: PARENTAL PERSPECTIVES CHILD PSYCHIATRY &amp; HUMAN DEVELOPMENT (OCTOBER 2015)</u></b> .....	<b>109</b>
<b><u>SECTION 3 CHAPTER 3. FACTORS IMPACTING THE QUALITY OF PEER RELATIONSHIPS OF YOUTH WITH TOURETTE'S SYNDROME BMC PSYCHOLOGY, (SEPTEMBER, 2015)</u></b> .....	<b>146</b>
<b><u>SECTION 3 CHAPTER 4. "THE IMPORTANCE OF SCREENING FOR EARLY INDICATIONS OF EMOTIONAL SYMPTOMS IN YOUTH WITH TOURETTE'S SYNDROME"</u></b> .....	<b>191</b>
<b><u>SECTION 3 CHAPTER 5. YOUTH WITH TOURETTE SYNDROME: PARENTAL PERCEPTIONS AND EXPERIENCES IN THE AUSTRALIAN CONTEXT</u></b> .....	<b>219</b>
<b><u>SECTION 3 CHAPTER 6. TOURETTE'S SYNDROME AND MOTHER-CHILD ATTACHMENT: A QUALITATIVE EXPLORATION</u></b> .....	<b>250</b>
<b><u>SECTION 3 CHAPTER 7. "DIFFERENTIATING BETWEEN THE IMPACT OF TOURETTE'S AND COMORBID DIAGNOSIS ON QUALITY OF LIFE AND FUNCTIONING"</u></b> .....	<b>287</b>
<b><u>SECTION 4 CHAPTER 1. THE STABILITY OF ATTACHMENT IN THE CONTEXT OF TOURETTE'S SYNDROME</u></b> .....	<b>305</b>
<b><u>SECTION 5 CHAPTER 1. IMPLICATIONS AND INTEGRATION OF THE FINDINGS AND RECOMMENDATIONS FOR PRACTICE AND POLICY</u></b> .....	<b>322</b>
<b>OVERVIEW OF THE RESEARCH AND MAJOR FINDINGS</b> .....	<b>322</b>
RESEARCH GOALS AND METHODOLOGY .....	322
MAJOR FINDINGS .....	324
<b>IMPLICATIONS AND RECOMMENDATIONS FOR PRACTICE AND POLICY: CLINICAL PROFILE &amp; CORRELATES OF TS</b> .....	<b>328</b>
<b>IMPLICATIONS OF THE INTEGRATED FINDINGS: A PREDICTIVE QUALITY OF LIFE MODEL</b> .....	<b>333</b>
<b>RECOMMENDATIONS FOR PRACTICE</b> .....	<b>336</b>
THERAPEUTIC APPROACH.....	336
<b>IMPLICATIONS AND RECOMMENDATIONS FOR PRACTICE: IMPROVING THE MOTHER-CHILD RELATIONSHIP</b> .....	<b>337</b>
ASSESSMENT .....	337
PSYCHOLOGICAL INTERVENTIONS .....	338
<b>IMPLICATIONS AND RECOMMENDATIONS FOR PRACTICE: IMPROVING PEER RELATIONSHIPS</b> ..	<b>341</b>
ASSESSMENT .....	341
PSYCHOLOGICAL INTERVENTIONS .....	342
<i>IMPROVING PERSONALITY FUNCTIONING AND PERSONALITY- RELATED BEHAVIOURS</i> .....	342
<i>FACILITATING ADJUSTMENT TO DIAGNOSIS OF TS</i> .....	343
<i>"COPING WITH" TICS - PSYCHOLOGICAL INTERVENTIONS</i> .....	344
<i>MINIMISATION OF PROBLEMS ASSOCIATED WITH "NON-TIC" TS SYMPTOMS AND COMORBID OR SECONDARY DISORDER</i> .....	345
<i>MANAGING AGGRESSIVENESS AND DISRUPTIVE BEHAVIOURS</i> .....	346
<i>TREATING ANXIETY AND DEPRESSION</i> .....	347

<i>PROVIDING SOCIAL SKILLS TRAINING</i> .....	347
<i>FACILITATING OPPORTUNITIES FOR FRIENDSHIP AND SOCIALISATION</i> .....	348
<i>CHANGING THE ATTITUDES AND BEHAVIOURS OF PEERS</i> .....	349
<b>LIMITATIONS AND FUTURE STUDIES</b> .....	<b>349</b>
LIMITATIONS .....	349
FUTURE STUDIES .....	353
<b>CONCLUDING COMMENTS</b> .....	<b>355</b>
<b>REFERENCES</b> .....	<b>357</b>

<b>APPENDIX A.</b> THE AUSTRALIAN TOURETTE'S SURVEY (STUDY 1) & SEMI-STRUCTURED INTERVIEW QUESTIONS (STUDY 2) .....	417
<b>APPENDIX B.</b> DATA DICTIONARIES AND DATA CODING PROFORMA.....	429
<b>APPENDIX C.</b> MATERNAL NARRATIVE DESCRIPTIONS OF THE CHILD (STUDY 2)...	452
<b>APPENDIX D.</b> EXTENDED REPORTS OF QUALITATIVE FINDINGS (STUDY 2).....	488
<b>APPENDIX E.</b> ABSTRACT OF CONFERENCE PRESENTATIONS.....	496

## **LIST OF TABLES**

<b>TABLE 1.</b> EXAMPLES OF SIMPLE AND COMPLEX TICS (ADAPTED FROM ROBERTSON & CAVANNA, 2012) .....	30
<b>TABLE 2.</b> PREVALENCE OF COMMON COMORBID DISORDERS (ADAPTED FROM FREEMAN ET AL., 2000) .....	33
<b>TABLE 3.</b> FREQUENCY AND TYPE OF PARENT-REPORTED COMORBID DIAGNOSES FOR TS GROUP (N = 63).....	102
<b>TABLE 4.</b> GROUP DIFFERENCES IN EMOTIONAL AND BEHAVIOURAL FUNCTIONING.....	104
<b>TABLE 5.</b> FREQUENCIES, RANKS AND PERCENTAGES OF MOTOR AND VOCAL TICS REPORTED FOR TS GROUP.....	105
<b>TABLE 6.</b> REPORTED LEVELS OF ACCESS, SATISFACTION AND UTILISATION OF SERVICES AND RATES OF STIGMATISATION FOR TS GROUP .....	107
<b>TABLE 7.</b> DEMOGRAPHIC CHARACTERISTICS AND BETWEEN-GROUP DIFFERENCES.....	139
<b>TABLE 8.</b> GROUP DIFFERENCES IN QUALITY OF LIFE OUTCOMES AND EMOTIONAL AND BEHAVIOURAL FUNCTIONING .....	141
<b>TABLE 9.</b> IMPACT OF PEER ATTACHMENT ON QUALITY OF LIFE AND STRENGTHS AND DIFFICULTIES OUTCOMES FOR TS GROUP (N = 86) .....	143
<b>TABLE 10.</b> CORRELATIONS BETWEEN TIC SEVERITY, QUALITY OF LIFE AND STRENGTH'S AND DIFFICULTIES OUTCOMES FOR TS GROUP (N = 86). .....	144
<b>TABLE 11.</b> IMPACT OF COMORBIDITY ON QUALITY OF LIFE AND STRENGTHS AND DIFFICULTIES OUTCOMES TS GROUP (N = 86). .....	145
<b>TABLE 12.</b> DESCRIPTIVE STATISTICS AND ANALYSIS OF VARIANCE RESULTS FOR TS AND CONTROL GROUP PARENTS ON SOCIAL FUNCTIONING AND PEER PROBLEMS.....	184
<b>TABLE 13.</b> DESCRIPTIVE AND CHI-SQUARE RESULTS FOR DIFFERENCES BETWEEN TS AND CONTROL GROUPS IN ATTACHMENT SECURITY AND ABILITY TO FORM FRIENDSHIPS.....	185
<b>TABLE 14.</b> FREQUENCY AND Z-TEST OF PROPORTIONS BETWEEN TS GROUP YOUTH AND	

CONTROL GROUP YOUTH IN PERSONALITY TRAITS WITH POSITIVE AND NEGATIVE IMPACT ON FRIENDSHIP.....	186
<b>TABLE 15.</b> FREQUENCY AND Z-TESTS OF PROPORTIONS BETWEEN TS GROUP YOUTH AND CONTROL GROUP IN NON-PERSONALITY FACTORS WITH POSITIVE AND NEGATIVE IMPACT ON FRIENDSHIP.....	188
<b>TABLE 16.</b> DEMOGRAPHIC CHARACTERISTICS AND BETWEEN GROUP DIFFERENCES FOR CONTROL AND TS GROUPS.....	218
<b>TABLE 17.</b> GENERAL AND TS SPECIFIC STRESSORS EXPERIENCED IN PARENTING A CHILD OR ADOLESCENT WITH TOURETTE SYNDROME .....	247
<b>TABLE 18.</b> STATEMENT OF NEEDS OF MOTHERS OF YOUTH WITH TOURETTE SYNDROME...	248
<b>TABLE 19.</b> MOTHER'S GENERAL EMOTIONAL RESPONSE TO THE CHILD & CLASSIFICATION OF EMOTIONAL RESPONSES & MATERNAL NARRATIVE REPRESENTATIONS (POSITIVE, NEGATIVE, AMBIVALENT).....	273
<b>TABLE 20.</b> MATERNAL INTERNAL REPRESENTATIONS OF THE MCR- DESCRIPTIONS AND ROLES AND FUNCTIONS. ....	275
<b>TABLE 21.</b> DIRECT THREATS TO MOTHER- CHILD ATTACHMENT ASSOCIATED WITH THE CHILD'S TS. ....	278
<b>TABLE 22.</b> MOTHER'S EMOTIONAL & BEHAVIOURAL RESPONSE TO HER CHILD'S DISTRESS.....	281
<b>TABLE 23.</b> PARTICIPANT DEMOGRAPHICS ( <i>N</i> = 86) .....	301
<b>TABLE 24.</b> IMPACT OF TS ONLY VERSUS TS PLUS COMORBID DIAGNOSIS (PARENT REPORTED) ON QUALITY OF LIFE (PEDS QL) AND STRENGTHS AND DIFFICULTIES OUTCOMES (SDQ) FOR TS GROUP YOUTH.....	302
<b>TABLE 25.</b> SIGNIFICANT FINDINGS FOR IMPACT OF INDIVIDUAL COMORBID DISORDERS (PARENT REPORTED) ON QUALITY OF LIFE (PEDS QL) AND STRENGTHS AND DIFFICULTIES OUTCOMES FOR TS GROUP YOUTH .....	303
<b>TABLE 26.</b> METHODOLOGY FOR ESTIMATING ATTACHMENT SECURITY- MATERNAL ATTACHMENT STYLE, MOTHER CHILD-RELATIONSHIP & CHILD -PEER ATTACHMENT .....	316
<b>TABLE 27.</b> SUMMARY RESULTS OF THE ANALYSIS OF INDICATORS OF QUALITY OF MOTHER'S ATTACHMENT, MOTHER CHILD-ATTACHMENT & CHILD-PEER ATTACHMENT .....	318

## **LIST OF FIGURES**

<b>FIGURE 1.</b> FLOW CHART OF THE RESEARCH: STUDY 1 (PART A & B) & STUDY 2.....	183
<b>FIGURE 2.</b> METHODOLOGY FOR EXPLORING THE SECURITY OF MOTHER-CHILD ATTACHMENT RELATIONSHIP (MCR) BASED ON CODED INTERVIEW DATA. ....	284
<b>FIGURE 3.</b> CONCEPTUAL MAP OF THE TRANSMISSION OF ATTACHMENT ACROSS GENERATIONS, TIME AND RELATIONSHIPS .....	306
<b>FIGURE 4.</b> TRANSMISSION OF ATTACHMENT SECURITY (ESTIMATED) ACROSS GENERATIONS, RELATIONSHIPS AND TIME. ....	321
<b>FIGURE 5.</b> ROLES FOR PSYCHOLOGISTS IN THE CARE & MANAGEMENT OF YOUTH WITH TS .....	332
<b>FIGURE 6.</b> PREDICTIVE QUALITY OF LIFE MODEL FOR YOUTH WITH TOURETTE'S BASED ON THE INTEGRATED FINDINGS FROM STUDIES 1 & 2.....	335

## **SECTION 1. Introduction to the Study**

Tourette's Syndrome (TS) is a complex neurodevelopmental disorder of unknown aetiology for which there is no known cure (Felling & Singer, 2011; Jankovic & Kurlan, 2011; Leckman, Bloch, Scahill & King, 2006; Robertson & Cavanna, 2011). It is characterised by the presence of multiple involuntary motor and vocal tics, often occurring in bouts over a period of at least one year (DSM-V, APA, 2013; ICD-10, WHO, 1990). Tics are described as "sudden, rapid, recurrent, non-rhythmic stereotyped movements or vocalizations" (APA, 2013). There is wide heterogeneity in the clinical presentation and severity of TS. Whilst some may experience very mild, almost undetectable symptoms, many others will express highly visible tics, particularly during waxing phases of the disorder (Robertson, 2011; Zinner & Coffey, 2009). In its most severe form, TS can be physically disabling, limit the individual's ability to function across multiple domains and in extreme cases, lead to the requirement of institutional care. The adverse consequences of TS, however, have been demonstrated for all levels of severity of the disorder (Stern, Burza & Robertson, 2005).

For as many as two-thirds with the disorder, TS tends to follow a similar pattern of onset and course, with early to middle childhood onset, a peak in tic symptomology during adolescence, and a significant remittance of symptoms by early adulthood (Jankovic, 2001; Leckman, Bloch, Scahill & King, 2006a). International prevalence rates for TS are estimated to be one percent of the pediatric and adolescent population (Robertson, 2008; Robertson, Eapen & Cavanna, 2009) whilst recent US figures suggest an incidence of 1 per 360 (Bitsko et al., 2014), with boys three to four times more likely to have the disorder than girls (Robertson, 2008).



With no biological makers, TS is diagnosed solely on the basis of clinical history and the observation of symptoms. Originally described by Itard in 1825, and eponymously named and clinically defined by the French neurologist Giles de La Tourette in 1885, TS is no longer considered to be a rare psychiatric curiosity (Jankovic & Kurlan, 2011; Leckman, 2001). It is currently included in both the DSM-V (American Psychiatric Association- APA 2013) and the ICD-10 (WHO, 1990) as the most severe of several major tic disorders. The criteria published in these manuals are employed to guide clinical diagnosis, which may be aided by the use of one of several standardized measures including the Yale Global Tic Severity Scale-YGTSS (Leckman et al., 1989)

Complicating the management, clinical presentation and diagnosis of TS are strikingly high rates of comorbidity (Cavanna, Servo, Manaco & Robertson, 2008a; Cohen & Leckman, 1994; Conelea et al., 2011; Eddy et al., 2011; Gaze, Kepley & Walk-up, 2006; Himle et al., 2007; Robertson 2006, 2006a, 2006b, 2011; Robertson, Eapen & Cavanna, 2009; Robertson & Cavanna, 2007; Singer, 2005). Upwards of ninety percent of those formally diagnosed have been found to have at least one of several commonly co-occurring psychiatric or behavioural conditions (Freeman et al., 2000; Robertson et al., 2009) Current research suggests some degree of shared aetiology occurring at the genetic and neural substrate levels between TS and its most prevalent comorbid disorders; Attention Deficit Hyperactivity Disorder (ADHD) and Obsessive Compulsive Disorder (OCD) (Freeman, 2007; Leckman et al., 2006a; Robertson, 2006a; Robertson & Cavanna, 2007; Termine et al., 2005). Other commonly noted comorbid diagnosis include, but are not restricted to, anxiety and mood disorders, oppositional defiance and conduct disorder, disordered impulse control and aggressiveness, learning disorders and an increased prevalence of TS is

noted for those on the autistic spectrum (Freeman et al., 2000). Disentangling the symptoms and effects of TS and comorbid disorders, many of which have been found to account for disproportionately high levels of adverse sequelae, presents challenges in clinical, research and educational settings (Gaze et al., 2006), and adds to the burden of those living with the syndrome and their loved ones (Carter et al., 2000).

As there is no cure for TS, current treatments aim to minimize symptoms. Evidence is emerging for the success of a promising, recently developed behavioural intervention for tic management (Comprehensive Behavioural Intervention for Tics - CBIT) (Himle et al., 2012; Piacentini et al., 2010; Woods et al., 2008). The current approach to treatment is, however, principally confined to the use of antipsychotic and neuroleptic medications to reduce tics, and to treat symptoms of comorbid disorders (Robertson, & Stern, 2000; Scahill et al., 2013; Scahill et al., 2006; Woods, Piacentini & Himle, 2007). Supportive psychotherapy, behavioural interventions such as CBIT, which includes contingency management and habit reversal training (HRT), family therapy and psychoeducation, play an important adjunctive role (Himle, Woods, Piacentini & Walk-up, 2006; Hollenbeck, Woods, Piacentini & Walkup, 2007; Robertson, 2011; Piacentini & Chang, 2005; Stewart et al., 2006; Woods, Conelea & Himle, 2010). Although effective, the iatrogenic risks, adverse side effects and challenges involved in titrating appropriate therapeutic doses for individuals accompanying the prescription of psychoactive drugs, limit their use (Eddy, Rickards & Cavanna, 2011b; Woods, Conelea & Himle, 2010). This is particularly so in paediatric populations, where parents and clinicians may express reservations about their short and long-term use, particularly for youngsters with milder forms of TS (Conelea et al., 2011; Robertson, 2011, Himle et al., 2012; Scahill et al., 2013).

Despite the long-term recognition and relatively high level of prevalence of TS, it remains an under-diagnosed and poorly understood syndrome (Collins, 2005; Comings, 1994; Dedmon, 1990). Researchers have documented many barriers to the diagnosis and management of TS (Eapen & Crncec, 2009). As far back as the mid-nineteen eighties, Dedmon (1990) expressed the need for professionals from multiple disciplines to become better informed about TS. However, the most recent review by a Canadian based social worker (Collins, 2005) revealed little change in the level of awareness and understanding of the disorder. Stern and colleagues (2005) suggested that the greatest service provision problems for those with TS in the UK are delayed diagnosis, lack of specialist management and inadequate educational support. Misdiagnosis or diagnostic delay creates intense frustration and distress for those with the disorder, and their families (Dedmon, 1990; Collins 2005; Shimberg, 1995, 2012). It also increases the risk to the child by preventing the initiation of interventions aimed at minimizing the adverse psychological, social and neurological consequences of TS, thereby compromising their development (Comings, 1994; Dedmon, 1990; Collins, 2005; Shimberg, 1996; Stern, Burza & Robertson, 2005).

A lack of understanding and acceptance of the bizarre, sometimes frightening, and generally disruptive symptoms of TS also leads to many individuals, and their families, feeling stigmatized, socially isolated and rejected (Collins, 2005). Media portrayals continue to sensationalize and misrepresent TS, thereby perpetuating myths and negative stereotypes that promote misunderstanding and stigmatization (Davis, Davis & Dowler, 2003). The popular media, for example, usually presents only the most bizarre, extreme or relatively rare tics such as Coprolalia (Freeman, 2014), whilst negative, exaggerated portrayals of TS posted for their “comedic value” are

more frequently viewed online than positive representations (Fat, Buchner & Lang, 2009).

Factors contributing to poor current levels of recognition of TS amongst professionals and the public include the failure to accommodate TS within the academic curriculum, and the lack of adequate advocacy, funding and interest in TS from other disciplines prior to the 1990's (Collins, 2005; Jankovic, 2001; Stern et al., 2005). Perhaps of most significance has been the dominance of the biomedical research paradigm in TS scholarship, with its focus on aetiology and pharmacotherapy. Although substantial advances are being made in identifying possible casual mechanisms for TS, bio-medical models cannot adequately explain the individual variability in impairment related to the disorder (Hendren, 2002; Swerdlow, 2005; Woods, 2005).

A review of the literature also reveals a relative dearth of information regarding interventions aimed at improving the social, educational, vocational and family functioning of those with TS (Hendren, 2002). The expansion of the TS research agenda and the education of professionals has, therefore, become a major goal of organizations such as the Tourette's Society of America (TSA) and their Australian counterpart, the Tourette's Syndrome Association of Australia (TSAA).

An additional gap in the TS literature, which is of concern in the context of the current study, is the relative lack of research specific to the Australian TS population. Several smaller scale studies, a larger scale genetic study of Australian TS-affected families, and an efficacy study of CBIT intervention have commenced, however, this author could identify few previously published studies based on Australian data. Amongst those identified were small-scale neurological studies profiling the clinical features of Australian children and adults with TS (Yun-Chee & Sachdev, 1997;

Sachdev, Yun-Chee & Wilson, 1996) and a recent study by Eapen and Crncec (2009) documenting the special considerations involved in diagnosis and management of youngsters with TS.

A growing body of biopsychosocially orientated research, predominantly focusing upon pediatric and adolescent TS populations, consistently reveals high levels of adverse short and long-term psychological, behavioural and social consequences associated with TS (Conelea et al., 2011; Cavanna et al., 2012; Robertson & Cavanna, 2011; Wood, 2005). To date, the search for correlates of psychological and social dysfunction associated with TS has been largely restricted to the role played by increased tic severity and the presence of comorbid diagnosis, both of which have been associated with adverse outcomes (Robertson, 2011). Results are sometimes contradictory, and also reveal significant variability among individuals, suggesting that a large proportion of variance associated with TS remains unexplained. It would appear that some children, adolescents and adults are more resilient to the negative consequences of TS than others, independent of both the severity of their disorder and the presence or absence of co-morbid diagnosis (Conelea et al., 2011).

Promising findings from the small body of psychologically oriented TS research suggest a significant explanatory role for psychosocial factors in the variability demonstrated between TS affected individuals. The increased rates of social dysfunction and lower levels of social abilities reported for the TS population suggest that young people with the syndrome face significant challenges in the social realm. Recent research has identified social support, particularly in the form of functional families and supportive and accepting peer relationships, as key factors in the psychosocial wellbeing of children and teens with TS (Cooper, Robertson &

Livingston, 2003; Packer, 1997; Wilkinson, Marshall & Curtwright, 2008; Wilkinson, al., 2001; Woods & Marcks, 2005; Woods et al., 2011). In one study, researchers found that having a well-functioning family was associated with better outcomes, even for those with more severe tics and co-morbid diagnoses (Carter et al., 2000).

Building on findings for the positive role played by social support, the main hypothesis for the current study was that the ability to form close relationships with peers would be associated with improved quality of life and functioning, in addition to reduced psychopathology, behavioural and social problems for children and adolescents with TS. In recent years, attachment theory has been increasingly employed as a conceptual framework within which close relationships are examined. According to this theory, children develop a style of attachment within the biologically determined primary attachment relationship between mother and infant, which can be classified on the basis of observable behaviour of the child (Ainsworth, 1982, 1985, 1989, 1991; Bretherton, 1992; Bowlby, 1982).

Over the course of development, attachment functions are gradually transferred to other family members, thence increasingly to childhood and adolescent friends, in a process culminating in the formation of adult romantic relationships and close friendships (Bowlby, 1982; Feeney & Noller, 1996; Fraley & Shaver, 2000; Hazan & Shaver, 1987, 1994; Hazan & Zeifman, 1994; Main, Kaplan & Cassidy, 1985; 1988). The style of attachment developed in the primary attachment relationship appears to remain relatively stable across time, relationships and generations, though it may change in response to high levels of stress (Sroufe, 2005; Sroufe & Waters, 1977; Sroufe, Egeland, Carlson & Collins 2005, 2009). It is hypothesised that attachment stability is a function of the child's development of "internal working models" of relationships during interactions with their primary

caregiver. These models guide their expectations and behaviours in all future close relationships (Ainsworth, 1985, 1989; Bartholomew & Horowitz, 1991; Bowlby, 1982; Hazan & Shaver, 1987).

A large body of research attests to the importance of security of attachment in determining a child's psychosocial wellbeing, behaviour and optimal development. Secure attachment is also key to developing the positive social skills and confidence required to initiate and maintain close friendships (Bowlby, 1982), and has been most closely related to the increased social competence of the developing child (Allen & Land, 1999; Booth-LaForce et al., 2006; Kerns, Klepak & Cole, 1996; Simpson, Collins, Tran & Hayden, 2007). In the context of the current study, secure peer attachment was hypothesised to predict improved outcomes for youth with TS. It was further hypothesized that secure peer attachment might moderate or mediate the adverse effects of increased tic severity or the presence of comorbid disorder, thereby enhancing the child's quality of life and functioning.

In order to achieve the multiple goals of the current research, two complementary studies that adopted a mixed method approach were conducted. Due to the ethical and practical implications of studying children as young as seven within the design of the current study (remote mode - via written survey and telephone interview), participation was limited in both studies to the primary caregiver, the vast majority of whom were the youth's biological mother.

#### *Study One, Part A - Quantitative*

This quantitative controlled study (Study One, Part A) surveyed the primary caregivers of a nation-wide sample of Australian children and adolescents with TS

( $n = 86$ ), with those of a comparative sample of Australian children with no known psychiatric or medical diagnosis ( $n = 108$ ). It employed a pen and paper survey assembled for this study, which included four empirically validated and reliable psychometric instruments, a demographic questionnaire and purpose authored questions.

The primary goal of the quantitative study was hypothesis testing. It was predicted that peer attachment security, tic severity and comorbid disorder would be associated with variability in quality of life outcomes and the psychological, behavioural and social functioning of Australian children and teens with TS, as reported by their primary caregiver. It was further hypothesised that secure peer attachment would mediate or moderate the adverse impact of tic severity and the presence of comorbid disorder. Although insecure attachment was predicted to be associated with adverse outcomes for all participants, higher rates of insecure attachment were expected for the TS group, given the elevated levels of psychosocial dysfunction associated with the disorder.

A secondary goal was to test the hypothesis that parents of youth with TS would report lower quality of life, higher rates of behavioural and psychological dysfunction for their children than control group parents.

In order to facilitate comparison with international paediatric and adolescent TS populations, the study also aimed to investigate the clinical profile of TS in a community-based sample of Australian youth. It was expected that findings would mirror those of overseas populations, supporting the universal nature of the disorder.

Finally, data were collected from the TS group regarding their experiences within the Australian health and education systems, their utilization of mental health and educational services, and perceived levels of stigma associated with TS. Findings



relating to this goal aimed to identify targets for therapeutic intervention, and to provide essential information to those advocating for improved services and supports for Australians with TS and their families.

*Study One - Part B. and Study Two - Qualitative*

Due to the novel and exploratory nature of the current research, and the lack of a multidimensional psychometric measure of peer attachment for middle childhood, two qualitative studies were also conducted. Study One, Part B, gathered qualitative data (in the form of limited written responses from parents) in order to compare the factors identified by each parent group, which were perceived to enhance or impair the ability of youth to form secure peer relationships. It was hypothesized that these factors would vary between groups, and reflect the ability of TS to shape the quality of the youth's peer relationships.

To augment these findings, a further interview-based qualitative study (Study Two) was conducted employing a subset of TS group participants (n = 22) from Study One. The primary goal was to enrich understandings of the attachment related functioning of Australian youth with TS by exploring key attachment relationships, and the impact TS had upon the quality of the attachment relationships under study. These included the child's peer attachments, the mother – child relationship and the mother's childhood and current attachment relationships. Of particular interest were factors relating to TS that affected mother-child and peer attachment security.

Study Two also tested the hypothesis that TS would represent a significant stressor with the capacity to change or otherwise disrupt the security of the attachment relationships of diagnosed youth. This was achieved by examining the stability of attachment demonstrated across generations, time and relationships for each mother-

child dyad in the sample. This was facilitated by a novel methodology grounded in attachment theory, which was designed to explore and provide estimates of the security of the relationships included in the study.

Finally, it was hoped that the integrated findings from this research project could be employed to develop psychological interventions to improve outcomes for youth with TS and their caregivers. It was also hoped that results could be used to inform recommendations for improved policy, practice and services for the Australian TS community.

## **SECTION 2. Chapter 1. Literature Review**

### **Current Definition and Diagnosis of TS**

Tourette's Syndrome (TS) is a complex neurodevelopmental disorder of unknown aetiology for which there is no known cure (Felling & Singer, 2011; Leckman et al., 2006; Robertson, 2011; Robertson & Cavanna, 2008). It is characterised by the presence of multiple involuntary motor and vocal tics, often occurring in bouts over a period of at least one year (APA 2013; WHO 1994). Tics are described as "sudden, rapid, recurrent, non-rhythmic stereotyped movements or vocalizations" (APA, 2013). As the aetiology of TS is currently unknown, differential diagnosis is based solely upon an individual's observed and historical tic symptomology.

Diagnostic criteria and guidelines for TS are included in both the ICD-10 (WHO, 1990) and DSM-5 (APA, 2013), although slight variation between the two is apparent. The DSM-5 limits the diagnostic process to the identification of differences in type, combination and temporal characteristics of observed tics. The ICD-10 also makes specific reference to patterns of onset of tics, links between underlying emotional states, such as stress and anxiety, and the relationship between TS and obsessional thinking. The ICD-10 also refers to some degree of behavioural control over tics. It emphasises the differences between TS tics, and the repetitive behaviours sometimes seen in Autism and mental retardation, as well as the ritualistic behaviours associated with Obsessive Compulsive Disorder (OCD). It does however

note that the line between TS and emotional disorders associated with tics, such as OCD and hypochondriasis, can be very unclear.

An additional difference is noted in the consideration of adult tic behaviours. Although both the ICD-10 and DSM-5 refer to TS as a neurodevelopmental disorder, they differ in how they describe the phenomenology of adult tic symptoms, with only the ICD observing that tics will usually resolve to subclinical levels for most patients after adolescence. Finally, both systems stipulate that tics must not be the result of substance use or a medical condition (such as Huntington's Chorea).

### **History of Tourette's Syndrome**

The current conceptualization of TS reflects a long and complex history, which has seen TS rise in significance from a rare syndrome based upon a few curious cases presented by Itard and Giles De la Tourette in the 1800s, to its current status as one of several Tic Disorders included in the DSM-5 and ICD-10 classification systems (Rickards & Cavanna, 2009). In the absence of any conclusive theories regarding the aetiology of TS, arriving at the current iteration of the syndrome has been problematic. Although the cardinal features of TS have long been recognised, there have been many challenges associated with defining the disorder (Walkup, Ferrao, Leckman, Stein & Singer, 2010). These have included the uncertainty created by the fluctuating nature of tics, the presence of comorbid symptoms and a history of changing theoretical approaches to the disorder (Jancovic & Kurlan, 2011; Zinner & Coffey, 2009). The current debate regarding recommended revisions of criteria for the recent DSM-5 reflects its continuing conceptual evolution (Walk-up et al., 2010).

Originally defined as a neurological disorder by Giles De la Tourette, a radical departure from this medically orientated perspective occurred in the early twentieth

century with the adoption of psychoanalytic explanatory models (Kushner & Kiessling, 1996; Rickards, Wolfe & Cavanna, 2010). Freudians conceptualized TS as psychogenic in origin, with tics representing the manifestation of unrestrained sexual urges. Interpreted within this model, the individual's inability to suppress and manage tics reflects narcissism, weak will or obsessive-compulsive neurosis (Leckman, 2001; Sacks, 1992). This theory dominated the approach to TS for the first half of the 20<sup>th</sup> century despite the failure of psychoanalytically orientated treatment interventions (Leckman, 2001; Woods, 2005). Remnants of this conceptualization may inform current misperceptions regarding tic behaviours and contribute to the stigmatization of those with the disorder.

By mid-century, dissatisfaction with this approach coupled with discovery that haloperidol effectively reduced tics, created a renewed interest in the neurobiological basis of TS and a return to the medical model (Hyde & Weinberger, 1995; Gaze et al., 2006). Indeed, the use of psychoactive medication remains the first line of treatment for symptom management (Robertson, 2011), whilst pioneering Deep Brain Stimulation (DBS), a technique developed to treat Parkinsonian tremors, is being trialled for the most severe and disabling cases, with mixed results (Walkup, Mink & Hollenbeck, 2006; Neimat, Patil & Lozano, 2006; Welter et al., 2008).

Noticeably absent from the research into TS during the later half of the twentieth century was input from psychology (Woods, 2005). As a result, research regarding the way in which TS impacts upon the child's normal development and psychosocial functioning, or how these effects extend into adulthood is somewhat limited. In recent years, psychologists have begun to play a significantly larger role, often as part of multidisciplinary teams adopting a biopsychosocial approach to the disorder. This research is revealing significant positive associations between TS and

dysfunction across multiple domains (Leckman et al., 2006; Zinner & Coffey, 2009). A review of the literature, however, reveals that the search for factors that may help to explain the variability of the highly heterogenous experience and expression of TS is lacking. Tic severity and the presence and type of comorbid disorders are the only two variables to have been systematically included in published studies. The role played by additional variables such as social skills, stress, contextual factors, parental stress and caregiver burden, quality of family functioning and school related experiences are beginning to be more comprehensively explored (Conelea & Woods, 2008; Cooper & Livingston, 2003; Woods, Himle & Osmon, 2005; Packer, 2005).

Recent behaviourally orientated research is also extending understandings of TS (Himle et al., 2012; Himle, Woods, Piacantini & Walkup, 2006; Woods, 2005; Woods, Conelea & Himle, 2010). Learning theory suggests that tic repetition may be the result of negative re-enforcement, in an “urge -tic -relief cycle” (Himle et al., 2006; Specht et al., 2013; Woods et al., 2010). These authors have demonstrated the efficacy of a manualised comprehensive behavioural therapy for tic reduction (CBIT) (Woods et al., 2008), and a recent trial found that positive long-term effects of behavioural treatment for children with TS were detected six months following intervention (Woods et al., 2011). These positive results offer support for a behavioural approach, and CBIT provides an effective psychological alternative or adjunctive therapy to pharmacological intervention (Woods et al., 2010). Yet it remains an underutilized form of treatment. The authors suggest that this may be due to a lack of trained providers, in addition to low levels of awareness amongst consumers and providers of this approach to treatment (Woods et al., 2010). An efficacy trial for CBIT is currently underway in Australia.

Another area of current debate concerns the definition of TS as a disorder characterised by multiple phenotypes (Cavanna et al., 2009; Grados & Mathews, 2009; Robertson & Cavanna, 2007; Robertson et al., 2009) versus its current classification in both the ICD 10 and the DSM-V manuals as a spectrum disorder (Comings, 1994; Comings & Comings 2005). Results of recent cluster and factor analyses on the phenomenology of tics and co-morbid symptoms (Alsobrook & Pauls, 2002; Grados & Mathews, 2009; Robertson & Cavanna, 2007; Robertson, Althoff, Hafez & Pauls, 2008; Leckman et al., 2006) revealed clusters of symptoms (factors) emerging from the data that provide initial support for the multiple phenotype approach. Results identified a range of TS phenotypes such as “TS with only simple motor and vocal tics”, “TS with complex tics”, “TS with Attention Deficit Hyperactivity Disorder (ADHD/ADD)” and “TS with OCD” and “TS with OCD and ADHD (inattentive type)”. These “TS Types” are hypothesised to share some common aetiological pathophysiology (Alsobrook & Pauls, 2002; Robertson & Cavanna, 2007; Robertson, 2011). Other researchers call for the cautious interpretation of these findings, noting methodological problems inherent in these studies (Walk-up et al., 2011). Limitations included the use of small samples in many of the biomedical studies, non-random sampling strategies that typically involved large kindreds and population isolates, as well as possible ascertainment bias introduced by the overrepresentation of clinic based subjects.

The manner by which TS is conceptualised is of fundamental importance. It guides research design, as well as the differential diagnosis and treatment of those with the disorder. It also determines the way in which those with the disorder are perceived and accepted by others. A general lack of understanding and acceptance of the bizarre, sometimes frightening and disruptive symptoms of TS lead to individuals

with the disorder and their families feeling stigmatized, socially isolated and rejected (Collins, 2005). Media portrayals continue to sensationalize and misrepresent TS thereby perpetuating myths and negative stereotypes that promote misunderstanding and stigmatization (Davis, Davis & Dowler, 2003).

### **The Diagnostic Experience**

Despite the long-term recognition and relatively high level of prevalence of TS, and despite significant recent advances in understanding, it remains an under-diagnosed and poorly understood syndrome (Collins, 2005; Bruun & Budman, 1997). As far back as the late nineteen eighties, Dedmon (1990) expressed the need for professionals from multiple disciplines to become better informed about TS, however a more recent review by Collins (2005) revealed that little had changed. Stern and colleagues (2005) suggested that the greatest service provision problems for those with TS in the UK are delayed diagnosis, lack of specialist care and inadequate educational support. The correct diagnosis of TS has been found to take up to several years to achieve, and on average requires consultations with seven professionals (Collins, 2005, Dedmon, 1990; Shimberg, 1995).

Misdiagnosis or diagnostic delay creates intense frustration and distress for those with the disorder and their families. It also prevents the initiation of interventions aimed at minimizing the adverse psychological, social and neurological consequences of the disorder during critical stages of child or adolescent development (Comings & Comings, 1993; Dedmon, 1990, Collins 2005, Shimberg, 1995; 2012; Stern et al., 2005).



## **Aetiology, Epidemiology and Phenomenology of Tourette's Syndrome**

### **Limitations of Previous TS Research**

The methodological difficulties associated with the study of this complex syndrome are reflected in the high rates of inconclusive and contradictory findings in the TS literature (Felling & Singer, 2011). The most frequently noted methodological limitations are ascertainment bias and nonrandom sampling, both of which limit the generalizability of results to wider TS populations. An over-reliance on samples drawn from clinical populations is commonly demonstrated, and is attributable to the difficulties associated with identifying and recruiting subjects with milder forms of the disorder. Research subjects therefore often represent those with more severe forms of TS, who are more likely to have comorbid disorders.

Other limitations refer to the inconsistency in assessment and measurement of TS across studies. Variations in the operational definition of TS, variability in the way comorbid disorders have been incorporated into study design, and variability in the measures used to determine tic severity have been noted. Measurement of tic severity ranged from the employment of a variety self or other reports, to the use of standardized clinical rating scales such as the Yale Global Tic Severity Score that are available to professionals only (Swerdlow, 2005). In addition, some researchers collapse boundaries between types of tics disorders, whilst other studies lack transparency regarding selection criteria and allocation of subjects to groups.

Another limitation commonly noted in the context of studying a small, widely distributed clinical population such as TS, is the frequent use of small sample sizes. TS studies have been found to include as few as four or five subjects, with a comparatively small number of studies including sample sizes greater than sixty.

Consequently, many studies report low levels of statistical power and findings with small effect sizes and low levels of statistical significance. Highly unequal sample sizes have also been noted, thereby limiting parametric analysis.

The confounding variables introduced by the high rates of co-morbidity and TS are particularly limiting and hard to avoid, rendering it difficult to disentangle effects of these disorders from those of TS. Other noted confounds include the lack of control for age, severity of TS and comorbid symptoms, use of medication, and consideration of other demographic variables (Felling and Singer, 2011). Finally, in view of the wide temporal variability of tic symptomology, a lack of longitudinal studies has prevented optimal study of the syndrome.

### **Aetiology**

The medical paradigm, with its focus on aetiology and biomedical interventions, continues to dominate the TS literature. This is reflected in the rapidly growing body of research based on findings from neuroimaging and histopathology, clinical trials of psychoactive medications, animal models of TS, genetic research and studies exploring relationships between TS and its comorbid disorders. The results illustrate multiple possible causal mechanisms for the disorder (Jankovic & Kurlan, 2011; Leckman, Bloch, Smith, Larabi & Hampson, 2010; Lombroso & Scahill, 2008; Robertson, 2008). Robertson and Cavanna (2008) speculate that interactions between differing causal mechanisms, which are grounded in genetics and pathophysiological processes, account for heterogeneity in the clinical presentation of TS.

A recent population-cohort based study of 4,826 individuals with TS revealed the strongly familial and heritable nature of tic disorders, including TS (Mataix-Cols et al., 2015). There is strong anecdotal and experimental evidence for the genetic

transmission of TS, albeit the mechanisms involved appear to be highly complex and poorly understood (Abelson et al., 2005; Bornstein, Stepl & Hammond, 1990; Eapen, Pauls & Robertson, 1993; Jankovic & Kurlan, 2011; Keen-Kim & Friemer, 2006; McMahon et al., 2007; Singer, 2003; 2005; Stern et al., 2005; Swerdlow, 2005; Walkup, et al., 1996; Zausmer & Dewey, 1987). Results of pedigree studies support theories of autosomal dominant genetic transmission (Carter, Pauls, Leckman & Cohen, 1994; Eapen, Pauls & Robertson, 1993; Swerdlow, 2005 check), whilst other researchers favour multiple susceptibility models of transmission and expression (Walk-up et al., 1996). Candidate genes include those located on the human genome in area 17q25 (Pashou et al., 2004), genes related to dopamine such as DRD1, DRD2 and D2 receptors and DAT1 (Chou et al., 2004), and sequence variants in SLITRK1 (Abelson et al., 2005). The first large scale genetic study of TS, which is linked to the work of an international consortium, is currently underway in Australia.

Support for the genetic links between TS, ADHD and OCD is also emerging, with significantly increased rates of each of these disorders reported within families (Alsobrook, Leckman, Goodman, Rasmussen & Pauls, 2000; Goodman, Storch, Geffken & Murphy, 2006; Hanna, Janjua, Contant & Jankovic, 1999; O'Rourke et al., 2011). Robertson and Cavanna (2008) however conclude that only a variant form of OCD has been strongly genetically linked to TS. There appears to be an increased risk for ADHD and tic disorders in TS affected families, however this may reflect overlapping neurophysiology or neurobiology (Stewart et al., 2006).

The pathophysiology of TS appears to be complex and multifactorial, involving neuroanatomical pathways, physiologic abnormality and neurotransmitter or synaptic components (Frey & Albin, 2006; Singer & Minzer, 2003). Of specific interest are the mechanisms controlling the neurocircuitry involved in executive

functioning, and in particular, behavioural regulation and disinhibition (Comings, 1987; Coffey & Shechter, 2005). Imagery and lesion studies suggest that this pathway consists of links between the prefrontal cortex, the basal ganglia and its projections, and the thalamus (Cavanna et al., 2009; Felling & Singer, 2011; Vloet, Neufang, Herpertz-Dahlmann & Konrad, 2006). These pathways would normally inhibit or “gate” neurological information from reaching other brain areas (Comings, 1987; Leckman et al., 2006; Vloet et al., 2006). Tics, and symptoms associated with common co-morbid disorders (inattention, impulsivity, obsessiveness, aggressiveness and dysfunctional motor inhibition), are hypothesized to result from dysfunctional regulation attributable to shared or overlapping pathophysiological pathways within this circuit (Banaschewski, Neale, Rothenberger & Roessner, 2007; Leckman et al., 2006). Inhibition may also be occurring at the cellular level for those with TS. Lower levels of the medium spiny neurons located at the head of the caudate nucleus, whose function is neural signal inhibition, have been found in those with the disorder (Kalanithi et al., 2005).

A strong role for neurotransmitters has also been suggested in the expression of tics in TS. Dopamine, serotonin and epinephrine all appear to affect TS symptoms, with dopamine appearing to be the leading candidate (Felling & Singer, 2011). Drugs that act on dopamine receptors, the neuroleptics, are effective agents for the control of TS tics, though how they reduce tic severity and frequency is not understood (Sprecher & Kurlan, 2009). One explanatory hypothesis is “super-sensitivity” to dopamine within the striatum, and that this is the abnormality within the dopamine system underlying TS (Bower, 1996; Comings & Comings, 1987; Leckman et al., 2010).

Gerard and Peterson (2003) and Leckman and colleagues (2006; 2010) attribute TS, at least in part, to impaired developmental processes in the central nervous system. The authors suggest that because most children express transient tics as a normative part of their development, the persistence and development of tics in TS may reflect absent or delayed normative maturational changes in those with the disorder.

Finally, gestational environmental stressors (Leckman et al., 1990) and streptococcal infection (PANDAS) have been implicated in the onset of TS in some cases, although the latter is a highly contentious finding based upon inconsistent and variable results (Cavanna et al., 2009b; Church, Dale, Lees, Giovannoni & Robertson, 2003; Mell, Davis & Owens, 2005; Harris & Singer, 2006; Singer, Hong, Yoon & Williams, 2005; Leckman et al., 2011).

## **Prevalence**

Tics disorders are relatively common developmental phenomenon, with estimates of prevalence for children and adolescents ranging between 7-28% (Kurlan et al., 2001; Khalifa & von Knorring, 2005). Robertson, Eapen & Cavanna's (2009) comprehensive review resulted in an international estimated rate of prevalence for TS of between 0.3-1% for young people between the age of five and eighteen, whilst Bitsko and colleagues found an incidence of 1:360 in the US paediatric population (Bitsko et al., 2014). Epidemiological data have yielded prevalences for child and adolescent populations (0.64% - 3.8%) when undiagnosed cases were included in the analyses (Stern et al., 2005; Christie & Jassi, 2002; Robertson, 2008).

The epidemiology and phenomenology of TS has been replicated in the Americas, Europe, Asia, the Middle East and Australasia, attesting to its ability to

cross cultural, socioeconomic and racial boundaries (Eapen & Robertson, 2009; Freeman et al., 2000; Robertson et al., 2009; Staley, Ward & Shady, 1997). The lower prevalence of TS in American and sub-Saharan Africans were the only exceptions to this finding, however it is unclear if the differences are attributable to under-diagnosis, genetic or other unknown factors within these populations (Robertson, 2008, Robertson et al., 2009).

Significant gender differences exist in prevalence rates for TS, which demonstrates a male to female bias ranging between three or four to one (Sukhodolsky, Williams & Leckman, 2004). Males with TS are also four times more likely than females to have a comorbid disorder (Robertson & Cavanna, 2008). TS has also been found to be over-represented in special educational settings (Comings & Comings, 1987; Eapen, Robertson, Zeitlin & Kurlan, 1997) and for children with Autistic Spectrum Disorder (ASD) (Robertson & Cavanna, 2008).

### **Onset and Course of TS**

Although there is wide variability in the presentation of TS, multiple studies have identified common developmental patterns of onset and course (Harris & Singer, 2006; Steinberg, King & Apter, 2010). The mean age of onset is between six and seven years, with simple motor tics usually preceding the expression of vocal or phonic tics by one to three years. (Bloch & Leckman, 2009; Leckman, Bloch, Scahill & King 2006b; Robertson, 2011; Zinner & Coffey, 2009). Occasionally, early signs of tic behaviours may appear in children as young as the age of two (Leckman et al., 2006b), and on very rare occasions the onset of tics may not occur until late adolescence or adulthood (Chouinard & Ford, 2000). Some researchers have observed that a younger age of tic onset is predictive of more severe disorder (Bloch &

Leckman, 2009; Cavanna et al., 2009a), whilst others have failed to find this relationship (Goetz, Tanner, Stebbins, Leipzig & Carr, 1992).

For the majority, tics typically occur in bouts that occur many times a day, nearly every day or intermittently. Their anatomical location, number, frequency, complexity, type, and severity usually change over time. Following onset, tics generally progress from simple and transient, to become more chronic and complex. Tics are expressed in characteristically waxing and waning patterns, with high levels of intra-individual variability in the frequency and intensity of bouts of tics. For the majority, tics peak in intensity and complexity during late middle childhood to mid-adolescence before remitting to a significant degree in early adulthood (Jankovic & Kurlan, 2011; Jankovic, Gelineau, Kattner, & Davidson, 2010; Leckman, 2001; Robertson, 2011; Zinner, 2000).

The presence and severity of TS symptoms in adulthood, however, appears to have been underestimated, with recent studies suggesting that tics persist beyond adolescence in much greater number than previously estimated, albeit in more stable and less severe form (Singer, 2005). Accounting for these findings are findings that adults demonstrate little awareness of their tics, and experience less subjective distress, perhaps by having learned to accommodate their tics over time (Pappert, Goetz, Louis, Blasucci & Leurgans, 2003). Singer (2005) suggests a “rule of thirds” that may be applied to the course of TS, with one third experiencing remittance of tics by early adulthood, one third a marked decrease in tic symptomology and the final third experiencing little change from childhood patterns (Singer, 2005). Zinner and Coffey (2009) suggest that this evidence establishes TS as a neurodevelopmental disorder that must be considered across the life cycle.

Over the course of the disorder, changes in tic behaviours remain highly unpredictable. However, some psychological and contextual factors have been associated with an exacerbation or reduction in tic frequency and intensity. Conelea & Woods (2008) reviewed nineteen studies that considered the impact of antecedent and consequent contextual factors on tic behaviour. They concluded that there was evidence to support the significance of these factors, but the relationships were unclear and required further systematic research. Exacerbation has been linked with fatigue, excitement, being alone and elevations in core body temperature (Leckman et al., 2006a; Lombroso Mack, Scahill, King & Leckman, 1991). Although controversial, there is some evidence indicating that stress, in particular stressors associated with day-to-day functioning, may mildly intensify tics (Hoekstra et al., 2004). Similarly, tic reduction has been noted during tasks requiring attention such as drawing, playing computer games or practicing a musical instrument (Leckman et al., 2006a).

Although this “average” pattern of the onset and course of TS can be described, and an understanding of this “average” presentation appears to be very useful in the context of psychoeducation regarding TS (Leckman et al., 2006a), the fluctuating nature of TS renders accurate prognostications for individuals with the diagnosis impossible. This presents a unique challenge for those adjusting to life with such an unpredictable and chronic disorder.

### **Tic Phenomenology and Classification**

The ability to identify tics that are characteristic of TS is essential to its differential diagnosis, particularly from those of other tic disorders (Transient Tic Disorder, TTD; Chronic Motor or Vocal Tic CTD, a frequent “gateway” disorder for



TS, and Tic Disorder not otherwise specified) (APA 2013). Tics must also be differentiated from the ritualistic behaviours of OCD (Comings, 1994), from those attributable to the physiological effects of a substance (e.g. stimulants commonly used to treat ADHD) or a general medical condition (e.g. Huntington’s Chorea or post viral encephalitis) (APA, 2013) (Sprecher & Kurlan, 2009).

Tics are classified along several dimensions, from “Motor” (Muscular) to “Vocal / Phonic” (sounds produced by vocals cords or other mechanisms such as the breath), from “Transient” to “Chronic”, and from “Simple” to “Complex”. Table 1 provides examples of common and less common characteristic, simple and complex motor and vocal / phonic tics.

**Table 1.** Examples of Simple and Complex Tics (adapted from Robertson & Cavanna, 2012)

Common	Common	Less	Common	Common	Less
Simple	Complex	Common	Simple	Complex	Common
Motor	Motor Tics	Complex	Vocal tics	Vocal Tics	Complex
Tics		motor Tics			Vocal Tics
Eye-blinking	Jumping	Corpropraxia	Throat clearing	Words/phrase out of context	Coprolalia
Facial grimacing	Touching	Echopraxia	Grunting	Combination s of sound	Echolalia
Should- shrugging	Squatting	Palipraxia	Sniffing	-	Palilalia
Head jerking	Licking/ smelling objects	Mental forms of above	Snorting	-	Mental forms of above

Commonly experienced motor tics include jumping, squatting, touching or licking objects, whilst eye-blinking, facial grimacing and shoulder shrugging are very

commonly noted simple vocal /phonic tics (Robertson & Cavanna, 2008). TS is also associated with a range of much less commonly experienced complex tics. These are associated with clinical TS populations and are not generally detected in those with milder forms of the disorder (Jankovic, 2001; Rickards & Robertson, 2003; Robertson & Cavanna, 2008). Coprolalia, (the uttering of obscenities) a relatively rare but much sensationalized complex tic, is reported in approximately one-third of patients referred to specialist clinics, and usually has a mean age of onset of fourteen.

Importantly, its prevalence is much lower in non-selected samples. Copropraxia (the making of obscene gestures) is reported in 3% to 21% of cases, whilst some report “mental” forms of both these compulsions. Echolalia (the imitation of the sounds or words of others) and echopraxia (repeating the actions of others) occur in 11% to 44% at some stage during the course of the disorder. Palilalia (repeating one’s own words) and palipraxia (repeating one’s own gestures) have also been reported in a substantial proportion of patients, along with other tic-related symptoms such as stuttering and forced touching of objects / body parts (Cavanna et al., 2009). Tic phenomenology appears to change by adulthood, with lower rates of phonic tics, and more facial and trunkal motor tics (Jankovic et al., 2010).

Tics are usually preceded by intense subjective experiences referred to as either a “premonitory urge” or a “sensory tic”, which is temporarily relieved following the expression of the tic, and have been reported in up to 90% of adults with TS (Cavanna et al., 2009a; Steinberg et al., 2010). This “urge”, however, is rarely identifiable by children under the age of ten (Banaschewski, Woerner & Rothenberger, 2007; Singer, 2005; Woods et al., 2008). Many with TS report the ability to voluntarily suppress their tics for variable periods of time. Tic suppression requires significant effort, particularly in social situations, and may provoke anxiety

that is relieved when the tic is finally discharged (Scahill, Ort & Hardin, 1993). It is argued by some that such “control” appears to represent the postponement of ticking, as tics have been found to later rebound later for many individuals (Robertson & Cavanna, 2008; Cavanna et al., 2009). Challenging the observation of the universality of rebound effects are recent findings of Meidinger and colleagues (2005), and from those developing CBIT (Himle et al., 2007; Woods et al., 2008, Specht et al., 2013). Not all individuals demonstrated tic rebound following suppression; in particular adults with TS and adolescents and adults who received training in CBIT. This suggests that tics remain largely involuntary or “unvoluntary”, but may be subject to some degree of modification (Meidinger et al., 2005). An understanding of ability to exercise voluntary control over tics is of great importance. The belief that tics are largely controllable leads to inappropriate blame and self-recrimination, whilst the possibility that tics may be behaviourally modified is of great importance in context of treatment (Himle et al 2007: Robertson & Cavanna, 2008).

### **Prevalence and Type of Comorbid Disorders**

TS is commonly accompanied by co-morbid psychiatric disorder, with up to 90% of clinic and 80% of community based TS populations in the United Kingdom and North America having been found to have least one additional diagnosis (Cavanna et al., 2009; Robertson, 2006a, 2006b, 2010). Gaze and colleagues (2006) estimated that only 50% of youth with TS experienced comorbidity, attributing higher rates to differences between clinical settings, clinical versus community based populations, and age groups surveyed. Although percentages vary among studies, there is continuity across studies in the type of co-morbid disorders identified. These include Attention Deficit Hyperactivity Disorder (ADHD), Obsessive Compulsive

Disorder / Behaviour (OCD/B), disorders of mood, anxiety, impulse control, learning, intermittent rage and aggression, conduct, personality and Autistic Spectrum Disorders (Cavanna et al., 2009; Carter et al., 2000; Ghanizadeh & Mosallaei, 2009; Robertson, 2006a, 2006b, 2008, 2011; Robertson & Cavanna, 2007, 2011; Robertson Banjeree, Eapen & Fox-Hilley, 2002; Robertson, Channon, Baker & Flynn, 1993; Robertson, Williamson & Eapen, 2006; Rickards & Robertson, 2003; Scahill et al., 2005; Termine et al., 2006).

A large-scale international study of 3,500 clinic based TS patients found that 88% had comorbid psychopathology (Freeman et al., 2000), and others have found that males are more likely to have co-morbid diagnosis than females (Robertson, 2011). Table 2 presents the results of Freemans et al's (2000) survey.

**Table 2.** Prevalence of Common Comorbid Disorders (Adapted from Freeman et al., 2000)

Conditions comorbid with TS	Rates of Co-morbidity
TS Only	12%
TS & ADHD	60%
TS & Impulsive Control / Aggression	37%
TS & OCD	27%
TS+ Mood Disorder	20%
TS & Anxiety Disorder (non-OCD)	18%
TS & Conduct/Oppositional Defiant Disorder	15%

Studies including milder TS cases reported similar patterns of comorbidity, but with slight variation in order, frequency and type of disorder. All, however, confirmed that ADHD and OCD were the two most prevalent comorbid diagnoses

(Carter et al., 2000; Cavanna et al., 2009; Comings & Comings, 1987; Freeman, 2007; Jankovic, 2001; Rickards & Robertson, 2003; Robertson, 2008b, 2011; Robertson & Cavanna, 2007; Robertson et al., 2009; Scahill et al., 2005). Although less severe TS was associated with a lower rate of ADHD (38.4%) in a community-based sample (Kurlan et al., 2002), the overall rate of comorbid ADHD/ADD remains high (Robertson, 2006, 2008b). The pervasiveness of obsessive-compulsive symptomology has also been demonstrated, with findings of up to ninety percent of those with TS experiencing sub-threshold obsessive compulsive symptoms (e.g. need for ordering, getting things “just right”, completion, and compulsive urges to tic), although these may vary from the thoughts and behaviours traditionally associated with OCD (Comings, 1994; Comings & Comings, 1987, 2005; Robertson et al., 2009; Zinner & Coffey, 2009).

Freeman and colleagues (2000) also found that anger control difficulties, sleep disturbances, coprolalia and self-injurious behaviours were associated with comorbidity, with anger being the most strongly correlated to comorbid disorder. Self-injurious behaviour (SIB), of varying degrees of severity, have however also been reported in those with milder forms of TS (Robertson, 2012).

Autistic Spectrum (Baron – Cohen, Scahill, Izaguirre, Hornsey & Robertson, 1999; Kurlan et al., 2002) and personality disorders (Robertson, Banerjee, Hilley & Tannock, 1997) are also overrepresented in the TS populations. Rates of disordered impulse control and aggression, particularly in the form of tantrums and rage attacks, are also elevated in those with TS (Leckman et al., 2006a). Some individuals with TS have been found to have specific neurological deficits in visual- motor functioning and fine motor control (Schultz et al., 1998).

Results regarding depression are inconclusive. Evidence, however, suggests that clinical and subclinical levels of depression and Bipolar Affective Disorder commonly co-occur with TS, particularly in adulthood, and the aetiology is likely to be multifactorial (Robertson, 2006a, 2006b; Robertson et al., 2006). Depression and depressive symptoms have been found in 13 % and 76% respectively for clinic based TS patients (Robertson, 2006b). An increased risk of substance use and depression has also been noted for adults with TS (Jankovic et al., 2010). Young clinic-based people with TS have also been found to have an increased rate of depression, however, there appeared to be a complex interplay between depression, long-standing and more severe tics and the presence of comorbid ADHD or OCD (Robertson et al., 2006). It is, therefore, unclear if depression is a primary comorbid disorder linked to endophenotypes of TS; is secondary to the difficulties associated with living with a chronic, stigmatizing disorder; compounded by the impact of comorbid disorder such as ADHD or OCD; or attributable to side effects of psychopharmacological intervention (Robertson, 2002; Robertson et al., 2002).

Learning Disorders (LD) are also disproportionately higher for a significant number of children and adults with TS (Denckla, 2006; Dykens et al., 1990). Burd and colleagues (Burd, Kauffman & Kerbehsian, 1992) found that 22.7% of the 5,450 international TS subjects had learning disorders as classified by the DSM-V. The authors caution that high rates of comorbid ADHD and TS limit the interpretation of this finding, as disordered learning may be more closely associated with the comorbid ADHD than with TS.

Gaze and colleagues (2006) and Gilbert (2006) highlight the need for a comprehensive approach to assessment for those with TS. TS severity, co-occurring diagnoses and secondary psychosocial complications must all be considered in order

to prioritize areas for intervention and tailor treatments meeting the unique needs of the individual.

### **The Impact of Tourette's Syndrome**

A growing body of evidence reveals that TS is associated with adverse short and long-term psychological, behavioural, social and academic consequences, although wide variability of experience has been noted between individuals with the disorder (Conelea et al., 2011; Carter, Pauls, Leckman & Cohen, 1994; Cavanna et al., 2009a; Robertson & Cavanna, 2007; Woods, 2005). An increasing number of studies are being conducted on adults with TS, however the majority of research regarding the impact of TS has been conducted on pediatric populations. The following section summarizes findings from the TS literature regarding the quality of life, psychosocial functioning, family functioning and intellectual / academic functioning primarily of children and teens with a diagnosis of TS or Chronic Tic Disorder, the gateway diagnosis for TS.

#### **TS and Quality of Life (QoL)**

Measures of “quality of life” (QoL) commonly reflect a biopsychosocial approach to the assessment of an individual’s wellbeing and functioning across the major realms of life (Schipper, Clinch & Olweny, 1996). The development of health-related quality of life measures has facilitated the examination of the differential impact of physical and psychological disorders upon an individual’s level of overall wellbeing, and their ability to function across specific life domains. Most recently, disease specific quality of life measures have been developed that incorporate key

symptoms and difficulties that are excluded from more general instruments. These may generate more accurate QoL assessments in specific clinical populations. Cavanna, Robertson and colleagues have recently developed and validated the GTS-QoL for adults (Cavanna et al., 2008), and a recently published paediatric version of the measure (Cavanna et al., 2012). The outcomes of QoL studies have become critical to the process of holistic comparisons between groups, the identification of areas of difficulties and strengths and identify the multidimensional needs of populations of interest. Multi-disciplinary responses can include services, supports, treatments, facilities and policies operating at the individual, societal and institutional levels.

A review of the recent TS literature revealed several published studies (English language or available in translation) that have explored the impact of the disorder on global (overall) wellbeing, and across major life domains. These include studies on adult TS populations (Cavanna et al 2008; Elstner, Selia, Timble & Robertson, 2008; Jalenques et al., 2012; Muller-Vahl et al., 2010) and several involving paediatric and adolescent populations (Bernard et al., 2009; Cooper & Livingston, 2003; Conelea et al., 2011; Cutler, Murphy, Gilmour & Heyman, 2009; Eddy et al., 2011a; Eddy et al., 2011c; Marek, 2006; Storch et al., 2007a; Storch et al., 2007b). With minor exceptions, the majority of these studies have been conducted within the past ten years, have recruited from clinic based populations, have employed relatively small samples sizes and applied a diverse range of measurement tools, including various health-related quality of life and general quality of life instruments. Most studies did not recruit control groups, but compared results against normative data or data from other studies. With the exception of single studies



conducted in France, Italy and Germany, research has been restricted to USA and United Kingdom TS populations.

The findings of many prior studies have been limited to correlations and non-parametric statistics, due to the abnormal distribution of data or small samples. Those with larger samples and greater statistical power have been able to employ multivariate analyses to examine more complex relationships and interactions between QoL outcomes, and factors hypothesised to influence them (Jalenques et al., 2012; Muller-Vahl et al., 2010).

The universal finding of these studies is that, when compared with healthy peers, global measures of QoL are significantly reduced in all TS samples (Robertson, 2011). One study found that TS was associated with more favourable global QoL ratings in comparison to adults with another chronic neurological disorder, epilepsy (Elstner et al., 2008), however these results were reversed in a paediatric sample (Eddy et al., 2011c). Another study also found that children with TS had higher global QoL of life scores than a general paediatric psychiatric sample, but lower QoL than healthy peers (Storch et al., 2007b).

QoL studies have also examined the impact of TS on specific domains of functioning. These have consistently found that adults with TS reported adverse effects in at least one functional domain including the physical, psychological, family, social and /or vocational realms (Robertson, 2011). Children and adolescents with TS have been found to have significantly impaired psychosocial functioning, and difficulties in at least one of four specific areas: family, academic, emotional and/or social life, when compared with typically developing peers (Bernard et al., 2009; Carter et al; 2000; Cutler et al., 2009; Eddy et al., 2011c; Marek, 2006; Storch et al., 2007a, 2007b).

The domains of functioning most adversely affected by the diagnosis of TS have, however, been found to vary. In general, impaired emotional functioning is the domain most adversely impacted for adults with TS (Elstner et al., 2008; Jalenques et al., 2012; Muller-Vahl et al., 2010), whilst another study found the greatest impairment in vocational functioning (Elstner et al., 2008). Most children and adolescents with TS demonstrate impaired functioning across multiple domains, with some identifying the most adverse impacts in the realm of school (Cutler et al., 2009; Storch et al., 2007), and others in the youth's social (Eddy et al., 2011a; Eddy et al., 2011c; Eddy et al., 2011c; Mareck, 2006) or emotional functioning (Culter et al., 2009). No studies have found that TS strongly negatively affects the youngster's physical functioning or ability to perform activities of daily life.

Findings of the largest QoL study of the youngsters with TS to date, Conelea and colleagues (2011) recent Internet survey of 740 North American parents of youngsters with TS or CTD and 232 youth with CTD or TS, are generally consistent with the literature. Results indicated that global QoL life was lower for the great majority of those with chronic tic disorder, and that functioning was impaired across the psychosocial realm (psychological, emotional and social domains). Physical functioning was not significantly different to normative peers. Taken together, the results of these studies suggest that the overall functioning and quality of life of the majority of adults, children and teens with TS appears to be significantly impaired in comparison with healthy peers.

### **TS and Socio-Emotional Functioning of Children and Adolescents**

As indicated by the QoL literature, many children and teens with TS experience social and emotional difficulties. Several studies have documented high levels of psychosocial stress for youngsters diagnosed with TS, particularly in the context of their relationships with peers. Psychosocial stress is an additional burden for those with TS. It appears to interact in a circular way with the youth's Tourette's, increasing the severity of symptoms, further elevating their level of psychosocial stress (Silva, Munoz, Barickman & Friedhoff, 1995). Lin and colleagues found that parent-rated psychosocial stress for their child with TS as measured by the Parent Perceived Stress Scale strongly predicted future depressive symptomology and future tic severity (Lin et al., 2007).

Sources of psychosocial stress identified in the literature include the increased incidence of bullying, teasing, peer victimisation, and stigmatization that have been recorded for youth with TS (Conelea et al., 2011; Storch et al., 2007c). Caregiver and self-reports revealed that those with TS often experienced difficulty interacting with peers. Those with TS reported feeling misunderstood, experienced rejection, were often socially ostracised and regularly practiced social withdrawal, They also encountered difficulty making and maintaining friends, and reported both lower quality and numbers of close friendships than peers with no diagnosis (Boudjouk, Woods, Miltenberger & Long, 2000; Kurlan et al., 1996; Stokes, Bawden, Backman, Dooley & Camfield, 1991; Woods, Koch, & Miltenberger, 2003; Woods et al., 2011).

The beliefs and expectations that the TS youth hold regarding the way their peers perceive them may also affect their social interactions. Although the impact of childhood experiences of stigma associated with mental and behavioural disorders are not well researched, a recent review identified important outcomes (Mukolo,

Helfinger & Wallston, 2010). This included the adverse effects of self – stigmatization, which was the consequence of the youth adopting the negative stereotypes and experiences of devaluation and discrimination that accompany their disorder.

Difficulties forming peer relationships may be in part attributable to peer perceptions of the TS youth. Studies have demonstrated that peers rated youth with TS less positively, and viewed them as less socially acceptable, less popular, less attractive, more aggressive and more withdrawn than peers with no diagnosis (Bawden, Stokes, Carol, Camfield & Salsbury, 2003; Boudjouk et al., 2000; Friedrich, Morgan & Devine, 1996; Sukhodolsky et al., 2005; Storch et al., 2007a, 2007b; Stokes et al., 1991). On examination of the consequences of negative peer evaluations of TS diagnosed youngsters, Storch and colleagues found that peer victimisation mediated the relationship between tic severity and loneliness (Storch et al., 2007c).

The characteristics and behaviours of the youth with TS may also contribute to difficulty in forming and maintaining friendships. Some research has found that children with TS had poor socialization skills when compared with age and gender-matched peers (Dykens et al., 1990), and others found that youth with TS were to 2-4 years behind peers in their level of social skills (Champion, Fulton & Shady, 1989; Cohen & Leckman, 1994). Some youth with TS may also avoid or restrict their level of social interaction. Kurlan and colleagues found that the fear associated with their own impulsive and socially inappropriate behaviour, particularly the presence of coprophenomena, inhibited the sociability of adolescents with TS (Kurlan et al., 1996).

Children with TS may also have less opportunity to meet other children and develop social skills. Having TS has been found to limit social opportunities, leisure activities, invitations to participate, outings and holidays (Conelea et al., 2011; Elstner et al., 2008; Robertson, 2011). The importance of social relationships for the emotional functioning of the youth with TS has also been demonstrated. Factors such as bullying and rejection by peers were strongly associated with higher rates of internalizing symptoms such as depression, feelings of loneliness, anxiety and increased tic severity (Storch et al., 2007a, 2007c). Youth with TS have also been found to experience elevated rates of social isolation, anxiety, low self-esteem, high levels of self – consciousness and depression (Termine et al., 2006; Walter & Carter, 1997). Although the majority of children and teens with TS are not diagnosed with clinical levels of depression, the incidence of depressive symptomology increases with adolescence. Those with TS appear to be at higher risk for adult psychopathology, with mood disorders being the strongest predictor of negative life outcomes for adults with TS (Carter et al., 2000; Muller-Vahl et al., 2010; Rickards & Robertson, 2003; Robertson, 2006a; Robertson, Williamson & Eapen 2006; Robertson, 2011; Storch et al., 2007b). Depression is the strongest predictor of adverse quality of life outcomes and higher rates of suicidality, substance abuse and inpatient hospital admissions by late adolescence and early adulthood for those with TS (Muller-Vahl et al., 2010; Robertson, 2006a).

### **TS and Family Functioning**

Other research has focused upon the impact of TS on family functioning. Parents have reported increased caregiver burden and increased rates of psychopathology, guilt relating to genetic transmission of TS, impairment in parent-

child and sibling relationships and elevated levels of family stress in several studies of families with a TS affected child (Carter et al., 2000; Cohen, Ort, Leckman, Riddle & Harding, 1988; Cooper, Robertson & Livingston, 2003; Lee, Chen, Wang & Chen 2007; Schroeder & Remer, 2007; Stewart, Greene, Lessove-Schlaggar, Church & Schlaggar, 2015; Wilkinson et al., 2001; Wilkinson et al., 2008; Woods, Himle & Osmon, 2005). Most recently, the results of a large-scale studies based on parent reported data from the 2007 National Survey of Children's Health in the USA, illustrated the increased challenges and elevations in rates of parental aggravation associated with parenting a child with TS (Bitsko et al, 2014; Robinson et al, 2013). These were further exacerbated by the youth's comorbid mental, emotional and behavioural disorders, with comorbid ASD, OCD, disruptive and conduct disorders particularly problematic in the family context (Robinson et al, 2013; Stewart et al, 2015). In a prospective longitudinal study, Carter and colleagues (1994) determined that family functioning was more closely associated with ADHD or Anxiety disorders, than severe tics or learning disorders.

Researchers have also found that caregiver burden increased with the presence of comorbid disorders (Cooper, Robertson & Livingston, 2003; Robertson, 2011; Stewart et al, 2015; Wilkinson et al., 2001). When compared with parents of children with another chronic disorder (severe asthma), parents of children with TS reported higher rates of burden and stress across multiple domains including relationships, daily activities, and psychological and physical wellbeing (Cooper, Robertson & Livingston, 2003). Woods and colleagues (2005) found that comorbid ADHD and internalizing behaviours were related to increased family stress. A qualitative study by Trificante (2007) found that mothers reported high levels of stress associated with efforts to successfully parent and alleviate the distress of their TS diagnosed child.

The author concluded that the high rate of stress for children with TS and their families was related to a perceived lack of TS specific services and supports.

In regard to their TS diagnosed child, behaviour problems, conflict at school and in the home (Cooper and Livingston, 2003), anger control problems and episodic rage caused the most concern for parents (Budman, Rockmore, Stokes & Sossin, 2003; Dooley, Bryna & Gordon, 1999). When present, episodic rage appears to have the most adverse impact on family relationships. DeLange and Olivier (2004) found that such episodes throw mothers into crisis, to the point where many require counselling support. In turn, family conflict can have an adverse impact on the TS affected child, increasing the severity of their TS symptoms (Silva, Munoz, Barickman & Friedhoff, 1995). Similarly, the child's disruptive behaviours add to family stress, however these adverse impacts have been shown to respond positively to short parent-training interventions (Scahill et al., 2006).

The quality of family life may also be adversely impacted by TS, as the effect of stigmatization and social rejection extends to the family. Some families resort to curtailing activities and impose restrictions on their social lives, in an effort to reduce their exposure to these negative experiences (Cohen, Ort, Leckman, Riddle & Hardin, 1988; Davis, Davis & Dowler, 2004; Dedmon, 1990; Kushner, 2008). Mukolo and colleagues (2010) found that "stigma by association" was commonly experienced by parents of children with mental and behavioural disorders, and contributed significantly to increased caregiver burden and family stress.

### **TS and the Intellectual and Educational Functioning of Children and Adolescents**

The experience of children with TS in the school context is highly variable, however, a significant number of children and teens do experience academic and /or

social difficulties in the school setting (Packer, 1997, 2005; Shady, Fulton & Champion, 1989; Stefl, & Rubin, 1985), and as previously noted, this was the domain of functioning most negatively impacted by TS (Storch et al., 2007a; Storch et al., 2007b).

Findings regarding intellectual ability and academic achievements of TS affected children are also inconclusive. Whilst some have found that the IQ of children with TS is average (Shultze et al., 1998), WAIS scores for 266 TS-affected children were slightly lower than unaffected peers (Debes, Hjalgrim & Skov, 2010). The authors noted, however, that these differences may have been due to co-morbid Attention Deficit Hyperactivity Disorder (ADHD) or Obsessive Compulsive Disorder (OCD), and also noted that age of onset of TS appeared to effect scores (Debes et al., 2010). Behavioural problems accompanying comorbid diagnoses, in particular obsessive-compulsive, disruptive and anti-social behaviours associated with comorbid OCD, ADHD and conduct disorders (Hanson, 1992; Robertson & Cavanna, 2008; Packer, 2005) have well documented adverse effects on learning and classroom interactions.

The high prevalence of comorbid learning disorders, both acknowledged and below threshold, also contribute to academic underperformance and classroom difficulties for some TS diagnosed children (Packer, 1997; 2005). Dyslexia, dysgraphia and acalculia are, for example, more prominent in TS populations (Dyken et al., 1990). Classroom functioning may also be impaired by the interference and fatigue caused by tics, or efforts to suppress them (Hanson, 1992; Packer, 2005; Robertson & Cavanna, 2008, 2011).



Social and emotional difficulties may also impact negatively on academic performance and classroom relationships (Hanson, 1992; Robertson & Cavanna, 2008). Fifty percent of parents of youth with TS in one study reported that their child experienced moderate to significant tic-related academic impairment, and problems relating to classmates (Packer, 2005). There is some evidence that providing school based psychoeducation and intervention aimed at changing peers attitudes and behavioural intentions towards those with TS, improved classroom interactions and social outcomes (Woods, et al., 2003; Woods & Marks, 2005). These studies reveal the key importance of acceptance and understanding to the wellbeing of those diagnosed.

### **Variables affecting the Impact of Tourette's Syndrome**

Although the association of TS and adverse consequences has been clearly demonstrated in the aforementioned studies, it is important to note that there remains significant variability between those with the disorder. Not all children with TS report lower quality of life, or display significant differences in psychosocial or behavioural dysfunction when compared with typically developing peers. Conelea et al (2011) for example found that 12% of children and teens with TS had higher QoL ratings than normative peers.

Some research has been directed towards trying to locate variables that could help to explain these within group differences. Social skills of youth with TS, school functioning and classroom factors, contextual variables associated with stress, quality of family functioning, caregiver burden and caregiver stress are some examples of factors that have been included in recent studies. A study of 40 male children and

teens with mild TS illustrated the potential complexity of factors impacting QoL. Correlations between several variables indicated factors that appeared to affect QoL outcomes in a circular manner. These included the child's age, TS symptoms, level of social competence, quality of their family relationships and the youth's behaviour in the home (Marek, 2006).

A review of the literature however reveals that, excluding age and gender, only two potential independent variables have been systematically included in published studies: tic severity and the type and presence of comorbid disorder. Studies that include these variables reveal more nuanced findings regarding the variability of the impact of TS.

### **Relationships between Tic Severity, Comorbidity, Functional Outcomes and Quality of Life**

Disentangling the effects of comorbid disorder from the effects of TS presents a challenge in both research and clinical settings (Conelea et al., 2011; Rickards & Robertson, 2003; Robertson, 2006, 2010). Some studies have been able to include comparison groups of subjects with "TS only" and "TS with comorbid disorder" to examine between group differences (Robertson, 2006; 2010). It can however be very difficult to recruit "TS only" subjects, and small, uneven groups often limit the interpretation of the results of such studies. Rather than attempting to create two groups, several researchers have measured the symptoms of all participants in order to determine the clinical correlates of TS and the impact these have on outcomes of interest, including QoL.

Findings from the majority of these studies suggest that the presence of comorbid symptoms is associated with an expansive range of long-term adverse

quality of life outcomes, and psychosocial dysfunction, particularly by late adolescence and adulthood (Bernard et al., 2009; Cavanna et al., 2009; Conelea et al., 2011; Cutler et al., 2009; Elstner et al., 2008; Gorman et al., 2010; Muller-Vahl et al., 2010; Robertson, 2006; Storch et al., 2007a; Storch et al., 2007b). Whilst Eddy and colleagues (2011a) determined that “TS only” was associated lower global QoL and adverse effects in the environmental domain (access to resources, income, food, housing, etc.), the negative effect of comorbidity extended to all QoL domains (Eddy et al., 2011b; Eddy et al., 2011c). Comorbidity may therefore be hypothesized to exacerbate or compound the adversity experienced by those with TS.

The differential effects of various common comorbidities have, however, not been well explored. As previously noted, depressive symptoms, often in addition to those of anxiety, have been found to be the strongest predictor of poor QoL for adults with TS. Specifically, depression was the strongest predictor of impairment in the quality of the individual’s social interactions and work performance (Jalenques et al., 2012). There is also some evidence that depressive symptoms in pediatric populations strongly predict negative outcomes on all QoL scales, an effect that increases over the course of adolescence (Eddy et al., 2011a). Mood disorders may affect a range of other domains including social functioning, the quality of relationships, work performance and physical health.

For pediatric TS populations, the focus of the research has been on the two most common comorbid disorders, ADHD and OCD. Storch and colleagues found that non-tic related impairment, as measured by the PedsQL parent proxy, primarily in the form of ADHD (inattentive type in particular) and OCD or obsessive-compulsive behaviours, was associated with over 70% of the youth’s difficulties (Storch et al., 2007a). Findings regarding the specific domains of functioning most

adversely effected by OCD and ADHD are however inconsistent. Whilst Cutler and colleagues found the strongest negative relationships between these disorders and emotional wellbeing and school functioning (Culter et al., 2009), others found that OCD was related to poor relationships with self, and that a combination of both ADHD and OCD was associated with poorer and more widespread dysfunction, across multiple domains (Eddy et al., 2011c). These authors also noted that both TS and comorbidity had the strongest effect in the social domain.

A related study, which employed the Child Behaviour Checklist CBCL (Achenbach, 1991) to explore a wider range of paediatric psychopathology, found that obsessive, attentional, and emotional symptoms were the strongest predictors of poor QoL (Eddy et al., 2011a). Whilst global QoL was predicted by both anxiety and depression, only depression predicted all QoL subscales, whilst the effects of obsessiveness were limited to the relationship domain. Anxiety predicted lower scores in the “self” domain, especially on the “harm” scale.

Of all of the comorbid disorders, the impact of comorbid ADHD has received the most attention in the paediatric TS literature (Carter et al., 2000; Debes et al., 2010; Dykens et al., 2009; Robertson, 2006, 2010; Spencer et al., 1998; Storch et al., 2007a, Storch et al., 2007b; Sukhodolsky et al., 2005). Several studies have consistently found that children with TS and comorbid ADHD have recorded the highest deficits on all measured areas of functioning, when compared with children with “TS only” and normative peers (Carter et al., 2000; Rizzo et al., 2007; Sukhodolsky et al., 2003). In another study, Pringsheim and colleagues found that the psychosocial health of children with TS only did not vary from that of normative peers, but was significantly lower for children with TS and comorbid ADHD and OCD (Pringsheim, Lang, Kurlan, Pearce & Sandor, 2009).

Others, however, have found that children with “TS only”, and those with comorbid ADHD, ranked similarly across multiple measures of intellectual, academic and behavioural performance, with the exception of lower performance IQ rankings for those with TS and ADHD (Dykens et al., 1990). In contradiction, when Rizzo and colleagues compared three groups of children (“TS only”, “TS plus ADHD” and “ADHD only”) with normative controls, all were significantly different on most measures (Rizzo et al., 2007). Maladaptive behaviour and impaired cognitive functioning was most strongly associated with ADHD, with or without TS, whilst the only difference between “TS only” children and controls was higher parent rated “delinquent” behaviour. Difficulties inherent in these studies were illustrated by Cavanna, Cavanna and Monaco (2008) who noted that specific TS associated anger symptoms are often overlooked, and mistakenly identified as symptomatic of ADHD and delinquency, thus challenging findings by Rizzo and colleagues (2007).

Higher levels of executive function were found to differentiate children with “Chronic Tic Disorder (CTD) only”, a gateway diagnosis for TS, from those with “CTD plus ADHD” (Roessner et al., 2007). Sukhodolsky and colleagues (2003) linked lower level social skills and competence for children with TS plus ADHD, with the disruptive behaviour accompanying ADHD. Similarly, the symptoms of comorbid ADHD predicted lower global psychosocial functioning than other comorbid disorders, or the presence of severe tics (Pringsheim et al., 2008).

Different patterns of psychopathology have also been found between children with tic disorder and those with tic disorders plus ADHD (Carter et al., 2000; Roessner, Becker, Banaschewski & Rothenberger, 2007a; Roessner, Becker, Banaschewski, Freeman & Rothenberger, 2007b). Although both groups have been reported as having higher rates of internalising disorders than non-psychiatric

controls, those with CTD plus ADHD had higher rates of externalizing symptoms than those with CTD or controls (Carter et al., 2000; Roessner et al., 2007a). Related studies that have explored the additive effects of OCD and ADHD without TS have revealed the increased burden on the child's emotional and adaptive functioning, when both are present (Sukhodolsky et al., 2005). Carter and colleagues (2000) concluded that children with TS plus ADHD, and those with "TS only" had very different socio-emotional profiles. It would appear that having a comorbid disorder is associated with adverse outcomes for those with TS, but the relationship between TS and these disorders is complex and unclear.

### **The Impact of Tic Severity**

As with the findings for comorbidity, the impact of tic severity on functional impairment is mixed and contentious. Some results report no significant effects for tic severity as an independent variable (Bawden et al., 1998; Carter et al., 2000; Eddy et al., 2011a; Stokes et al., 1991), whilst the majority of more recent studies have determined the negative effect of increased tic severity on QoL outcomes and levels of psychopathology, behavioural and social difficulties (Conelea et al., 2011; Eddy et al., 2011b; Eddy et al., 2011c; Elstner et al., 2008; Storch et al., 2007a; Storch et al., 2007b; Schoeder & Remer, 2007; Wilkinson et al., 2001; Wilkinson et al., 2008; Zhu et al., 2006). The size of the effect of tic severity has, however, varied between studies. Some have found tic severity moderately predictive of negative QoL and functional impairment (Storch et al., 2007b), whilst others determined minimal impact on measured outcomes (Muller-Vahl, 2010). Furthermore, when the effects of tic severity and type of comorbidity as independent variables are compared, the results from findings are inconclusive.

Complicating the study of the relative contribution of tic severity to adverse outcomes has been the high prevalence of comorbid psychiatric disorder. Some research has been conducted to compare the impact of tic severity on those with “TS only” and those with “TS plus” comorbid disorder. The results have been inconclusive. Whilst some have found that children with “TS only” had elevated rates of behavioural problems (Comings & Comings, 1987; De Groot, Janus & Bornstein, 1995), others found no such relationship (Stokes et al., 1991). More recently, a study compared sixty-nine youth with TS with healthy peers using the Child Behaviour Checklist - CBCL. Results indicated elevated levels of delinquency, cognitive and attentional difficulties, aggression and externalizing behaviours for youngsters with TS, and that adverse findings were positively related to increased tic severity (Zhu et al., 2006). Others have linked specific disorders such as ADHD or OCD with increased tic severity (Comings & Comings, 1987; Randolph, Hyde, Gold, Goldberg & Weinberger, 1993).

Research has also demonstrated that ADHD and OCD have been more powerfully associated with dysfunction across multiple domains than tic severity alone (Bernard et al., 2009; Comings & Comings, 1987; Eddy et al., 2011; Stewart et al., 2015). Muller-Vahl and colleagues (2010) found that tic severity contributed minimally to QoL of adults with TS in comparison to the effect of anxiety and depression. For those with mild to moderate TS, quality of life was negatively related to ADHD and OCD (Bernard et al., 2009), with inattention as opposed to hyperactivity associated with lower quality of life outcomes. Woods, Himle and Osman (2005) found that parental perceptions of the adverse impact of TS on the domain of family functioning were more related to externalizing (ADHD) and internalizing problems than to tic severity. Conelea and colleagues (2011) also found

that tic severity and comorbidity both had adverse consequences for youth with CTD and TS however impairment ratings were higher for those with CTD plus a comorbid disorder.

Other research has identified the additive effect of tic severity and comorbid disorder on outcomes. For Cutler and colleagues (2009), the combination of severe tics and co-occurring ADHD or OCD was associated with the highest levels of dysfunction and poorest overall quality of life. Others have noted that adverse social, emotional and behavioural functioning for children was related to having comorbid ADHD and complex tics (Himle et al., 2007), whilst it was this combination that was most associated with negative family functioning in a study by Wilkinson and colleagues (2001).

The combined effect of the tic severity and comorbidity may also vary as a function of age. ADHD, OCD, and tic severity were all found to negatively impact children's QoL, whilst the best predictors of increased psychopathology and poor psychosocial functioning in later adolescence and adulthood were tic severity, depression, ADHD and OCD (Eddy et al., 2011a; 2010; 2009; Elstner et al., 2008; Gorman et al., 2010; Jalenques et al., 2012; Muller-Vahl et al., 2010; Pringsheim et al., 2009). In a recent QoL study of 46 adults with TS, both tic severity and depression emerged as the strongest predictors of adverse quality of life outcomes. In this case, results were assessed using a TS specific QoL measure (GTS-QoL, Cavanna et al., 2008), which may have been more sensitive to detecting difficulties for those with TS than the general QoL measures used in other studies.

A very small number of studies have attempted to understand how tic severity and co-morbidity may interact to increase risk of adverse outcomes. Muller-Vahl and colleagues (2010) found no significant interactions between comorbidity and tic



severity, with both depression and tic severity exerting independent effects on QoL, and that the impact of depression was more powerful than tic severity. Similarly, Lewin and colleagues determined that functional impairment in adults with TS was moderated by depression and anxiety when tic severity was controlled (Lewin et al., 2011). When Storch and colleagues (2007) considered interactions between tic severity and various comorbid disorders in children, the researchers found that the negative impact of tic severity on QoL was stronger when self-reported, than with the results of parental proxy reports. Parents also reported that the negative impact of tic severity was moderated by type of comorbid disorder. The child's externalizing, but not internalizing behavioural problems, more strongly associated with adverse outcomes than tic severity. Few such multivariate analyses are, however, evident in the TS literature.

Given their demonstrated adverse impacts on youth with TS, both tic severity and comorbid diagnosis were, therefore, included as independent variables in the current study. In an attempt to disentangle the effects of tics from those of comorbidity, the current study attempted to include a greater number of youth with "TS only" than is evident in prior clinic-based TS studies. It did so by recruiting a community-based sample, although, given the clinical profile of TS, a relatively high risk of comorbidity for the group remained.

### **The Impact of Peer Attachment on Children and Adolescents with TS**

Some individuals do, however, appear to be more resilient to the negative consequences of TS than others, independent of both tic severity and the presence or absence of co-morbid diagnosis. As Conelea and colleagues (2011) found, a

proportion of children with TS (12%) rated higher quality of life than healthy controls. Whilst tic severity and comorbidity were significantly correlated with dysfunction, their moderate level of impact did not fully explain the variance between individuals.

A unique contribution and primary research goal of the current study was to test the hypotheses that a third, previously unexplored factor - the quality of the youth's peer relationships - may account for a portion of this individual variability. Given the previously discussed preliminary evidence from QoL studies indicating the adverse effects that a diagnosis of TS had upon the quality of social interactions and relationships, it was hypothesized that positive peer relationships would enhance the quality of life of youngsters with the disorder. There is considerable evidence supporting the important role played by both general and close friendships in the adaptive psychological and behavioural functioning of children, particularly by the time they enter adolescence (Wilkinson, 2010). Close relationships with peers are conceptualized as having a buffering effect against psychosocial distress and enhance the psychological wellbeing of the child (Berndt, 2002; Wilkinson, 2010).

As noted earlier in this review, there is evidence for the adverse impact of TS on the youth's relationships within the family and with peers, and the disorder is also linked with impaired social and emotional functioning. The positive impact of social relationships and psychosocial support for those with TS has, however, been under-researched. TS studies that have included social and relationships factors have generated promising results. As mentioned in the discussion of the effects of comorbidity and tic severity, positive family functioning were related to improved social and emotional functioning in children with TS, even for those with comorbid ADHD (Carter et al., 2000), and peer victimisation mediated the relationship between

tic severity and loneliness of the child with TS (Storch et al., 2011c). Others have found that family and peer relations that offer support and acceptance are positively associated with global wellbeing, increased quality of friendships and improved school performance in TS affected youth (Cooper et al., 2003; Packer, 1997; Wilkinson et al., 2001; Wilkinson et al., 2008; Woods, 2005).

In recent years, attachment theory has been increasingly employed as a conceptual framework within which close relationships and affection bonds are examined, and was therefore chosen as the current study's guiding theoretical approach. The importance of the quality of a youth's close relationships with their primary caregivers and peers as a key determinant of childhood and future socio-emotional wellbeing, social competence and optimal childhood and adolescent development, has been robustly demonstrated (Ainsworth, 1982, 1985, 1989; 1991; Berndt, 2002; Bowlby, 1982; Contreras, Kerns, Weimer, Gentzler & Tomich, 2000; Hazan & Shaver, 1987; Lee & Hankin, 2009; Kerns, 2008; Kerns, Klepac & Cole, 1996; Shaffer & Kipp, 2010; Sroufe, 1977, 2005). The quality of close relationships may assume an even more significant role for children and teens with TS, as they face the challenges of growing up with a poorly understood and often stigmatizing chronic disorder. For this study, secure attachment was conceptualized as a factor that might reduce the adverse impact of increased symptom severity and comorbidity. Specifically, it was hypothesised that secure attachment to peers would be associated with improved outcomes for diagnosed youth. It was further predicted that secure attachment might mediate or moderate the adverse impact of tic severity and the presence of comorbid disorders, resulting in higher quality of life ratings, socio-emotional functioning and adaptive skills for those with TS.

## **Attachment Theory: The Development of Attachment Style**

Attachment theorists posit that the ability to form close bonds with others across the life span is rooted in the key relationship of infancy; the enduring bond established between infant and primary caregiver (Ainsworth 1989; Bowlby 1982). The primary attachment figure, usually the infant's mother, becomes the secure base from which the child can explore the environment, and also provides a safe haven and comfort in times of distress (Ainsworth, 1989; Bowlby, 1982). Within this model, attachment is viewed as a normative developmental process that represents an innate behavioural system acquired through natural selection. It is biologically driven and therefore occurs in all contexts, including situations of inadequate care. This results in qualitative differences in the level of security experienced within the attachment relationship (Ainsworth, 1989; 1991; Bowlby, 1982; Siebert & Kerns, 2009).

According to attachment theory, it is within this primary attachment relationship (usually with the biological mother) that the child develops a sense of self and learns how to regulate emotions (Ainsworth, 1989; Bowlby, 1982; Kerns et al., 1996; Kerns, Tomisch, Aspelmeier & Contreras, 2000; Shaffer & Kipp, 2010). Through repeated interactions with the mother, the child is hypothesised to form expectations and beliefs upon which they build their internal representations - "working models" - of relationships, which they employ as prototypes for all future close relationships (Ainsworth, 1985; 1989; 1991; Bowlby, 1982).

The transactional nature of attachment is highlighted by findings that maternal attachment style strongly predicts that of the child (Ainsworth, 1989; Bowlby, 1982; Main & Goldwyn, 1984). The mother's own working models of close relationships and aggregated attachment history are hypothesised to guide interactions with her child, thereby determining the quality of the mother-child attachment and transmitting

attachment style across generations (Button, Pianta & Marvin, 2001; Main & Goldwyn, 1984; Van Ijzendoorn, 1992, 1995). Researchers have successfully measured the mother's own childhood attachment representations and found them to be strongly associated with the mother's behaviour towards her child, in ways predicted by attachment theory (Van Ijzendoorn, 1995).

Secure maternal attachment history has been associated with available caregiving, greater sensitivity to the changing needs of their developing child, appropriate parent-child boundary maintenance and an increased ability to cope with the burden of caregiving, stress and daily hassles (Main, 1996; Pianta, Marvin, Britner & Borowitz, 1998; Sroufe & Waters, 1977). Slade, Grienberger, Bernbach, Levy & Locker (2005) described the ability of the mother to hold her child's mental state in mind as reflective functioning, and determined that this played the vital role in the intergenerational transmission of attachment. The way a mother represents negative affective experiences with her child, in addition to her ability to emotionally regulate her responses, was of particular importance in determining maternal availability and sensitivity to her child's needs.

Attachment relationships are differentiated from other human relationships by several characteristics; they are usually emotional bonds of long-standing, are not interchangeable, involve proximity seeking behaviours as well as distress on separation, and grief upon the permanent loss of the attachment figure (Ainsworth, 1989; Bowlby, 1982). Ainsworth's (1989) and Sroufe, Egeland and Carson's (2005; 2009) extensive research, including the longitudinal Minnesota studies, have successfully operationalized and demonstrated Bowlby's theory.

Building on Bowlby's theory, Ainsworth (1989) developed a widely employed classification system based on the child's observed behaviour, which identified the

child as “securely” or “insecurely” (with subcategories of insecure attachment: “Ambivalent” and “Anxious -Avoidant”) attached. Ainsworth pioneered the “Strange Situation” procedure (Ainsworth, 1982; 1989), a method devised to enable the observation of the child’s attachment related behaviours following separation and reunion with the mother. This research found that secure children were able to explore their environment, and while they displayed distress when their mothers were absent, reacted positively toward the mother upon her return. Secure children were also noted to respond flexibly to threats, and to be able to acknowledge and turn to others for support and comfort when distressed. The reciprocal attachment behaviours of mothers were also observed, with mothers of secure children being consistently available, appropriately responsive to, and aware of their child’s needs (Ainsworth, 1989).

“Insecure-avoidant” children displayed detachment from and avoidance of their mothers, and were often unable to either acknowledge their distress or turn to others for comfort or support (Cassidy, 1994). Mothers for this group have been observed to be rejecting or even hostile. “Insecure-Anxious Ambivalent” children displayed distress and protest when their mothers were absent, but unlike secure children, responded with anger or ambivalence on reunion. Mothers of these children responded to their needs inconsistently or insensitively. These children often displayed elevated levels of distress and negative affect (Ainsworth et al., 1978; Cassidy, 1994; Feeney and Noller, 1996; Main & Cassidy, 1988). Within this model however, insecure attachment styles are not viewed as pathological, but instead (Main 1994) represent adaptive responses on the part of the child to suboptimal caregiving.

A fourth category, “Insecure- disorganized” was later identified by Main and Solomon in 1986 (Main, 1996). Here the child’s responses lacked coherence,

combined ambivalent and avoidant responses, occasionally demonstrated stereotypes on mother's return, such as rocking or freezing, and in some cases reversed the parental role. This style was found to be associated with psychologically unavailable parenting, whereby the caregiver's resources were limited by their own stress (e.g. death of significant other, divorce, chronic illness) or resulted from serious levels of child abuse or neglect (Schuengel, Bakermans-Kranenburg & Van Ijzendoorn, 1999).

In general, secure attachment is thought to benefit the developing child by facilitating exploratory behaviours, and protecting the child against future unsupportive or disappointing interactions with others. Insecure attachment is disadvantageous as it is based upon negative beliefs and expectations of self, others and relationships, all of which can negatively affect behaviour and the quality of relationships (Bowlby, 1982). A large body of research has confirmed that children can be classified according to their attachment related behaviours, and that these help to predict future social and emotional functioning (Ainsworth, 1989; Bakermans-Kranenburg, Van Ijzendoorn, & Juffer, 2003; Sroufe, 2005). Research has included the consideration of the child's attachment relationships with fathers, additional caregivers, and other important figures in the life of the child including friends and peers (Van Ijzendoorn & Bakermans-Kranenburg, 1996; Van Ijzendoorn & Wolff, 1997).

### **Attachment Stability and Continuity across Time and Relationships**

As previously discussed, attachment style demonstrates stability between the mother and child, however, an individual's attachment style also remains constant across close relationships and time (Bartholomew & Horowitz, 1991; Fraley, 2002; Fraley & Davis, 2005; Hazan & Shaver, 1987; Waters, Hamilton & Winfield, 2000;

Weinfield, Waley & Egeland, 2004). Longitudinal studies, in particular the large-scale prospective Minnesota Study by Sroufe and colleagues (Sroufe, 2005; Sroufe, Egeland, Carlson, & Collins, 2005, 2009) and the National Institute of Child Health and Development (NICHD) Study of Early Child Care (Belsky, 2005), have demonstrated the temporal stability of attachment for infants, children and adolescents. Berlin, Cassidy & Appleyard's (2008) recent comprehensive review of the attachment literature also led them to conclude that early attachment bonds were causally linked to the individuals' ability to build relationships with others, particularly for those relationships involving close emotional bonds.

It was the work of Hazan and Shaver beginning in the 1980s that provided the impetus for exploring attachment relationships beyond early childhood. These researchers were able to identify adult attachment categories that closely resembled those of Ainsworth's childhood system, a system subsequently revised and extended by Bartholomew and Horowitz (1991). Researchers developed psychometric instruments to assess late adolescent and adult attachment, including the widely employed Adult Attachment Inventory (A.A.I.) (George, Kaplan & Main, 1985). A growing body of research suggests that attachment persists into adulthood, that adult attachment in close relationships can be classified, and that these classifications can predict the attachment related behaviour of adults (Bartholomew & Horowitz, 1991; Fraley & Shaver, 2000; Hazan & Shaver, 1994; Heffernan, Fraley, Vicary & Brumbaugh, 2012; VanIjzendor, 1992).

The causal mechanisms for the stability of attachment are, however, not well understood. Some research indicates that the attachment style of the youth can change, particularly in response to highly stressful negative life events that impact adversely upon family (Waters, Hamilton, & Weinfield, 2000). The loss of a parent,



divorce, or chronic illness of the primary caregiver are examples of stressors that have been associated with a shift from secure to insecure attachment in children and adolescents (Waters et al., 2000; Weinfield et al., 2004). The NICHD ECCRN (2005) identified continuity in parenting quality as the factor most closely associated with changes in security of attachment for children, and these effects could be bidirectional.

Adult attachment related behaviour might also change in response to stress and high emotion (Berlin, Appleyard & Cassidy, 2008). Insecure mothers reporting high, but not low levels of self-reported daily parenting stress were associated with less positive mother-child interactions (Phelps, Belsky & Cronic, 1998) whilst others found that a mother's positive parenting was related to her adult attachment style only in those reporting high levels of psychological distress (Mills-Koonce et al., 2007; Hill-Soderlund et al., 2008). Roisman and colleagues determined that an adult's working model of attachment might be modified in response to the quality of their current attachment relationships (Roisman, Madson, Hennighausen Sroufe & Collins, 2001; Roisman, Padron, Sroufe & Egeland, 2003). In this model, adults with poor quality parental attachment history may move from insecure to secure attachment in the context of a close adult attachment relationship. These findings support Bowlby's proposition that the attachment system is open and dynamic, although the model suggests that change is expected to be more likely during early development.

The diagnosis of TS and the difficulties associated with life in its aftermath can be construed as a significant stressor that may affect change in the quality of the youth's parental, family and peer relationships. A significant literature has documented common stressors associated with parenting a child with a chronic illness, though no such studies have included the parents of children or teens with TS.

In a meta-synthesis of eleven published qualitative studies of this phenomenon, Coffey (2006) identified common themes reflecting the stressors associated with parenting a child with a serious medical or neurological condition (e.g. Spina Bifida, Cerebral Palsy and Juvenile Diabetes). These included the mother's sense of omnipresent worry; feelings of helplessness and difficulty in maintaining optimism; assuming the burden of care; and having to take charge and become the expert in their child's illness. In addition, mothers struggled to keep their families together; to find ways of connecting with the outside world; and to be able to cope at particularly stressful and crucial times in the life of the child such as diagnosis, and during developmental transitions.

The diagnosis of a child with a chronic condition is not only a stressful experience and time of great crisis for parents, it is also time of grief, and is often described in the literature as the "loss of their ideal child" (Coffey, 2006). Grief and loss are important concepts within attachment theory. It is hypothesized that the inability to resolve the grief associated with early childhood loss of the primary attachment figure results in insecure future attachment relationships (Bowlby, 1982; Main et al., 1985). Although limited research has considered the impact of diagnosis on attachment functioning, some studies have explored the quality of maternal attachment and changes in mother – child attachment following the diagnosis of developmental or chronic neurological disorder in the child (Barnett et al., 2006; Oppenheim, Koren-Karie, Dolev & Yimiya, 2009). These have found a relationship between an increased rate of child insecurity and the mother's adverse response to diagnosis. A mother's ability to resolve her grief over time, higher level of insight and acceptance of the child's condition and an understanding of the child's experience were related to increased security and less family stress (Barnett et al., 2006;

Oppenheim et al., 2009). In a study of 70 youngsters with Cerebral Palsy, mothers with secure attachment style were able to resolve their grief more successfully than insecure peers (Marvin & Pianta, 1996). The authors hypothesized that for the insecure mother, diagnosis mirrored her early experiences of loss, thereby activating fearful, avoidant or dissociative responses. These responses in turn perpetuated her anxiety, preventing her from successfully resolving her grief and impairing her ability to provide sensitive caregiving and secure base functions for her child.

### **Developmental Stages and the Extension of Attachment**

Bowlby's model also stated that children respond to the complex social networks into which they are born by forming multiple attachment relationships, hierarchically arranged in level of importance. In addition, a developmental approach suggests that these attachment relationships are formed in response to the goals of each life stage (Bowlby, 1982).

In infancy and early childhood, attachment extends first from mother to father, thence to siblings and other family members or important caregivers. Although attachment to primary caregivers may remain strong throughout childhood and adolescence, as children mature they appear to sequentially transfer attachment-related functions from parents to peers (Kerns et al., 2000; Mikulincer & Shaver, 2007; Nickerson & Nagle, 2005; Sroufe, 2005). Although there is a large body of research regarding attachment in infancy and early childhood, it is only in recent years that attention has been turned toward attachment to parents and peers during middle childhood years (7-12 years of age) and adolescence.

There is also a current debate regarding the time at which children begin to form the type of friendships with peers that can be identified as attachment relationships. This involves a wider discussion in the attachment literature relating to the “narrow” and “broader” views of attachment (Belsky & Cassidy, 1994). Research adopting the “narrow” view only considers the quality of the individuals closest (“affectional”) relationships, whilst a “broader” view of attachment encompasses both affectional (close or intimate) and non-“affectional” relationships (friendships) (Dykas & Cassidy, 2011). Rubin and colleagues (2004) argue that it is the attachment related function played by the youths peer friendships that is of relevance to attachment theory, not exclusively the “closeness” of the affectional bond. Hazan and Shaver (1987) also explained that whilst attachment functions (proximity seeking, safe haven and secure base) remain a central feature of development, they change in form to reflect the differences between the attachment relationships of children, adolescents and adults.

During middle childhood and early adolescence, the change in the child’s focus from relationships with their parents, to friendships with peers, is dramatic (Nickerson & Nagle, 2005). Attachment behaviours such as proximity maintenance (desire to be physically close) and safe haven (relying on a person for comfort) that were once directed toward parents, are increasingly directed toward peers (Nickerson & Nagle, 2005; Mikulincer & Shaver 2007; Schneider, Atkinson & Tardif 2001). It is an important time in the child’s social development, in which friendships become deeper and perform more complex functions (Ainsworth, 1989; Bartholomew & Horowitz, 1991; Fraley 2002; Kerns, 2008; Rubin et al., 2004). By integrating the developmental theories of H. S. Sullivan (1953, cited in Youniss 1982) and Piaget (1965, cited in Youniss, 1980) Youniss (1982) posited that it is during their

interactions with friends that children achieve a major developmental goal - by learning the principals of reciprocity, a characteristic associated with lifelong interpersonal adjustment and socio-emotional wellbeing (Youniss, 1982).

Findings for attachment behaviours directed towards peers for children in middle childhood years have, however, been mixed. Kerns (2008) found that the function of peer relationships in middle childhood were oriented towards companionship goals and not to attachment. However, Seibert and Kerns (2009) found evidence that children's attachments increasingly extended in a hierarchical fashion to others at certain times, directing secure base behaviour towards significant caregivers (e.g., teachers, grandparents) and other family members, particularly older siblings, as well as their peers, as they matured. Proximity seeking and safe haven functions for peer relationships have also been found to increase by early adolescence (Nickerson & Nagle, 2005). Gender differences were however noted, with females more securely attached to peers than males. Furthermore, children and adolescents with less secure parental relationships were more likely to seek out peers to fulfill attachment functions. Youngsters between 8 -14 years of age increasingly seek safe haven, comfort and support from peers (Hazan & Zeifman, 1994). Fraley and Davis (2005) found on examination of the working models of attachment of teens, those who were securely attached reported using a best friend as an attachment figure. In contradiction, Freeman & Brown (2001) found that ninety percent of those with secure attachments to parents reported the parent as their primary attachment figure, but the large majority of those with insecure parental relationships identified peers as their primary attachment figure.

It is important to note that research into middle and later childhood is fledgling; results are inconsistent and characterised by a lack of adequate instruments

suitable to assess attachment at this age (Kerns et al., 2000; Kerns 2008; Dwyer 2005). There is currently only one a brief, single-item measure of attachment to peers currently available for middle childhood (Murriss, Meesters, van Melcik & Zwambag, 2001)

The transition of attachment to peers has been found to intensify during adolescence, as teens strive to achieve their developmental goal of autonomy, build deeper relationships with peers and initiate fledgling romantic partnerships (Nickerson & Nagle, 2005; Waters et al., 2000). This does not mean that secure attachment to parents is no longer of importance to teens. Even as adolescents increasingly turn to peers to fulfil attachment needs, they continue to rely on their parents to provide the key attachment functions such as of the provision of a secure base (Allan & Land, 1999). Research has found that secure attachment to both parents and to peers was associated with successful adolescent transition (Laible, Carlo & Raffaelli, 2000). Moderate to strong correlations have also been found between attachment to parents and attachment to peers for adolescents (Laible, 2007), and both types of attachment relationships appear to have similar effects on adolescent psychosocial adjustment. Papini & Roggman's (1992) longitudinal study found that secure attachment to parents had a buffering effect on the transition to adolescence, with secure attachment at age twelve predicting perceived self-competence and emotional wellbeing at age thirteen. However the increased importance of peer attachments in adolescence is evident in the findings that teens with insecure relationships with parents, but positive relationships with peers, had higher levels of emotional adjustment than those with secure parental but insecure peer relationships (Laible et al., 2000).

Given the difficulties youth with TS experience in their relationships with peers, diagnosed adolescents may be particularly disadvantaged by the barriers they encounter as they attempt to form secure peer attachments. The process of the transfer of attachment from the primary caregiver that began in childhood is thought to culminate in the formation of romantic relationships and close friendships of adulthood (Hazan and Shaver, 1987). Although the study of the extension of attachment to other relationships in middle childhood and adolescence is in a relatively early stage, researchers have identified multiple relationships that appear to qualify as attachment bonds by adulthood. This is particularly so in cases whereby the criteria for determining attachment was not overly restrictive and reflected age appropriate functioning (Doherty & Feeney, 2004; Hazan & Zeifman, 1994; Trinke & Bartholomew, 1997). As Doherty and Feeney (2004) determined, adult attachment networks reflect normative life events such as forming a sexual relationship or becoming a parent.

By young adulthood, Hazan and Zeifman (1994) determined a hierarchy of attachment relationships with the romantic partner (if present) as the primary attachment figure, followed by relationships with mother, thence with father, siblings and close friends. Others have examined the sequencing of various forms of attachment behaviours that emerge during the formation of attachment relationships. Proximity seeking was the first noted behaviour in the formation of adult attachment relationships, followed by safe haven thence secure base, and that attachment relationships may form more quickly in adulthood than had previously been thought (Heffernan, Fraley, Vicary & Brumbaugh (2012).

## **The Impact of Attachment on Child and Adolescent Psychological and Behavioural Functioning**

A significant body of research confirms the importance of attachment security for the adaptive psychological and behavioural functioning of the developing child from infancy to adulthood (Bartholomew & Horowitz, 1991; Kerns et al 1996; Mikulincer, Shaver, Gillath & Nitzberg, 2005; Rubin et al., 2004). Although the primary focus has been upon the quality of parent (usually maternal) - child attachment, the quality of peer relationships on outcomes is increasingly subject to study, particularly during adolescence and occasionally during middle childhood years.

In summary, secure attachments in childhood have been associated with positive outcomes for the individual. These include increased self-confidence, self-understanding and self-esteem; enhanced cognitive functioning; emotion regulation ability and psychological adjustment; adaptive behaviours; academic achievement; social competence; and relationship satisfaction. In contrast, the negative consequences of insecure attachment in childhood include lower self-esteem; behavioural problems; difficulty regulating emotions; higher rates of externalizing and internalizing symptoms; learning difficulties; and difficulties in forming and maintaining close social relationships (Ainsworth, 1985, 1989, 1991; Berlin et al., 2008; Bowlby, 1982; Goodvin, Meyer, Thompson & Hayes, 2008; Jacobsen & Hoffman, 1997; Warren, Houston, Egeland & Sroufe, 1997).

Attachment has been most strongly related to the social competence of the developing child, from infancy to adulthood (Allen & Land, 1999; Booth –LaForce et al., 2006; Kerns et al., 1996). Simpson, Collins, Tran and Hayden (2007) conducted a longitudinal study of 78 subjects from infancy to early adulthood and found a double



meditational role for security of attachment at three pivotal developmental times. Secure attachment in infancy predicted greater social competency in early middle childhood, which in turn predicted having more secure peer relationships at sixteen. This predicted higher rates of positive emotional experiences and lower levels of conflict in adult romantic relationships (Simpson et al., 1997). Secure attachment has also been found to enhance intimacy in close relationships in early adulthood (Grabill & Kerns, 2005). Secure attachment to parents also predicts the youth's competence in forming friendships (Frietag, Belsky, Grossman, Grossman & Sheuer-Englisch, 1996).

Children of all ages who were securely attached to their parents have also demonstrated increased empathy, lower levels of hostility and aggression and increased ability to show more positive affect with peers (Sroufe et al 2005). In early childhood, increased self-reliance and decreased attention seeking and dependent behaviours have been noted for securely attached children. Conversely, insecure children, have been found to be less ego-resilient, have lower self-esteem, and less flexible than secure peers (Kerns, 2008). Secure attachment to mother, and mother's positive affective response to the child have been linked to the child's ability to view themselves as psychological beings, with enhanced and consistent self-concept (Goodvin et al., 2008)

Secure attachment to parents also predicts psychosocial wellbeing into adulthood (Fraley & Davis, 2005), whilst insecure attachment has been consistently associated with increased risk for psychopathology (Allen, Moore, Kuperminc & Bell, 1998; Mikulincer & Shaver, 2007; Rutter, 1995), particularly the increased risk of depression and anxiety in adolescence (Contreras et al., 2000; Granot & Mayseless, 2001; Lee & Hankin, 2009). In a review of the literature, Brumariua and Kerns (2010)

concluded that insecure attachment to mothers is linked with internalizing disorders in young people, particularly for depression and anxiety in late childhood and adolescence. As adolescence is a period of great change, it can be associated with increased risk for problems such as depression, conduct disorder, suicide and substance use (Adams, 2005). Although most cope well, Mason and colleagues (Mason, Cauce, Gonzales & Hiraga 1994) found that a significant minority of adolescents experienced distress and uncertainty. Liable and colleagues found that secure attachment to parents and to peers was associated with successful adolescent transition (Liable et al., 2000). Others have identified the link between relational competence and attachment to parents, which predicted emotional adjustment in early and mid-adolescence (Engles, Finkenauer, Meeus, & Dekovic, 2001; Booth-Laforce et al., 2006).

Bradford & Lyddon, (1993) found that secure attachment to parents in adolescence was associated with higher self-esteem, improved life satisfaction and lower levels of psychological distress. Secure attachment to peers has also been related to lower levels of anxiety and depression in younger children (middle childhood) and teens (Muris & Maas, 2004; Muris & Meesters, 2002; Muris, Mayer, Meesters, 2000; Muris, Meesters, Merckelbach & Hulskenbeck, 2000; Muris, Meesters, Vandenberg, 2003; Muris et al., 2001). The results of a large nonclinical sample of adolescents ( $N = 476$ ) concurred with these findings, with the authors suggesting that secure peer attachments appeared to buffer the distress experienced during adolescence (Nelis & Rae, 2009).

The relationship between psychopathology and attachment has been demonstrated in clinical populations studies. The overwhelmingly majority of 14 year old boys ( $N = 66$ ) with a history of psychiatric admission for (having excluded

organic and thought disordered youth) were found to be insecurely attached and that this was associated with criminality and the use of hard drugs by age twenty-five (Allen, Hauser & Borman-Spurrell's, 1996). Secure parent-child attachment also predicted lower parent-rated aggression, lower levels of social stress and higher self-esteem in boys in middle childhood with disruptive behaviour disorders (n=91) referred to a Singaporean clinic, mirroring attachment findings in Western populations (Phaik Ooi, Ang, Fung, Wong & Cai, 2006).

Security of attachment has been found to uniquely explain variance in behavioural, as well as emotional difficulties, of teens when controlling for parenting style, a factor also identified as being associated with adolescent and child psychosocial functioning (Scott, Briskman, Woolgar, Humayun & O'Connor, 2011). In their meta-analysis of fifty-three studies, Faeron, Bakermans and Anderson (2010) revealed a strong relationship between early maternal insecure attachment and later externalizing behaviour, particularly for boys.

In their longitudinal study, researchers (Booth, 1994; Booth, Rubin, & Rose-Krasno, 1998) found that the quality of mother-child attachment at age four was the strongest predictor of internalizing problems and social engagement or acceptance at age eight. Externalizing problems at age eight, however, were not found to be associated with insecure attachment, but were most strongly and negatively predicted by the experience of maternal warmth.

The impact of attachment style was also demonstrated when environmental risk was controlled. For a sample of moderately "at risk" adolescents, secure attachment was related to competence in peer relationships, lower levels of internalizing and deviant behaviours, while insecure teens reported higher levels of internalization and deviance (Allen et al., 1998). The adolescent's ability to cope

positively with stress was also related to secure attachment (Howard & Medway, 2004). Secure attachment was associated with the teens' adaptive coping strategies, such as the use of family communication, whilst insecure attachment was related to negative avoidance behaviours such as substance use. Antisocial behaviour in early adolescence has also been modestly related to insecure attachment to parents, but not to peers (Marcus & Betzer, 1996)

Insecure attachment, however, does not always indicate disorder or maladaptation. Within normative samples, as many as 30 % of children are insecurely attached (Ainsworth, Blehar, Waters & Wall, 1978), yet many appear to function well (Ein-Dor et al., 2010). Similarly, attachment style for children and teens is not always stable, and may alter in response to changes in life's trajectory (Iwaniec & Sneddon, 2001). As Yirmiya (2009) suggests, secure attachment may be best viewed as a protective factor that helps the youth actualize their developmental potential, whilst insecure attachment inhibits it.

### **Attachment and Peer Relations**

In addition to its association with adaptive psychological functioning, secure attachment has been positively correlated with increased social competence and the ability to form friendships, both of which are key factors in determining the child's adjustment and future socio-emotional wellbeing (Collins & Laursen, 2004; Dykas et al 2004; Kerns et al., 2000; Rubin et al., 2004; Suess, Grossman & Sroufe, 1992). Children with poor peer relationships are at increased risk for aggressiveness, academic problems, mood disorders, and loneliness (Wentzel & Caldwell, 1997). As the child matures, the negative consequences of poor peer relationships intensify.

Rubin and colleagues hypothesized that the ability to form qualitatively rich friendships was of particular significance in late childhood and adolescence because of the central role these relationships play in the developmental process (Rubin et al., 2004). In a longitudinal study of the quality of early adolescent friendships, those who reported having no friends had lower self-esteem, and more psychopathological symptoms in adulthood (Bagwell, Newcombe & Bukowski, 1998). Although self-esteem has been positively related to secure attachment to peers, it appears to be a complex relationship mediated by empathy and pro-social behaviour (Laible, Carlo & Roesch, 2004).

Laible et al (2000) demonstrated that youngsters benefit optimally by having secure relationships with both their parents as well as their peers. The researchers studied adolescents (n=98) to explore the relationship between security of attachment to parents and security of attachment to peers. They found that those who reported secure attachments to parents and secure attachments to peers demonstrated the highest level of adjustment, across all adjustment measurement indices. As these children aged, improved adjustment was more closely associated with secure attachment to peers.

A large literature indicates that securely attached children have many advantages over insecure peers in terms of their ability to make and maintain positive peer relationships. Using observation, structured interviews and various parent, teacher and self-rated psychometric instruments, cross-sectional and longitudinal studies have revealed that securely attached youth are more socially adept than insecure peers (Benson, McWey & Ross, 2006; Schneider et al., 2001).

Securely attached children and teens demonstrate greater social competence from infancy to later adolescence (Kerns et al., 2000) and have more positive peer

group status than insecure peers (Feeney & Cassidy, 2003; Feeney, Ziv & Cassidy, 2003; Kerns et al., 2000; Rubin et al., 2004). Conversely, insecurely attached children had been found to experience more problematic and disturbed relationships with peers than secure youth (Sroufe, 2005).

Security of attachment in infancy has predicted increased social competence in early childhood, and securely attached children are reported to have higher levels of general and social competence and quality friendships than insecure peers (Sroufe, 2005). Secure youth demonstrate more confidence in their explorations of new relationships, better relationship development skills that render them more attractive to peers, and hold higher expectations that friendship will have positive consequences when compared with insecure peers (Kerns, 1996; Kerns et al., 2000). Bohlin and colleagues (Bohlin, Hagekull and Rydekk (2000) found that secure infants were more socially active, positive and popular at school age, and tended to report less social anxiety than insecure children. Securely attached children and teens also view themselves as worthy of care and affection, regard friendships as worthwhile, are more trusting of adults and peers and are more satisfied with their relationships than insecure children and teens (Laible, 2007; Bowlby, 1982; Dykas et a 2006; Kuperminc, Allen & Arthur 1996). On the other hand, insecure youth may hold beliefs that they are unworthy of care and unlovable (Laible et al., 2004), and have been found to express less positive and more negative emotions in their relationships (Simpson et al., 2007). Insecure attachment has also been related to interpersonal conflict, hostility and manipulative behaviour with peers (Feeney & Cassidy, 2003). Secure attachment has been associated with higher rates of reciprocity in peer relationships (Laible, 2007), and number of reciprocated classroom friendships (Kerns et al., 2006). Feeney and colleagues found that an adolescents attachment style was

associated with reciprocal interactions, even with strangers, in ways consistent with attachment theory (Feeney, Cassidy, Ramos-Marcuse, 2008).

Some research has been directed towards identifying which peer relationships are most influenced by security of attachment. Recent meta-analyses suggest that attachment security may have the strongest effect on the child's ability to develop close relationships (Kerns, 1996). One of these meta-analyses included 63 studies on the link between parent-child attachment and peer relations (Schneider et al., 2001). It suggested that although attachment style was related to social competence and general sociability, it might be even more strongly related to close friendships. Although secure attachment was related to competence in peer relationships, the effect size was higher for the quality rather than the quantity of friendships.

Although both peer and parental attachment were important to adolescent friendships, Benson, McWey and Ross's (2006) meta-analysis (53 studies, 12,482 participants) found stronger effects for parent-adolescent attachment on the quality of the adolescent's closest friendships. A recent Australian study, employing a newly developed adolescent friendship attachment measure, found that secure attachment to close friends was positively correlated with self-esteem, self-competence, attitude towards school and lower levels of depression (Wilkinson, 2010). These findings tentatively suggest that close peer relationships contribute more to the child's psychosocial functioning than peer relationships in general (as measured by the Parental and Peer Attachment- IPPA, Short Form inventory- Armsden & Greenberg, 1987).

## **Conclusion - Literature review**

In summary, the rationale for the inclusion of security of peer attachment as an independent variable in the current study was based upon the vast body of research attesting to the importance of secure attachment in determining child and adolescent psychosocial wellbeing, behaviour and optimal development. Peer attachment security was conceptualised as an important psychological predictor of wellbeing and functioning; and TS as a stressor with the potential to disrupt or otherwise impair the attachment relationships of diagnosed youth. The present research study also sought to examine whether peer attachment security mediated or moderated the effects of increased tic severity, and the presence of a comorbid disorder, on outcomes such as quality of life for youth with TS. Finally, given the stability of attachment demonstrated across generations, time and relationships, the study was extended to include the impact of TS on both peer and mother-child attachment. The multiple research goals and hypotheses for the current study will now be presented in the following Section 2, Chapter 2.



## **SECTION 2 CHAPTER 2 OVERVIEW OF THE RESEARCH**

### **Primary Research Objectives**

Tourette Syndrome is a highly visible, poorly understood and surprisingly common neurodevelopmental disorder with well-documented adverse psychosocial consequences for those diagnosed. Yet psychologists are under-represented in both the provision of care and the advancement of research regarding this distressing disorder. The primary objective of the current research, therefore, was to explore the role of a psychological variable – the security of peer attachment - in determining individual variability in outcomes such as quality of life and functioning demonstrated by youth with the diagnosis. It was hoped that findings from the study could be used to inform psychological interventions designed to address the unmet needs of this clinical population.

In order to achieve the multiple objectives of the current research, two separate studies employing both quantitative and qualitative techniques were conducted (Barker & Pistrang, 2005). The quantitative component of Study 1 (Study 1, Part A) comprised a controlled, survey-based study (n = 194) designed to test multiple hypotheses. The main objective was to reveal hypothesised relationships between the quality of peer attachment and variability in the quality of life (QoL), psychopathology, and behavioural and social outcomes reported for TS diagnosed youth. The study also examined the impact of two additional independent variables - comorbid disorder and tic severity - on measured outcomes. To address the problem of overreliance on small, clinic-based samples apparent in the TS literature, a nation

wide community-based youth sample was recruited. In view of the ethical and practical constraints involved in surveying children as young as aged seven, participation was restricted to the youth's primary caregiver. Multivariate statistical analyses were employed to examine hypothesised differences between the responses of TS group parents ( $n = 86$ ), and those of a broad sample of parents of peers with no known psychiatric or medical diagnosis ( $n = 108$ ); and to investigate differences within the TS group.

An additional, major research objective for this project was to explore and expand upon current understandings of the adverse impact of TS on the youth's peer attachment relationships. Due to the lack of a multidimensional psychometric measure of attachment in middle childhood, and in light of the novel and exploratory nature of the study, qualitative methodology was employed (Study 1, Part B). All parents in Study 1 were invited to provide written responses regarding their child or adolescent's ability to form peer attachments. The factors emerging from the implicit theories of parents were analyzed, and findings compared between groups.

A second major study (Study 2) was conducted with the goal of gaining detailed insights into the lived experience of TS, and deeper understandings of the manner by which TS influenced important attachment relationships, and functioning within these relationships. Specifically, Study 2 extended the research to include an exploration of the quality of the youth's primary attachment relationships (the mother – child relationship - MCR), in addition to child-peer attachments. The sample comprised a subset of mothers ( $n=22$ ) from the TS group in Study 1. The stability of attachment across time and relationships (maternal attachment history, the MCR and the youth's peer attachments) was also explored for each dyad in this study, and any estimated change in attachment associated with TS noted. Due to the novel and

exploratory nature of this research, a qualitative design based on semi-structured interview data was employed. A methodology grounded in attachment theory was designed for the purposes of the research, and employed to explore and estimate the security of the attachment relationships under study. In order to remain open to the voices of participants, and to facilitate the emergence of unexpected data, all interview questions were open ended.

The final research objective addressed an additional gap in the TS literature - the relative dearth of research conducted for the Australian TS population. Results from Study 1, Part A were therefore employed to reveal the clinical profiles and correlates of TS in the Australian context for first time, thereby facilitating comparison with international findings. Secondly, data relating to experiences within the health and education systems of youth with TS and their parents was collected. Such information is critical to identifying the needs of the Australian TS community, to support advocacy, and to enable the design and implementation of appropriate services and supports in response. It can also inform the training programs of undergraduates and professionals, including psychologists, to optimally service the multidimensional needs of the Australian TS community.

### **Hypotheses for Study 1**

1. That youth with TS would be reported as having lower quality of life, higher rates of psychological, behavioural and social difficulties, and higher rates of insecure peer attachment than undiagnosed peers (as reported by the youth's primary caregiver)
2. Secure attachment to peers would be associated with improved quality of life outcomes and lower rates of psychological, behavioural and social difficulties,

with inverse results expected for insecure peer attachment for all children and adolescents (as reported by the youth's primary caregiver)

3. That insecure attachment, increased tic severity and comorbid disorder would be associated with lower quality of life and higher rates of psychological, behavioural and social difficulties for children and teens with TS (as reported by the youth's primary caregiver)
4. That secure peer attachment would moderate or mediate the adverse impact of tic severity and comorbid disorder on quality of life outcomes (as reported by the youth's primary caregiver)
5. That there would be variability in factors impacting the child or teens' ability to form peer attachments identified by TS group parents and those of undiagnosed peers
6. That the clinical phenomenology and clinical correlates of TS presenting in this Australian sample would mirror international findings

### **Hypotheses for Study 2**

As this study was largely exploratory, only two *a priori* hypotheses were tested.

1. That TS would impact the security of the MCR and peer attachments of youth in the study.
2. That stability of attachment would be demonstrated across time, generations and relationships.
  - a. Specifically, the mother's ability to form a secure attachment relationship with her TS diagnosed child would be related to her own childhood attachment history and current attachment style.

- b. That security experienced within the Mother-child relationship would predict the child's ability to form secure peer attachments.

### **Outline of the Presentation of the Research**

The current research is presented in the form of seven research reports written for publication in peer reviewed journals (*Published, In Press, Under Review or Ready for Submission*) and presented in Section 3, Chapters 1-7, and one thesis chapter (Section 4, Chapter 1). All findings are based upon the two major studies (Study 1, Parts A & B; and Study 2) comprising the current research project. Research reports are formatted to meet the requirements of the journal targeted for publication. References for individual reports are, however, presented in APA style to enhance readability, and a combined reference list is provided at the end of this thesis.

The research began by determining the clinical profile and correlates of TS in a broad community-based sample of Australian youth, and identifying the adequacy of services and supports currently reported, with results reported in Section 3, Chapter 1. The findings from Study 1 also revealed the challenges and difficulties associated with living with TS for this sample of Australian youth in comparison with undiagnosed peers, which were discussed in Section 3, Chapter 2.

The primary goal of this research project, however, was to test the main hypotheses that peer attachment, tic severity and the presence of comorbid disorder would account for variability in quality of life outcomes and rates of psychological, behavioural and social difficulties for those diagnosed. This is discussed in Section 3, Chapter 2.

The current analyses also attempted to disentangle the impact of TS on outcomes such as quality of life and functioning from those associated with individual comorbid disorders included in the TS group youth, with results presented in the Section 3, Chapter 7. Furthermore, during the process of determining the clinical profiles of this sample of youth with TS, the high rate of emotional disorder associated with the diagnosis was revealed. Given the clinical significance of this finding, an additional study examined the nature of the relationship between TS and increased signs of anxiety and depression in diagnosed youth. These findings are reported in Section 3 Chapter 4.

Factors to emerge from the quantitative and qualitative analyses regarding the impact of TS on the quality of peer relationships and social functioning are reported in Section 3, Chapters 3. Qualitative data from the Study 2 was analysed to explore the impact of TS on the mother – child relationship. Findings are presented in Section, Chapter 6. In addition to the impact TS had upon mother-child attachment, qualitative analyses also identified a range of more general stressors associated with parenting a child or teen with TS in the Australian context. Section 3, Chapter 5 examines these stressors and to compares them with those experienced by parents of youth with other serious chronic disorders.

Finally, the hypothesis that attachment security would demonstrate stability across generations, time and relationships was explored in Section 4, Chapter 1.

Section 5 presents the integrated findings from both studies, and the recommendations for practice and policy emerging from this major research project. Two abstracts of conference presentations (Australian Psychological Society 50<sup>th</sup> Anniversary Conference, 29th September – 2<sup>nd</sup> October, 2015) based on the current research are included in Appendix E.

**SECTION 3 CHAPTER 1. Recognising and Treating Tourette's in Young Australians: A Need for Informed Multidisciplinary Support**  
*Australian Psychologist (In Press)*

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**This chapter is a report on the study investigating the clinical profile of the current sample of Australian youth with TS. The study also explored the types, adequacy and accessibility of services currently available for the Australian Tourette's community.**

### Abstract

*Objective:* This study aimed to evaluate the complex clinical phenomenology of Tourette's syndrome within a community-based Australian sample. It also aimed to determine the service needs of this population. *Method:* Participants included parents of individuals with Tourette syndrome (n=86; Mean age = 11.4, sd = 2.8) and control group peers (n=108; Mean age = 11.3, sd = 2.6). Clinical phenomenology was assessed using the Parent Tic Questionnaire, Strengths and Difficulties Questionnaire. A comprehensive family history and reports of service needs were also gathered. *Results:* Findings mirrored the profile of Tourette syndrome reported in international populations, revealing heterogeneous tic and comorbidity profiles. High rates of comorbidity (73%), often involving multiple co-occurring diagnoses were reported for the Tourette syndrome group. These included Obsessive Compulsive Disorder (44%), Anxiety Disorder (36%), Attention Deficit Hyperactivity Disorder (32%) and Learning disorders (18%). Results of the Strengths and Difficulties Questionnaire determined significant elevations in symptoms of conduct and mood disorders for individuals with Tourette syndrome ( $p = .01$ ). Parents also reported a lack of health and education services, low levels of understanding of Tourette syndrome by health professionals and high rates of stigmatisation within the Australian context. *Conclusions:* The study confirmed the challenges confronting youth with Tourette syndrome and revealed pronounced deficits in Australian health and educational services for individuals with Tourette syndrome. Findings indicate the urgent need for increased education for health professionals and the provision of multidisciplinary services to meet the complex needs of this clinical population.

*Key words:* Comorbid disorder, diagnosis, intervention, service provision, stigma, Tourette syndrome



## Introduction

Tourette's syndrome (TS) is a complex, childhood onset neurodevelopmental disorder of unknown aetiology (Felling & Singer, 2011; Robertson, 2012). It is characterised by the presence of multiple involuntary motor and vocal tics, usually occurring in bouts over a period of at least one year (American Psychiatric Association, 2013). With prevalence rates of 1-3% of the paediatric population (Eapen & Robertson, 2008; Robertson, 2008), clinical presentation is highly heterogeneous and greatly complicated by high rates (up to 90%) of comorbid diagnoses, most common of which are Attention Deficit Hyperactivity Disorder (ADHD) and Obsessive Compulsive Disorder (OCD) (Cavanna, Servo, Monaco, & Robertson, 2009; Freeman et al., 2000; Kurlan et al., 2002; Robertson, 2006a, 2006b, 2008; Termine et al., 2006). TS has been consistently associated with a wide range of highly adverse outcomes including reduced quality of life and impaired psychosocial, behavioural, academic and family functioning (Eapen & Črnčec, 2009; Robertson, 2012).

Comparative analyses of core clinical features and correlates of TS have documented the frequencies of facial and limb tics, complex motor tics, echolalia, coprolalia, corpropraxia, ADHD and OCD (Bruun & Budman, 1997; Eapen & Robertson, 2008). Others have identified various TS "types" by examining relationships between clusters of simple or complex tics and clinical correlates such as tic severity, age of onset and externalising or internalising behaviours (Alsobrook & Pauls, 2002; Grados & Mathews, 2009; Mathews et al., 2007). These findings stimulate a currently unresolved debate regarding the conceptualisation of TS as either a spectrum or multiple phenotype disorder (Robertson, 2008, 2012). Many researchers hypothesise that relationships between TS and comorbid disorders

indicate a degree of shared aetiology, with TS phenotypes resulting from the interplay of various genetic, neurobiological, social and the environment factors (Eapen & Črnčec, 2009; Robertson, 2012).

With no biological makers, TS is diagnosed solely on the basis of clinical history and the observation of symptoms. Differential diagnosis of TS however represents a significant clinical challenge given its heterogeneous presentation and high rates of comorbidity (Eapen & Črnčec, 2009). Poor understanding and acceptance of TS by clinicians, educators and the public in general has however resulted in serious diagnostic delays, limited service provision for those affected and the social isolation and stigmatisation of those with the disorder (Collins, 2005; Dedmon, 1990; Stern, Burza, & Robertson, 2005). The primary mode of treatment is the prescription of psychoactive medication to reduce tic severity (Robertson, 2012). Although often effective, the use of antipsychotic medications is limited by their attendant iatrogenic risks, adverse side effects and inconsistent levels of efficacy, particularly when prescribed for paediatric and adolescent populations (Robertson, 2012; Woods, Conelea, & Himle, 2010).

Where available, non-medical interventions such as supportive psychotherapy, psychoeducation and behavioural interventions play a secondary, adjunctive role in the treatment regimes (Hendren, 2002; Woods, Piacentini, & Walkup, 2007). Despite promising results from a recently developed comprehensive behavioural intervention for tics (CBIT) to reduce symptoms (Woods et al., 2010; Woods et al., 2008), relatively few guidelines for psychological interventions have been developed to address the complex needs of those with TS, thereby leaving many individuals inadequately supported (Collins, 2005).

The current research aimed to address a significant gap in knowledge regarding the understudied Australian TS population. As part of a larger project, the current aim was to reveal the clinical phenomenology of TS in a broad community-based paediatric sample, the experience of those diagnosed within the Australian health and education systems, and the incidence of stigmatisation. It was hoped that the results of this study could be employed to enhance the awareness, understanding and recognition of TS by professionals, and prompt the increased participation of psychologists in research and service provision.

## **Method**

### **Participants**

Two groups of participants were included in this nation-wide survey. The Tourette Syndrome Association of Australia (TSAA) and the Tourette Syndrome Association of Victoria (TSAV) provided support in the recruitment of the TS group. The control group was recruited from multiple Australia-wide sites via local and Internet advertising, James Cook University faculty staff and the TSAA and TSAV. The total sample included 194 children aged between 7-16 years. The TS group consisted of 86 individuals (Males = 72, Females = 12; Mean age = 11.4, *SD* = 2.8) formally diagnosed with Tourette syndrome. The control group included 108 children and individuals (Males = 79, Females = 29; Mean age = 11.3, *SD* = 2.6) with no history of psychiatric or serious medical disorder. The majority of the children and adolescents included in the sample were male (78%), Caucasian (93%) with no significant between group differences in sex and age determined. No significant differences were found between TS and control group parents in demographic characteristics, with the great majority (90%) being the biological mothers of youth in the study, married (82%) and middle income or above (92%).

Written informed consent was required prior to participation, which was voluntary and confidentiality was guaranteed. The study was approved by James Cook University Human Research Ethics Committee, and conducted in compliance with NHMRC's Australian Code for Responsible Conduct of Research.

Randomisation and the calculation of response rates could not be conducted 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.

### **Measures**

Parents were administered the "Australian Tourette Survey", a postal survey designed for the purposes of a larger controlled study of Australian youth with TS that explored differences in quality of life, rates of psychopathology, behavioural and social difficulties and factors impacting peer attachment and close relationships of those diagnosed. Additional questions addressed demographics, formal diagnosis of comorbid disorders, family history of TS and comorbid disorders, experiences within the health and education systems, current treatment and stigmatisation. Two psychometric measures relevant to the current study were included.

**The strengths and difficulties questionnaire (SDQ).** The Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997) is a widely utilised brief screening questionnaire for emotional and behavioural disorders in children aged 4 to 16 years. It consists of 25 items across five subscales assessing emotional symptoms, conduct problems, hyperactivity / inattention, peer problems and prosocial behaviour, with a total summary score calculated by summing the 20 items assessing emotional, conduct, inattention and peer problems. The SDQ has robust psychometric properties

(Mellor, 2005) and demonstrated good to excellent internal consistency for the subscales relevant to the current study (Cronbach's Alpha  $\alpha = .80 - .90$ ).

**The parent tic questionnaire (PTQ).** The Parent Tic Questionnaire (PTQ; Chang et al., 2009) is a parent-report measure designed to assess the presence, frequency, and intensity of their child's motor and vocal tics during the previous week. This period was extended to one month in order to facilitate consistency with the observation period for other measures in the study and to minimise the risk of respondent error. Parents were asked to identify whether fourteen common motor and fourteen common vocal tics occurred within the previous month (yes/no). In addition, parents indicated the intensity and frequency for each tic endorsed by rating these 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 psychometric properties in prior studies (Chang et al., 2009) using clinical samples. Measures of internal consistency across all items as well as the motor and vocal subscales were adequate in a recent large community sample (Conelea et al., 2011) and were good to excellent in the current study (Cronbach's Alpha  $\alpha = .80 - .90$ ).

## **Results**

### **Data Coding**

Data were entered into a single SPSS spreadsheet, the completeness of the data minimised missing data, and demographic data were coded and entered for analysis. Homogeneity of variance (Levene's test) was found to be adequate for all variables. Outliers were examined, and while a couple of meaningful outliers were identified, it was decided to retain them for analysis, given their clinical legitimacy

and importance. Tests for normality and homogeneity of variance were satisfactory with the expected abnormal distribution of the PTQ satisfactorily resolved by applying a Log 10 transformation. To protect against inflated family-wise error rate, a per comparison critical significance level of  $\alpha = .01$  was applied.

### **Clinical Characteristics of the TS Youth**

Significantly more TS Group parents (N=36, 41%) reported a possible family history of TS than control group parents. TS group siblings did not demonstrate a higher rate of TS, and were reported to have significantly higher rates of only one comorbid disorder – Learning Disorder- than control group children/adolescents ( $\chi^2$  (1, N=176) = 8.31,  $p = .004$ ).

High rates of comorbidity were reported for children/adolescents with TS (n= 63, 73.3%). In order of decreasing frequency formally diagnosed disorders were reported as OCD/B (n= 38, 44.2%); Anxiety (n=31; 36.0%); ADHD/ADD (n= 28, 32.6%); Learning Disorder (n=16,18.6%) Impulse Control Disorder (n=6, 7.0%); Autistic Spectrum Disorder (n=4, 4.7%); Depression (n=2, 2.3%); Conduct Disorder (n=2, 2.3%) and “Other” unspecified disorders (n=10, 9.3%). High rates of multiple comorbidity (67%) were also reported (Table 3) with a minority (33%) of those with TS and comorbidity (total n =63) reporting only one occurring disorder.

### **Strengths and Difficulties Questionnaire (SDQ)**

To evaluate any significant differences in subscales and total scores for the SDQ between the TS Group and Control Group youth, a series of single-factor between-subjects MANOVAs and follow-up ANOVAs were conducted. Significant differences were noted between the TS Group youth and Control Group youth on

every measured outcome, indicating higher levels of dysfunction for the TS group. Results are displayed in Table 4.

### **Tic Phenomenology**

The type and frequency of motor and vocal tics reported for the group are presented in Table 5. Results for the group indicated that motor tics were reported more frequently (60%) than vocal tics (40%). Means and standard deviations for the three Tic Severity scales for the TS group ( $n=86$ ) were Motor Tic Severity ( $M = 61.03$ ,  $SD = 52.56$ ); Vocal Tic Severity ( $M = 35.27$ ,  $SD = 46.55$ ) and Total Tic Severity ( $M = 97.91$ ,  $SD = 86.65$ ). The number of motor and vocal tics displayed by individuals ranged between 0-14, with a modal number of Motor Tics = 7 and modal number of Vocal Tics = 2.

### **Experiences in the Australian Health and Education Systems and Social Stigma**

Table 6 presents the frequencies and percentages calculated for TS group responses to questionnaire items related to experiences within the health and education sectors and perceived stigma.

## **Discussion**

### **Clinical Profiles**

Present findings reveal the complex and highly variable clinical profiles of a broad-based community sample of young Australians with TS, with core clinical features generally agreeing with those identified in recent comparative analyses (Bruun & Budman, 1997; Eapen & Robertson, 2008).

The genetic underpinnings of TS were reflected in the 3-1 male gender-bias and positive family history of TS determined for the TS group. The present high rate of comorbidity (77%) concurs with the rate reported for non-clinic based TS samples (Kurlan et al., 2002). It is in agreement with multiple prior TS studies (Cavanna et al., 2009; Freeman et al., 2000; Kurlan et al., 2002; Robertson, 2006a, 2006b, 2008; Termine et al., 2006) on the reported rates of formal diagnoses. The results are also supported by findings from the SDQ, which indicated the high prevalence of OCD, non-OCD anxiety disorders, ADHD and learning disorders in the TS group. However other assessed disorders previously noted to commonly co-occur with TS (Freeman et al., 2000), including conduct disorder (CD), oppositional defiance (ODD), impulsivity, depression and Autistic Spectrum Disorder (ASD), were reported in low numbers that did not differ significantly from the control group. This may have been attributable to under-diagnoses in the current sample or a lack of statistical power in the study to detect rare disorders.

Results from the SDQ however indicated that TS group members experienced significant elevations in symptoms of mood and conduct disorder, which may have reflected a high level of subclinical symptomatology or under-diagnosis of these pathologies. A divergent finding from the study was the higher than usual prevalence of non-OCD anxiety disorder, which exceeded the rate reported for ADHD.

Challenges associated with disentangling symptoms of TS from those of comorbid disorders such as ADHD (Eapen & Črnčec, 2009) may result in under or misdiagnosis for some individuals. Alternatively, anxiety (particularly social or separation anxiety) may be exacerbated by factors uniquely experienced within the Australian social context, a hypotheses that is supported by current findings of the high (50%) rate of stigmatisation experienced by those with TS.



Further emphasising the clinical complexity of the group was the high reported rate of multiple comorbidity (67%). As can be seen in Table 3, over two thirds attracted more than one comorbid diagnosis, with combinations most frequently involving OCD, ADHD and Anxiety. Those with the most serious pathology had four or more diagnoses, with ASD the common thread in this small group.

The present findings also revealed the variability of tic profiles exhibited by this youth sample, mirroring the diversity of tic phenomenology presented in the TS literature (Robertson, 2012). Motor tics were more varied and reported in greater number than vocal tics for the group (ratio of 7:2), with the most commonly reported motor tics including eye blinking, facial grimacing and head jerking, whilst sniffing, grunting, coughing and echolalia were the most frequent vocal tics (Table 5). Coprolalia, the tic most strongly associated with poor psychosocial outcomes (Eddy & Cavanna, 2013; Kurlan et al., 1996), was experienced by 20%. Whilst complex motor tics were relatively common (50%), a very small minority expressed complex vocal tics (14%).

### **Access, Utilisation and Service Satisfaction in Australia**

The concerns of primary caregivers of TS youth (presented in Table 6), which echoed those expressed by their international counterparts (Collins, 2005; Conelea et al., 2011; Dedmon, 1990; Stern et al., 2005), revealed pronounced deficits in TS specific services and supports within the Australian health and educational sectors, low levels of understanding of TS and high rates of stigma for those diagnosed.

Over half of the TS group parents encountered difficulty accessing health and mental health services, with over 80% expressing dissatisfaction with the level of knowledge in regard to TS demonstrated by health professionals. Almost 80% of youth with TS were receiving treatment from a health professional with a minority

(33%) currently being medicated for their TS. Less than a third had consulted a psychologist, counsellor or was receiving special educational support. Medical doctors (Paediatricians, general practitioners and neurologists) currently appear to carry the major responsibility for the care of youth with TS. These findings suggest that the majority of youth with TS are not receiving the level of multidimensional support required to optimally manage this complex disorder.

Over two thirds of the TS group parents also expressed dissatisfaction with the level of support received within the education system. Deficiencies within this sector have serious adverse consequences for those with TS and their families with prior research highlighting the critical role of the school in determining academic and psychosocial outcomes for diagnosed youth and impacting the level of stress experienced by their primary caregivers (Packer, 1997, 2005; Woods, Koch, & Miltenberger, 2003).

### **Implications**

The high rate of comorbidity and diverse tic profiles demonstrated by the majority of youth in this study highlight the challenges associated with differential diagnosis and intervention in the context of such complex clinical phenomenology. The findings also emphasise the importance of conducting initial and ongoing assessments for psychopathology, behavioural disorder and severity of tic symptomatology in all youth with TS. This requires that clinicians acquire a high level of understanding and awareness of TS and comorbid disorders and develop the ability to design and implement interventions that address all of the difficulties exhibited. Adopting a multidisciplinary approach would help to provide the range of services to address the needs of this clinical population, services which are clearly not yet available or accessible to the majority of those included in the current study.

**Limitations**

Several limitations of the current study are noted, including the reliance on parental reports to assess clinical phenomenon necessitated by the study design (survey), and the demographic homogeneity of the sample. Recruitment from the TSAA and TSAV may also have biased the sample towards those with more severe forms of TS.

**Future Studies**

To inform the multiple-phenotype versus spectrum disorder debate, a future study could apply data reduction techniques to current results in an attempt to replicate the TS “types” emerging from prior studies. The current findings also indicate the urgent need for improvement in services and supports for the Australian TS community. Future studies might examine attitudes and knowledge regarding TS amongst Australian psychologists, medical staff, allied professionals and educators and identify areas for intervention to address gaps in knowledge and resources.

**Conclusions**

The current findings may be useful to employ in the context of TS specific psychoeducation, professional education, research and advocacy. Results also confirm the importance of encouraging a much greater level of involvement by psychologists, allied health and educational professional as providers of research and services for this clinical population with substantial, currently unmet needs.

## **Key Points**

### **What is Already Known About this Topic?**

1. TS is a poorly understood, under-diagnosed and stigmatising disorder with a surprisingly high rate of prevalence.
2. TS has highly adverse psychological and behavioural consequences yet psychologists are under-represented in TS research and the clinical setting.
3. Two notable gaps in the TS literature include a lack of Australian TS research and an over-reliance upon clinic based (more severe forms of TS) populations.

### **What this Topic Adds**

1. Knowledge documenting the complex clinical phenomenology and adverse psychological and behavioural difficulties accompanying Tourette's syndrome (TS) in a community-based Australian youth sample.
2. Knowledge essential to improving recognition, differential diagnosis and treatment of Tourette's.
3. Evidence indicating the urgent need for the increased education of and involvement by psychologists in both TS research and the provision of multidimensional services to meet the needs of this clinical population.

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**Table 3.** Frequency and Type of Parent-Reported Comorbid Diagnoses for TS Group (n = 63)

One comorbid disorder	n	Two comorbid disorders	n	Three comorbid disorders	n	Four or more comorbid disorders	n
OCD	8	OCD ANX	10	ADHD OCD CD	1	ADHD OCD ANX LD	3
ADHD	6	CD LD	2	ADHD OCD ANX	4	ADHD OCD ANX Other	1
LD	5	ADHD OCD	2	OCD ANX ICD	2	ADHD OCD ANX CD	1
ANX	2	ADHD ANX	2	ADHD OCD ASD	1	ADHD OCD ICD LD	1
		OCD LD	2	OCD ANX LD	1	ADHD OCD ANX ASD LD	1
		OCD ASD				Other	
		ANX DEP	1	ADD ODD LD	1		
		ASD Other	1	ANX ASD LD	1		
		ADHD DEP	1	ADHD OCD LD	1		
		OCD ASD	1				
		ADHD LD	1				
<b>%</b>		<b>%</b>		<b>%</b>		<b>%</b>	

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33.3%	21	36.5%	23	19.1%	12	11.1%	7
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*Note.* ADHD = Attention Deficit Hyperactively Disorder; ICD= Impulse Control; OCD= Obsessive Compulsive Disorder/Behaviour; ANX= Anxiety Disorder; LD = Learning Disorder; CD= Conduct Disorder; ASD= Autistic Spectrum Disorder

**Table 4.** Group Differences in Emotional and Behavioural Functioning

	Controls (n=108)		TS Group (n=86)		Effect size
	Mean	SD	Mean	SD	
<b>Strengths and Difficulties Questionnaire</b>					
Emotional symptoms	0.9	1.3	4.3**	2.5	0.44
Conduct problems	0.7	1.1	2.7**	2.2	0.27
Hyperactivity / inattention	2.3	2.0	6.7**	2.6	0.49
Total score	4.7	3.9	16.8**	7.0	0.55

\*\*p < 0.01

**Table 5.** Frequencies, Ranks and Percentages of Motor and Vocal Tics Reported for TS Group

Motor Tics	Frequency (%)	Rank (Group %)	Vocal Tics	Frequency (%)	Rank (Group %)
Total	502 (100%)		Total	329 (100%)	
Eye Blinking	64 (12.7%)	1 (74.4%)	Sniffing	49 (14.9%)	1 (57%)
Facial grimace	57 (11.3%)	2 (66.2%)	Grunting	42 (12.7%)	2 (48.8%)
Head jerk	56 (11.1%)	3 (62.8%)	Coughing	26 (7.9%)	3 (30.2%)
Arm/hand movements	51 (10.2%)	4 (59.3%)	Echolalia	26 (7.9%)	3 (30.2%)
Other motor tics	48 (9.6%)	5 (55.8%)	Phrases	25 (7.6%)	5 (29.1%)
Complex (combined) motor tics	45 (9%)	6 (52.3%)	Other noises	23 (7%)	6 (26.7%)
Shoulder shrugs	42 (8.4%)	7 (48.8%)	Other types of vocalisation	22 (6.7%)	7 (25.6%)
Mouth/tongue movements	38 (7.6%)	8 (44.2%)	Snorting	20 (6.1%)	8 (23.3%)
Eye rolling / darting	38 (7.6%)	8 (44.2%)	Words	19 (5.8%)	8 (22.1%)

Leg/feet movements	38 (7.6%)	8 (44.2%)	Corporalia	19 (5.8%)	10 (22.1%)
Echopraxia	20 (4%)	11(23.2%)	Blocking /Stuttering	18 (5.5%)	11 (20.1%)
Corpropraxia gestures	18 (3.6%)	12(13.9%)	Complex vocal tics	15 (4.6%)	12 (17.4%)
Chest Stomach movements	16 (3.6%)	13(18.6%)	Animal noises	13 (3.9%)	13 (15.1%)
Pelvic tensing	9 (1.8%)	14(10.5%)	Syllables	12 (3.6%)	14 (13.9%)

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**Table 6.** Reported Levels of Access, Satisfaction and Utilisation of Services and Rates of Stigmatisation for TS Group

Survey Items	Response = Yes	Response = No
	Satisfied	Dissatisfied
	Frequency (%)	Frequency (%)
Difficulty accessing Health/Mental Health services?	41 (47.7)	45 (52.3)
Satisfaction with level of support from Education system?	28 (32.9)	57 (67.1)
Satisfaction with level of knowledge of TS by Health Professionals?	16 (18.6)	70 (81.4)
Child/teen receiving treatment from a Health /allied health Professional?	67 (77.9)	19 (22.1)
TS Child receiving Medication for TS?	30 (35.3)	55 (64.7)
Perception of Stigmatisation of the Child/teen due to TS?	37 (44.6)	46 (55.4)
Current service provider/s for TS child/ adolescent	Frequency (%)	
Paediatrician	34 (39.5)	
Psychologist	28 (32.6)	
Special education Assistant	24 (27.9)	
Neurologist	22 (25.6)	

General Medical Practitioner	22 (25.6)
Psychiatrist	16 (18.6)
Counsellor/School counsellor	14 (16.3)
Family Therapist	1 (1.2)

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**SECTION 3 CHAPTER 2. The impact of tic severity, comorbidity and peer attachment on quality of life outcomes and functioning in Tourette's Syndrome: Parental perspectives** *Child Psychiatry & Human Development* (October 2015)

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**This chapter is a report on the study designed to test the main hypotheses of Study 1. It examines the impact of three variables - peer attachment, tic severity and comorbidity - on quality of life and functional outcomes for youth with TS. Results are compared with those a control group sample of typically developing peers.**



**Abstract**

The aim of this controlled, community-based study based on data from parents of youth (aged 7-16 years) with Tourette's syndrome (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.

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 (APS, 2103; Robertson & Eapen, 2014). 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 (Cavanna, Servo, Monaco & Robertson, 2009; Robertson, Cavanna & Eapen, 2015). 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 (Robertson, 2012). Other commonly identified psychopathologies include anxiety, mood and conduct disorders, impulsivity, aggression, learning (LD) and autistic spectrum disorders (ASD) (Freeman et al., 2000; Robertson et al., 2105; Termine et al., 2006).

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 (Conelea et al., 2011; Eddy et al., 2011a; Leckman, Bloch, Scahill & King, 2006; Robertson, 2012). Findings from paediatric TS QoL studies universally demonstrate impaired global QoL and reduced psychosocial functioning, with the strongest adverse impact in academic (Storch et al., 2007a), social (Eddy et al., 2011a; Eddy et al., 2011b), and emotional domains (Culter, Murphy Gilmore & Heyman, 2009). 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 (Eddy et al., 2001b; Robertson, 2012)

and comorbidity (Cavanna et al., 20009; Conelea et al., 2011; Eddy et al., 2011b; Jalenques et al., 2012; Muller-Vahl et al., 2010; Storch et al., 2007a; Storch et al., 2007b), particularly when seen in combination (Conelea et al., 2011; Eddy et al., 2011b; Robertson, 2012). However, a substantial proportion of variance in QoL and functional outcomes remained unexplained (Conelea et al., 2011).

Differential diagnosis in the context of TS is a challenging and protracted process, even for highly experienced clinicians (Boudjouk, Woods, Miltenberger & Long, 2000; Eapen & Črnčec, 2009). 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 (Achenbach & Rescalora, 2000), along with various instruments to assess tic severity (e.g. the Parent Tic Questionnaire (Chang et al., 2009).

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 (Boudjouk et al., 2000; Conelea et al., 2011). Peer relationships and family functioning have been identified as key factors in determining the wellbeing and functioning of children and teens with TS (Copper, Robertson & Livingston, 2003; Packer, 1997, 2005; Wilkinson Marshall & Curtwright, 2008; Woods, 2005). Relationship factors have also been shown to moderate the impact of tic severity and comorbidity on outcomes. For example,

Storch and colleagues (Storch et al., 2007c) found that peer victimisation mediated the relationship between tic severity and loneliness. Carter and colleagues (Carter et al., 2000) also found that positive family functioning was also 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 (Silva, Munoz, Barickman & Friedhoff, 1995). Problems encountered in peer relationships include experiencing difficulty making and maintaining friendships (Stokes, Bawden, Backman, Dooley & Camfield, 1991; Woods et al., 2010; Woods Koch & Miltenberger 2003) being subjected to negative attitudes and behaviours from peers (Boudjouk et al., 2000; Conelea et al., 2011; Storch et al., 2007c) and having reduced social skills and fewer opportunities to socialise (Champion, Fulton & Shady, 1989; Cohen & Leckman, 1994; Dykens et al., 1990). 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 (Kurlan et al., 1996). 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 interaction with their primary caregiver, which remains relatively stable across time and relationships (Bowlby, 1982; Ainsworth, 1982, 1989, Hazan & Shaver, 1987; Sroufe, 2005). Ainsworth (1982) observed differences between the behaviours of securely and insecurely attached children. Children displaying secure attachment seek out their caregiver for comfort,

and while they show distress at separation, they become soothed upon their return (Ainsworth, 1982). Those displaying insecure attachment are either avoidant or ambivalent, tending to be either unresponsive to their caregiver and show little distress when left alone, or to become clingy and distressed when separated from their caregiver (Ainsworth, 1982).

The importance of secure attachment to the healthy psychological, behavioural, social and physical development of the child is supported by an extensive body of research (Sroufe, 2005). Secure attachment to the caregiver facilitates positive representations and expectations of relationships and guides future social behaviour (Bowlby, 1982; Ainsworth, 1982, 1989, Hazan & Shaver, 1987; Sroufe, 2005). It 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 (Seibert & Kerns, 2009; Sroufe, 2005). There is increasing evidence of the important role played by secure attachment to peers and the psychosocial functioning of the child, particularly in adolescence (Liable, Carlo & Roesch, 2004; Muris & Maas, 2004; Rubin et al., 2004). 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  $\chi^2$  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 7.

### *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) (Varni & Limbers, 2009) 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 to 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 (Chan, Mangione-Smith, Burwinkle, Rosen & Varni, 2005; Varni & Limbers, 2009; Varni, Seid & Kurtin, 2001) 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. Parents have the most opportunity to observe a child’s tics, particularly in the home environment when many release tics after periods of attempted suppression (Chang et al., 2009). 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 one 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 (Chang et al., 2009) in clinical samples, adequate internal consistency in a community sample, indicating adequate results (Conelea et al., 2011) and good internal consistency for all summary and subscales in the current study (Cronbach's  $\alpha = .82 - .88$ ).

*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) (Goodman, 1997). This is a widely utilised questionnaire (suitable for children aged 4 to 16 years), that 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 (Mellor, 2005), which was good to excellent for the total difficulties score and all subscales employed in the current study (Cronbach's  $\alpha = .75 - .87$ ), with the exception of the prosocial scale, which was poor ( $\alpha = .41$ ).

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) (Goodman & Scott, 1999), which has been used to

identify symptoms of psychopathology in prior published TS studies, including recent quality of life research by Storch and colleagues (Storch et al., 2000b).

*Attachment Questionnaire for Children (AQC)*: Security of peer attachment was assessed via the Attachment Questionnaire for Children (AQC) (Muris, Meesters, van Melick & Zwambag, 2001). 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 (1997) 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 (Bartholomew & Shaver, 1998; Muris & Maas, 2004; Muris et al., 2001). While establishing the reliability of any single item measure can be challenging, inter-rater reliability for the AQC has been shown to be acceptable (Muris & Meesters, 2002).

## **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 versus 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 indicted 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 indicted significant between group differences on the combined SDQ subscales,  $\Lambda = .42$ ,  $F(5, 188) = 52.42$ ,  $p < .001$ ,  $\eta_p^2 = .58$  with ANOVAs revealing significantly higher dysfunction for the TS group on all sub-scales and the total score (see Table 8).

*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 9.

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 two of the univariate outcomes were not significant at  $\alpha = .01$ : the physical functioning sub-scale of the PedsQL and the conduct problems sub-scale 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 10 provides the correlations between tic severity (motor, vocal, and total) and the sub-scales and total scores 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 vocal tics were significant, with nine of 12 significant at  $p < .001$ , whereas five of the 12 correlations involving motor tics were not significant.

*Comorbidity:* Table 11 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 11. 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.

*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 (Baron & Kenny, 1986) 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 (Bernard et al., 2009; Carter et al., 2000; Eddy et al., 2011b; Storch et al., 2007b) with effects extending to all functional domains. Mirroring recent findings (Conelea et al., 2011; Storch et al., 2007b), 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 (Conelea et al., 2011; Dyken et al., 1990; Packer, 1997, 2005; Robertson, 2012; Woods et al., 2003), 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 (Eapen & Črnčec, 2009; Robertson 2012), 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 (Robertson, 2008, 2011). 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 7) suggests that differences in peer attachment were not related to structural variables often implicated in the development of insecure attachment (Main & Solomon, 1990). 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 (Stokes et al., 1991; Woods et al., 2011; Woods et al., 2003) and the multiple documented barriers to friendship youth with TS encounter (Boudjouk et al., 2000; Champion et al., 1989; Cohen & Leckman, 1994; Dykens et al., 1990; Woods et al., 2003; Woods et al., 2011).

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 9). 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 (de Lange & Olivier, 2004).

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 activities, 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 (Liable et al., 2004; Muris & Maas, 2004; Rubin et al., 2004; Siebert & Kerns, 2009;). 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 (Bowlby, 1982; Ainsworth 1982, 1985; Sroufe, 2005; Seibert & Kerns, 2009). 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 (Conelea et al., 2011; Storch et al., 2007c), 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 (Kurlan et al., 1996; Woods, 2005; Woods et al., 2011).

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 (Cavanna et al., 2009; Conelea et al., 2011; Eddy et al., 2011b; Jalenques et al., 2012; Muller-Vahl et al., 2010; Storch et al., 2007a; Storch et al., 2007b).

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 (Conelea et al., 2011; Eddy et al., 2011b; Robertson, 2012), 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 (Conelea et al., 2011; Storch et al., 2007b) 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 (Freeman et al., 2000; Robertson et al., 2015). 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.

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**Table 7.** Demographic Characteristics and Between-Group Differences

		Control (n = 108)			TS (n = 86)		
		<i>f</i>	%	<i>M (SD)</i>	<i>f</i>	%	<i>M (SD)</i>
Age (years)				11.30 (2.58)			11.44 (2.78)
Relationship to youth	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 residence	City	67	62		50	50	
	Regional	33	30.6		20	23.8	
	Rural-remote	8	7.4		14	16.7	
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	Male	79	73.1		71	85.4	
	Female	29	26.9		12	2.4	
Ethnicity **	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	

Sibling	Yes	101	93.5	76	88.4
	No	7	6.5	10	11.6

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

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

**Table 8.** Group Differences in Quality of Life Outcomes and Emotional and Behavioural Functioning

Outcome Measure	Groups				ANOVA		
	TS ( <i>n</i> =86)		Control ( <i>n</i> =108)		<i>F</i>	<i>p</i>	$h_p^2$
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>			
<b>Pediatric Quality of Life Inventory</b>							
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
<b>Strengths and Difficulties Questionnaire</b>							
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

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*Note.*  $df(1, 192)$  for all analyses.

**Table 9.** Impact of Peer Attachment on Quality of Life and Strengths and Difficulties Outcomes for TS Group (n = 86)

Outcome Measure	Attachment Style				ANOVA		
	Secure ( <i>n</i> =49)		Insecure ( <i>n</i> =37)		<i>F</i>	<i>p</i>	$h_p^2$
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>			
<b>Pediatric Quality of Life Inventory</b>							
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
<b>Strengths and Difficulties Questionnaire</b>							
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

*Note.* *df* (1, 84) for all analyses.

**Table 10.** Correlations between Tic Severity, Quality of Life and Strength's and Difficulties Outcomes for TS Group (n = 86).

	Motor Tics		Vocal Tics		Total Tics	
	<i>r</i>	<i>p</i>	<i>r</i>	<i>p</i>	<i>r</i>	<i>p</i>
<b>Pediatric Quality of Life Inventory</b>						
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
<b>Strengths and Difficulties Questionnaire</b>						
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

**Table 11.** Impact of Comorbidity on Quality of Life and Strengths and Difficulties Outcomes TS Group (n = 86).

Outcome Measure	Comorbid Disorder				ANOVA		
	Present ( <i>n</i> =63)		Absent ( <i>n</i> =23)		<i>F</i>	<i>p</i>	$\eta_p^2$
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>			
<b>Pediatric Quality of Life Inventory</b>							
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
<b>Strengths and Difficulties Questionnaire</b>							
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

Note. *df* (1, 84) for all analyses.

**SECTION 3 CHAPTER 3. Factors Impacting the Quality of Peer Relationships of Youth with Tourette's Syndrome *BMC Psychology*, (September, 2015)**

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**This chapter is a report on the study of the impact of TS on the security of the peer attachment relationships and social functioning of youth with TS. It is based on the qualitative and quantitative findings from Study 1 and Study 2.**

**Abstract**

**Background:** Tourette's syndrome (TS) is a poorly understood neurodevelopmental disorder consistently associated with impaired peer relationships. This research aimed to investigate the relationship between TS and the ability of diagnosed youth to form secure attachment relationships with peers. A quantitative study examined differences between youth with TS and typically developing peers in social functioning, relationship problems and attachment security. Qualitative studies sought to identify factors that enhanced or impeded the ability to form secure peer relationships, including the impact of tic severity, comorbidity and personality traits. All research was conducted from the parental perspective.

**Methods:** The research consisted of a controlled, survey-based qualitative and quantitative study (Study One) of parents of youth with TS ( $n = 86$ ) and control group peers ( $n = 108$ ), and a qualitative telephone interview-based study of TS group parents (Study Two,  $n = 22$ ). Quantitative assessment of social functioning, peer problems and peer attachment security was conducted using the Paediatric Quality of Life inventory, the Strengths and Difficulties Questionnaire and the Attachment Questionnaire for Children. Qualitative data relating to personality was classified using the Five Factor Model.

**Results:** Results revealed significantly higher rates of insecure peer attachment, problems in peer relationships, difficulty making friends, stigmatisation and lower levels of social functioning for the TS group. Significant between-group differences in number and type of factors impacting peer relationships were also determined with 'personality' emerging as the most prevalent factor. Whilst Extraversion and

Agreeableness facilitated friendships for both groups, higher rates of Neuroticism were barriers to friendship for individuals with TS. The TS group also identified multiple ‘non-personality’ factors impacting peer relationships, including TS and comorbid symptom severity, the child’s psychological and behavioural adjustment to their disorder, coping strategies and the behaviour and attitudes of peers.

**Conclusions:** The findings from the study may help clinicians, parents and individuals with TS to better understand and cope with the difficulties experienced in interactions with peers. While it is important to remember that these findings are based on parental report and not the perceptions of youth themselves, this study may also help clinicians to identify youth at increased risk of developing insecure peer relationships and guide the development of targeted supports.

## **Background**

Tourette’s syndrome (TS) is a complex neurodevelopmental disorder characterised by the presence of multiple motor and vocal tics occurring for a period of at least one year (American Psychiatric Association 2013). The severity of TS varies widely between individuals and is complicated by high rates of comorbid diagnoses (90%), the most common of which are Attention Deficit Hyperactivity Disorder (ADHD) and Obsessive Compulsive Disorder (OCD) (Freeman et al., 2000). Exhibiting a male gender bias of 3:1, the onset of symptoms typically occurs in childhood and peaks during the developmentally sensitive period of early adolescence (Robertson, 2012).

Recent quality of life studies (QoL) on paediatric TS populations have revealed strong relationships between the disorder and decreased QoL, with the highest level of impairment evident in psychosocial functioning (Bernard et al., 2009;

Cavanna et al., 2013; Conelea et al., 2011; Cutler et al., 2009; Eddy et al., 2011a; Eddy et al., 2011b; Storch et al., 2007b; Storch et al., 2007c). Further evidence suggests that TS has a particularly adverse impact on peer relationships. Prior research has found that youth with TS experience an increased incidence of bullying, teasing, peer victimisation and social rejection (Storch et al., 2007c), have difficulty making and maintaining friends, have lower quality and numbers of close friends (Boudjouk et al., 2000; Shady et al., 1989) and are more likely to be negatively evaluated by peers (Bawden et al., 2003; Stokes et al., 1991).

There is considerable individual variability in the level of difficulty youth with TS experience in their peer relationships and social functioning. However the factors contributing to these differences are not well understood. In previous studies, increased tic severity and the presence of comorbidity accounted for a significant proportion, but not all the variance in psychosocial outcomes (Bernard et al., 2009; Cavanna et al., 2013 ; Conelea et al., 2011; Cutler et al., 2009; Eddy et al., 2011a; Eddy et al., 2011b; Storch et al., 2007b; Storch et al., 2007c). Other factors with adverse effects on peer relationships include characteristics of TS such as impulsivity, aggressiveness, episodic rage and coprophenomena (Budman et al., 2003; Robertson & Eapen, 2014) and the lower levels of social competence that some youth with TS exhibit (Dykens et al., 1990). Stigmatisation and social rejection also create limited opportunities for friendship and the development of social skills (Collins, 2005; Conelea et al., 2011), and diagnosed youth may limit their interaction with peers in response to fears associated with their own socially inappropriate symptoms or the negative behaviour of others (Kurlan et al., 1996).

The emergence of peer relationships as a key determinant of a wide range of outcomes for youth in recent TS studies highlights the importance of improving



current understanding regarding the way in which TS impacts friendships. Having supportive and accepting friends has been associated with increased wellbeing, improved socio-emotional functioning and improved school performance (Carter et al., 2000; Cooper et al., 2003; Storch et al., 2007c; Wilkinson et al., 2008; Wilkinson et al., 2001). Conversely, negative peer behaviours and social isolation have been linked to higher rates of mood disorder, loneliness, poor self-esteem, self-consciousness and increased tic severity (Storch et al., 2007c; Termine et al., 2006). The major goal of the current research was to develop a greater understanding of the peer relationships of youth with TS, and how TS itself may shape these relationships. As Attachment Theory has become the dominant model within which close relationships are examined (Hazan & Shaver 1994), it was adopted for the purposes of the current exploratory study.

Ainsworth (1989) and Bowlby (1982) proposed that a classifiable style of attachment (secure or insecure) is developed during an infant's interactions with their primary caregiver. This attachment style remains relatively stable across time and guides both expectations and behaviour in future relationships (Ainsworth, 1989; Bowlby, 1982; Sroufe, 2005; Weinfield et al., 2004). Secure attachment is contingent upon the primary caregiver's positive representation of the child, their availability to provide a reliable source of safety and comfort in times of distress and a secure base for their child (Ainsworth, 1989; Bowlby, 1982). As children develop, they gradually transfer these attachment functions from parents to peers, in a process that culminates in the development of romantic relationships and close friendships in adulthood (Kerns et al., 2000; Rubin et al., 2004; Seibert & Kerns, 2009; Sroufe, 2005).

The literature reveals the critical role that the security of the child's attachment relationships play in determining optimal development, childhood and

future wellbeing (Ainsworth, 1989; Bowlby, 1982; Sroufe, 2005), with secure attachment emerging as the strongest predictor of the child's emotional and social competence. The relationship between attachment style and TS has not been explored in any prior published studies. Given the aforementioned psychosocial and peer relationship difficulties experienced by youth with TS, the current study hypothesised that youth with TS would be at increased risk of forming insecure relationships with peers.

In order to achieve the goals of the current research, two complementary studies that adopted a mixed method approach were conducted. Study One included a nation-wide survey-based study of parents of youth with TS ( $n = 86$ ) and a group of parents of children without a diagnosis of TS ( $n = 108$ ). The quantitative component of Study One examined differences in social functioning, problems in peer relationships and rates of insecure peer attachment between youth with TS, and age and gender matched peers. Given the novel and exploratory nature of this study, as well as difficulty identifying a multidimensional psychometric measure of peer attachment suitable for use across this age range, two qualitative studies were also conducted. Within Study One, qualitative data was gathered to identify factors that were perceived to enhance or impair the ability of youth to form secure peer relationships and to investigate differences in findings between the TS and control group. To augment these findings, a further interview-based qualitative study (Study Two) was conducted employing a subset of the TS group participants ( $n = 22$ ) from Study One. Study Two aimed to collect qualitative data to identify the types of friendships experienced by youth with TS, as well as develop an understanding of motivation and other factors that shaped the security of peer relationships within this sample. Due to the ethical and practical implications of surveying children as young

as seven within the design of the current study, information in both studies was provided by the primary caregiver, the majority of whom were the youths' biological mothers.

As reported below, a key finding to emerge from the qualitative analyses related to the youths' personality traits. Data relating to personality were classified according to the "Big-Five" Factor model (FFM) (Costa & McCrae, 1992; Goldberg, 1993), which consist of Extraversion, Agreeableness, Neuroticism, Conscientiousness and Openness (to experience). Previous research has found direct associations between Extraversion, Agreeableness and Openness and the ability to form and maintain friendships and wider social networks, and to develop social competence (Scholte et al. 1997; Selfhout et al. 2010), whilst Neuroticism has been found to have the opposite effect (Selfhout et al. 2010). Personality traits may also indirectly affect peer relationships of youth with TS, with correlational studies demonstrating links between specific "Big Five" traits and a range of psychological and developmental disorders that adversely affect socio-emotional functioning. For example, autism has been correlated with low levels of Extraversion, Conscientiousness and Openness (Fortenberry et al., 2011), while anxiety and depression have been consistently associated with Neuroticism (Kotov et al., 2010). The processes by which such links are established are, however, poorly understood. Furthermore, no research documenting the relationship between personality traits and TS has been published.

In addition, a broader literature exists regarding the impact of personality traits on resilience, coping abilities and strategies adopted by the individual in response to stressors, including those associated with chronic and developmental disorders, all of which may moderate or mediate the impact of TS on peer relationships. Extraversion, Agreeableness, Contentiousness and Openness have been associated with increased

psychological resilience, in addition to improved problem focused coping and cognitive restructuring, whilst neuroticism has consistently predicted low resilience and maladaptive emotion-focused coping in youth (Connor Smith & Flachsbart, 2007). In addition, personality traits influence the response of others, with agreeableness increasing an individual's ability to enlist support and acceptance from others (Shiner & Masten, 2012).

In summary, the current study sought to investigate the impact of TS on the peer relationships and the factors that enhanced or impeded their ability to experience secure peer relationships. Although primarily an exploratory study, several hypotheses were proposed. It was predicted that youth with TS would experience higher rates of insecure peer attachment, increased peer problems, and decreased social functioning in comparison with undiagnosed peers. It was further hypothesized that the qualitative analyses would reveal differences in factors identified by parents of youth with TS and controls as impacting their child's ability to form secure relationships with peers, and that TS would be associated with unique barriers to secure peer relationships.

It is hoped that the findings from the current research have the potential to help clinicians, parents and young people with TS to understand the psychosocial difficulties of those diagnosed, particularly in the context of their peer relationships. It is also hoped that results have the potential to be employed to inform clinical intervention and encourage further research into this important aspect of TS youths' psychosocial functioning

## **Methods**

The research was conducted with the approval of the James Cook University Human Research Ethics Committee (Approval number H4380), in compliance with the Helsinki Agreement and within the guidelines for research ethics outlined in the National Statement on Ethics Conduct in Research Involving Humans (2007).

Informed consent was obtained from all parents/guardians of participating minors in the study. Participation was confidential, all records were de-identified and stored in compliance with JCU guidelines, and no incentives to participate were offered.

### **Participants**

Two groups of volunteers participated in Study One ( $N = 194$ ). The TS group ( $n = 86$ ) consisted of a community based, national sample of parents with a child aged between 7 and 16 years formally diagnosed with TS. The control group ( $n = 108$ ) comprised parents of age and gender matched peers with no reported medical or psychiatric diagnosis. A subset of TS group parents volunteered to participate in Study Two ( $n = 22$ ).

### **Procedure**

TS group participants for Study One and Study Two were recruited with the assistance of the Tourette Syndrome Association Australia (TSAA) and the Tourette Syndrome Association Victoria (TSAV) following advertising and invitations to participate. Controls were recruited with the help of the TSAA and TSAV, research assistants in several nation-wide locations, and JCU University staff and students. Surveys were posted to the entire membership base of both societies, and mailed or distributed to controls. Accurate response rates could not be calculated due to the lack of information maintained on databases and the inability to track distribution of control group surveys. Figure 1 presents a flow chart of the present research.

## Measures

### Study one.

The Australian Tourette's Survey was designed by the primary author for the purpose of a larger study of Australian youth with TS. In addition to demographic questions, the survey included questions that asked parents to identify their child's formally diagnosed comorbid disorder and the experience of stigma. It also employed three widely used and well-validated psychometric measures (parent proxy versions) relevant to the current project to assess social functioning, peer relationship problems and security of peer attachment of youth in the study.

***Social functioning.*** Social functioning was assessed via the social functioning subscale of the Pediatric Quality of Life Inventory (PedsQL) (Varni & Limbers, 2009; Varni et al., 2001). This five-item subscale is a measure of quality of life related to social functioning. For example, parents are asked 'How much of a problem has your child had (in the past month) getting along with other kids?' Extensive reliability and validity data exist for the PedsQL (Bastiaansen et al., 2004; Chan et al., 2005; Varni et al., 2001). Internal consistency for the social functioning subscale in the current study was excellent (Cronbach's  $\alpha = .90$ ).

***Peer relationships.*** Difficulties experienced in peer relationships was assessed via the peer problems subscale of the Strengths and Difficulties Questionnaire (SDQ) (Goodman, 1997; Goodman, 1999). This five-item measure forms part of a widely employed brief behavioural screening questionnaire assessing psychopathology, behavioural, and social problems. For example, parents are asked if the statement that their child 'has at least one good friend' (in the past 6 months) is not true, true or certainly true. The measure has robust psychometric properties (Goodman, 1999;

Goodman & Scott, 1999; Mellor, 2005). Internal consistency in the current study for the peer problems subscale was excellent (Cronbach's  $\alpha = .87$ ).

**Peer attachment.** The study adopted a broad operational definition of attachment that encompassed affectional (close or intimate) and non-affectional (friendships) components (Belsky & Cassidy, 1994; Dykas & Cassidy, 2011). This decision was guided by findings that have demonstrated the differing functions attachment figures perform at different developmental stages (Robertson et al., 2015). The availability of attachment figures to provide these functions at age appropriate times rather than the 'closeness' of the affectional bond is thought to be most relevant in the context of childhood and adolescent peer attachments (Robertson et al., 2015; Trinke & Bartholomew, 1997).

The single-item Attachment Questionnaire for Children (AQC) (Muris et al., 2001) was employed to assess peer attachment. The use of this age downward version of the widely employed single-item measure of self-reported adult attachment (Hazan & Shaver, 1987) was necessary due to the absence of multi-item measures of peer attachment suitable for use in middle childhood. Parents were asked to choose which of three narrative descriptions based on the three attachment styles constituting Ainsworth's (1982) typology (secure, insecure-ambivalent and insecure-avoidant) best described their child's peer attachment. The reliability of the AQC and Hazan and Shaver's measure has been demonstrated in multiple studies (Bartholomew & Shaver, 1998; Muris & Maas, 2004; Muris et al., 2001). As the current research sought to determine the impact of secure versus insecure attachment, the two insecure attachment styles were combined to create a dichotomous variable evaluating secure or insecure attachment.

**Qualitative studies.**

**Study one.** To gather data for the qualitative study, two questions were asked at the end of the demographics section of the survey. Parents were asked, “Does your child have any trouble making friends?” Participants were then asked to comment with a very brief written response to the question “What do you feel makes it easy or difficult for your child / teen to make friends?”

**Study two.** A series of open-ended questions were composed to form the basis of the semi-structured interviews that comprised Study Two. Questions were reviewed and refined by a senior academic supervisor before implementation. Example items include “How would you describe your child/ teen’s friendships?” “How do you feel TS affects your child’s ability to make friends?” Questions were kept to a minimum to facilitate openness of responses from participants and to provide space for the participants’ voices and unique experiences (Bloomberg & Volpe, 2008).

All interviews were conducted via telephone by the principal researcher once consent had been obtained. Interviews varied in duration from one hour to 90 minutes. Participation was limited to one interview per parent, one parent per family, and one child or adolescent with TS. Each recorded interview was then transcribed in full for the purposes of data analysis.

### **Coding of Qualitative Data**

#### **Study one.**

The principal researcher conducted a content analysis of the survey, employing a method described by Biddle and colleagues (2001). A deductive approach to coding was adopted, with the text read multiple times by the researcher to identify major themes, which were code-named as they emerged.



Data were further reduced by classifying each item related to the youth's personality according to the "Big Five" (FFM) traits of extraversion, agreeableness, openness, conscientiousness and neuroticism (Costa & McCrae, 1992; Goldberg, 1993). The validity of the findings regarding personality classification was examined by conducting third party trustworthiness checks (by a rater blind to the purposes of the study) on all of the coded personality data. This resulted in 100% agreement.

### **Study two.**

The researcher employed both an inductive approach to detect emerging themes, as theoretically described by Strauss and Corbin (1998), and a deductive approach to content analysis (guided by open-ended research questions grounded in the relevant literature) as described by Miles and Huberman (1994). The major analytic category of the current report was parental perception of the child's peer attachments. Manual coding of verbatim transcripts was conducted with multiple line-by-line reviews to create a hierarchy from lower to higher order themes (Biddle et al., 2001). Manual coding with the aim of data reduction continued in an iterative process over multiple months until completion. Data dictionaries, which provided definitions and examples of all of the emerging codes, were composed for each study.

### **Inter-rater reliability.**

Two trained raters blind to the purposes of the study conducted inter-rater reliability checks. Using three randomly selected cases from each study, both raters checked all of the coded text data that emerged from Study One and all of the coded interview data from Study Two by referencing the respective data dictionaries.

Percentage of agreement and Krippendorff's alpha coefficient- KALPHA (Krippendorff, 2004) were calculated using ReCAL (Freelon, 2013). 100% agreement was determined for the overall sample, and all of the examined codes in Study One

and Study Two achieved 100 percent agreement with KALPHA  $\alpha = 1$ . The high degree of agreement between raters may have been attributable to the clarity and simplicity of the variables in the study, which required minimal interpretation.

### **Statistical Procedure**

Data were analysed using SPSS Version 19 (IBM Corp 2010). All continuous outcome and predictor variables were assessed for their suitability for parametric analyses by testing for normality and homogeneity of variance (Levene's test). This was shown to be adequate for all variables. Outliers were examined and a per comparison critical significance level of  $\alpha = .01$  was applied for all comparisons. For all inferential test results, an  $R^2$ -type effect size measure is reported (i.e.,  $\eta_p^2$  for ANOVA analyses).

In Study One, differences between the TS and control groups on the PedsQL and the SDQ subscales were evaluated with univariate ANOVAs. Chi-square analysis was used to test for significant relationships between group membership and responses to the AQC, demographic variables and the ability to form friendships.

The qualitative findings from both studies were cross-tabulated to generate frequency data. Z-tests of proportions were conducted to reveal between group differences in the qualitative findings from Study One, with a critical significance level of  $\alpha = .01$

## **Results**

### **Participant demographics**

TS group parents ( $n = 86$ ) and Controls ( $n = 108$ ) participated in the quantitative components of Study One ( $n = 196$ ). All TS group and 92 control group parents provided text for the qualitative analysis (TS,  $n = 86$ ; Controls,  $n = 92$ ; total,  $n = 178$ ). The majority of participants in Study One were the biological mothers of the

child or adolescent TS group = 91 %, Controls = 89%), resided in metropolitan areas (TS group = 59%, Controls = 62%), were married (TS Group = 84%, Controls = 82%) and reported an average or above level of income (TS Group = 67%, Controls = 70%). Mean age of the children for the total sample was 11 ( $SD = 3$  years), (TS Group,  $M = 11.44$ ,  $SD = 2.78$ ; Controls,  $M = 11.31$ ,  $SD = 2.58$ ). The majority of the youth included in the study were male (TS Group = 85%, Controls = 73%). Only one significant demographic difference was reported, with a slightly higher level of racial diversity reported within controls ( $p < .01$ ).

Study Two participants included the child's biological mother ( $n = 22$ , 100%), were mostly married ( $n = 18$ , 82%) and resided in metropolitan areas ( $n = 13$ , 59%). The mean age of the children in Study Two was 12 ( $SD = 3$  years). 90% of the youth in Study Two were male ( $n = 20$ ).

## **Quantitative Findings**

### **Study one.**

The results of the main quantitative analyses for Study One (presented in Table 12 & Table 13) support the hypotheses that youth with TS experience higher rates of parent reported peer problems, impaired social functioning, insecure peer attachment and difficulty forming friendships than control group peers. Almost half ( $n = 37$ , 45%) of the TS group believed their child was stigmatised by their TS and a high rate of comorbidity was reported for the TS group ( $n = 66$ , 77%). The most commonly reported co-morbidities were OCD (44%), other anxiety disorders (36%), ADHD (33%) and Learning Disorder (LD) (19%).

## **Qualitative Findings**

### **Characteristics of the friendships of youth with TS.**

Coding and analysis of data in Study Two provided maternal descriptions of the friendships of the current sample of youth with TS ( $n = 22$ ), with findings suggesting fewer than 20% enjoyed a 'typical' social life. Mothers defined 'typical' as having at least one close friend, several peripheral friends and the ability to socialise with classmates and acquaintances. Mothers attributed reduced motivation for peer interaction to factors associated with TS symptoms including social anxiety ( $n = 3$ , 14%), fear of bullying, teasing and rejection ( $n = 4$ , 18%), difficulty maintaining friendships ( $n = 5$ , 23%), inability to spend long periods of time with friends due to efforts to suppress tics ( $n = 6$ , 27%) and having a low level of interest in classmates ( $n = 2$ , 32%). Almost a third of the youth in the sample had overtly expressed the desire for more friends to their mother ( $n = 7$ , 32%). Motivation for friendship and romantic relationships was reported to increase for three of the four older adolescents.

### **Factors impacting peer attachment – parental perspectives.**

Findings from both studies revealed multiple factors impacting the peer relationships of youth. Parents in Study One identified twenty-two factors; The first being 'Personality' (FFM Traits) followed by twenty-one discrete 'Non-Personality' factors. Study Two revealed eighteen factors including 'Personality' that enhanced peer attachment and seventeen that negatively impacted peer attachment. There was a high degree of homogeneity across the findings from both studies in the factors identified by parents of youth with TS.

### **Between group differences in factors impacting peer relationships.**

As hypothesised, Study One revealed variability in the type, frequency and number of factors identified by TS and control parents affecting their child's peer

relationships. 'Personality' (FFM Traits) emerged as the most frequently identified factor for both TS and control group parents (see Table 14). The analyses of data from Study One also revealed the increased complexity of the factors identified by TS group parents in comparison with controls. No significant difference was found between the proportion of parents in the two groups who made reference to personality factors (TS = 74%, control group = 88%,  $z = 2.13$ ,  $p < .05$ ). However, a significantly larger proportion of parents in the control group (37%) attributed the ability to form friendships exclusively to their child's personality compared with the TS group (11.6%) ( $z = 4.14$ ,  $p < .001$ ). TS group parents (88%) identified a significantly increased number of 'non-personality' factors compared with controls (63%) ( $z = 4.14$ ,  $p < .01$ ).

As presented in Table 14, the major findings for 'Personality' were that Extraversion, low Neuroticism and Agreeableness had positive impacts for both groups, but were identified by a significantly higher percentage of control group parents. Extraversion, Neuroticism and low Agreeableness were associated with significant negative impacts for the TS group (see Table 14). The major findings for non-personality factors (Table 15) included the positive role of high Social Skills and Activities for controls in comparison with TS group youth, and the ability to cope with tics, a Positive School Environment and the Positive Behaviour of Others benefiting the friendships of TS group youth. The main negative 'Non-Personality' factors for TS versus control youth included Maladaptive Symptoms, the Negative Impact of Tics, low Social Skills, and the Negative Behaviour of Others (see Table 15)

## **Discussion**

To the best of our knowledge, this is the first study to explore the ability of TS diagnosed youth to form secure relationships with peers. The quantitative results from Study One, a large community-based survey of parents of youth with TS and age and gender matched peers, confirmed the hypothesis that youth with TS are at increased risk of forming insecure peer attachment relationships. Between group differences in the measure of peer attachment security reveal a threefold increase in insecure peer attachment for youth with TS, with the rate of insecurity for those diagnosed exceeding that expected in a normative population sample (Ainsworth, 1989).

Additional quantitative results from Study One illustrate the adverse consequences of TS for the peer relationships of diagnosed youth. As hypothesized, peer relationships are likely to be negatively influenced by the highly significant elevation in impaired social functioning parents reported for youth with TS in comparison with controls. This finding has been reported in several recent studies of TS (Conelea et al., 2011; Eddy et al., 2011a; Eddy et al., 2011b; Storch et al., 2007a). Consistent with previous research (Boudjouk et al., 2000; Shady et al., 1989; Storch et al., 2007c), parental reports confirmed that youth with TS experience a greatly increased number of problems (such as bullying and social rejection) within their peer relationships, and increased difficulty forming friendships in comparison with undiagnosed peers. Almost half of the parents in the TS group believed their child to be stigmatised by their diagnosis. This aligns with the rates reported in recent studies (Conelea et al. 2011), and indicates barriers to positive peer relationships occurring at the societal level.

It is however important to note two significant limitations associated with the design of the current research when interpreting these findings. First, the study was limited to parental responses due to the ethical and practical constraints associated

with surveying children as young as seven in remote mode (i.e., written survey and telephone interview). Second, in order to maximise participation from the difficult to access TS population, the study included a wide age range of youth, and therefore was unable to adequately control for the developmental stage of the youth under study.

### **Characteristics of the friendships of youth with TS.**

Whilst determining quantitative differences between the type or number of friendships youth with TS and peers experienced was beyond the scope of the current research, evidence from Study Two suggests the restricted nature of friendship this sample of diagnosed youth appears to experience. Further, with the exception of an increased motivation for friendship and romantic relationships amongst older adolescents in the study, the findings did not reflect any change in the nature of friendship that might be expected at different developmental stages. Friendships appeared to be largely limited to a circle rarely extending beyond one to three individuals, with the majority experiencing impaired or restricted interaction with classmates and wider peer acquaintances. While some attachment theorists suggest that peer attachment is most relevant in the context of the child's close 'best' friends (Kernset al., 1996), others stress the importance of the attachment functions played by less intimate peer relationships and more extensive social networks (Robertson et al., 2015; Trinke & Bartholomew, 1997). The results of the current study indicate that these wider social networks may not be available to the majority of youth in this study.

Whilst these qualitative findings are limited by the subjectivity of maternal beliefs regarding 'typical' friendships, participant's definitions were highly consistent. Future studies should build upon these findings by employing objective

measures to assess friendship, as well as be extended to include the youth's self-reported interpersonal experiences.

### **Factors impacting the peer attachment relationships of youth with TS.**

The combined qualitative findings from Study One and Study Two revealed multiple factors that parents perceived as impeding or enhancing their child's ability to form secure peer relationships. These fell into two broad categories, those related to the FFM personality traits (Costa & McCrae, 1992) (Table 14) and those representing a broad range of 'non-personality' factors (Table 15). As hypothesised, findings indicated substantial variability in the factors identified by parents of youth with TS and control group parents, as well as the barriers to friendship specifically associated with diagnostic status.

*Non-personality factors.* Parents identified many 'non-personality' factors that they believed impacted the quality of their children's peer relationships. These included the adverse effects of increased tic severity and the presence of comorbid disorders on the peer relationships of youth, although standardised assessment of tic severity and the quantitative impact of these variables were not goals of the current study. This is consistent with previous research (Bernard et al., 2009; Cavanna et al., 2013 ; Conelea et al., 2011; Cutler et al., 2009; Eddy et al., 2011a; Eddy et al., 2011c; Storch et al., 2007a; Storch et al., 2007c).

Current findings also revealed a highly complex role for tics and increased tic severity in impairment in peer relationships. Whilst this research found that simply having tics and increased tic severity are detrimental to peer relationships, distress and dysfunction in peer relationships was more closely related to the youth's negative cognitive appraisal and their affective and behavioural responses to their tics. Specifically, the degree of self-consciousness experienced in regard to tics, rather than



tic severity, appears to be most damaging to the youth's peer relationships. This is an important finding as it may help to explain some of the individual variability in results of the impact of tic severity on social functioning evident in prior studies.

In addition, parents in Study Two linked increased self-consciousness and an inability to adjust to or accept a diagnosis with highly adverse behavioural and psychological consequences that further alienated individuals with TS from their peers. These included responses such as denial, rage, depression, social anxiety and social withdrawal. Supporting the acuity of these parental observations is the finding that a diagnosis of chronic disorder places youth at a significantly increased risk of adjustment disorder (Wallander & Varni, 1998).

Parents also identified three more factors with negative impacts directly attributable to the 'non-tic' symptoms of their child's TS and comorbid diagnoses. These included maladaptive symptoms of TS such as aggressiveness, impulsivity, a tendency to dominate peers, to behave bizarrely, incongruently, or to withdraw from or fail to participate socially. These factors have all been shown to have notable adverse effects on interpersonal relationships in previous TS studies (Budman et al., 2003; Robertson & Eapen, 2014).

Parents also indicated that some of the behaviours associated with comorbid diagnoses, including inattention, impulsivity, anxiety, obsessiveness, defiance and antisocial behaviour had a significant impact on their child's relationships. High rates of comorbid diagnoses were evident in both studies. Disorders such as OCD and ADHD have previously accounted for a disproportionate amount of social adversity for individuals with TS (Bernard et al., 2009). It is however important to note that disentangling behaviours attributable to TS from comorbid disorders, the child's underlying personality traits, and behaviours that would generally be construed as

misbehaviour, was reported as being very challenging for parents in the current study (Study Two). This is also a challenge in both clinical and research settings.

As identified in prior studies (Conelea et al., 2011), negative peer behaviours such as bullying, teasing, social rejection and stigmatisation were reported as being a major impediment to secure peer relationships for many TS youth. The concerns of youth regarding peer responses towards them in this study also extended to a fear of being seen as “different”, “uncool”, “weird”, receiving unwanted peer attention and anxiety that they may frighten or irritate peers with their symptoms. For youth who experience socially embarrassing tics such as coprolalia, these self-perceptions are understandable. However, there is also some suggestion that some individuals with TS may “self-stigmatise” by internalising negative TS stereotypes and adverse social experiences, including being stigmatised by others (Mukolo et al., 2010).

Other less frequently identified factors to negatively affect peer relationships included poor social skills and competence, which parents often attributed to comorbid disorders and low social interest, that is a manifestation of the youth’s “eccentricity”, “shyness” or comorbidity. The ability to participate in sport was also important and appears to be of great cultural significance in the Australian context, particularly for males (Daly, 2011). Non-participation for youth with TS appears to be associated with low interest, physical limitations associated with diagnosis, psychological barriers (e.g., social anxiety), the stress of competition and the youth’s cognitive rigidity. Parents also identified barriers to participation, such as social exclusion by peers, other parents, schools and social organisations.

Both studies also identified multiple ‘non-personality’ factors that enhanced the ability to form secure peer relationships. The most common factor was the youth’s ability to cope with their tics. This referred to a suite of factors including the youth’s

successful psychological adjustment to the diagnosis of TS, as well as a lower level of self-consciousness in regard to their tic symptomology. Parents reported that the ability to camouflage, suppress or otherwise manage tics at important times (such as during class or whilst playing with other children), and being in a waning phase of the tic cycle both positively impacted the development of secure relationships with peers.

Other important positive factors included the youth's ability to be open with others about their TS and comorbid disorders, to self-advocate and educate peers regarding their TS and their ability to defend themselves against the adverse behaviour of others (such as bullying teasing and social rejection). Some of these abilities form components of recent interventions designed to improve outcomes for children and adolescents with TS, particularly in the school setting (Packer, 2005; Woods & Marcks, 2005).

Finally, external factors including the acceptance, understanding and support of peers emerged as a key determinate of positive peer relationships, with parents in Study Two identifying this factor more frequently than any other. Some participants also acknowledged that peers needed occasional respite and support in order to preserve friendship with youth with TS. The importance of the attitudes of classmates to the relationships and wellbeing of youth with TS has been demonstrated during early trials of school-based interventions (Woods & Marcks, 2005).

**Personality factors.** Personality was the most frequently identified factor impacting peer relationships to emerge from the current research, with over 80% of Study One respondents attributing the quality of their child's peer relationships to at least one personality dimension. Although there were no between-group differences in the frequency with which parents nominated personality, the control group were more likely to refer exclusively to FFM personality traits.

Of the FFM traits, Extraversion and Agreeableness, and to a limited degree Openness (to experience), appear to help youth with TS to overcome the significant barriers to friendship that they experience, as well as counterbalance the stigmatising, alienating and disturbing impact of their symptoms. As noted in the literature, Extraversion and Agreeableness have direct positive effects on peer relationships, with both linked to more positive peer representations. Extroverts are valued for their sociability, drive and energy, and this trait is the strongest predictor of friendships longitudinally (Scholte et al., 1997; Selfhout et al., 2010). Those high in Agreeableness are appreciated for their caring, loving and empathic qualities, and Agreeableness predicts the highest rate of peer acceptance and reciprocity of friendship (Scholte et al., 1997; Selfhout et al., 2010).

Both Extraversion and Agreeableness may also indirectly enhance peer relationships by increasing resilience, adaptive coping and attracting social support. An extrovert's assertiveness and social skills may help them to overcome the often reported negative behaviour of peers experienced by individuals with TS (Collins, 2005; Conelea et al., 2011; Stokes et al., 1991; Storch et al., 2007c), and enhance self advocacy skills. Agreeableness may help youth to recruit peer support and understanding (Scholte et al., 1997; Selfhout et al., 2010). However, extraversion was also linked to problems with peers by some TS group parents. Furthermore, it has been suggested that the increased drive of extroverts for friendship may expose individuals high in this trait to increased psychological risk associated with rejection (Ashton et al., 2002). Openness to experience may extend the social networks of youth, which were found to be limited in this study, and the opportunities these provide for relationships facilitated by shared values and interests (Jensen-Campbell et al., 2002; Scholte et al., 1997; Selfhout et al., 2010).

Consistent with the literature, the results of the current study indicated that Neuroticism was identified by parents as being commonly associated with the inability to form friendships in individuals with TS. It appears to share the closest theoretical link to insecure attachment, as both insecure attachment and Neuroticism are characterised by emotional dysregulation and negative affect (Ainsworth, 1989; Bowlby, 1982; Costa & McCrae, 1992; Goldberg, 1993). Previous research has also suggested that Neuroticism has the most adverse impact on the ability to maintain friendships (Selfhout et al., 2010), a problem that was identified for a significant minority of youth in Study Two. Emotional dysregulation, as well as a decreased ability to interpret social cues associated with Neuroticism, has also been shown to result in increased relationship conflict (Penke & Denissen, 2008). Finally, Neuroticism has been associated with decreased resilience and the impaired ability to cope with adversity in the context of chronic disorders (Shiner & Masten, 2012), both of which may adversely affect social functioning and peer relationships.

### **Limitations and Future Research**

Several limitations have been noted for this study that may be addressed in future research. The generalisability of the results of the qualitative study may be limited by recruitment from TS support groups, the presence of unmeasured respondent characteristics, the demographic homogeneity of participants, and the dominant participation by biological mothers of male children and teens. Further, social desirability may have biased participants' responses, while the researcher may have introduced bias at any or all levels of the qualitative analyses. Quantitative assessment of attachment was limited to a single-item measure due to apparent lack of

availability of an appropriate multi-item measure for middle childhood. However, to some extent this was compensated for by the inclusion of the two qualitative studies.

A high priority is to compare the current findings with youth self-reports, which have been found to vary slightly from parent-reported outcomes in prior research on TS (Conelea et al., 2011). Future studies could also examine variability in the impact of TS on peer relationships at different developmental stages. The reliability and validity of the current qualitative findings would also benefit from replication in quantitative studies employing standardised measures of variables such as tic severity and psychopathology.

## **Conclusions**

The current study explored and confirmed the positive relationship between TS and the increased risk of developing insecure peer attachment relationships. Findings also provided detailed insights into multiple factors parents identified as either impeding or enhancing the development of effective peer relationships in youth with TS. These included the impact of TS and comorbid diagnoses, emotional, cognitive and behavioural response to diagnosis, the attitudes and behaviour of peers, as well as a number of personality traits.

The findings from the study may help clinicians, parents and those with TS to better understand and cope with the difficulties experienced in their interactions with peers. They may also help clinicians to identify those more at risk for developing poor peer relationships and guide the development of targeted supports, although it must be remembered that these findings are based on parental reports and not the perceptions of youth themselves.

Finally, the emergence of personality as an important variable suggests the value of including personality assessment in future research examining individual differences in youth with TS.

**List of abbreviations**

ADHD, Attention deficit hyperactivity disorder

AQC, Attachment questionnaire for children

FFM, Five Factor Model

OCD, Obsessive compulsive disorder

PedsQL, Pediatric quality of life inventory

QoL, Quality of life

SDQ, Strengths and difficulties questionnaire

TS, Tourette's syndrome

TSAA, Tourette Syndrome Association Australia

TSAV, Tourette Syndrome Association Victoria

**Competing interests**

The authors declare that they have no competing interests

**Authors' contributions**

DO designed the study, conducted the survey, carried out the interviews and performed the statistical analysis, and drafted the manuscript. EH, VE, JR and KM participated in the design of the study and assisted with the statistical analysis. RG contributed to the data analysis and preparation of the manuscript. All authors read and approved the final manuscript.

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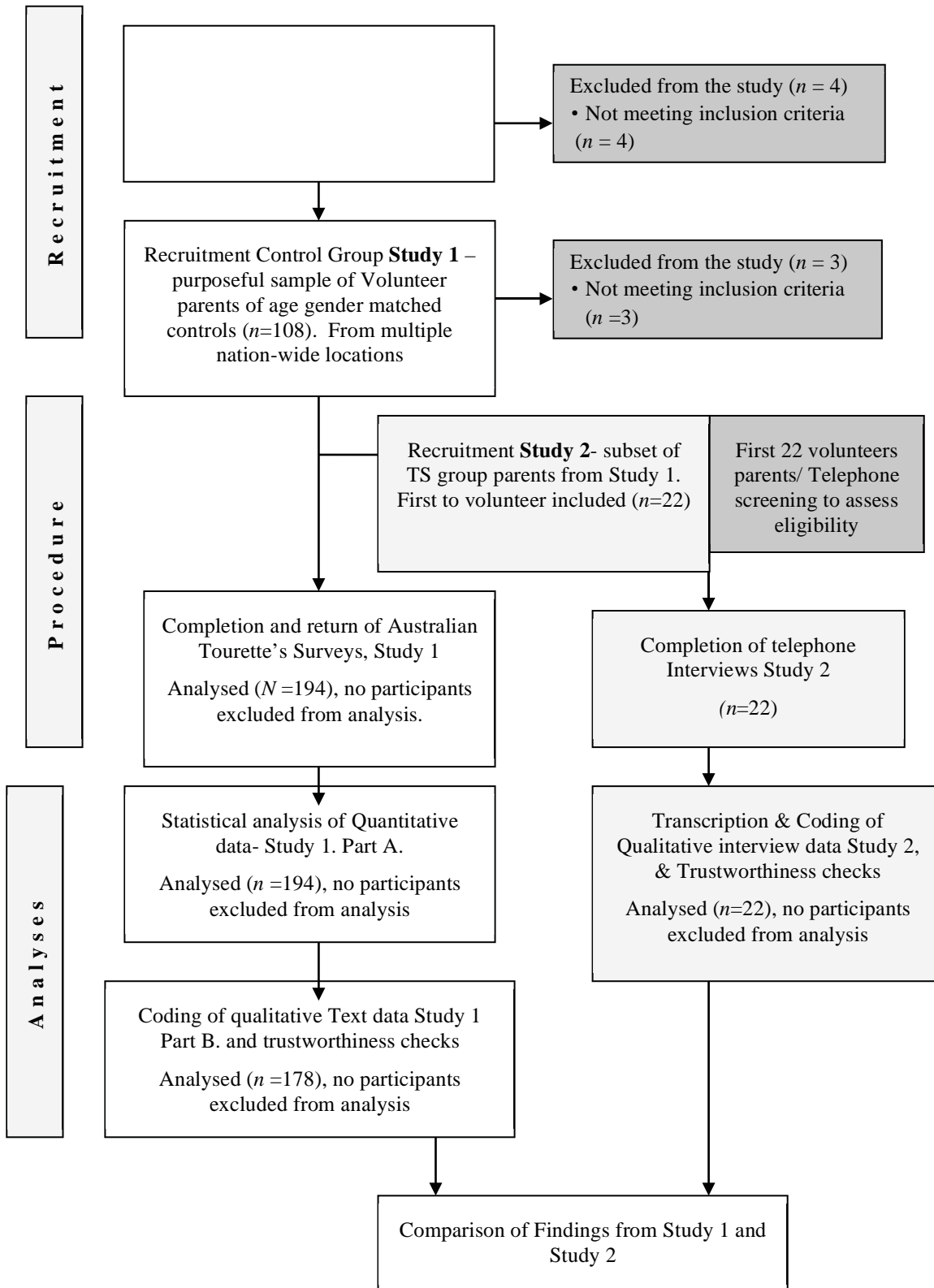


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**Figure 1.** Flow Chart of the Research: Study 1 (Part A & B) & Study 2.

**Table 12.** Descriptive Statistics and Analysis of Variance Results for TS and Control Group Parents on Social Functioning and Peer Problems

Outcome Measure	Groups				ANOVA			
	TS Group ( <i>n</i> =86)		Control Group ( <i>n</i> =108)		<i>F</i>	<i>df</i>	<i>p</i>	$\eta_p^2$
<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>					
Social Functioning (PedsQL)	60.76	24.76	87.73	14.92	84.74	1, 192	< .001	.31
Peer Problems (SDQ)	3.05	2.41	.87	1.37	62.91	1, 192	. < .001	.25

**Table 13.** Descriptive and Chi-Square Results for Differences between TS and Control Groups in Attachment Security and Ability to Form Friendships

Variable	Group		Total	$\chi^2$	df	<i>p</i>
	TS Group ( <i>n</i> = 86)	Control Group ( <i>n</i> = 108)				
Attachment (AQC)						
Secure	49 (57%)	102 (94%)	151 (78%)	36.5	1	<.001
Insecure	37 (43%)	6 (6%)	45 (23%)			
Difficulty Forming Friendships						
Yes	33 (38%)	4 (4%)	37 (19%)	37.3	1	<.001
No	53 (62%)	106 (96%)	159 (81%)			

**Table 14.** Frequency and Z-Test of Proportions Between TS Group youth and Control Group Youth in Personality Traits with Positive and Negative Impact on Friendship

Personality Trait With Positive Impact	Group		Test of Difference in Proportions (two-tailed)	
	TS Group <i>n</i> = 86 <i>n</i> (%)	Control Group <i>n</i> = 92 <i>n</i> (%)	<i>z</i>	<i>p</i>
High Extraversion	48 (56)	84 (91)	5.77	< .001
Low Neuroticism	9 (11)	45 (49)	6.18	< .001
High Agreeableness	24 (28)	42 (46)	2.48	.01
High Openness	15 (17)	15 (16)	0.19	.85
High Conscientiousness	0	2 (2)	1.40	.16

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Personality Trait				
With Negative Impact				
Extraversion	8 (19)	1 (1)	4.12	< .001
Introversion	10 (12)	12 (13)	0.28	.77
High Neuroticism	25 (29)	1 (1)	5.55	< .001
Agreeableness	0	1 (1)	1.01	.31
Low Agreeableness	12 (14)	2 (2)	2.89	.004
Low Openness	0	5 (7.7%)		
Conscientiousness	0	4 (4)	2.02	.04
Low Conscientiousness	0	3 (3)	1.76	.08

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**Table 15.** Frequency and Z-Tests of Proportions Between TS Group Youth and Control Group in Non-Personality Factors with Positive and Negative Impact on Friendship

“Non – Personality” Factors	Positive Impact				Negative Impact			
	Group		Test of Difference in Proportions (two-tailed)		Group		Test of Difference in Proportions (two-tailed)	
	Control Group ( <i>n</i> =92)	TS Group ( <i>n</i> =86)	<i>z</i>	<i>p</i>	Control Group ( <i>n</i> =92)	TS Group ( <i>n</i> =92)	<i>z</i>	<i>p</i>
Social skills - High	14 (17)	-	4.30	< .001	-	-		
Social skills - Low	-	-	-	-	1 (1)	20 (23)	4.69	< .001
Social interest - Low	-	-	-	-	2 (2)	3 (4)	0.52	.6
Sports participation	18 (20)	9 (11)	1.71	.08	1 (1)	-	1.01	.3
Humour	7 (8)	2 (2)	1.65	.10	-	-	-	-
Activities & interests	11 (12)	2 (2)	2.57	.01	-	-	-	-

Parents	14 (15)	5 (6)	2.07	.04	-	-	-	-
School - Positive	-	7 (8)	2.73	.01	-	-	-	-
School - Negative	-	-			2 (2)	5 (6)	1.21	.2
Maladaptive symptoms	-	-			-	46 (54)	9.89	< .001
Tics - OK	-	17 (20)	4.58	< .001	-	-	-	-
Tics - Negative	-	-	-		-	20 (23)	5.07	< .001
Negative behaviours from others	-	-	-		1 (1)	23 (27)	5.20	< .001
Positive behaviours from others	-	9 (11)	3.24	.01	-	-	-	-
Understanding & acceptance	-	4 (5)	2.02	.04	-	-	-	-
Lack of understanding & acceptance	-	-	-		-	7 (8)	2.74	.006
Opportunity to socialize	5 (5)	1 (1)	1.59	.1	-	-	-	-
Child as “different”	-	-	-		-	8 (9)	2.95	.003
Age-passage of time - Positive	1 (1)	7 (8)	2.23	.02	-	-	-	-



Age-passage of time - Negative	-	-	-	-	2 (2)	6 (7)	1.52	.12
Preference for one or a small group friends	4 (4)	4 (5)	0.10	.9	-	4 (5)	2.02	.04
Trouble maintaining friendships	-	-	-	-	-	5 (6)	2.29	.02
Having long-term friends	1 (1)	3 (4)	1.06	.29	-	-	-	-
Preferring older or younger friends; not peers	1 (1)	2 (2)	0.61	.54	-	-	-	-
Context	2 (2)	2 (2)	0.04	.96	2 (2)	-	-	-
“Alike” kids	1 (1)	2 (2)	0.61	.54	-	-	-	-
Sibling	4 (4)	1 (1)	1.27	.20	-	-	-	-

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*Note.* Raw scores indicate frequency of references made to the “Other” factor. Total positive or negative impact attributable to each factor by group membership expressed as raw score and percentage.

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**SECTION 3 CHAPTER 4. “The importance of screening for early indications of emotional symptoms in youth with Tourette’s Syndrome”**

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**This chapter is a report based on the findings from the current research of the high level of emotional symptomatology determined for youth with TS in Study 1. Given the clinical importance of this finding, and the relative lack of research examining the impact of depression in youth with TS, it was determined that this warranted a dedicated report.**

**Abstract**

**Background:** Tourette's syndrome (TS) is a neurodevelopmental disorder often accompanied by emotional disorder in adult clinical populations; however, the prevalence and aetiology of emotional difficulties in paediatric TS populations is under-researched. **Aims:** This study aimed to determine the prevalence of emotional symptomatology and dysfunction in a community-based sample of youth with TS, and to examine relationships between emotional difficulties, tic severity and comorbid diagnoses. **Methods:** This controlled, nation-wide study employed survey methodology and included robust psychometric instruments to measure emotional difficulties and tic severity. Voluntary, community-based participants comprised parents of youth with TS (n= 86) and matched controls (n=108). **Results:** Between-group parametric analyses revealed that youth with TS were at significantly ( $p < .01$ ) increased risk of emotional symptomatology and impaired emotional functioning, which within TS group analyses revealed were exacerbated by increased tic severity and comorbidity. **Conclusion:** Youth with TS are at increased risk of emotional difficulties, particularly if they experience more severe tics or comorbid disorder. Clinical implications include the importance of screening for early signs of emotional disorder in all youth with TS; facilitating early remedial intervention where indicated; and considering the complex factors that may contribute to emotional difficulties for individuals with TS.

**What this paper adds**

While recent studies have determined the prevalence and correlates of emotional disorders in adults with TS, this is under-studied in the paediatric TS population. The current research represents one of the largest controlled studies of

youth with TS, and benefitted by the inclusion of a greater number of youth with milder forms of TS than typically observed in the TS literature. As anxiety and depression are the strongest predictors of adverse outcomes for adults with TS, understanding the impact of emotional disorder and dysfunction in childhood and adolescence is of significant clinical importance. Present results therefore advance such understandings by revealing the high prevalence of clinical and subclinical level symptoms of emotional disorders, including depression and anxiety, and the impaired emotional functioning of youth with TS. The finding that tic severity and comorbidity increased the risk of emotional disorder, but did not account for all of the variance in outcomes, suggests the multifactorial aetiology of emotional difficulties in the context of TS. Clinicians and researchers therefore need to explore the role of additional variables, such as psychosocial factors, that may contribute to emotional difficulties. Results also indicate that symptoms of depression may be overlooked or under-recognised in youth with TS and indicates the importance of screening and ongoing assessment of emotional symptomatology. In conclusion, results indicate that improving emotional outcomes for all youth with TS is an important goal of treatment, ranking alongside the traditional targets of tic reduction and the management of comorbid disorder.

## 1. Introduction

Tourette syndrome (TS) is a childhood onset neurodevelopmental tic disorder complicated by high rates of comorbidity, reaching 90% in clinic-based samples (Freeman et al., 2000). Although obsessive compulsive disorder (OCD) and attention deficit hyperactivity disorder (ADHD) are the most frequently identified (Freeman et al., 2000; Robertson, 2006a), TS is also associated with an increase in comorbid emotional difficulties. Clinical and subclinical level symptoms of other anxiety disorders (excluding OCD) and depression are those most commonly reported in clinical and community-based TS populations (Cavanna et al., 2009; Embry & Rabian, 1998; Robertson, 2006b). Impaired emotional functioning has also been associated with TS in paediatric quality of life studies (Conelea et al., 2011; Eddy et al., 2011; Storch et al. 2007).

Elevated rates of separation anxiety, general anxiety disorder, social anxiety, and phobias have all been described in studies of clinical and community-based TS populations (Coffey et al., 2000; Freeman et al., 2000; Kurlan et al., 2002; Robertson 2000; Robertson, 2006a). Research suggests that the aetiology of comorbid disorders such as anxiety may be related to neurobiological processes shared with TS (Kurlan et al., 2002) including common cortico-striato-thalamocortical pathways, in addition to physiological and neurotransmitter abnormalities, particularly those involving dopamine (Singer & Minzer, 2003). However, increased tic severity has also been strongly associated with anxiety disorders such as generalised anxiety disorder (GAD) and social phobia (Coffey et al., 2000; Eapen & Črnčec, 2009). Some anxiety symptoms have also been accounted for as a reactive response to the psychosocial stress accompanying this stigmatising and unpredictable disorder (Lin et al., 2007).

Although the reported prevalence of depression in the TS literature is not always consistent, a recent comprehensive review of the literature suggests that it is very common, with up to 13% of individuals with TS attracting a diagnosis of clinical depression and as many as 76% experiencing depressive symptomatology (Robertson, 2006b). Higher rates of major depression and bipolar disorder have also been found in clinic-based paediatric samples (Coffey et al., 2000) as well as increased levels of self-reported depressive symptoms (Robertson et al., 2006).

However, some uncertainty regarding the prevalence and the aetiological relationship between TS and disturbed emotional functioning remains, particularly in the case of depression in community-based paediatric TS populations (Robertson, 2000, 2006b; Robertson & Orth, 2006; Robertson et al., 2006). Evidence suggests that the aetiology of depression in the context of TS is multifactorial and is more likely to occur in individuals with more severe forms of the syndrome (Cavanna et al., 2009; Robertson, 2006b, 2012). One exception is bipolar affective disorder, which appears to be associated with having comorbid OCD and ADHD (Robertson, 2012).

Several factors have been shown to contribute to an increase in depressive symptoms in TS. These include increased tic severity (Cardona et al., 2004; Elstner et al., 2001), comorbid diagnoses of OCD and ADHD (Carter et al., 2000; Cutler et al., 2009; Eapen et al., 2004; Hoekstra et al., 2004; Robertson & Orth, 2006; Robertson et al., 2006; Sukhodolsky et al., 2003) and the presence of coprophenomena (Robertson, 2012). Depression has also been associated with the side effects of psychoactive medications employed to treat TS (Robertson, 2000). The complex nature of the relationship between TS and depression has also been illustrated through the emergence of symptoms in response to an interplay between tic severity and comorbid ADHD and OCD (Robertson et al., 2006).

Although not amongst the most commonly reported comorbidities, researchers have also linked several impulse control disorders (ICD), such as self-injurious behaviours (SIB) and episodic rage, to increased depression, anxiety and emotional dysfunction (Frank et al., 2011; Mathews et al., 2004; Wright et al., 2012). The finding that ICDs tend to co-occur in individuals with ADHD, OCD, and more severe tics, compounds these negative affective outcomes (Wright et al., 2012). Finally, whilst not the focus of the current study, psychosocial variables such as poor quality peer relationships and impaired social functioning are also highly likely to increase anxiety and depression and have consistently been found to be compromised in individuals with TS (Conelea et al., 2011; Eddy et al., 2011; Storch et al., 2007).

The need to understand the risk of anxiety and depression, and the ability to screen for clinical and non-clinical level symptoms of these disorders in TS populations has been highlighted previously (Robertson et al., 2006). Depression in particular has emerged as a predictor of highly adverse outcomes in late adolescence and adulthood for individuals with TS. Adverse outcomes associated with depression include an elevated risk for inpatient psychiatric treatment, suicide, substance abuse and decreased quality of life and functioning (Coffey et al., 2000; Eddy et al., 2011; Jalenques et al., 2012 ; Muller-Vahl et al., 2010; Robertson 2006b). The ability to identify early signs of anxiety and depression across a wide variety of settings is therefore essential for our understanding of outcomes for individuals with TS.

### *1.1. The present study*

The current study aimed to explore the prevalence and correlates of emotional difficulties in a nation-wide community sample of children and adolescents with TS using survey methodology. This required the use of brief, psychometrically robust screening measures suitable for use in a non-clinical context, and precluded the ability

to conduct formal psychiatric evaluations. A control group was also included to facilitate comparison with peers. The design was limited to parental responses due to ethical considerations associated with surveying children as young as aged seven.

It was hypothesised that individuals with TS would experience higher rates of emotional symptomatology and impaired emotional functioning compared with their peers. It was also predicted that this would be exacerbated by increased tic severity and the presence of comorbid disorders including ADHD, OCD, and ICD. The final aim of the study was to test the ability of a brief psychometric measure to aid in identifying individuals most at risk of emotional problems, thereby facilitating referral to clinical services for formal evaluation, differential diagnosis, and early intervention.

## **2. Methods**

### *2.1. Participants and procedure*

The Tourette Syndrome Association of Australia (TSAA) and the Tourette Syndrome Association of Victoria (TSAV) provide non-financial support by advertising the study and assisting in the recruitment of participants. A control group (matched for age and gender of youth) was recruited from multiple Australia-wide sites via local and Internet advertising, James Cook University faculty staff, research assistants and the TSAA and TSAV.

The total sample included 194 participants aged between 7 to 16 years. The TS group consisted of 86 individuals (males = 72, females = 12) with a formal diagnosis of Tourette syndrome ( $M$  age = 11.4,  $SD$  = 2.8). The control group included 108 individuals (Males = 79, Females = 29) who reported no diagnosis of a psychiatric disorder ( $M$  age = 11.3,  $SD$  = 2.6). TS participants were recruited via the Tourette Syndrome Association of Australia (TSAA) and the Tourette Syndrome



Association of Victoria (TSAV) by mail and Internet invitation. Control participants were recruited from multiple Australia wide sites following local advertising and with the assistance of the TSAA, TSAV and James Cook University. The majority of the participants included in the sample were male (78%). There were no significant differences in demographics between the TS and control groups.

Following recruitment, all participants were given (post or in person) the survey pack. Written informed consent was required prior to voluntary participation, all material and data de-identified and confidentiality was guaranteed. The study was approved by James Cook University Human Research Ethics Committee, and conducted in compliance with NHMRC's Australian Code for Responsible Conduct of Research and the declaration of Helsinki. The calculation of response rates could not be conducted for the TS group due to restricted information maintained on member databases and the inability to accurately record survey distribution for controls over multiple Australia-wide locations precluded response rate calculation.

## *2.2. Measures*

The Australian Tourette Survey was designed for the purpose of a larger study of Australian youth with TS. Along with demographics, the questionnaire included items regarding history of formal comorbid diagnoses and current treatment providers. It included three well-validated measures relevant to the current project, as detailed below.

### *2.2.1. Pediatric Quality of Life Inventory*

Emotional functioning associated with anxiety and depression was assessed via the emotional functioning subscale of the 15-item Pediatric Quality of Life Inventory Short Form (PedsQL) (Varni & Limbers, 2009; Varni et al., 2001). Parents rated the items on a five-point Likert scale ranging from 'never a problem' to 'almost

always a problem' within the past month. The PedsQL shows good reliability and validity estimates (Bastiaansen et al., 2004; Chan et al., 2005; Varni et al., 2001). The emotional functioning subscale of the PedsQL consists of four items assessing symptoms of anxiety and depression. Internal consistency in the current study was good to excellent for this subscale ( $\alpha = .87$ ).

### *2.2.2. Strengths and Difficulties Questionnaire*

The SDQ is a widely employed measure consisting of 25 items evaluating five subscales including emotional symptoms, conduct problems, hyperactivity / inattention, peer problems and prosocial behaviour (Goodman, 1997; Goodman, 1999). Parents are asked if a statement is true, somewhat true or always true. The extended version includes an impact supplement that asks whether the respondent thinks the child or adolescent has a problem with emotions, concentration, behaviour, or getting along with others. If these problems are present, parents are then asked about chronicity, distress, social impairment, and burden to the family. The 25 items are summed to form total scale and subscale scores.

The SDQ has robust psychometric properties, with numerous studies demonstrating its strong structural and construct validity (Goodman, 1997; Goodman, 1999; Mellor, 2005; Palmieri & Smith 2007). It has also been shown to be a sensitive clinical outcome measure (Mathai et al., 2003), with the ability to screen for disorders in community samples (Goodman et al., 2000), and correlates strongly with more established measures of child psychopathology including the Child Behaviour Checklist (Achenbach & Rescorla, 2000; Goodman, 1997; Goodman, 1999). The five-item emotional symptoms subscale that assesses general symptoms of anxiety, depression and somatisation (a behavioural manifestation of childhood anxiety and

depression) was utilised in the current study and displayed good internal consistency ( $\alpha = .80$ ).

### *2.2.3. Parent Tic Questionnaire*

The Parent Tic Questionnaire (PTQ) (Chang et al., 2009) is a parent-report measure designed to assess the presence, frequency, and intensity of motor and vocal tics during the previous week. To ensure continuity and minimise response error, this observation period was extended to one month in order to mirror the PedsQL. Frequency and intensity of each tic is rated on four-point Likert scale, with greater scores indicating greater frequency and stronger intensity of tics. A weighted score for each tic, ranging from 0-32, was calculated by combining the frequency and intensity ratings. Motor and vocal tics were then summed separately to produce the motor tic severity subscale score, a vocal tic severity subscale score, and combined to form a total tic severity score. The PTQ has good psychometric properties across clinical (Chang et al. 2009) and community samples (Conelea et al., 2011). It demonstrates excellent convergent validity with the clinician administered Yale Global Tic Severity Scale (Chang et al. 2009). Internal consistency in the current study was good for all three scales: Total tic severity ( $\alpha = .88$ ); vocal tic severity ( $\alpha = .82$ ) and motor tic severity ( $\alpha = .84$ ).

### *2.3. Statistical Design and Analyses*

Frequency data and percentages were generated where relevant. Between-group differences in demographic data were analysed using  $\chi^2$  for categorical variables and ANOVAs for continuous variables. For the main analyses, ANOVAs were conducted to reveal between group differences (TS group and controls) in the emotional symptoms (SDQ) and emotional functioning subscales (PedsQL). ANOVA was also used to examine the relationships between motor, vocal and total tic severity

and individual comorbid disorders. Because of the highly unbalanced nature of the analyses in association with some concerns over homogeneity of variance, a non-parametric Mann-Whitney  $U$  test was applied to examine differences in SDQ and Peds subscales associated with comorbid diagnostic status. Simple regression models tested the prediction that increased tic severity would predict emotional symptoms (SDQ) and emotional functioning (PedsQL) outcomes. Non-parametric tests examined relationships between individual comorbid disorders and impact scores of the SDQ.

### **3. Results**

#### *3.1. Data screening*

Data were entered in an SPSS spreadsheet. Due to the completeness of the data set, imputation of missing values was not required. With the exception of some comparisons involving the presence of comorbid disorders, homogeneity of variance for all comparisons was supported by Levene's tests. A small number of meaningful outliers were retained for analysis given their clinical legitimacy. With the exception of the PTQ, which was successfully corrected by applying a Log 10 transformation, normality was satisfactory. To protect against inflated family-wise error, a per comparison critical significance level of  $\alpha = .01$  was applied.

#### *3.2. Demographic information*

Demographic findings are presented in Table 16. Between group differences for categorical variables were analyzed with  $\chi^2$  and for continuous variables with ANOVAs. With one exception, the groups were homogenous. Although both groups were overwhelmingly Caucasian, the TS group was slightly more ethnically diverse than the controls,  $\chi^2(3, N = 193) = 12.02, p = .007$ .

### 3.3. Prevalence of comorbid disorders

The majority (73%) of the TS group was reported as having a formally diagnosed comorbid disorder. These included OCD (44%), “other” anxiety disorders, a category that could include GAD, separation anxiety, panic disorder, social anxiety and specific phobias (36%), ADHD (33%), learning disorders (19%), impulse control disorders (7%), autism spectrum disorder (5%), depression (2%), conduct disorder (2%) and “other” unspecified disorders (9%). Of those with comorbidity, 66% ( $n = 63$ ) had more than one co-occurring disorder.

### 3.4. Between group differences in emotional symptoms and emotional functioning

To determine significant differences between the TS and control group on these measures (SDQ; emotional symptoms subscale, PedsQL: emotional functioning subscale), two single-factor between-subjects ANOVAs were conducted. Results revealed that individuals with TS experienced significantly higher rates of emotional symptoms (SDQ),  $F(1, 192) = 148.77, p < .001, \eta_p^2 = .44$  and impaired emotional functioning (PedsQL),  $F(1, 192) = 85.29, p < .001, \eta_p^2 = .31$ , than controls.

### 3.5. Relationship between tic severity and emotional symptoms (anxiety and depression) and emotional functioning

Simple regression analyses revealed that total tic severity predicted both emotional symptoms as measured by the SDQ  $F(2, 83) = 8.80, p = .004, R^2 = .18$  and emotional functioning as assessed by the PedsQL,  $F(2, 83) = 6.04, p = .001, R^2 = 0.11$ . These relationships were in the hypothesised direction. ANOVAs revealed significant relationships between tic severity and two parent-reported comorbid disorders. Comorbid ADHD was associated with increased vocal tic severity,  $F(1, 85) = 6.70, p = .01, \eta_p^2 = .27$ , and comorbid learning disorder was associated with increased motor,

$F(1, 85) = 9.98, p = .002, \eta_p^2 = .11$ , and total tic severity,  $F(1,85) = 7.76, p = .007, \eta_p^2 = .11$ .

### *3.6. The impact of formally diagnosed comorbid disorders on emotional symptoms, emotional functioning and impact scores.*

Results of the Mann-Whitney  $U$  tests determined that individuals with ‘TS only’ displayed lower levels of emotional symptoms (SDQ),  $U = 432.00, z = 2.88, p = .004$ , and reported better emotional functioning (PedsQL) than those with a comorbid diagnosis,  $U = 378.50, z = 3.39, p = .001$ .

Decreased emotional functioning (PedsQL) was limited to two comorbid disorders. Increased impairment was found for individuals with OCD,  $U = 383.00, z = 4.63, p < .001$ , and “other” anxiety disorders,  $U = 451.00, z = 3.63, p < .001$ .

Significant relationships were found between two comorbid disorders and impact scale scores of the SDQ, indicating that some types of comorbidity were associated with increased levels of distress and impairment as well as increased burden for the family. The presence of autism,  $U = 24.00, z = 2.82, p = .002$ , and “other” comorbid disorders,  $U = 113.00, z = 2.74, p = .006$ , was associated with significantly higher levels of distress.

### *3.7. Current treatment providers*

Within this sample, individuals with TS were receiving treatment from a paediatrician ( $N = 34, 39.5\%$ ), psychologist ( $N = 28, 32.6\%$ ), special education assistant ( $N = 24, 27.9\%$ ), neurologist ( $N = 22, 25.6\%$ ), general medical practitioner ( $N = 22, 25.6\%$ ), psychiatrist ( $N = 16, 18.6\%$ ), counsellor/school counsellor ( $N = 14, 16.3\%$ ) and family therapist ( $N = 1, 1.2\%$ ).

#### **4. Discussion**

The results of the current study indicated that this broad community-based sample of children and adolescents with TS displayed increased rates of emotional symptomatology and impaired emotional functioning in comparison with controls. Comorbidity and tic severity were also associated with increased emotional symptomatology and impairment in emotional functioning in individuals with TS.

The SDQ was employed to screen for the presence of emotional symptoms, which assessed both subclinical and clinical level general symptoms of anxiety and depression (Goodman & Scott, 1999), with results indicating a significantly increased rate of symptoms for youth with TS in comparison with their peers. Findings also indicated an increased level of impaired emotional functioning for the TS group, as measured by the PedsQL. This is consistent with multiple prior studies associating TS with symptoms of anxiety, depression and impaired functioning in the emotional domain (Cavanna et al., 2009; Coffey et al., 2000; Conelea et al., 2011; Eddy et al., 2011; Kurlan et al., 2002; Robertson, 2006a; Robertson, 2006b, 2012; Storch et al., 2007)

The study also assessed the prevalence and impact of clinically diagnosed anxiety disorders and depression as reported by parents. Consistent with prior studies of community-based populations, the current results revealed a substantial rate (73%) of comorbidity for individuals with TS (Freeman et al., 2000; Robertson, 2006a; Robertson, 2006b). Formally diagnosed “other” anxiety disorders, a category including GAD, separation anxiety, panic disorder, social anxiety and specific phobias, was the second most prevalent comorbidity reported by parents behind OCD, with ADHD and learning disorder also strongly represented. This pattern of comorbidity is consistent with prior research (Freeman et al., 2000) with the

exception of the slightly lower than expected prevalence of ADHD in the current study. These results support the conclusion that anxiety represents a significant problem, even in community-based samples of youth with TS. Given the findings from prior research identifying the adverse role of anxiety in increasing the risk of distress, dysfunction, and depression in individuals with TS (Lin et al., 2007; Silva et al., 1995), this result is of substantial clinical interest and reinforces the need to screen for anxiety symptoms in children and adolescents with TS.

However, the current results revealed a discrepancy between the presence of emotional symptoms indicated by the SDQ and impaired emotional functioning identified by the PedsQL, and the low level of formally diagnosed comorbid depressive disorder (2%) reported for the TS group. This discrepancy may be interpreted in several ways. The finding may suggest the presence of elevated rates of subclinical level symptoms of depression that fail to meet the DSM-5 criteria for diagnosis of a depressive disorder within the TS sample. Indeed a lower rate of clinical level depressive disorder would be expected in a community-based sample if, as has been hypothesised, significant depressive symptomatology is more likely to be experienced by individuals with more severe forms of TS (Robertson, 2006b; Robertson et al., 2006).

Alternatively, this discrepancy may indicate the presence of undiagnosed depression in this group of children and adolescents with TS. Under-diagnosis of emotional disorder may be attributable to several factors, including the well-recognised challenge associated with differential diagnosis in the context of TS and high rates of comorbidity (Eapen & Črnčec, 2009). Moreover, clinicians may underestimate the risk of depression in younger TS populations or fail to routinely include assessment of such symptomatology. A clinical approach that necessarily



prioritises tic reduction and the management of comorbid disorder may also result in an unintentional tendency to overlook less obvious signs of emotional dysfunction. The presence of any level of depressive symptomatology is of particular clinical importance given the demonstrated role that depression plays in predicting highly adverse outcomes by late adolescence and adulthood for those with TS (Coffey et al., 2000; Jalenques et al., 2012; Muller-Vahl et al., 2010; Robertson, 2006b). Further research is needed to establish the prevalence of symptoms of depression in TS as well as to understand best practice for incorporating an evaluation of depressive symptoms into clinical assessments of TS.

The current study also examined the role of tic severity and the presence of comorbid disorders (particularly ADHD, OCD and ICD). As hypothesised, tic severity predicted an increase in emotional symptomatology (SDQ) and impaired emotional functioning (PedsQL) in individuals with TS. Effect sizes however revealed that a substantial proportion of variance was not attributable to tic severity. This supports the theory that the aetiology of depression in particular is likely to be multifactorial (Robertson, 2012), and implies the need to consider other factors.

As expected, comorbidity also emerged as a predictor of adverse emotional outcomes for youth with TS. Results of nonparametric analyses revealed that having a formal comorbid diagnosis was significantly associated with increased symptoms of anxiety and depression (SDQ) and impaired emotional functioning (PedsQL). This is consistent with prior studies linking comorbid disorders with adverse emotional outcomes (Carter et al., 2000; Cutler et al., 2009; Eapen et al., 2004; Hoekstra et al., 2004; Robertson & Orth, 2006; Robertson et al., 2006; Sukhodolsky et al., 2003).

Impact scale scores of the SDQ also suggested that the presence of a comorbid disorder, had an adverse impact on parental perceptions of the level of distress and

dysfunction their child experienced and increased family burden. This is consistent with the findings reported by Cooper and Livingston (2003).

As found for tic severity, comorbidity is likely to explain some but not all of the variability in the emotional status of the current sample of children and adolescents with TS. As Robertson and colleagues show (2006), in the context of depression, symptoms appear to emerge from complex interactions of factors such as tic severity and comorbidity. Coprolalia, a severe tic symptom, which has also been associated with increased social anxiety and depression in the literature (Robertson, 2012), was reported at a rate of 20% in the current study. This may also have had a significant impact on the level of emotional symptoms reported in the TS sample. The study was, however, unable to determine the relationship between ICD and emotional outcomes. This may be reflective of the lack of power associated with the low number of cases of ICD reported (7%), a rate consistent with the decreased severity of TS expected in a community sample. Further research is needed in regard to these findings.

Finally, the study demonstrated the utility of employing a brief, well-validated and reliable psychometric measure such as the SDQ or PedsQL to screen for emotional difficulties as part of the multidimensional assessment of young people with TS. Such measures can be employed across a wide range of clinical, non-clinical and research settings, and include options for use by multiple raters. Although brief measures such as the SDQ and the PedsQL collapse anxiety and depression into a single 'emotional' category, their use is supported by the consistency between the current results and the major findings of studies employing longer form or disorder specific measures. For example, the present findings are consistent with those of Robertson and colleagues (2006) who determined an increased prevalence of

depressive symptoms using the Children's Depression Inventory (Kovacs, 1992) and the Birlson Depression Self Report Scale (Birlson, 1981) for individuals with increased tic severity and comorbid diagnoses in a clinical sample. While brief assessments cannot replace thorough clinical assessment, initial screening could be potentially useful in the identification of at risk youth in primary care, educational or other non-clinic based settings, and assist in the process of appropriate referral for differential diagnosis and early intervention. The finding that only a small minority of TS youth were currently under the care of a psychiatrist or psychologist highlights the potential benefit of such instruments.

#### *4.1. Limitations and future research*

Several limitations should be noted when interpreting the current findings. Although community drawn and mirroring the lower rate of comorbidity associated with clinic-based TS samples, the high incidence of multiple comorbidity reported in the current sample suggests that individuals with more severe TS may be over-represented. Whilst a future study employing an epidemiologically based sample could address this limitation, current resources did not extend to such a study.

The need to capture data from a nation-wide, non-clinic based sample precluded the ability to control for clinical assessment of TS and comorbid disorders. Whilst reliable measures of general emotional functioning, the SDQ and the Peds QL are not diagnostic tools, and therefore cannot differentiate between symptoms of specific anxiety or depressive disorders, nor determine symptom severity, factors that may variably impact emotional outcomes. Although a relatively large sample, the study also lacked the statistical power to assess the impact of individual anxiety and depressive disorders and less frequently diagnosed comorbidities. As in multiple prior TS QoL studies, this research was unable to determine the relationship between

impairment, comorbidity and variability in rates of the diagnosis. Future large-scale longitudinal studies that include systematic psychiatric evaluation would be required to address these limitations.

The reliance upon parental reports of comorbidity may have introduced error and whilst prior TS studies found that parental reports more accurately identified behavioural problems of youth with TS than youth self-reports (Termine et al., 2011), discrepancies have been observed between self and parent reported QoL outcomes (Conelea et al., 2011; Storch et al., 2007). This suggests the benefit of employing multiple raters, including youth self-reports in future TS studies.

The inability to assess the impact of gender and age due to the high percentage of similar aged males in the current sample could be assessed in future studies, which could also address the lack of the cultural and SES diversity currently demonstrated.

Finally, the use of a generic health-related QoL measure facilitated comparison with healthy peers; however, future studies may benefit by employing disease specific measures, such as the newly developed Gilles de la Tourette Syndrome Quality of Life Scale – Child and Adolescent (Cavanna et al., 2013). The inclusion of TS specific items (such as tics and obsessive phenomena) is likely to increase the sensitivity and accuracy of such measures for individuals with TS.

#### **4.2. Conclusion**

The present study found that when compared with undiagnosed peers, children and adolescents with TS are at significantly increased risk of clinical and sub-clinical level symptoms of emotional disorders such as anxiety and depression and impaired emotional functioning. Whilst tic severity and comorbidity disorder increase the risk of emotional disorder, it is likely that the aetiology of emotional symptomatology in TS is multifactorial. This knowledge may inform assessment and clinical intervention.

Given the highly adverse outcomes associated with comorbid emotional disorder for individuals with TS, detection and prevention of anxiety and depression in childhood is critical. Brief screening instruments including the PedsQL and the SDQ may provide simple and efficient early identification of children at risk across multiple settings and facilitate appropriate clinical referral and early intervention.

### **Study Funding**

None to declare

### **Conflicts of Interest**

The authors have declared that they have no competing or potential conflicts of interest.

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### **Highlights**

- This controlled study found that Tourette's (TS) greatly increased the risk of emotional disorder and dysfunction in diagnosed children and adolescents
- Signs of depression in youth with TS in this large community sample were poorly recognised or overlooked
- Tic severity and comorbid disorders exacerbated the increased the risk of emotional difficulties for youth with TS

- The aetiology of emotional symptomatology in TS is likely to be multifactorial and this should inform assessment and intervention
- As the strongest predictors of adverse outcomes for adults with TS, prevention, detection, and treatment of signs of anxiety and depression in youth with TS is of considerable clinical importance
- Employing brief psychometric instruments may enhance detection of at risk youth across multiple settings and facilitate clinical referral and early intervention

### **Key Points**

1. TS is a poorly understood, under-diagnosed and stigmatising disorder with a surprisingly high rate of prevalence.
2. TS has highly adverse psychological and behavioural consequences yet psychologists are under-represented in TS research and the clinical setting.
3. Two notable gaps in the TS literature include a lack of Australian TS research and an over-reliance upon clinic based (more severe forms of TS) populations.

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**Table 16.** Demographic Characteristics and Between Group Differences for Control and TS Groups

Demographics	Control Group			TS Group		
	<i>n</i>	<i>n</i> (%)	<i>Mean SD</i>	<i>n</i>	<i>n</i> (%)	<i>Mean SD</i>
Age of child	108		11.31 (2.58)	86		11.44 (2.78)
Relationship with Child	108	Biological Mother	97 (88.8%)	85	77 (90.6%)	
		Adoptive mother	2 (1.9%)		1 (1.2%)	
		Biological father	9 (8.3%)		7 (8.2%)	
Place of Residence	108	City	67 (62%)	86	50 (59%)	
		Regional Centre	33 (30.6%)		20 (23.8%)	
		Rural/Remote	8 (7.4%)		14 (16.7%)	
Marital Status	107	Never married	6 (5.6%)	86	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	108	Low	6 (5.6%)	82	8 (9.8%);	
		Low-Middle	26 (24.1%)		19 (23.2%)	
		Middle & Above	76 (70.4%)		55 (67.1%)	
Gender of Child	108	Male	79 (73.1%)	86	72 (85.4%)	
		Female	29 (26.9%)		12 (14.3%)	
Ethnicity	108	Caucasian	107 (99.1%)	85	74 (87.1%)	
		Aboriginal TSSI	0		2 (2.4%)	
		Asian	0		3 (3.5%)	
		Other	0 1 (0.9%)		6 (7.1%)	
Sibling	108	Yes	101 (93.5%)	86	76 (88.4%)	
		No	7 (6.5%)		10 (11.6%)	

\*\*  $p \leq .01$

**SECTION 3 CHAPTER 5. Youth with Tourette syndrome: Parental perceptions and experiences in the Australian context**

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**This chapter is a report on the findings from Study 1 and Study 2, which documents the stressors and difficulties associated with parenting a child or adolescent with TS. Results are compared with those of parents of children with other serious chronic medical or psychiatric disorders, which have been documented in the disability literature.**

**Abstract**

**Objective:** To enhance understandings of the impact of TS on parents of diagnosed youth. Specifically, the current study aimed to explore and identify the multidimensional stressors associated with parenting a child or adolescent with Tourette Syndrome (TS) in the Australian context.

**Method:** As part of a larger qualitative and quantitative community-based study, semi-structured telephone interviews with 22 mothers of youth with TS were conducted regarding their experiences.

**Results:** The study identified parent, child and contextual factors that contributed to parental stress, with many mirroring the experiences of parents of children with other chronic paediatric disorders. However several TS specific factors also emerged from the data analysis, highlighting the unique difficulties encountered by parents of diagnosed youth. Serious deficits in professional expertise and services currently available for the TS community were also identified.

**Conclusions:** Findings indicate the generally unacknowledged challenge of parenting a child with TS, which equates with that experienced in the context of other serious chronic paediatric disorders. Results also indicate the need for psychosocial support for both child and parent, and greatly improved access to well-informed mental health and educational services in the Australian context.

**What is already known about this topic**

- Tourette's Syndrome (TS) is a complex neurodevelopmental disorder that has been associated with increased parental stress, caregiver burden and family conflict.
- Although evidence suggests that characteristics of TS, comorbidity and poor professional and public understanding of TS adversely impact parents, the factors contributing to parental stress in the context of TS are understudied.
- Deficits in available services and supports for those diagnosed and their families have been identified in the international literature.

**What this topic adds**

- Findings indicate the unacknowledged challenge of parenting a child with TS in the Australian context, which equates with the difficulty experienced by parents of youth with a range of chronic paediatric disorders.
- Present findings substantially expand upon current understandings of the many complex factors, including those unique to TS, which contribute to parental stress.
- Results also indicate the need for psychosocial and psychotherapeutic support for both youth diagnosed with TS and their parents, and greatly improved access to well-informed mental health and educational services in the Australian context.



Tourette Syndrome (TS) is a surprisingly prevalent (1:360 youth in the United States) (Bitsko et al., 2014) neurodevelopmental disorder characterised by the presence of multiple motor and vocal tics occurring for a period of at least one year (American Psychiatric Association, 2013; Leckman, Bloch, Scahill, & King, 2006). TS is greatly complicated by high rates of comorbidity (80-90%), particularly Attention Deficit Hyperactivity Disorder (ADHD) and Obsessive Compulsive Disorder (OCD) (Freeman et al., 2000).

The adverse impact of TS has been consistently demonstrated in quality of life studies; with diagnosed youth experiencing impaired physical, psychological, social, and academic functioning (Conelea et al., 2011; Cutler, Murphy, Gilmour, & Heyman, 2009; Eddy et al., 2011a; Eddy et al., 2011b; Storch et al., 2007a; Storch et al., 2007b). The impact of TS on the parents of those diagnosed however has not been thoroughly examined. Given the increase in parental stress accompanying a wide range of chronic paediatric disorders (Wallander & Thompson, 1995), it is likely that parents of children with TS are at similar risk. The importance of understanding parental stress is illustrated by research linking it with adverse consequences for parents, children and family functioning as well as exacerbation of the child's symptomatology (Streisand, Braniecki, Tercyak, & Kazak, 2001).

Theoretical models have been developed to predict the processes and variables associated with increased parental stress, the most popular being Abidin's ecological model (1992). This model describes an additive, multidimensional process that

conceptualises child, parental, and contextual (situational or demographic life events) factors as mediators or moderators of parental stress. The stress in turn predicts negative parenting behaviour and consequent adverse child outcomes. Transactional models of stress and coping have also been tested to determine the impact of specific disorders on parental adjustment (Wallander & Thompson, 1995). These emphasise psychosocial factors such as parent-child interactions and family functioning, parental cognitions (such as stress appraisal and self-efficacy) and coping responses, and demographic and illness parameters as mediators of maternal and child psychological adjustment to chronic disorder.

Studies employing these models have revealed factors contributing to parental stress that transcend underlying diagnosis as well as those that are disorder specific. Commonly experienced stressors identified in a metasynthesis of qualitative paediatric studies targeting a range of disorders (Coffey, 2006) include the perception of increased caregiver burden, omnipresent fear and anxiety, the need to adopt a proactive role in the management of the child's condition, the impact of critical times and events such as diagnosis, and social isolation (Coffey, 2006). It follows that parents of a child with TS may experience these same difficulties in addition to characteristics unique to TS.

A review of the TS literature provides some evidence for increased parental stress and identifies several challenges confronting parents of those diagnosed. High levels of caregiver burden and stress were found for mothers of children with TS, which exceeded the rate reported by parents of children with chronic asthma (Cooper, Robertson, & Livingston, 2003). When compared with unaffected families, parents of a child with TS experienced higher rates of parental stress and frustration (Bitsko et al., 2014; Robinson, Bitsko, Schieve, & Visser, 2013; Stewart, Greene, Lessove-Schlaggar,

Church & Schlaggar, 2015), substance abuse, and marital dysfunction (Stefl, 1983). Most caregivers also encounter low levels of acceptance and understanding of TS, high rates of delayed and mistaken diagnosis, a lack of adequate specialist TS services, with the child often having unmet mental health care needs (Bitsko et al., 2014), in addition to stigmatisation, negative stereotyping and social isolation related to TS (Collins, 2005; Conelea et al., 2011; Davis, Davis, & Dowler, 2004; Dedmon, 1990; Eapen & Črnčec, 2009).

Given its waxing and waning course, unknowable prognosis, and the lack of treatment options currently available, living with TS is inherently stressful (Bruun & Budman, 1997; Collins, 2005). Other sources of stress include the increased incidence of psychopathology amongst parents of youth with TS, parental guilt relating to genetic transmission of TS, impairment in parent-child and sibling relationships, and elevated levels of overall family stress (Carter et al., 2000; Cohen, Ort, Leckman, Riddle, & Hardin, 1988; Cooper et al., 2003; Lee, Chen, Wang, & Chen, 2007; Schoeder & Remer, 2007; Storch et al., 2007a; Storch et al., 2007b; Wilkinson, Marshall, & Curtwright, 2008; Wilkinson et al., 2001; Woods, Himle, & Osmon, 2005). There is also evidence to suggest that increased symptom severity (Lee, Chen, Wang et al, 2007) and comorbidity may exacerbate parental stress, with disorders such as co-occurring ADHD associated with elevations in parental and family stress and conflict (Bitsko et al., 2014; Cooper et al., 2003; Stewart et al., 2015; Sukhodolsky et al., 2003; Wilkinson et al., 2008; Wilkinson et al., 2001). Traficante (2007) concluded that parental stress is related to the frustrations experienced in the efforts to alleviate distress and successfully parent a child with TS in the absence of adequate professional and informational support. Finally, the role of socio-demographic factors was illustrated in a study of

Taiwanese parents of children with TS. Low family income and limited access to childcare were strong predictors of increased parental stress (Lee et al, 2007).

The above findings illustrate several challenges confronting parents of youth with TS. Theoretical modelling (Abidin, 1992; Wallander & Thompson, 1995), however, suggests the possibility that many more factors may impact parental stress. These may include the general stressors reported by peers in the context of chronic paediatric disorder, and those uniquely associated with TS. Given the central and under-supported role that many parents play in the care and welfare of their child diagnosed with TS, an improved understanding of the unique pressures they experience is of substantial concern. It is also likely that the current status of TS in the Australian context, which until recently has been under-researched, and the availability of services and supports influence parental stress. Although Australia has universal health care, a recent survey by the current authors (O'Hare et al, In Press) found that parents of youth with TS (n=86) encountered low levels of TS specific knowledge and difficulty accessing services within the Australian health and education sectors; high rates of stigma (50%) and the majority were reliant upon general medical practitioners for diagnosis and ongoing management of the child's TS. Less than two thirds of youth in the study were currently receiving any form of intervention for their TS. With a population of less than twenty four million spread over a vast geographical area, Australian demographics present unique challenges to the provision of accessible specialist services. These are clustered in a few capital cities, and waiting lists can be long. Furthermore, advocacy groups such as the Australian Tourette's Association, which numbers around one thousand active members, lack the funds and resources of

their international counterparts to adequately advance the cause of the Australian TS community.

The primary goal of the current study, therefore, was to address a current gap in the TS literature by exploring and identifying the stressors that the primary caregivers of youth with TS perceive as contributing to parental stress. In order to best describe and understand these subjective experiences, a qualitative approach to research design was adopted. Whilst the processes by which these factors affected parenting were not the subject of the current study, it was hoped that findings could provide a foundation for future research, facilitate more informed approaches to clinical intervention, and help providers and policy makers respond more effectively to the needs of the TS community.

## **Methods**

### **Participants**

The sample consisted of 22 biological mothers of youth diagnosed with TS. The majority were married and living with their partner (82%); had an average of two children; resided in urban areas (68%); and were middle income or above (86%). Children ranged in age from 7 to 19 years ( $M$  age = 12, 20 males, 2 females). Modal age of TS diagnosis was 7 years, and 21 youth had comorbid diagnoses including OCD ( $n = 6$ ); obsessive-compulsive behaviours ( $n = 6$ ); anxiety ( $n = 6$ ); learning disorder ( $n = 6$ ); ADHD ( $n = 4$ ); oppositional defiant disorder ( $n = 2$ ); impulse control ( $n = 2$ ); Asperger's syndrome ( $n = 2$ ); autism ( $n = 2$ ) and depression ( $n = 1$ ).

### **Measures and Procedure**

Semi-structured telephone interviews, ranging in duration from 60 and 90 minutes, were conducted as part of a larger study evaluating attachment relationships and functioning of Australian youth with TS. Questions were open-ended to allow space for the voices of participants. Many provided unsolicited insights into their experiences as parents of youth diagnosed with TS, which extended beyond the interview questions (Appendix A for a copy of the interview guide).

A purposeful sample of participants was recruited via email invitation issued to all members of the Tourette Syndrome Associations of Australia and Victoria. The first 22 volunteers to respond were included in the study. Oral informed consent for participation and audio recording was required prior to all interviews, which were conducted by the principal researcher. Participation was voluntary, confidentiality guaranteed, and the study was conducted with the approval of the JCU University Human Research Ethics Committee, in compliance with the latest Declaration of Helsinki (2008). No incentives to participate were offered.

On completion of each interview, all audio files were transcribed verbatim. An eclectic approach to content analysis and data coding was adopted for the purposes of the study, by employing both an inductive approach to detect emerging themes and a deductive approach to content analysis (Miles & Huberman, 1994; Strauss & Corbin, 1998). An initial round of coding was conducted with the reading and rereading of all transcripts several times, to identify initial themes and categories. Three *a priori* themes grounded in attachment theory were employed to code data related to the impact of TS on the attachment relationships included in the study, the findings of which are reported elsewhere (O'Hare et al, 2015). Coding also identified a fourth major theme, which

emerged inductively from the initial round of coding. This was labelled “Parental Stress” and is the sole focus of the current report.

Line- by-line manual coding of all transcripts then continued in an iterative process, leading to the reduction of data into subcategories and categories within the theme of “Parental Stress”, which were modified over a period of six months (Biddle, Markland, Gilbourne, Chatzisarantis, & Sparkes, 2001). Twelve subcategories, each representing a distinct parental stressor described by participants, emerged inductively from this process. These were further grouped into two major categories labelled “General stressors” and “TS specific factors” (presented in Table 17).

Definitions and sample text from multiple participants for the major theme, two categories and twelve subcategories were included in a data dictionary composed for the study. Third party trustworthiness checks of data coding by two trained coders blind to the purposes of the study resulted in high percentage agreement and significant inter-rater agreement as measured by Krippendorff’s Alpha, ( $p < .01.$ ) and an audit trail was maintained.

One direct question was posed at the conclusion of each interview. “*What do you feel the TS community needs the most?*” Responses to this question were recorded and data coded by a simple content analysis (Table 18). Finally, the frequencies and sample percentages for responses in the study were calculated.

## **Results**

### **Experiences Related to Parenting a Child with Tourette Syndrome**

Twelve distinct themes representing the major stressors associated with parenting a child with TS emerged from the data analysis. Responses across the sample

demonstrated a high level of homogeneity. The quotes included in this report are widely sourced from the twenty-two participants included in the study. Many findings are consistent with the results of Coffey's (2006) recent metasynthesis whilst others were disorder specific (TS). Results are presented in Table 17.

### **Mother assumes the burden of care.**

Over 90% of the mother's described the stress associated with assuming primary responsibility for the care of their TS diagnosed child, even for those with an available and supportive partner. This demanding role was characterised by self-sacrifice and a determination to "do the best" by the youth.

*"We made the decision that I'd be a stay at home mum. I think it's been the best thing for my children but it's so hard, so all encompassing (caring for a child with TS)."*

### **Serious / Credible - Battling ignorance, fighting for understanding and acceptance**

The widespread lack of understanding and acceptance of TS experienced by those diagnosed and their families was another critical contributor to parenting stress for 95% of participants. This had wide-ranging adverse consequences such as delayed or mistaken diagnosis, inadequate school support, relationship breakdowns, misattribution of blame and responsibility for symptoms and behaviours, dismissal or minimisation of maternal concerns regarding her child, and the experience of negative stereotypes and stigmatisation.

*"The doctor suggested his behaviour was 'Tourettish' but it was behaviourally based and recommended parenting lessons! I was in stunned silence and I just felt so angry."*



### **Mother forced to take charge, advocate, become the expert**

Participants (95%) also identified the increased burden associated with “taking charge”- becoming the expert, educator and advocate for the diagnosed child and family.

*“I feel sorry for the child that doesn’t have a parent that wouldn’t have a voice to be able to speak up...you don’t always come out (after talking to school) with brownie points but I think, well I have to do it!”*

### **“Living worried” & “Staying in the struggle”**

Mothers (90%) also described struggling to manage the many anxieties and fears they experienced in response to their child’s pathology and behavioural, social, emotional and academic difficulties. Many had difficulty maintaining a sense of optimism regarding a “normal” future for both child and mother in the face of the youth’s symptoms. Low self-efficacy in regard to coping with the child’s high level needs was common. Most challenging were the youth’s self-injurious behaviours, aggressiveness, episodic rage, impulsivity, very severe tics, or multiple comorbid diagnoses.

*“The stress is never ending...life was so simple and easy and everybody was happy, but now everyone around me is sort of falling to bits and it’s hard. And so if I lay in my bed and think about it then I get so sad, so I just hop out of bed and just do it!”*

### **“Tangled”**

Participants (78%) also identified parenting dilemmas that were a function of the difficulties they experienced when attempting to disentangle TS and comorbid symptoms from behaviour that would usually be construed as childhood or adolescent misbehaviour.

*“It’s like you’ve got to learn how to draw the line between treating this child like a normal teenager and... keeping in mind that he’s not quite the same as everybody else but he’s got to be treated the same as everyone. The balance becomes harder as he gets older...you really double think, is this the teenager talking, or the TS, ADHD, or is he just being a pain in the ass?”*

### **Critical times**

The fluctuating nature of parenting stress also emerged as a significant theme for 95% of the sample, with stress peaking around critical times and events in the life of the child and family. These included the time of diagnosis, developmental transitions such as puberty and adolescence, changing schools, changes in family routine or circumstances, apparently minor events such as holidays, and major life events such as a death in the family. Whilst attempts to anticipate and prepare for such events provided a sense of control for mothers, this was also challenging and stress provoking.

*“Adolescence! It’s my big fear (when he enters high school) because violence is just not going to cut it!” “Going on family outings or holidays... it’s just a nightmare...we have to plan ahead and it’s really hard... we try to prepare him... teach him strategies”*

### **The diagnostic experience.**

The process of diagnosis was highly traumatic for the majority (90%). Many (55%) had to drive the diagnostic process by proactively assuming the role of educator for the professionals they consulted, and described being invalidated and dismissed during this process.

*“ I just don’t want other parents to have the same nightmare!...it took years...we’ve been to so many specialists... one psychologist said really weird things... like they said he had ODD, that he might be homosexual!”*

As a result of the diagnostic experience, the majority of mothers (68%) felt some sense of relief amidst the trauma.

*“I was happy because you know, like I was just sick and tired of people constantly telling me that he can stop it when I knew he couldn’t. We had a psychiatrist who told me to give him two dollars every time he stopped. That’s how bad it got!”*

### **Grief and loss of the “ideal child”**

Others overtly referred to their experience of distress and grief regarding the loss of their “ideal” child (32%) after the diagnosis of a chronic, incurable disorder. This was compounded by diagnosis occurring at an unexpected time (i.e., middle childhood or teens), which for many followed the child’s “normal” infancy and early childhood development.

*“We had to grieve for the sense of loss- at the same time guilt because he didn’t have leukaemia and he was not going to die from this ... but it is going to affect his quality of life... when he was born he was perfect and I’d had this perfect child, so it never occurred to me that something could kick in at 5 or later!”*

### **Social isolation (child / mother/ family)**

Mothers (86%) described the social isolation experienced as a consequence of their child's diagnosis. Most reported the loss of important relationships, a drop in social support, and difficulty maintaining a 'normal' social life due to restriction in activities, voluntary social withdrawal or social exclusion post diagnosis.

*“With family it's really, really quite difficult so in the end I've had lots of arguments... I've realized that I've tried and tried and I've talked until I'm blue in the face and I think well they are really quite ignorant!” “Families with 'normal' kids, just don't get it. They don't want their child to be associated with yours.”*

### **Family first - Home as sanctuary**

Many (73%) responded to social isolation and reduced social support by focusing upon and surviving as a nuclear family. Home became a place of “sanctuary”, where the child could ‘let it all hang out’ and parents didn't have to monitor or manage the response of others to the youth's symptoms.

*“I suppose with our family (nuclear) unit, we are very strong with each other, we rely on each other a lot which I think helps in lots of ways.” “We said honey you just tic as much as you want and as often as you want and as loud as you want because we don't care (mum, dad and siblings).”*

### **“Bridge to the outside world”**

The importance of having some sort of “bridge to the outside world” to counter the isolation, distress and difficulty associated with the lack of acceptance and understanding of TS by others was also reported (73%). Many stressed the importance

of connecting with those who “get it” - for example peers (Tourette Syndrome Associations of Australia and Victoria, TS Mothers support groups), parents of children with other disorders, understanding medical professional, or teachers.

*“Our TS mothers group was a godsend.” “This new doctor... I said ‘do you know anything about Tourette’s’ and he said ‘I’ve got three brothers with Tourette’s!’ so I was like Yes! Now you’re my new doctor!”*

### **School as a key player**

Finally, 95% of the mother’s identified the critical role of the school in determining the youth’s academic and social wellbeing and maternal stress.

*“Teachers are going to be my best friends - I’m not going to be in their face but I’m there whenever they want me for anything.” “The school- I can’t thank them enough! They’ve worked so hard to get him to where we are now”. “The school is fantastic... curriculum support. He’s slowly catching up so everything’s good... they handled the bullying well...”*

### **Statement of needs**

Table 18 summarises the response to the final interview question regarding the needs of the TS community. No data interpretation was required. The most frequently identified needs (78% of the group) were for acceptance, understanding and support for those diagnosed and their families in addition to comprehensive informational support. These were followed by a need for improved TS knowledge amongst professionals in the education (50%) and health sectors (47%). Table 18 also includes several less frequently identified specific needs reported by individuals.

## Discussion

To the best of our knowledge this is the largest qualitative study of parents of youth with TS and the first to be conducted in Australia. It achieved its primary goal of identifying the major factors contributing to stress in the context of parenting a child or adolescent diagnosed with TS. Findings also indicate the substantial and largely unacknowledged level of stress experienced by the present sample. In summary, and as predicted by theoretical models of parenting stress (Abidin, 1992; Wallander & Thompson, 1995), participants shared many problems and described similar levels of stress as those reported by parents of children with a wide range of serious, chronic paediatric disorders (Coffey, 2006). Also as expected, not all of the parenting experiences reported in the present study transcended the child or adolescent's underlying diagnosis, with results demonstrating a role for TS in exacerbating and creating several unique challenges for parents.

Specifically, the study revealed twelve distinct major stressors confronting this sample of parents of children with TS (See Table 17). As predicted by Abidin (1992) and Wallander and Thompson's (1995) models, stressors included parent, child and contextual factors, in addition to illness parameters, with parent and contextual factors emerging as those most frequently identified. As reported in prior TS studies (Cooper et al., 2003; De Lange & Olivier, 2004; Wilkinson et al., 2001) and the parenting literature (Coffey, 2006), the majority of mothers assumed the burden of care of their child in addition to the role of child advocate and TS expert (Collins, 2005; Dedmon, 1990; Packer, 1997, 2005). This is consistent with Ray's (2002) notion of the "invisible work" that parents of children with chronic conditions undertake. Consistent with

anecdotal reports, both the child and mother also endured a lack of understanding, acceptance, and stigmatisation (Collins, 2005; Conelea et al., 2011; Davis et al., 2004; Dedmon, 1990; Packer, 1997, 2005).

Mothers also identified difficulties occurring around specific times or events, such as adolescence or the time of TS diagnosis. As previously noted (Collins, 2005; Dedmon, 1990), the later was experienced as a highly traumatic major life event for mothers. Compounding the trauma was the difficulty half of the participants experienced in achieving the diagnosis. Many encountered high levels of professional ignorance regarding TS, found it necessary to educate consultants, and were often invalidated and dismissed during the diagnostic process. This is an unfortunate and pervasive experience that has been reported to commonly occur within general medical settings (Balling & McCubbin, 2001). Impaired transactions with providers and the health system were therefore significant barriers to maternal adjustment to their child's diagnosis, and a source of significant stress.

The majority of respondents also described a range of highly adverse emotional experiences relating to their child's TS. These increased the risk of maternal stress in addition to psychopathology and its attendant negative parenting outcomes (Lovejoy, Graczyk, O'Hare, & Neuman, 2000). The fears, anxieties, and high levels of stress reported by participants had the potential to adversely affect maternal cognitive, emotional and behavioural functioning (e.g., self-efficacy, stress appraisals, coping responses, etc.). These are hypothesised to contribute to impaired interactions between mother and child thereby increasing parenting stress (Abidin, 1992; Wallander & Thompson, 1995).

Symptoms that caused the highest level of maternal concern were self-injurious behaviour, aggressiveness and impulsivity, which have been strongly associated with adverse family outcomes, stress, and conflict (Budman, Rockmore, Stokes, & Sossin, 2003; Cohen et al., 1988; Cooper et al., 2003; De Lange & Olivier, 2004; Dooley, Bryna, & Gordon, 1999; Sukhodolsky et al., 2003; Wilkinson et al., 2008; Wilkinson et al., 2001). Fears of the future, inability to parent adequately, persistent guilt and remorse regarding pre-diagnostic parenting, and grief at the loss of their “ideal child” were also deeply distressing.

Mothers also reported significant difficulty disentangling the symptoms and behaviours attributable to their child’s TS and comorbid disorders from normative “misbehaviour”. The current high rate of comorbidity created complex challenges and parenting dilemmas that undermined maternal confidence and exacerbated stress, and is an experience that has been reported in the TS literature (Bitsko et al, 2014; Cohen et al., 1988; Cooper et al., 2003; Lee et al, 2007; Stewart et al, 2015; Wilkinson et al., 2008). Future studies would however be required to assess the relative contribution of various co-occurring disorders to the level and type of stress parents experienced.

A contextual factor contributing to maternal distress was social isolation. The importance of social support has been illustrated in prior TS studies. Schoeder & Remer (2007) found that adequate social support for families of children with TS mediated the impact of tic severity and caregiver burden whilst Lee and colleagues (2007) determined that social support reduced parental stress. The buffering effect of emotional and material support (Green, Furrer, & McAllister, 2007) and its relationship with positive parenting experiences (Andresen & Telleen, 1992) is also well supported in the literature. That participants identified connecting with others who “get it” is therefore



unsurprising. The family's "bridge to the outside world" took the form of peer support and understanding professionals. Many participants also described keeping their focus upon and "surviving" as a nuclear family, which has also been previously described (Davis et al., 2004; Dedmon, 1990) and making home sanctuary.

The final contextual factor playing a critical role in determining child and maternal wellbeing was "the school". This is consistent with prior studies demonstrating the positive contribution of a supportive school environment to maternal and child wellbeing in the TS literature (Packer, 1997; Woods & Marcks, 2005). A limitation of the current study, however, was the inability to adequately examine the impact of important socio-demographic factors such as low income and family breakdown on rates of parental stress. This was due to the relative affluence and stable family characteristics of the majority in the current sample.

The present study also revealed the service and support needs identified by the current parent sample (Table 18), which reflected the deficits identified by O'Hare and colleagues (In Press). The need for improved understanding, acceptance, and support for the child with TS, mother and the family and the availability of immediate, accessible and comprehensive information regarding TS were most frequently endorsed. The latter has been noted in Canadian TS study (Traficante, 2007) and identified by parents of children with other paediatric disorders (Perrin, Lewkowicz, & Young, 2000). Parents also called for substantial improvement in knowledge and understanding of TS for key professionals in the Australian education and health sectors in addition to improved diagnostic processes and access to specialist treatment.

Participants also referred to a suite of specific needs, the most common of which included access to support and respite services. Unlike the study by Traficante (2007),

the present sample did not identify the need for psychological interventions. Perhaps the reason was that this sample of Australian mothers lack awareness of or has limited access to non-pharmacological intervention.

Present findings reflect the experiences of a small group of Australian parents, thereby limiting the ability to generalise to the wider TS population. The high level of agreement expressed between participants however and the relatively large size of this qualitative study ( $n = 22$ ) support the meaningfulness of the findings, which could be confirmed in a future quantitative study employing a psychometric measure of parental stress. Other limitations which could be addressed in future research include the inability to control for the relative effects of TS and comorbid disorder on parental stress and the inability to explore the impact of socio-demographic factors due to the relatively homogenous demographic characteristics of the current sample. The researcher may also have introduced bias at any or all levels of this qualitative study, although efforts were undertaken to reduce this risk. The current study was also limited to the identification of stressors and did not examine the processes by which these may moderate or mediate parental stress. This could be a goal of future theoretically driven quantitative research.

In conclusion, the study identified a wide range of factors with the potential to directly determine, mediate or moderate parental stress in the context of TS and revealed the generally unacknowledged challenges encountered by parents of youth with TS. Present findings also indicate the need for psychosocial and psychotherapeutic support for both child and parent, and greatly improved access to well-informed mental health and educational services.

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**Table 17.** General and TS Specific Stressors Experienced in Parenting a Child or Adolescent with Tourette Syndrome

<i>General Stressors - Shared with Parents of Youth with Chronic, Serious Disorders.</i>	Frequency (% of sample, <i>N</i> =22)	<i>Tourette's Specific Stressors</i>	Frequency (% of sample, <i>N</i> =22)
Mother assumes Burden of Care	20 (91%)	Take TS Seriously/Credibly	21 (95%)
Mother forced to take charge/ advocate/ become the expert	21 (95%)	Social isolation (child / mother/ family)	19 (86%)
Bridge to the Outside world	16 (73%)	School as a Key Player	21 (95%)
Family First (home as sanctuary)	16 (73%)	The Diagnosis Experience:	
		Difficult Process	Yes (12; 55%) No (10; 45%)
		Traumatic	Yes (20; 91%) No (2; 10%)
		Relief	Yes (15; 68%) No (7; 32%)
Living Worried-Struggling to remain optimistic	20 (91%)	Grief /Loss of Ideal Child	21 (95%)
Critical Times	21 (95%)	Tangled (TS vs. co-morbid disorders vs. normative behaviour)	17 (78%)

**Table 18.** Statement of Needs of Mothers of Youth with Tourette syndrome

<i>General Needs</i>	Frequency (% of sample, N=22)
Understanding, acceptance and support from all levels of society for the child, for the mothers themselves as parents of a child with TS & for the family	17 (78%)
Need for immediate, accessible and comprehensive information regarding TS for parents & concerned others (particularly at the time of diagnosis & later in relation to interventions parents could implement to help their child)	17 (78%)
Need for the education of those within the school system (classroom teachers, principals, classroom aids, school administrators and educational policy makers	11 (50%)
Urgent need to improve the low level of knowledge currently demonstrated by medical practitioners in particular.	11 (47%)
Improved diagnostic processes and access to specialist treatment -high priority	11 (47%)
<i>Specific Needs</i> (less frequently identified)	

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Access to formal and informal respite services

Increased funding- federal, state and local government level

Easier access to classroom aides and curriculum flexibility

Increased social networking opportunities for mothers, child and family to reduce feelings of isolation- particularly disadvantage experienced by mothers in non-metropolitan areas

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**SECTION 3 CHAPTER 6. Tourette's Syndrome and mother-child attachment: A qualitative exploration.**

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**This chapter is a report on the study of the qualitative findings from Study 2, regarding the impact of TS on the security of the mother-child attachment relationship.**

### **Abstract**

This novel qualitative study explored the impact of Tourette's syndrome (TS) on the security of the mother-child attachment relationship ( $N = 22$ ). As expected, TS appeared to influence maternal representations of the diagnosed child, the mother's internal working model of the relationship, and her role as caregiver. Although the diagnostic event appeared to strengthen mother-child bonds, the analysis revealed multiple stressors accompanying diagnosis of TS and life in its aftermath that threatened the security of the mother-child relationship. These stressors disrupted or impaired the mother's ability to consistently provide attachment functions, altered the mother's positive representation of the child and her role as caregiver, and appeared to contribute to a tendency towards maternal over-involvement. It was concluded that understanding the impact of TS on the mother-child relationship has implications for clinicians working to enhance child and maternal wellbeing and functioning, and is a subject that warrants further investigation.

### **Introduction**

TS is a childhood onset neuropsychiatric disorder characterised by involuntary motor and vocal tics, which persist for a period of at least one year (American Psychiatric Association, 2013), and high rates of comorbidity (Freeman et al., 2000; Robertson, 2012). It is also surprisingly common, with estimated prevalence rates ranging between 1 per 360 and 1 per 1,000 in the paediatric population (Bitsko et al.,

2014; Robertson, 2012). TS has been strongly associated with adverse outcomes for youth including lower quality of life, impaired psychosocial and academic functioning, increased psychopathology, negative peer relationships, social isolation, and stigmatisation (Bawden et al., 1998; Bernard et al., 2009; Conelea et al. 2011; Collins, 2005; Davis, Davis & Dowler, 2004; Dedmon, 1990; Packer, 1997; Stern et al., 2005; Storch et al., 2007).

A small but developing literature also reveals the adverse impact of TS on primary caregivers, with elevated rates of psychological stress and burden, emotional distress, substance abuse, psychopathology and social isolation reported by parents of youth with TS (Cooper et al., 2003; Lee et al., 2007; Robinson et al. 2013; Stewart et al, 2015). Although TS also appears to disrupt marital, sibling and family relationships (Carter et al., 2000; Cohen et al., 1988; Cooper et al., 2003), few studies have systematically explored the impact of TS on the mother-child relationship (MCR). Understanding the impact of TS on this relationship is important given the likelihood that mothers will assume the burden of care for their TS diagnosed children (Coffey, 2006). A secure mother-child relationship is also critical to optimal child and adolescent wellbeing and development (Allen & Land, 1999; Kerns, 2008; Laible, Carlo & Raffaelli; 2000; Papini & Roggman, 1992; Sroufe, 2005). The aim of the current research, therefore, was to explore the quality of the mother – child relationship in the context of TS. As attachment theory has emerged as the dominant model within which close relationships are examined, it was adopted for the current study.

Attachment theorists have identified classifiable styles of attachment based on the child's observable behaviour during interactions with their primary caregiver (most frequently the child's biological mother). It is hypothesised that during these

interactions, children develop internal working models of self and relationships, which guide their behaviour and expectations in future close relationships (Ainsworth, 1989; Ainsworth, 1982; Bowlby 1982; Hazan & Shaver, 1987; Kerns, 2008; Main & Cassidy, 1988). The reciprocal behaviours of the mother have also been observed. Secure attachment is dependent upon the availability of the mother to consistently provide attachment functions such as a secure base from which the child can explore the world, and safe-haven and comfort in response to the child's distress (Ainsworth 1982; Bowlby 1982; Main & Cassidy, 1988) (Ainsworth, 1982; Bowlby, 1982). Secure attachment is also contingent upon the mother's positive representation of both her child and her role as a willing and competent caregiver, in addition to the ability to respond sensitively to the needs of her child (Ainsworth 1982; Bowlby 1982; George & Solomon, 1989; Main & Cassidy, 1988; Main et al., 1985; Solomon & George, 1996).

Conversely, insecure attachment is associated with controlling, over or under involved, unresponsive, neglectful, inconsistent or abusive caregiving (Ainsworth, 1989; Bowlby 1982; Hazan & Shaver 1987; Main & Cassidy 1988). Insecure mothers may view their child, and their role as caregiver, negatively or ambivalently, and display insensitivity towards the child's emotional distress and needs (Main & Cassidy, 1988; Solomon & George, 1996).

Although understudied, research has linked the impact of the diagnosis of a child with a developmental or neurological disorder with an increased risk of insecure attachment, and that this appeared to be related to higher levels of parental grief and stress (Barnett et al., 2006; Oppenheim et al., 2009). This is consistent with attachment theory, which argues that significant stress increases the risk of insecure attachment (Bowlby, 1982; Sroufe, 2005). The current study therefore hypothesised



that the emotional distress associated with a child's diagnosis of TS, and the stress accompanying life in its aftermath, would threaten to the security of the mother-child relationship.

The goal of this novel and exploratory qualitative study was to gain detailed insight into the lived experience of biological mothers of youth with TS. Interviews were conducted to explore the impact of TS on several key aspects of the mother-child attachment relationship including the mother's representation (internal working models) of her TS diagnosed child, and her roles and functions within the MCR. The threats TS posed to the mother's ability to serve as her child's primary attachment figure, and the mother's ability to provide safe-haven and comfort in response to her child's distress were also investigated. It was hoped that findings would enhance understandings of the needs of mothers and their TS diagnosed children, and inform the development of improved psychological interventions and supports.

## **Methods**

### **Participants**

Participants included 22 biological mothers of youth formally diagnosed with TS, aged between 7-19 years (mean age = 12, 20 male and 2 female; Modal age of TS diagnosis = 7 years). The majority of the mothers were currently partnered (90%), and widely geographically distributed. Formal comorbid diagnoses were reported for 21 children. These included OCD (n = 6); obsessive-compulsive behaviours (n = 6); anxiety (n = 6); learning disorder (n = 6); ADHD (n = 4); oppositional defiant disorder (ODD) (n = 2); impulse control difficulties (n = 2); Asperger's syndrome (n = 2); autism (n = 2) and depression (n = 1). In order to facilitate participant disclosure and trust, collection of demographic data was limited.

## **Measures**

A series of open-ended questions grounded in attachment theory were developed. These were refined following review by a supervisory academic colleague prior to implementation. Sample questions included:

“How would you describe your child?”

“How do you feel when your child is upset?”

“How did you feel when your child was diagnosed with TS?”

“How did diagnosis affect your relationship with your child, if at all?”

## **Model for exploring attachment**

A model for exploring the security of mother-child attachment for dyads in the study based on the interview data is presented in Figure 2. In summary, maternal representations of the child were explored by constructing narrative descriptions for each child based upon their mother’s words. These were augmented by findings regarding the mother’s general emotional response to her child. Maternal internal working models of the MCR were explored by analysing each mother’s description of her MCR, and her roles and functions within this relationship. Threats posed to the security of the MCR associated with TS were also identified. The mother’s emotional (positive, ambivalent or negative) and behavioural response (modes of comfort classified as active or passive) to her child’s distress were employed to explore the mother’s availability to perform the key attachment functions of “safe-haven” and “comfort” for her child. Finally, the mother’s perception of change in the quality of her MCR post diagnosis was explored.

## **Procedure**

A purposeful sample of mothers of children and adolescents with TS was recruited via email invitation issued by national TS support groups, with the first 22 volunteers included in the study. As part a larger study of the impact of TS on quality of life and key attachment relationships, semi-structured interviews were conducted by telephone following informed consent for participation and recording. All audio files were transcribed verbatim for the purposes of data coding and analysis. Confidentiality was maintained, and the study was conducted with the approval of James Cook University Human Research Ethics Committee, in compliance with the Helsinki agreement. All participants were offered counselling support, however none was required. No inducements to participate were offered.

## **Data coding**

An eclectic approach to data coding was employed, using both a deductive approach to content analysis and an inductive approach to detect emerging themes (Miles & Huberman 1994; Strauss & Corbin 1998). The *a priori* major theme entitled “the Mother-Child Relationship” was established prior to coding. An initial round of coding was then conducted, with multiple readings of the transcripts undertaken to identify major categories and subthemes. Line- by-line manual coding of all transcripts then continued over a period of six months in an iterative process, and a data dictionary was written.

Data was further reduced by coding maternal emotions using Parrot’s (2001) Classification of Emotions System, a tree structured list of primary (Love, Joy, Sadness, Fear, Anger), secondary and tertiary emotions. These were then

dichotomised as positive (Love and Joy) and negative (Sadness, Fear and Anger-Shutdown) emotions based on the recommendations of Laros and Steenkamp (2005).

A supervisory academic colleague conducted an initial review of the data coding and data dictionary, resulting in agreement. Third party trustworthiness checks of the final coding system were conducted by two trained coders, blind to the purposes of the study, on three randomly selected cases. Percentage agreement was high and Krippendorff's Alpha (Krippendorff, 2004) results indicated highly significant levels of inter-rater agreement ( $p < 0.01$ ). An audit trail was maintained. Maternal narrative descriptions of each child were constructed from the mother's words. Member checks of narratives could not be conducted due to the sensitive and potentially disconcerting nature of the subject matter. Frequency data and percentages were generated by cross tabulating responses.

## **Results**

### **Maternal Representations of the child with TS**

Findings relating to the mother's representations of their child with TS, frequency data for responses and illustrative quotes are summarised in Table 19, and a sample narrative is available in Appendix A (All narratives are included in Appendix C of this dissertation). Findings indicated that the majority of the mother's narrative descriptions of their child were positive. Findings for the mother's affective response to the child indicated that all participants expressed positive emotions in addition to high rates of negative emotions, the most prevalent being anxiety (90%).

### **Mothers' descriptions of the MCR- Internal working models of the MCR**

Findings relating to mothers' internal working models of their MCRs, frequency data and illustrative quotes are presented in Table 20. Quotations in each table presented are widely sourced from all participants in the study. Eleven dominant themes emerged, representing the participants' descriptions their MCRs, and their roles and functions within this relationship. Findings provided detailed insight into complex and distinctive MCRs that were to a large extent shaped by the youth's diagnoses.

### **The Impact of Diagnosis of TS on the MCR**

Findings also revealed that half of the participants (50%) reported a significant change in their MCR on diagnosis of TS. *“Everything fell into place, we had more understanding and acceptance”*. *“I think it drew us in, it changed (the MCR sic) in the sense that I think I was more protective of him and more aware of his needs...”*

Remaining mothers noted that the relationship did not change with diagnosis and had always been close. This was attributed to the child's pre-existing comorbid disorder *“He has always a needy, anxious child...poor little thing...he was always anxious. We thought he was shy. The diagnosis of TS didn't change that.”* or to the mother and child sharing personal characteristics *“he really gets me and I really get him”*.

### **TS Specific Threats to the Security of the Mother-Child relationship**

Four major themes (with multiple subthemes) emerged from this analysis. Each represented an adverse emotional experience that mothers encountered within the MCR, and each was related to their child's TS (presented in Table 21). These included maternal fears associated with their child's diagnosis; conflicts and ruptures

within the MCR associated with TS; maternal self doubt regarding her adequacy as a parent in the context of TS; and high levels of stress, burden and exhaustion accompanying her caregiver role in the context of TS.

**Mother as a source of Safe Haven / Source of Comfort in response her child's Distress.**

Findings regarding the mother's emotional and behavioural response to her child's distress are summarised in Table 22.

***i. Emotional Response to Child's Distress.***

All participants (100%) expressed positive emotions in response to her child's distress, however a large majority also commonly expressed negative emotions. A case-by-case analysis revealed that more mothers experienced predominantly negative emotional reactions to their child's distress, when compared with their general emotional response to their child.

***ii. Behavioural Response to Child's Distress.***

The mothers' modes of behavioural response to their child's distress were identified and classified as being "active-positive" or "passive-negative" modes of comfort. The majority "actively" responded to their child's distress, with the great majority relying upon "Talk" as their primary mode of comfort provision. The case-by-case analysis determined that active –positive responses were associated with positive maternal emotional responses to the child's distress, and higher levels of negative emotions with linked to the mother's "passive-negative" response.

## Discussion

Findings from this exploratory study revealed the complex relationships participating mothers share with their TS diagnosed children, the manner by which TS shapes maternal roles and functions within the MCR, and the many threats that TS poses to the security of the MCR.

Maternal descriptions of the MCR revealed internal working models that were greatly influenced by the youth's TS (Table 20). The majority embraced their role as the youth's caregiver, despite viewing this task as challenging. Participants universally described their relationship as uniquely close and qualitatively different from other MCRs. This "special" relationship was based upon a level of empathy, acceptance and understanding of the child that the participants believed only they were capable of providing. This was reinforced by the mothers' perception of low levels of support, acceptance and knowledge of TS, a common experience of parents of children with TS (Collins, 2005; Davis et al., 2004; Dedmon, 1990).

An unexpected finding was that diagnosis of TS appeared to increase, rather than decrease, the security of mother-child attachment. Mothers attributed this to their enhanced compassion for their child, and enlightenment regarding the child's increased needs for support following diagnosis. This is consistent with prior research identifying maternal insight and acceptance of a child's disorder with the ability to maintain secure attachment (Barnett et al., 2006; Oppenheim, Koren-Karie, Dolev & Yimiya, 2009). Whilst not assessed in this study, increased security of the MCR may also reflect the successful resolution of any grief that participants may have experienced in response to their child's diagnosis.

Another key to understanding the mother's internal working model of the MCR, and her willingness to adopt the role of caregiver, were her perceived roles and

functions within the relationship. Consistent with anecdotal reports in the TS literature (Packer, 1997), almost all mothers in the sample emphasised their position as the child's protector, advocate and defender. The great majority also prioritised their role as their child's emotional adjunct, in addition to assuming the task of acting as the child's "interpreter", ensuring that everyone (including the child) had an understanding of TS and associated behaviours.

As a group, the great majority expressed high levels of positive affective responsiveness to their child, in addition to a willingness and commitment to provide sensitive caregiving, protection and comfort in times of distress – attachment functions that are necessary for developing and maintaining secure attachment (Ainsworth, 1989; Bowlby, 1982; Kerns, 2008). However, the study also revealed the complex nature of the relationship between mothers and their TS diagnosed child. Mothers universally expressed compassion and empathy for their child's distress, and responded actively and positively to provide comfort, the most common mode being to "Talk" the child through their difficulties (Table 22). Conversely, several participants also reported experiencing highly negative emotions, which resulted in occasional passive maternal responses such as detachment, immobilisation and withdrawal from the child.

Similarly, whilst the majority of the mothers' narrative descriptions of their TS child were predominantly positive, almost a third were "ambivalent" (Table 19). Ambivalent views of the child appeared to be a consequence of fluctuating levels of stress and distress mothers experienced within the MCR, usually in response to negative aspects of their child's symptoms and behaviour. As expected, the study identified many threats to secure MCRs associated with TS (Table 21). These



contributed to adverse emotional experiences within the MCR, which many described as a “rollercoaster” of rapidly shifting and mixed emotions.

Relationship ruptures were most frequently attributed to child’s aggressiveness, particularly in response to episodic rage, a finding that has also been reported by prior researchers (De Lange & Olivier, 2004; Dooley et al., 1999; Robinson et al., 2013). Moodiness, and the desire for autonomy accompanying adolescence, was another source of conflict, as were unanticipated or difficult to understand changes in the child’s behaviour (such as the onset of coprolalia, impulsive or self-harm behaviours). Although these relationship ruptures were generally transient, several mothers described deterioration in the MCRs as their child’s challenging behaviours became more persistent and severe. Such disconnections inhibited the mother’s ability to respond and care for her child with consistency, an essential feature of secure attachment relationships (Ainsworth, 1982).

Almost all participants reported feelings of sustained stress and anxiety associated with their caregiver role, often in combination with a struggle to maintain a sense of optimism and hope. A significant minority also described being “over-vigilant”, which for some was an appropriate response given the seriousness of their child’s psychopathology. This was particularly so for children exhibiting self-harm, aggressiveness, impulsivity, very severe tics or multiple comorbid diagnoses. As predicted and consistent with previous research, mothers in the current study also reported being overburdened by their caring duties and advocacy roles in the context of TS (Collins, 2005; Dedmon, 1990; Packer, 1997; Stern et al., 2005). This finding is important, given that overburdened mothers of children with a disability have been found to provide less supportive and sensitive parenting (Button et al., 2001).

Furthermore, increased maternal burden has been associated with impaired family functioning in prior TS studies (Carter et al., 2000; Cooper et al., 2003; Wilkinson et al., 2001).

Several previously reported pervasive fears, anxieties and self-recriminations were also noted (Packer, 1997; Walkup, 1999). These included concern for the future, difficulty understanding or tolerating the child's pathology, and remorse for pre-diagnostic parenting. These appeared to erode the mother's perception of herself as a competent parent. All of these negative emotional experiences placed mothers at significantly increased risk for depression and anxiety. Furthermore, participants may already be at increased risk for psychopathology due to the genetic links between TS and common comorbid disorders (Eapen & Črnčec, 2009). These findings, therefore, have serious implications for both maternal wellbeing and the quality of the MCR. Maternal psychopathology, and depression in particular, is strongly associated with insecure mother-child attachment (Wan & Green, 2009).

The high rate of comorbidity in the study, which complicates understandings of a child's behaviour (Cooper et al., 2003; Walkup, 1999; Wilkinson et al., 2001) and exacerbates parental stress (Carter et al., 2000; Lee et al., 2007; Robinson et al., 2013; Woods et al., 2005), may also have added to maternal distress. Mothers in the study also reported a lack of social support, which buffers against stress (Green et al., 2011), enhances parenting ability (Andresen & Telleen, 1992), and reduces caregiver burden in mothers of children with TS (Schoeder & Remer, 2007).

An important finding from the current study was the emergence of maternal over-involvement and emotional enmeshment as potential threats to the security of the MCR. A small majority of participating mothers appeared to display some signs of involvement with their children that went beyond a level that might be considered to

be “age appropriate” in a contemporary western cultural context (Rothbaum, Rosen, Ujjié & Uchinda, 2002), at least on occasion. Many strove to maintain close mother-child contact and found it difficult to encourage their child’s independent social behaviour. This is an important finding given that maternal over-involvement has been closely associated with insecure attachment (Feldman, 2010). It is thought that maternal over-involvement limits sensitive and available caregiving, and inhibits the developing child’s autonomy and individuation from the mother (Button et al., 2001). Furthermore, a small minority of mothers in this study described blurring psychological and behavioural boundaries between mother and child. This can have adverse outcomes for young people, including an increased risk of defiance during adolescence, disrupted social interactions, low cognitive competence and substance use (Feldman, 2010).

In prior TS research, the presence of maternal over-involvement, overcompensation and overprotection have been attributed to a mother’s lingering guilt regarding her pre-diagnostic parenting, or to anxiety regarding her parenting ability (Dedmon, 1990; Walkup, 1999). The current study, however, has revealed the complexity of this feature of the MCR in the context of TS. Fear for the child’s future, as well as increased symptom severity, have been found to foster over-involved maternal behaviour in prior research (Button et al., 2001). Many mothers in the current study expressed explicit fears for their child’s futures, and many of the youth in the study exhibited complex pathology and serious tics, at least on occasion. The intimate roles that present participants assumed within the MCR - the child protector, advocate, emotional adjunct and interpreter- perhaps posed the most obvious threat to the maintenance of appropriate mother- child boundaries. Furthermore, the need to adopt multiple roles in the absence of adequate professional support also increased the

risk of maternal over-involvement for many participants. Balancing the adolescent's need for continuing support and increased autonomy posed a particular challenge for mothers of older youth. Finally, the social isolation reported by the majority of the mothers in the present study intensified the focus upon the MCR and the nuclear family.

Several limitations of the study must be considered when reviewing the current findings. These include the use of the novel methodology designed to explore attachment in the current research. Not all characteristics of the attachment relationship were targeted for exploration, and although a large sample for a qualitative study, the ability to generalise the findings to the wider TS community is restricted. As with all qualitative research, the researcher and participants may have introduced bias, although efforts were taken to reduce this risk.

Although the study achieved its' goals, findings would benefit from replication in future research employing standardised methodology to assess attachment. This study also pointed to additional variables which may be included in future research; demographic differences, length of time since diagnosis; child-centred factors such as TS severity and comorbidity; and mother-centred factors including maternal grief, stress, coping styles and psychopathology.

In conclusion, the current findings suggest that TS plays an important role in determining the quality of the mother-child relationship, and warrants further investigation. Given the crucial role that mothers play in the care and welfare of their diagnosed children, these insights may be employed to tentatively inform clinical interventions and services designed to assist mothers and young people with TS.

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There is no potential conflict of interest for any authors arising from this research

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**Table 19.** Mother’s General Emotional Response to the Child & Classification of Emotional Responses & Maternal Narrative Representations (Positive, Negative, Ambivalent).

<b>Mother’s General Emotional response to the child</b>	<b>Frequency of Responses</b> percentage of sample (%)	<b>Examples</b>
• Positive	Maternal Love, Joy, Pride, Relief- (22; 100%)	<p>“He’s got incredible energy and love and compassion. He’s so intuitive and insightful; it’s quite amazing”</p> <p>“He is caring and giving and people love him... it used to bother me (his bizarre symptoms) but now all of that is completely invisible to me. I don’t care what he’s doing!”</p>
• Negative	(Sadness (19; 86%) Sympathy (12; 54%) Neglect (6; 27 %) Disappointment (13; 59%) Suffering (15; 68%) Shame (13; 59%) Horror (17; 77%) Nervousness (20; 90%) Confusion (10 45%) Rage (9; 41%) Irritation (15;68%) Shutdown (5; 23%)	<p>“Oh I just feel really sad about it for him. I felt sorry.... I just wanted to cuddle him and not let him go”</p> <p>“I get very anxious at times...have meltdowns “...you don’t want him to try to deal with it on his own...I probably worry about it more than he does”</p> <p>“..one episode a friend told me about, was when the kids stopped taking the tram with X because he was staring at them, and it ‘weirded’ them out. That hurt me, because he’s such a lovely kid”</p> <p>“It’s so frustrating ...he’s got so much potential...but it’s so hard to get him through school (Gifted IQ 148)!” “He can just frustrate the hell out</p>

of me and we have some horrific arguments!”

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**Overall Classification of  
expressed emotions**

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- Predominantly Positive (5, 23%)
  - Ambivalent - Mixed (14, 64%)
  - Predominantly Negative (3, 14%)
- 

**Mother's Narrative  
Representations of  
Child**

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- Predominantly Positive (12, 54%)
  - Signs of Ambivalence – Mixed emotions (7, 32%)
  - Predominately Negative (2, 14%)
-

**Table 20.** Maternal Internal Representations of the MCR- Descriptions and Roles and Functions.

Themes	Response Frequency (%)	Illustrative Quotes
“Close”	22 (100%).	“Well I think we’re closer, (than with sibling sic), more in-tune, more switched on...but when I say that, he’s also like that with me. I have that with him- he’s got to read me too and he knows (me sic).”
Mother as “Lioness” protector, advocate, defender	21 (95.4%)	“I’m more protective of him (than siblings sic) and every now and then there was um a different teacher comes in and things are not working out so I just went to the school and said look you can’t do that with X. I find it extremely difficult to leave him alone”
Mother as Child’s “Emotional Regulator”	19 (86.4%)	“When he’s upset, I’ve just worked out heat packs and massage oil and soft music and fish tanks and talking about stuff and al that ‘til your black and blue in the face (giggling). When he’s really bad (his tics sic) like he was really angry so I got him to go up to the shed and punch a punching bag and he got stuck into it. I go to all these lengths to help him.”
Overinvolved/enmeshed	13 (59.1%)	“...but if I ‘over mum’ him also that wouldn’t be good. I have to watch myself also that I give him a good balance, to give him the freedom he needs, but at the same time to be there when he needs me. So I don’t want to over mum him...but it’s hard,

		and I do ‘over-mum’ him...’& “My heart is broken if he is upset...I know immediately if he’s upset and I just talk to him...I can’t, I don’t ignore him. If he’s upset I have to communicate with him- I have to or it will kill me.”
Mother as Child’s “Interpreter”	13 (59.1%)	“It was really hard to get that message through at the time, and then everybody else in the family, they all have to go through it, and even now they’re all at different levels of understanding (re the child TS sic).”
Mother at “Wits End”	11(50%)	“Well I used to get upset but now I’m at the point where I’ve just had enough. I used to comfort him...but now he can be so horrible. He’s slowly trying to ruin all of our relationships. As a mum I keep saying surely it’s got to ease...oh I just feel like I’m having a breakdown every day!”
Mother and Child on an “Emotional Rollercoaster”	10 (45.4%)	“He’s really such a good kind, and he tries so hard...but he can be moody and grumpy and its really so hard at times...”
Striving for a “Normal” relationship	6 (27.3%)	“ I just keep it like any normal family...”
Mother and Child “United” (against the world)	5 (22.7%)	“He just really gets me and I really get him...and its doesn’t matter (what everyone else does sic)”
TS as a “gift” for mother and family	4 (18.2%)	“It’s (TS sic) been a gift for us in some ways... I sometimes I look at my husband and I am so proud...there he is educating everyone about TS” “his sister is so supportive and she’s really grown up (because of her role in helping sic) she’s great...”

“Atypical” Mother Child relationship	2 (9.1%)	“We are just not like other mums and daughters you know?”
Change in MCR relationship since diagnosis.	Yes (10; 50%) No (10; 50%)	“We had more tolerance I think, and more understanding of what was happening in his life (After diagnosis sic)... “He needed me more than his brother.” “Everything
Recent Change	(3; 15%)	fell into place, we had more understanding and acceptance”. “I think it drew us in...he got, yeah it changed a bit in the sense that I think I was more protective of him and more just aware of his needs...”

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**Table 21.** Direct Threats to Mother- Child Attachment Associated with the Child's TS.

Themes and subthemes	Response frequency (% of sample)	Illustrative Quotes
<i>Maternal Fears Regarding TS</i>		
Dependence of the child	13 (59%)	“He needs to be able to learn to cope for himself...I think in a lot of ways I’m probably harder on him because I know that he’s going to find it a lot harder in his life and I think you know I want him to be able to cope and be independent.”
Future of the child	16 (72%)	“You don’t know how it’s going to affect his life, (TS sic) is he going to get married and have kids and have a normal job?”
Future of the MCR relationship & Mother’s future	8 (35%)	“It’s like oh my god, what am I going to do when he’s grown? I’ve only got a couple more years and I’m going to have to find me completely and figure out what I want to do”.
Being over-vigilant & constantly anticipating disaster	9 (41%)	“that’s my fear, that they’re (tics sic) going to come back again and I’ve seen how bad they can be”. “I’m very worried because of bipolar running in the family, so we’re on the watch for that as well. We just don’t know what’s beyond...”
<i>Conflict and ruptures within the MCR</i>		

<i>associated with TS</i>		
The child's aggressiveness	16 (73%)	“As he's getting older he's getting angry quicker, you know hitting walls, and he smashed all his own stuff and he got really, really furious...he was screaming at me and at one stage I though he was going to hit me, I could see it in his eyes
Behavioural change accompanying developmental transitions	15 (68%)	He's an aggro as a teenager, lots of temper tantrums, not his usual self". “He's pulling away from me now. He's becoming more distant and I'm putting that down to coming into puberty.”
Unanticipated or difficult to understand changes in the child's behaviour	10 (45%)	“ it was hard (onset of coprolalia sic), especially for my husband- it led to fights”
<i>Maternal Self Doubt</i>		
Mother confused (TS, Co-morbid signs and symptoms or usual child behaviour)	16 (73%)	“At the moment we are just dealing with teenage things ...sometimes it's hard to differentiate between just straight out teenage attitude as opposed to him not coping and sometimes it takes us a few days and you think, hang on a sec, that's a new tic...ahhh that's what it is!”
Neglect of siblings	8 (36%)	“I try hard to keep it even, because they compete for my love and attention” “I know that's not fair but it's just what happens I suppose”
Out of her depth (parenting a child with TS)	15 (68%)	“I haven't gone down to the school and done my 'splurb' (TS education sic) this year because I thought that's maybe why he does have friends, but I don't

		know if that's the right thing... He's on medications, and I'd rather he was not, I just don't know what to do..."
Guilt, Shame & remorse by pre-diagnostic parenting	15 (68%)	““We thought he was trying to get our attention and being irritating so we kept saying would you stop doing that and of course I mean you feel so guilty afterwards...”
<hr/> <i>Stress / Exhaustion/Burden</i>		
Stay strong, the need to stay strong and model calm for their children despite their own negative feelings	12 (55%)	“I think we cope pretty good. I try not to let him see me get upset or anything.” “I try to stay calm because I know when I lose it, it becomes worse.” “Sometimes I feel very sad and emotional just thinking about it...’gotta keep pulling myself together”
Burden of care	17 (77%)	“...my husband is great you know... but I'm the one that does it all really with X(son sic)... we made that decision”
Exhausted/struggling/stressed	20 (91%)	“You can't even think about it (the child's TS sic) but you do, you have to keep going. I mean I've had times where I was actually, got very anxious”
Social isolation/ alone to work things out	19 (86%)	“I'm the one...I get frustrated sometimes because I've got all the knowledge on it (TS sic) but it sometimes gets to the point that I get overloaded or depressed and have to pull back” “Sometimes it's just me and him (son sic) ”
<hr/> <i>Mother-Child Disconnect</i>		
	4 (18%)	“I used to get upset (her daughter's behaviour sic) but now I'm at the point where I've just had enough”

**Table 22.** Mother’s Emotional & Behavioural Response to her Child’s Distress

Mother’s Emotional Response to Child Distress	Frequency of Responses percentage of sample (%)	Illustrative Quotes
• Positive	Love, compassion, empathy (22;100%)	Respondent M. “ ...poor little thing...it’s his oversensitivity and I think he’s worn out all the time. He’s always using more energy just with all his tics and worries, (it’s why he’s sic) grumpy, frustrated and moody”
• Negative	Sadness (20; 90%) Fear (20; 90%) Anger (17; 77%) Restrained or effort to suppress distress (15; 68%) Blunted /overwhelmed shutdown (9; 41%)	<p>“Oh it’s pretty heartbreaking to watch isn’t it... oh it’s awful...”</p> <p>“..I feel exactly the same way I think (as her son does when he is distressed sic)..you know I get anxious I think and I start to feel a bit panicky and worried.” ....</p> <p>“I cry... I just stand there saying well I can’t do anything for you! .</p> <p>“ It’s hard but I try not to let him see it (mother’s distress and concern sic”)... Yeah it’s not easy at the best of times!</p> <p>“ I just went to bed!” ... “and it doesn’t matter what you say doesn’t help, it’s not good enough (when mother tries to help son with his obsessiveness sic)” ... “feel like I am going insane, can’t stand it (when he is distressed by his ticcing sic)” .“I feel like my brain’s exploding!”</p>
Mother’s Behavioural Response to		

## Child's Distress

- 
- **A Talk** (21; 95%) “...we have a lot of deep talks, sometimes I’m more of a talker...I keep giving him the positive mind concept...and I’m working on that so that he starts to get the idea well having TS isn’t a problem”
  - **A Mother provides and models “calm” and “strength** (16; 73%) “I hope I am instilling in her a sense that she can do anything...we accept it, it’s here to stay (TS sic) and essentially we just roll with it”
  - **A Physical Comfort** (13; 59%) “He still asks for a hug when no-one’s around (when he’s upset sic)”. “ Oh he is very cuddly and needs a lot of that physical attention”
  - **A “Mum to the rescue”** (13; 59%) “ok, yeah...I’d better go and save everyone.....he knows he’s got it, and he knows we know how to deal with it - we’ve got lots of little strategies and it’s just on-going learning.” “We just take it minute by minute, a day at a time, hour by hour and see how we go...I’m constantly using high level crisis management skills, assessment skills...”  
 “But there’s lots of little tools out there, it’s just that you’ve really got to think left of center”
  - **A Distraction** (6; 27%) “We try anything to help him take his mind off it all...”
  - **A Empathizing with the child** (5; 23%) “ I try to understand you know what it is like and tell him so...”

- **P** Logical Detached (2; 9%) “I just try to work out what’s going on...and just get on with it...”
- **P** Shutdown-immobilized (4; 18%) I used to do everything...try everything to help...but it got so bad now I just gave up left her alone.” “He’s just driving everyone away...and we just leave him to it”

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Key: **A**= represents an active maternal response. **P** = represents a passive maternal response

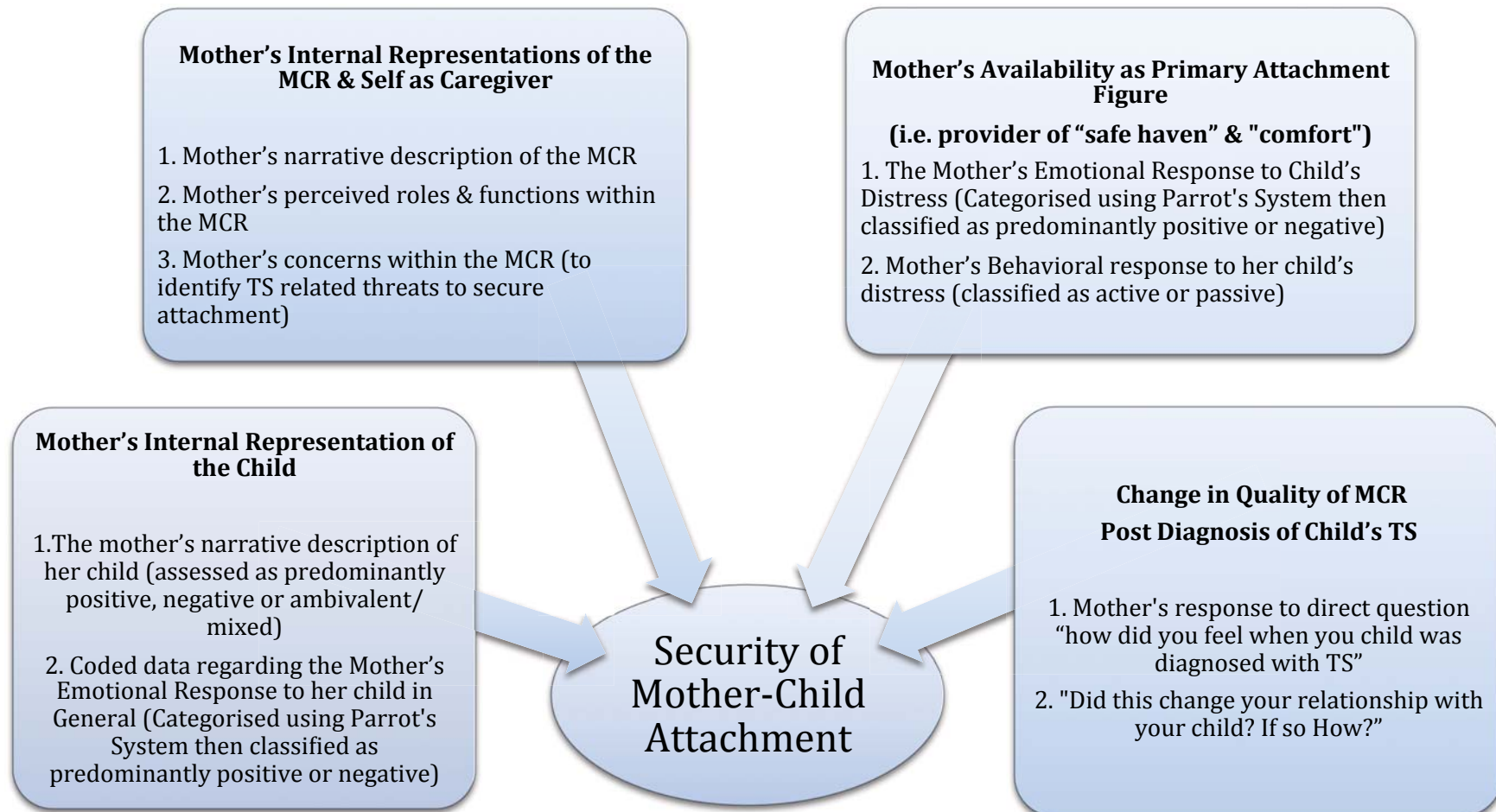


Figure 2. Methodology for Exploring the Security of Mother-Child Attachment Relationship (MCR) Based on coded Interview Data.

## Appendix A.

### Sample Narrative.

The following short narrative summarised the participant's description of her child and is based on the mother's own words, which are presented in *italics*.

#### Dyad 1.

Mother describes both positive and negative characteristics of her son. Difficulties that effected her representation of her child were primarily associated with changes accompanying adolescence, with conflict and ruptures being very difficult for them both to manage. There seems to be a developmentally appropriate transfer of attachment from parent to peers, however this is creating difficulties for both mother and son. The mother describes changes over the past 12 months in the youths increased aggressiveness and conflict within the MCR. Her tone of voice reflected variations in her feelings towards her son.

*“He gets on well with those older (adults) and especially well with younger kids... He’s lovely and got lots of young kids around him and he gets along fantastically with them all... I think he’s um look he’s one of those kids that um is really helpful and friendly. I love him to bits... I don’t think he’s got a nasty bone in his body at all. BUT...he has become so ‘aggro’ in his teens...we have big blues...he frustrates the hell out of me. Has OCD traits and can’t give up on things (These lead to distressing arguments with this mother, then afterwards he is very remorseful... “sorry mum”...). He has become a lot more aggressive I suppose in the last oh 12 months I would say but then like you know how much of it... I have often wondered you know how much of that is normal teenage behaviour and how much of it is the Tourette’s?”*



*“And I think it’s made even worse because his outside, outward appearance to everybody else is just you know loveable child, who’d do you know go out of his way to help you and he is generally like that, so he doesn’t have that (aggression) sort of behaviour for anybody else it seems to be reserved specifically for me. So people sort of look at me like I’m the fruit loop when I complain about him...(Mother feeling judged by others and isolated. Only she is privy to range of negative behaviour).”*

*“And he yeah he’s not afraid to sort of get out there. I think that well...he’s insecure in some ways but he’s quite confident in others. His self-esteem has been up and down the last few years...probably had enough positive people around him to sort of you know have some more ups than downs, but he does have and especially like when we’ve had arguments together. He has real concerns at school - he doesn’t mix a lot with them (his small group of friends) on the weekends...generally he has a fairly good group of friends and they’ve been pretty supportive but he has the odd one or two that like as I try to tell him they’re not really your friends. I think he’s going to have to be multi millionaire to have all the things he says he’s going to have in his life!*

All maternal narrative descriptions of the child are presented in Appendix C of this dissertation.

**SECTION 3 CHAPTER 7. “Differentiating between the impact of Tourette’s and comorbid diagnosis on quality of life and functioning”**

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**This Chapter reports on the study examining the differential impact of individual comorbid disorders on quality of life and functional outcomes for youth with TS in Study 1.**

### **Abstract**

Tourette's syndrome (TS) is a complex neurodevelopmental disorder consistently associated with adverse quality of life outcomes and functional impairment. High rates (80-90%) of comorbidity, however, greatly complicate outcomes for diagnosed youth and confound efforts to disentangle the impacts of TS from those attributable to co-occurring disorder. Results of the current correlational study suggest that the adverse effects of comorbid disorder are more restricted and disorder specific than indicated by prior research. Results also suggest that TS uniquely accounts for the impaired social functioning and increased peer relationship problems experienced by many diagnosed youth.

### **Introduction**

Tourette's syndrome (TS) is a childhood onset neurodevelopmental disorder characterised by the presence of motor and vocal tics that persist for a period of at least one year (American Psychiatric Association, 2013). Affecting as many as 1-3 per 1,000 of the paediatric population (Freeman et al., 2000; Robertson, 2008), the severity of TS varies widely and clinical presentation is further complicated by high rates of comorbidity, particularly in clinic-based populations (80-90%) (Cavanna, Servo, Monaco, & Robertson, 2009; Robertson, 2006a, 2006b, 2008). The most common comorbid diagnoses are attention deficit hyperactivity disorder (ADHD) and obsessive compulsive disorder (OCD), which are hypothesised to share aetiological links with TS occurring at the level of the neural substrate (Felling & Singer, 2011).

Others include non-OCD anxiety disorders, depression, bipolar affective disorder, disordered impulse control, intermittent rage, learning disorders, oppositional defiance and conduct disorders, and autistic spectrum disorders (ASD) (Freeman et al., 2000; Robertson, Cavanna & Eapen, 2015; Robertson, 2006a, 2006b, 2008; Termine et al., 2006).

TS has been consistently associated with poor quality of life (QoL) outcomes, and adverse psychological, behavioural, social and academic consequences (Conelea et al., 2011; Cavanna et al., 2009; Stern, Burza & Robertson, 2005). The current research reports on findings from a larger project involving the understudied Australian TS youth population (O'Hare et al., 2015; O'Hare et al., In Press). Results indicated that when compared with control group peers, youth with TS experienced significantly lower quality life and impaired functioning across all domains, in addition to increased psychopathology, behavioural and peer relationships difficulties. However, high rates of comorbidity confound efforts to disentangle the effects of TS and co-occurring diagnoses. This poses a serious challenge to clinicians and caregivers as they attempt to differentially diagnose and treat youngsters with complex pathology and multidimensional needs (Conelea et al., 2011; Eapen & Črnčec, 2009).

Quality of life research has determined that the presence of comorbidity predicts increased global impairment and adverse functioning across multiple domains, particularly by late adolescence and adulthood (Bernard et al., 2009; Cavanna et al., 2009; Conelea et al., 2011; Cutler et al., 2009; Eddy et al., 2011; Storch et al., 2007). The study of the impact of individual comorbid disorders on outcomes such as QoL has, however, been largely restricted to the roles of ADHD and OCD.

The current survey-based study, therefore, aimed to advance understandings of the differential effects of TS and comorbid disorders on quality of life (QoL) outcomes, and rates of psychological, behavioural and social dysfunction of diagnosed youth. It aimed to compare findings between individuals with “TS Only” and those with “TS Plus” a comorbid diagnoses, in addition to identifying the individual impact of each comorbid disorder included in the present study on measured outcomes. It was hoped that results would reveal those areas of functioning most adversely affected by TS, and those primarily impacted by co-occurring diagnoses. Such knowledge may benefit assessment, treatment planning, and interventions tailored to meet the variable, often complex needs of individuals with TS.

## **Methods**

### *Participants and Procedure*

A nation wide sample of 86 primary caregivers of youth (7-16 years of age,  $M=12.3$ ) formally diagnosed with TS volunteered to participate (Table 23). Recruitment followed advertising and invitation to participate, which was facilitated by the non-financial support of the Tourette’s Syndrome Association of Australia (TSAA). Response rate could not be calculated due to information deficits on the TSAA database. The research was conducted with the approval of James Cook University Human Research Ethics Committee (Approval number H4380). Written informed consent was obtained prior to completion of the postal survey.

### *Measures*

Parents were administered the “Australian Tourette Survey”, a pen and paper instrument assembled for the purposes of a larger controlled study of Australian youth with TS. Additional questions addressed demographics, formal diagnosis of comorbid disorder, family history of TS and comorbid disorder. Two psychometric measures relevant to the current study were included.

*Pediatric Quality of Life Inventory (PedsQL)* (Varni, Seid & Curtin, 2001; Varni & Limbers, 2009). A widely used 15-item measure assessed 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 to 100, with higher scores indicating better quality of life. Four subscales measure physical, emotional, social and school functioning. Reliability and validity of the PedsQL have been demonstrated (Chan et al., 2005; Varni et al., 2001; Varni & Limbers, 2009) and excellent internal consistency was determined for all scales in the current study (Cronbach’s  $\alpha = .86 - .92$ ).

*The Strengths and Difficulties Questionnaire (SDQ)* (Goodman, 1999): This is a widely utilised questionnaire consisting of 25 items across five subscales that screen for emotional symptoms, conduct problems, hyperactivity/inattention, peer problems, and prosocial behaviour. A total summary score is calculated by combining the 20 items assessing emotional, conduct, and inattention and peer problems. The SDQ has demonstrated robust psychometric properties and strong internal consistency (Goodman, 1999; Mellor, 2005), which was good to excellent for all scales in the current study (Cronbach’s  $\alpha = .75 - .87$ ), with the exception of the prosocial scale, which was poor ( $\alpha = .41$ ).

## Results

*Data screening and assumption testing*

Data were entered into a single SPSS 20 spreadsheet for analysis. Distance measures did not identify any outliers demanding deletion. Levene's test was conducted to examine homogeneity for all variables, and parametric analyses were conducted where data met the assumptions for normality. Due to the uneven size of groups associated with comorbid disorders, nonparametric Mann Whitney U tests were employed for comorbidity data. A per comparison critical significance level of  $\alpha = .01$  was applied to protect against family-wise error.

*Comorbidity.*

High rates of comorbidity were reported for youth with TS ( $N = 66$ , 73%). In order of decreasing frequency, parent reported formally diagnosed disorders included OCD ( $N = 38$ , 44.2%); Anxiety ( $N = 31$ ; 36.0%); ADHD/ADD ( $N = 28$ , 32.6%); Learning Disorder ( $N = 16$ , 18.6%) Impulse Control Disorder ( $N = 6$ , 7.0%); Autistic Spectrum Disorder ( $N = 4$ , 4.7%); Depression ( $N = 2$ , 2.3%); Conduct Disorder ( $N = 2$ , 2.3%) and "Other" unspecified disorders ( $N = 10$ , 9.3%). Of those with comorbidity ( $N = 66$ ), a majority was reported as having more than one additional diagnosis (67%).

*Impact of Comorbidity on Quality of Life and SDQ outcomes*

As presented in Table 24, Mann Whitney U tests determined that having a comorbid disorder was only associated with a significant decrease in global QoL, impaired emotional and school functioning, and increased emotional symptoms on the SDQ.

*Impact of Individual comorbid disorders on Quality of Life*

As presented in Table 25, Mann Whitney U tests revealed significant but restricted results for individual comorbid disorders. Significant associations were reported between ADHD and impaired School Functioning on the PedsQL and increased Hyperactivity/ Inattention on the SDQ; OCD and impaired Emotional Functioning on the PedsQL and increased Emotional Symptoms of the SDQ; Learning Disorder and impaired School functioning and global QoL on the PedsQL and Hyperactivity/ Inattention on the SDQ; Anxiety Disorder and impaired Emotional Functioning on the PedsQL and increased Emotional Symptoms on the SDQ; Autism to reduced prosocial skills on the SDQ and “Other” to Hyperactivity/ Inattention and reduced prosocial skills on the SDQ.

*Impact of Individual Disorder on diagnosed child and family.*

Mann Whitney U test did not reveal any difference in the impact Scale Score of the SDQ (which assessed level of distress and impairment for the child, and increased burden for the family) for youth with or without comorbid diagnosis. Only Autism and “Other” were significantly associated with increased distress and burden (Table 25).

**Discussion**

Present findings suggest that the adverse effects of comorbid disorder are more restricted and disorder specific than indicated by prior research. Furthermore, results suggest that TS uniquely accounts for impairment in the social functioning domain and the rates of peer relationships problems reported for youth with TS included in the current study.

As predicted, and in agreement with multiple prior studies, a high rate of comorbidity (73%) was reported for the current sample of youth with TS (Cavanna et



al., 2009; Freeman et al., 2000; Robertson, 2006; 2008; Termine et al., 2006). OCD, non-OCD anxiety disorders, ADHD and learning disorders were most frequently reported, whilst other commonly co-occurring disorders were found in very low numbers and as such, related findings will not be discussed.

In agreement with prior QoL studies, those with “TS Plus” a comorbid diagnosis experienced lower global QoL than youth with “TS Only” (Bernard et al., 2009; Cavanna et al., 2009; Conelea et al., 2011; Cutler et al., 2009; Eddy et al., 2011; Storch et al 2007). However, functional impairment associated with having a comorbid diagnosis in this study was restricted to the emotional and school domains. Comorbidity was not associated with an increased rate of physical or social impairment beyond that reported for all youth with TS. Restricted results were also found for the SDQ. Having a comorbid diagnoses was only associated with an increased rate of the youth’s emotional symptoms, however, parents did not perceive that comorbidity significantly increased the diagnosed youth’s level of psychological distress. In addition, and in contradiction to prior research, comorbidity did not elevate levels of perceived family burden (Wilkinson, Marshall, & Curtwright, 2008)

Findings for the impact of the individual comorbid diagnoses included in this study on QoL and the psychological, behavioural and social difficulties assessed by the SDQ were notable. Unlike prior studies that have associated comorbidity with a broader range of adverse outcomes (Bernard et al., 2009; Cavanna et al., 2009; Conelea et al., 2011; Cutler et al., 2009; Eddy et al., 2011; Storch et al., 2007), the current results revealed patterns of impairment that were highly restricted and logically related to each individual disorder (Table 3). Comorbid ADHD was, for example, only significantly associated with increased impairment in school functioning and increased symptoms of hyperactively and inattention identified by the

SDQ. This is as would be expected given the association between hyperactivity, inattention and learning. Similarly, learning disorders were logically associated with dysfunction in the context of school and an increase in hyperactive inattentive behaviours. This finding demonstrates the well-recognised relationship between LD and ADHD, which was prevalent in this sample (Pliszka, 2005).

Findings also indicated that youth with comorbid OCD and non-OCD anxiety disorders appeared to be particularly vulnerable to emotional difficulty. This is a finding of substantial clinical concern, given the increased risk of highly adverse outcomes associated with mood disorder in those with TS by late adolescence (Robertson, 2006b).

The current finding of no significant relationships between comorbid diagnosis and variability in social functioning or problems in peer relationships over and above those of youth with “TS only”, also varies from prior research (Bernard et al, 2009; Conelea et al, 2011). It may be hypothesised that these adverse social outcomes are a consequence of the unusual nature of TS symptomatology, which is supported by prior studies revealing the high rate of stigma, social rejection and lack of understanding that youth with TS frequently experience (Collins, 2005).

It may therefore be concluded that whilst having a comorbid disorder appears to increase the risk of adverse outcomes for youth with TS, these risks are domain specific. In order to optimally support youth with TS, it may be beneficial for clinicians, educators and caregivers to understand the specific risks associated with each comorbid disorder, and those associated with TS. Such information can be used to guide interventions and services targeted to the youth’s individual needs.

Limitations of the current study must be considered when reviewing these findings. These include the inability to imply causality due to the correlational nature

of the analyses, and the need to rely upon parental reports due to methodological constraints. Replication of the current findings in future studies that employ youth self reports and control for formal psychiatric evaluation are therefore strongly recommended.

### **Competing interests**

The authors declare that they have no competing interests

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**Table 23.** Participant Demographics ( $n = 86$ )

		<i>M</i>	<i>SD</i>
Child Age (years)		11.44	2.78
		<i>f</i>	<i>%</i>
Relationship to youth	Biological mother	77	90.6
	Adoptive mother	1	1.2
	Biological father	7	8.2
Place of residence	City	50	50
	Regional	20	23.8
	Rural-remote	14	16.7
Marital status Parent	Never married	1	1.2
	Married	72	83.7
	Separated/divorced	10	11.6
	Widowed	3	3.5
Family income	Low	8	9.8
	Low-middle	19	23.2
	Middle -above	55	76.1
Gender Child	Male	71	85.4
	Female	12	2.4
Ethnicity family	Caucasian	74	87.1
	A/TSI	2	2.4
	Asian	3	3.5
	Other	6	7.1
Siblings	Yes	76	88.4
	No	10	11.6



**Table 24.** Impact of TS Only versus TS Plus comorbid diagnosis (Parent Reported) on Quality of Life (PedsQL) and strengths and Difficulties outcomes (SDQ) for TS Group Youth

PedsQoL & SDQ scales	Mean rank	Mean rank	U	Z	p
	“TS Only”	“TS Plus” comorbid			
	(N= 23)	(N= 63)			
Emotional Functioning PedsQL	58.54	38.01	378.50	-3.39	<i>p</i> = .001
School Functioning PedsQL	55.52	39.11	448.00	-2.71	<i>p</i> = .007
Total QoL Score PedsQL	55.59	39.09	446.50	-2.71	<i>p</i> = .007
Emotional Symptoms SDQ	30.78	48.14	432.00	-2.88	<i>p</i> = .004

Note: higher PedsQL Scores = higher functioning. Higher SDQ scores = Increased difficulty

**Table 25.** Significant findings for Impact of Individual Comorbid Disorders (parent reported) on Quality of Life (PedsQL) and Strengths and Difficulties outcomes for TS Group Youth

PedsQoL & SDQ scales	Mean rank	Mean rank	U	Z	P
	No ADHD ( <i>N</i> =58)	ADHD ( <i>N</i> =28)			
School Functioning PedsQL	49.79	30.46	447.00	-3.81	<i>p</i> = .001
Hyperactivity /Inattention SDQ	36.13	58.77	384.50	-3.97	<i>p</i> = .000
	No OCD ( <i>N</i> = 48)	OCD ( <i>N</i> = 48)			
Emotional functioning PedsQL	54.52	29.58	383.00	-4.62	<i>p</i> = .000
Emotional Symptoms SDQ	35.02	54.21	505.00	-3.57	<i>p</i> = .000
	No Anxiety ( <i>N</i> = 55)	Anxiety ( <i>N</i> =31)			
Emotional functioning PedsQL	50.80	30.55	451.00	-3.63	<i>p</i> = .000
Emotional Symptoms SDQ	35.29	58.06	401.00	-4.09	<i>p</i> = .000
	No LD ( <i>N</i> = 70)	LD ( <i>N</i> = 16)			
School Functioning PedsQL	48.46	21.81	213.00	-3.87	<i>p</i> = .000
Total QoL PedsQL	47.49	26.03	280.50	-3.10	<i>p</i> = .002

Hyperactivity/Inattention SDQ	39.99	58.84	314.50	-2.74	$p = .006$
	No "Other"	"Other"			
	( $N = 78$ )	( $N = 8$ )			
Total QoL Peds QL	45.72	21.81			
Hyperactivity/Inattention SDQ	40.51	72.69	138.50	-2.58	$p = .010$
Prosocial SDQ	45.71	22.00	140.00	-2.60	$p = .009$
Impact Score SDQ a	36.64	59.38	112.00	-2.74	$p = .006$
	No Autism ( $N = 4$ )	Autism ( $N = 82$ )			
Prosocial SDQ	45.06	11.50	36.00	-2.67	$p = .005$
Impact Score SDQ b	37.33	69.50	24.00	-2.82	$p = .002$

Note: a = Impact score  $n = 77$ - No Autism  $N = 73$ ; other  $N = 4$ ) b. Impact score  $n = 77$ - No "other"  $N = 69$ ; "Other"  $N = 8$

Higher PedsQL Scores = higher functioning. Higher SDQ scores = Increased difficulty. U = Mann Whitney U

No significant findings for Impulse Control Disorder or Depression.

## **SECTION 4 CHAPTER 1. The Stability of Attachment in the Context of Tourette's Syndrome**

This chapter presents the qualitative findings emerging from Study Two ( $N = 22$ ), related to the final goal of the present research project, which was to explore the stability of attachment in the context of TS. Whilst an individual's attachment generally remains stable across generations, time and relationships (Ainsworth, 1997; Bowlby, 1982; Fraley & Davis, 2005; Hazan & Shaver, 1987; Sroufe, 2005; Sroufe et al., 2009), attachment may change in response to substantial stress (Mills-Koonce et al., 2007; Hill-Soderlund et al., 2008; Phelps, Belsky & Cronic, 1998; NICHD ECCR, 2005). The diagnosis of TS, and the challenges associated with life in its aftermath, were therefore conceptualised as major stressors with the potential to disrupt or change the quality of the attachment relationships of diagnosed youth.

In order to explore relationships between TS and change in attachment, the study tested the hypothesis that the security of the mother-child relationship would predict the security of the youth's peer relationships. It was further hypothesized that the mother's ability to form a secure attachment relationship with her TS diagnosed child would be related to her own childhood attachment history, and the quality of her current adult attachment relationship.

As no standardised method of assessing attachment was suitable for use within the current research design, a novel methodology based upon a conceptual model grounded in attachment theory was developed for the purposes of the study (Ainsworth, 1997; Bowlby, 1982; Hazan & Shaver, 1987). This provided a strong, developmentally orientated framework within which to analyse and interpret the emerging qualitative data.

### Estimating Attachment - Methodology

The reader is referred to the Methods section in Section 3, Chapters 3 & 6 for information regarding participants, procedure and data coding and analyses for Study Two.



**Figure 3.** Conceptual Map of the Transmission of Attachment Across Generations, Time and Relationships

The process of designing a methodology grounded in attachment theory for the current study began by developing a conceptual map modelling the transmission of attachment across relationships and time (Figure 3). This was followed by the development of a detailed methodology, which aimed to provide “estimates” of the security of attachment of the relationships under study on the basis of the findings emerging from the qualitative data analysis. This methodology is presented in Table 26.

*Estimating Maternal Over-Involvement in the MCR.*

Maternal over-involvement emerged as a notable theme during data analysis. The presence of maternal over-involvement was assessed for each participant by examining each mother's report of her roles and functions within the MCR, her emotional response to her TS child, and her mode of behavioural response to her child's distress.

**Findings***Estimates of the mother's attachment security*

A summary of the findings is presented in Table 27. Estimates of the quality of the mother's childhood and current attachments revealed a high rate of insecure childhood attachment (insecure = 74%) and a pronounced movement towards secure attachment in adulthood (insecure = 57%) for the group. A large majority (89%) reported adopting a different approach to parenting than the style they experienced as children.

*Estimates of mother-child relationship (MCR) security*

As presented in Table 27, the majority of mothers held representations of their child that were generally positive or mixed (86%), and were available to fulfill attachment functions of secure base-safe haven (86%). Results for the group revealed that 90% appeared to have secure MCRs, although 38% of these participants showed occasional signs of maternal over-involvement. Half of the mothers reported a change in the MCR on diagnosis. The data analysis also identified multiple roles and functions within the MCR, and threats to the quality of the MCR associated with the child's diagnosis. These were presented in full in Section 3, Chapter 6.

*Estimates of the security of child-peer attachments*

Results for the group are presented in Summary Table 27, with 41% of the group estimated as having secure peer attachment, 45% with insecure peer attachment, and 9% moving from insecure to secure peer attachment with maturity. Results of the case-by-case analyses for each mother-child dyad are presented in a summary in the Appendix D. Factors identified as impacting peer attachment were reported extensively in Section 3, Chapter 3.

*Hypotheses testing.*

Findings (illustrated in Figure 4) provide partial support for the stability of attachment style across time, generations and relationships. Many bidirectional changes in attachment are, however, also evident.

Maternal childhood attachment history did not strongly predict attachment style in future relationships. Whilst secure maternal childhood history perfectly predicted secure MCRs, over half of the mothers with an insecure childhood history developed apparently secure adult attachments, and 84% developed secure albeit complex relationships with their child (MCRs). Furthermore, having a secure MCR did not strongly predict secure peer attachment. Although 90 % of youth experienced secure MCRs, half of the youth in this study developed insecure peer attachments. Finally, a case-by-case analysis revealed that mothers with an insecure childhood history were more likely to demonstrate maternal over-involvement within the MCR. It was further observed that maternal over-involvement was associated with insecure peer attachment relationships.

## Discussion

As predicted, the current findings demonstrate the ability of TS to disrupt the security of the attachment relationships of diagnosed youth. The hypothesised stability of attachment across generations and relationships was, however, only partially supported. In agreement with attachment theory, a secure attachment style was transmitted across generations and relationships (Ainsworth, 1989; Bowlby, 1982; Button, Pianta & Marvin, 2001; Main & Goldwyn, 1984; Van Ijzendoorn, 1992) for the current sample of mothers and their TS diagnosed children, yet the results for insecure attachment were inconsistent. As illustrated by Figure 4, many bidirectional changes in attachment security were observed between the relationships in the study.

The present research also suggests several possible mechanisms by which change in attachment occurred within each relationship and the unique role performed by TS in shaping the quality of the MCR and the child's peer attachments. When interpreting these findings, it is important to note that the security of the attachment relationships under study is limited to estimations. These were derived from the methodology designed to meet the exploratory goals of this novel research project.

### *The Mother's Childhood and Adult Attachment*

The limited exploration of the mother's primary attachment relationship conducted for this study suggested a surprisingly high (75%) rate of insecure childhood attachment for the group. The adverse family of origin experiences described by many participants may explain this phenomenon. The mothers' accounts



were characterised by abusive, inconsistent or neglectful parenting, factors robustly linked to insecure attachment in the literature (Ainsworth, 1989; Cassidy, 1994; De Wolf & Van Ijzendoorn, 1997; Hazan & Shaver, 1987; Hess & Main, 2000; Main, 1996; Schuengel et al., 1999). Why so many mothers with apparently insecure attachment histories volunteered for this study may be explained by chance or unmeasured respondent characteristics. Alternatively, those with insecure histories may be more likely to seek the support of organisations such as the TSAA (the recruitment base for the current sample). Others may reach out for information and support in a manner consistent with the autonomous mode of coping described by many of the current participants.

The brief evaluation of the mother's adult attachment suggested that half overcame their insecure primary attachments to form apparently secure adult attachments (spousal) and a strong majority (75%) developed secure, albeit complex relationships with their TS diagnosed children. This shift towards security may have resulted from the participants ability to transfer attachment functions away from the unavailable mother to alternate attachment figures such as a father or older sibling (Seibert & Kerns, 2009), or to peers during childhood (Bowlby, 1982; Kerns et al., 2000; Mikulincer & Shaver, 2007; Nickerson & Nagle, 2005; Sroufe, 2005). Such relationships however were not explored in the current study. Change may also have been facilitated in the context of the close, supportive spousal relationships that many participants described (Roisman et al., 2001; Roisman et al., 2003). One prior qualitative TS study noted the benefit to parents of being able to rely upon a partner, particularly in regard to the shared distress of having a child with TS (Einarsdottir, 2008).

Finally, as discussed in Section 3, Study 6, change in attachment style may have been associated with the mother's ability to resolve the grief and loss resulting from her own childhood experiences (Bowlby, 1982; Main et al., 1985). In addition, those having experienced more enduring adverse circumstances were more likely to suffer enduring negative effects, including the preservation of an insecure attachment style. The current study, however, did not evaluate either the level of the mother's unresolved grief, or the seriousness of the trauma or neglect she experienced in childhood. Closer attention to the mother's past and current attachments may be suitable subjects for future studies.

#### *Mother-Child Attachment*

As illustrated in Figure 4, findings indicate that the overwhelming majority (90%) of participating mothers appeared to have secure MCRs, and willingly functioned as available attachment figures for their TS children, despite the fact that the majority experienced insecure childhood attachment. Several possible mechanisms associated with change in the security of the MCR were identified. Increased maternal empathy and commitment to care appeared to accompany the diagnostic event for half of the participants, thereby increasing the security of the mother-child attachment. This finding was thoroughly discussed in Section 3, Study 6 and will therefore not be repeated here.

The present study also identified a role for parenting style in the shift from insecure maternal history to security within the MCR. Aware of the damaging impact of their own mother's inadequate parenting, mothers reported adopting a positive and conscious approach to child rearing. Initial research has determined that the ability of the mother to reflect upon her experience as a parent, and her ability to cope with the

mixed feelings accompanying parenting, are important to the formation of secure MCRs (Slade, 2005; Slade et al., 2005). The current finding, therefore, indicates the value of considering the inclusion of parenting style in future TS studies, and in the context of therapy.

As hypothesized, the many stressors associated with parenting a child with TS threatened attachment security. As discussed in detail in Section 3, Chapters 5 and 6, the current research extends understandings of the multiple stressors experienced by mothers of children with TS. As hypothesized, mothers described transient, and in some cases persistent, breakdown in the MCR as a result of high levels of maternal fear, anxiety, stress and relationship ruptures. This is consistent with findings that high levels of sustained stress predict negative change in attachment (Green et al., 2007; 2011; Hill-Soderlund et al., 2008; Mills-Koonce et al., 2007; Phelps et al., 1998). In the current study, these stressors appeared to impair the ability of mothers to parent with continuity - a key factor in determining secure mother-child attachment (Berlin et al., 2008; Belsky, NICHD ECCRN, 2005; Waters et al., 2000; Weinfield et al., 2004).

Furthermore, research suggests that present participants with an insecure history may have been less able to cope with persistent stressors, and the ruptures and conflict experienced within the MCR, than their securely attached peers. Insecure mothers have been found to respond with higher levels of psychological distress to sustained stress than secure peers (Green et al., 2007; 2011; Hill-Soderlund et al., 2008; Mills-Koonce et al., 2007; Phelps et al., 1998), to encounter greater difficulty interpreting their child's affective state, and to be less able to help their child to regulate their emotions (De Oliveira, Moran & Pederson, 2005). Correspondingly, children of mothers with an insecure style have been found to respond less positively

to their mothers efforts to help them to deal with adversity than those with secure mothers (Abaied & Rudolph, 2010).

Another factor that appeared to effect negative change in the MCR was the role of maternal over-involvement. As discussed in detail in Section 3, Chapter 6, and briefly recapped here, a substantial minority of mothers described occasionally blurring psychological and behavioural mother – child boundaries deemed normative within a western cultural context (Feldman, 2010). The complex roles and functions mothers adopt within the MCR and the many fears and doubts that mothers in the study expressed (Minuchin, 1974; Rothbaum et al., 2002) were most likely to precipitate over-involvement. A mother's fear for her disabled child has been found to adversely affect maternal sensitivity and behavioural intrusion; particularly for those whose child has more severe pathology (Button et al., 2001).

Once again, mothers with a history of early insecure attachment may have been more vulnerable to becoming over-involved with their TS child. Researchers have found that mothers with an insecure history experience greater difficulty in resolving the grief associated with their child's diagnosis than secure mothers, and this results in insecure mother-child attachment (Marvin & Pianta, 1996). Whilst maternal insecure history did not predict insecure MCRs in the present study, it may have contributed to the mother's development of a slightly more anxious or overinvolved pattern of responding to her child (Marvin & Pianta, 1996). The findings of this study suggest the value of closely evaluating maternal over-involvement, and the factors that contribute to this phenomenon, in future TS studies.

*Peer Attachment.*

The current findings provide only partial support for the current hypotheses that the security experienced within the MCR would predict the quality of the youth's peer attachment relationships. Whilst almost all youth in the study enjoyed apparently secure MCRs, only half of the youth were estimated as having secure relationships with peers. As discussed in Section 3, Chapter 3 and therefore not repeated here, the results of the current research revealed the complex factors parents identified as either enhancing or impairing the ability of youth with TS to form secure peer attachments. The factor that appeared to influence the shift from a secure MCR to insecure peer attachment in the current analysis, however, was the presence of maternal over-involvement.

One interpretation of the finding that the security of the MCR was not a strong predictor of secure peer attachment is that the security of attachment experienced within the MCR is not the only mechanism by which mothers can affect the quality of their child's peer relationships. A metaanalysis of 200 quantitative studies revealed the small effect size for secure MCR as a predictor of secure peer attachment (Schneider et al., 2001). The authors (Schneider et al., 2001) suggested that other relative, cumulative or interactive maternal factors were liable to impact the quality of peer relationships. These included the mother's modelling of social behaviour, disciplinary practices and perhaps of most significance in the context of the tendency towards maternal over-involvement demonstrated in this study, the role mothers played in the regulation of their child's independence and contact with peers.

Overinvolved mothering is an issue of clinical concern. It has been linked to multiple adverse psychological, behavioural and social outcomes for the child (Feldman, 2010), and is believed to foster an insecure attachment style by limiting the developing child's autonomy and individuation from the mother (Button et al., 2001;

Minuchin, 1974; Rothbuam, Rosen, Ujiie & Uchinda, 2002). With the majority of participants in the present study describing the importance of their protective role, mothers may have variably prejudiced their child's ability to acquire the skills, opportunities, experiences and motivation necessary to interact optimally with peers. This has significant implications for children and teens with an overt and stigmatizing disorder. The development of strong social skills may be of particular importance in helping diagnosed youth manage the well-documented challenges and barriers to peer relationships that accompany TS.

### *Conclusion*

The findings of the current exploratory study suggest the benefit of understanding the impact of TS on the key attachment relationships of diagnosed youth, and the way in which attachment may either demonstrate stability or change across generations, time and relationships in the context of the child's TS.

The limitations associated with this research were discussed in Section 3, Chapters 3 and 6, and will also be noted in the limitations subsection of the following, final chapter. Replicating the current findings using standardised methods to assess attachment would be an important goal for future research.

The following final chapter will summarize and integrate the quantitative and qualitative findings from this major research project; discuss the implications of the results; include recommendations for practice and policy; and present a new, predictive quality of life model for youth with TS, based on the integrated findings of the current research. Finally, recommendations for psychological interventions aimed at improving the attachment relationships and outcomes for youth with TS will be made.

**Table 26.** Methodology for Estimating Attachment Security- Maternal Attachment Style, Mother Child-Relationship & Child -Peer Attachment

Process	Mother's Childhood and Current Attachment	Mother- Child Attachment (MCR)	Child-Peer Attachment
Step 1	<p>Mother's report of her childhood experience with her parents:</p> <ul style="list-style-type: none"> <li>• Secure = mother reports the presence of a positive relationship with her primary caregiver</li> <li>• Insecure = mother reports a history of abuse, neglect, inconsistency, or other adverse parenting-environment. Specifically: <ul style="list-style-type: none"> <li>– Mother's reported experience of secure base as a child (primary caregiver as available Y/ N)</li> <li>– Mother's reported mode of comforting when distressed as a child. Secure = Parental attachment figure used as source of comfort vs. Insecure = Autonomous i.e. self-comforting</li> </ul> </li> </ul>	<p>The mother's "representation" of her child, as reflected in the short narratives (Positive, Negative or Ambivalent).</p> <p>This incorporated coded data regarding the Mother's Emotional Response to her child in General (Positive or Negative)</p>	<p>Mother's report of the child's level of sociability (number and type / target of friendships)</p>
Step 2	<p>Mother's mode of comfort as an adult (a partial indication only of adult attachment style)</p>	<p>Mother's description of the relationship, her roles and functions within the MCR, and her</p>	<p>Mother's report of the child's desire/ motivation</p>

Secure = seeks comfort when distressed from partner or significant other (if applicable) versus  
 Insecure = no reference to comfort from significant others

concerns within the MCR

for friendship

### Step 3

The mother's availability as the child's "safe-haven and source of comfort". This referred to coded data pertaining to:

- The Mother's Emotional Response to TS Child's Distress (Available or Unavailable)
- Mother's Behavioural response to her TS child's distress (Active or Passive)

Examine Discrepancies between desired friendships and actual friendships, to indicate the youth's satisfaction with their current level of sociability

### Step 4

The factors that mothers identified as positively or negatively impacting upon their child's ability to socialise with peers

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Note: Raw Scores, Frequencies and Percentages were calculated and hypothesized relationships were tested for individuals and the group, referencing the frequency data



**Table 27.** Summary Results of the Analysis of Indicators of Quality of Mother's Attachment, Mother Child-Attachment & Child-Peer Attachment

Qualitative Analytical Themes (Mother-Child Attachment; Mother's Attachment; Child-Peer Attachment)	Total Responses and Percentage of Sample (%)	Totals
Mother's Perceived Childhood Attachment	Secure = 5 (26%) Insecure = 13 (74%) n/a = 3	18
Mother's childhood experience of Mother as available (safe-haven; secure base)	Present (Secure) = 4 (27%) Absent (Insecure) = 11 (73%) n/a = 7	15
Mother's childhood experience of mother as source of comfort when distressed	Available (Secure) = 4 (27%) Absent (Insecure) = 11 (73%) n/a = 7	15
Mother's source of Comfort As an Adult	Secure = 6 (43%) Insecure = 8 (57%) n/a = 8	14
Mother's Representation of child (narrative-based)	Positive = 12 (54%) Mixed - Ambivalent = 7 (32%). Negative = 3 (13 %)	22

Changes in Mother-Child Relationship	Change with Diagnosis Yes = 10 (45%) Change with Diagnosis No = 10 (45%) Recent change = 3 (14%) n/a = 2	20
Mother's Parenting Style - Comparison with own Parent	Different = 16 (89%) Same = 2 (11%) n/a = 4	18
Mother's Emotional Response to Child in General.	Positive = 5 (23%) Mixed-Ambivalent = 14 (64%) Negative = 3 (14%)	22
Mum's Emotional Response to Child's Distress	Positive = 1 (4%) Mixed-Ambivalent = 13 (59%) Negative = 8 (36%)	22
Mother's Behavioural Response to Child's Distress	Active and Positive = 12 (45%) Active & Positive - Occasional Disconnect = 9 (41%) Negative = 1 (4%)	22
Estimated Mother-Child Attachment	Secure = 11 (52%) Secure with signs of over-involvement = 8 (38 %) Insecure = 1 (5%) Insecure Overinvolved = 1 (5%)	22

	n/a = 1	
Estimated Child's Peer Attachment	Secure = 9 (41%)	22
	Insecure = 11 (50%)	
	Moving from Insecure to Secure with maturity = 2 (9%)	

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## **SECTION 5 CHAPTER 1. IMPLICATIONS AND INTEGRATION OF THE FINDINGS AND RECOMMENDATIONS FOR PRACTICE AND POLICY**

### **Overview of the Research and Major Findings**

#### **Research goals and methodology**

The research described in this thesis was conducted in response to the under-representation of psychologists in both the provision of care and the advancement of research regarding Tourette Syndrome. The major goal of the research was to improve current understandings of individual differences in the effects of this distressing disorder on diagnosed youth, and those involved in their care. With no known cure and treatment for many largely restricted to psycho-pharmacotherapy, the potential for psychology to contribute to understandings of the impact of TS and to the development and delivery of therapeutic interventions for this clinical population is evident. It was therefore hoped that the research findings could inform psychological interventions designed to address the unmet needs of this clinical population, and to encourage greater participation by the discipline of psychology as providers of care, research and advocacy for the TS community.

Specifically, the research sought to test the hypothesis that a psychological variable - the security of peer attachment - would account for individual differences in the quality of life and functioning of diagnosed youth. It was further hypothesised that TS would exert uniquely adverse effects on the security of the attachment relationships, and the attachment-related functioning of those diagnosed. Given the critical role that secure primary and peer attachment relationships play in determining

the optimal development and wellbeing of youth, the impact of TS on both relationships was examined.

The multiple aims of the research included (i) profiling the clinical phenomenology of a large community-based sample of Australian youth with TS, and ascertaining the adequacy of services and supports currently available, to facilitate comparison with international findings, (ii) comparing the quality of life and functioning of youth with TS with typically developing peers, (iii) determining the impact of tic severity and comorbid disorder on the quality of life (using the PedsQL) and functioning of diagnosed youth (employing the SDQ), (iv) testing the novel hypothesis that a psychological variable - the security of peer attachment - would account for individual differences in measured outcomes for youth with TS (employing the PedsQL & SDQ), (v) exploring the impact of TS on the lived experienced of those diagnosed and their primary caregivers, in particular how TS effected the security of peer attachment and the mother- child attachment relationship, and finally, (vi) determining the ability of TS to disrupt attachment security by examining the stability of attachment demonstrated between the relationships included in the study.

In order to achieve these research objectives, two separate studies were conducted. Study 1 (Study 1, Part A) comprised a quantitative, survey-based study ( $N = 194$ ) of a nationwide sample of parents of Australian youth with TS and parents of peers with no known diagnoses, in addition to a qualitative study based upon written parental responses (Study 1, Part B). Study 2 was a qualitative telephone interview-based study of biological mothers of youth with TS ( $N = 22$ ).

The research design aimed to address several methodological problems inherent to many prior TS studies. The recruitment of a comparatively large sample

facilitated the use of multivariate statistical analyses, which advanced current understandings of the role of the multiple variables included in the study. Although not an epidemiological sample, confirmation bias was minimised by employing a community-based sample. This research also represents one of the larger controlled studies of the difficult to access TS population. Finally the research addressed a significant gap in the TS literature by comprehensively examining the experience and expression of TS in the understudied Australian TS youth population.

### **Major Findings**

The study achieved all of its major goals, and the majority of the hypotheses were supported. As predicted, results revealed the complex and highly heterogeneous clinical profiles of this Australian community-based sample, which mirrored phenomena reported in international TS studies. Also as hypothesised, TS was strongly and consistently associated with adverse quality of life outcomes and higher rates of psychopathology, behavioural and social problems. Results also revealed deficits in health and educational services, and continuing stigmatization and social exclusion for many Australian youth with TS and their families (Section 3, Chapter 1)

In agreement with the major hypotheses, insecure peer attachment, increased tic severity and the presence of comorbid disorder were strongly and independently associated with decreased quality of life, increased impairment across each major life domain and higher rates of psychological, behavioural and social difficulties (Section 3, Chapter 2). The additional hypothesis, that secure peer attachment would moderate or mediate the adverse effects of increased tic severity and comorbid disorder on quality of life outcomes, was not supported. The importance of considering the quality of peer attachment on outcomes for TS youth was, however, emphasised by

the highly significant increased rate of insecure peer attachment reported for those diagnosed. This placed TS group youth at increased risk of the multiple negative consequences of insecure peer attachment demonstrated in the literature and confirmed by this study.

Although difficult to disentangle from the impact of comorbid disorder, the current research suggests a unique role for TS in impairing the social functioning and peer relationships of diagnosed youth. The type of difficulties and areas of impaired functioning associated with individual comorbid disorders were restricted and highly disorder specific. This diverges from prior studies that attributed a disproportionate amount of adversity experienced by youth with TS, including increased social and relationship difficulties, to the presence of common comorbid disorders such as ADHD and OCD (Section 3, Chapter 7)

The study also revealed the increased vulnerability of youth with TS to emotional disorder (Section 3, Chapter 4). This was considered to be of particular clinical relevance given prior research identifying depression and anxiety as the strongest predictors of highly adverse outcomes by late adolescence and early adulthood for youth with TS. These include reduced quality of life, increased rates of inpatient admission, and substance abuse. Whilst evidence suggests that the aetiology of mood disorder in youth with TS is multifactorial, impaired relationships and reduced social functioning are likely to contribute substantially to emotional difficulties.

The further hypotheses that TS would pose unique challenges to the security of the key attachment relationships (mother-child relationship and peer attachments), and the attachment-related functioning of diagnosed youth, were supported by the detailed findings from the qualitative studies. TS was found to exert complex effects



on the ability of youth to form secure relationships with peers (Section 3, Chapter 3). Whilst tic severity and symptoms of comorbid disorders were problematic, factors such as the youth's adaptive cognitive appraisal of their symptomology, a lack of self-consciousness, successful psychological adjustment to diagnosis, and the ability to defend themselves from negative peer attitudes and behaviours, ameliorated the adverse psychosocial consequences of their disorder. Other influential factors identified by the research included a strong role for the youth's personality traits (the "Big Five", Costa & McCrea, 1992), with Extraversion and Agreeableness facilitating friendships, and Neuroticism acting as an impediment. Furthermore, when compared with parents of undiagnosed youth, TS group parents expressed more complex implicit theories and identified different factors affecting their child's friendships.

TS also appeared to determine uniquely "close" mother-child relationships (MCRs), and directed maternal roles and functions within this relationship (Section 3, Chapter 6). Multiple threats to the security of the MCR attributable to the youth's TS were also identified; in particular, the manner by which TS increased maternal fear, anxiety and stress. These adverse emotional experiences limited the mother's availability to serve as her child's attachment figure, at least on occasions. Many general stressors associated with parenting a child with TS (Section 3, Chapter 5), and mechanisms by which change in attachment within this relationship could occur were also identified (Section 3, Chapter 6).

Finally, the strength of the impact of TS on attachment quality was explored by examining the consistency of attachment demonstrated across relationships (the mother's childhood attachment, the MCR and the youth's peer attachments), which theory predicts should remain stable in the absence of significant stress (Section 4, Chapter 1). Whilst secure maternal attachment predicted security in most future

relationships, the results for insecurity were inconsistent. Multiple mechanisms by which bidirectional change in attachment could occur, including a role for maternal over-involvement and several factors attributable to the youth's TS, emerged from the data.

Taken together, the results of the current research support the importance of giving priority to interventions aimed at improving the close relationships and social functioning of youth with TS. As discussed in each study and summarised in the final section of the current chapter, several limitations must be considered when interpreting the present findings. Of most significance were the limitations associated with the need to employ survey methodology in Study 1. This necessitated the exclusive reliance upon parental reports due to ethical and practical considerations involved in the inclusion of children as young as seven in the present research. Nation-wide survey methodology also precluded the ability to conduct formal clinical assessments of TS and comorbid disorders.

The rate of comorbidity reported for the TS group (72%) indicates that this community-drawn sample did include a greater number of youth with less severe TS ("TS only"), when compared with the 90% rate generally reported in clinic-based samples (Robertson, 2012). The majority of those with a comorbid condition ("TS Plus"), however, were also reported as having more than one additional diagnosis, suggesting that the sample may also have included a subgroup of youth with more serious forms of TS. Finally, due to the exploratory and novel nature of Study 2, and the lack of suitable standardised measures, findings were limited to "estimates" of the security of the attachment relationships under study. These "estimates" resulted from the employment of the unique methodology designed for the purposes of the current research. Therefore the following recommendations, particularly those emerging from

the qualitative studies, must be considered cautiously until replicated in future research.

The implications of these research findings for practice and policy in addition to recommendations for intervention will now be discussed. The latter will be restricted to recommendations for psychological practice aimed at improving the mother-child and peer relationships of diagnosed youth, as these were the major areas of interest in the current study, and represent an original contribution to the TS literature.

### **Implications and Recommendations for Practice and Policy: Clinical Profile & Correlates of TS**

This project began by determining the clinical profiles and correlates of TS for the present sample of young Australians with TS. In regard to the continuing evolution of the conceptualization of TS, whilst parental reports of the clinical phenomenology noted in this study provides measured support for the universality of the expression of TS and its' genetic, neurobiological aetiology (Eapen & Črnčec, 2009; Robertson, In Press; 2008, 2012), findings could not substantially advance the current “multiple-phenotype” versus “spectrum disorder” debate. This would require future research aimed at replicating the “TS Types” that emerged in prior studies, employing data reduction techniques such as factor analyses (Alsobrook & Pauls, 2002; Mathews et al., 2007; Grados & Mathews, 2009), and would also benefit from controlled diagnosis within a clinical setting. The prevalence of comorbidity for this community-based sample does however support Robertson's hypothesis (Robertson, 2006b; 2012) that comorbidity may be best conceptualized as a central feature of TS.

The complex, heterogeneous clinical profiles demonstrated in the study also highlight the challenges associated with differential diagnosis, treatment planning and intervention in the context of TS, all of which requires clinicians to have a substantial level of understanding and awareness of TS (Eapen & Črnčec, 2009).

Recommendations for practice regarding the clinical assessment of individuals with TS have been well documented (Woods et al., 2007), and the results of the current study add to the literature by demonstrating the utility of multi-rater, brief screening measures such as the PedsQL and the SDQ for youth with TS. These can optimise detection of early stage or unrecognised problems, and facilitate appropriate referral and early intervention. In the current study for example, the SDQ successfully identified a high rate of depressive symptomology, which was not reflected in rates of formal diagnoses.

In addition to clinical complexity, the wide-ranging adverse impacts of TS on QoL and psychological, behavioural and social functioning, demonstrated in the current and prior studies, suggest the need for a biopsychosocial (Engel, 1977), multidisciplinary response to the assessment and management of TS. The current findings, therefore, of the deficit in TS specific services and supports reported by parents, the over-reliance on medical practitioners (paediatricians, general practitioners, neurologists and psychiatrists) as the major providers of care for youth with TS (80%), and the low reported level of knowledge of TS demonstrated by professionals, are of significant concern. Furthermore, less than a third of TS youth in the study were currently prescribed medication to manage their TS, and an equally small proportion was receiving psychological or educational assistance. This meant that the majority of youth in this study were not currently receiving any form of

conventional therapeutic support or being adequately monitored for change in psychological or behavioural status.

The study also identified many other areas of need for the TS community. Parents placed a high priority upon improving the education of professionals, particularly those encountered during the diagnostic process, which the majority described as being protracted and highly traumatic. The need to extend training to GPs in particular was apparent, as they were often the first point of contact for those seeking a diagnosis for their child. Whilst an initiative to improve the training of psychologists and psychiatrists sponsored by the TSAA has recently commenced, a substantial effort will be required to design and implement professional development programs for all key practitioners. Greater inclusion of TS in undergraduate and post-graduate clinical training programs and academic curricula will also be required.

Having immediate access to comprehensive information regarding TS, its diagnosis and treatment, was also a high priority for parents. Whilst such resources are available through the TSAA, parents may not be aware of this organisation, be reluctant to make contact early in the diagnostic process when their need for information is highest, and the capacity of this self-funded, voluntary organisation to meet the needs of all Australians affected by TS is necessarily limited.

Increased access to specialist treatment, and a diverse array of more specific needs ranging from access to respite care to increased funding for multidisciplinary services, were also noted. These findings suggest the need to advocate for greater support from professional groups, institutions and government, and the results of the current research may provide useful evidence in that regard.

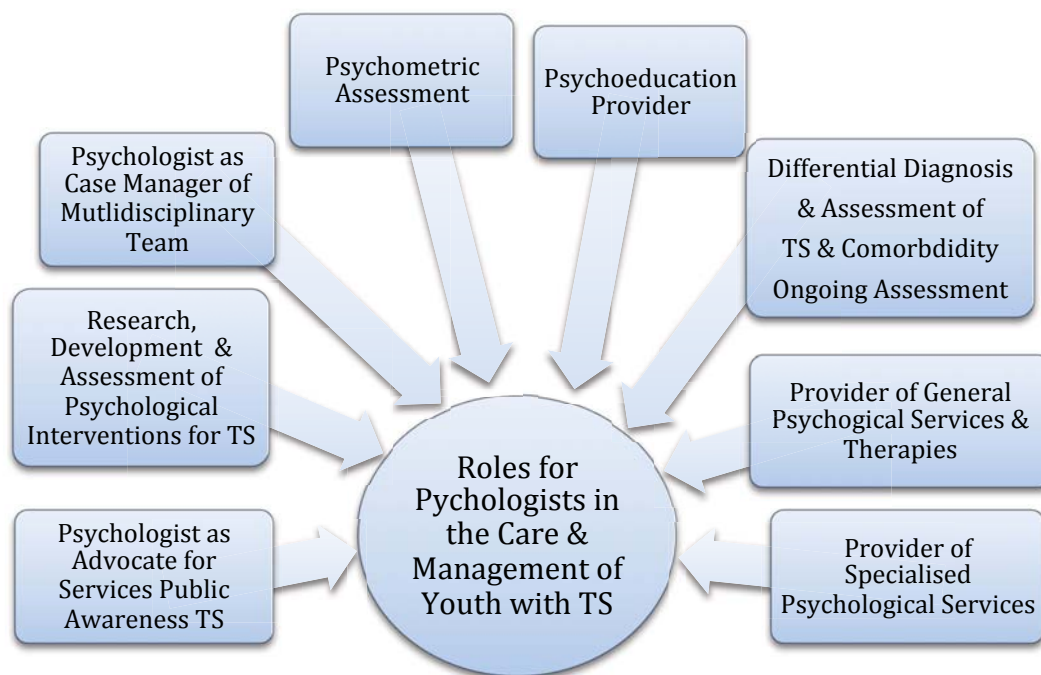
The need to extend knowledge of TS to the wider community was also indicated by the current results. As found in prior TS research (Collins, 2005;

Dedmon, 1990; Stern, Burza & Robertson, 2005), factors external to youth including the substantial rate of stigma, and low levels of understanding and acceptance, were likely to contribute to the social isolation, stress and distress experienced by both parents and their TS children. Somewhat alarmingly, parents in the current study prioritised rectifying widely held negative TS stereotypes and myths, and the lack of acceptance and understanding regarding TS, ahead of increased services or funding. This indicates the high level of suffering associated with the continuing stigmatization and misunderstanding of TS. Furthermore, this was experienced at the interpersonal, professional, institutional and societal levels, suggesting the need for an ongoing comprehensive awareness and education campaign to counter these difficulties. Australia may lag behind countries such as the UK, the USA and Canada where substantial recent efforts driven by patient advocacy groups (e.g. the Tourette Syndrome Association- USA; Tourette's Action UK; Tourette Syndrome Foundation of Canada) have raised the profile of TS, with positive outcomes. Again, such an effort will require funding and support at the governmental level.

Of particular interest in the context of the current study was the finding that unlike their Canadian counterparts (Trifricante, 2007), parents did not directly identify the need for psychological support for themselves or their child. Perhaps the reasons for this were that Australian participants lack awareness of the potential benefit of psychological services, have limited access to non-pharmacological forms of intervention for TS or this result may be a function of the under-involvement of psychology in the field of TS. The findings of the current research project, and the promising early results of efficacy trials of CBIT (Chang et al., 2009), emphasise the potential for psychologically oriented TS interventions. With so few treatment options currently available, the development of such therapies and psychological models of

care are an important goal for the future. Figure 5 illustrates the many possible roles for psychologists in the care and management of youth with TS.

In conclusion, the current findings indicate the urgent need for the increased education of and involvement by psychologists, allied health and educational professionals in TS research, and the provision of multidimensional services to meet the complex unmet needs of this clinical population. Present findings may also be useful to employ in the context of psychoeducation, professional education, research and advocacy.



**Figure 5.** Roles for Psychologists in the Care & Management of Youth with TS.

### **Implications of the Integrated Findings: A Predictive Quality of Life Model.**

The aforementioned results represent a meaningful contribution to, and address several gaps in the TS literature. The remaining findings, however, advance current understandings of individual differences in the adverse impact of TS, and facilitate greater opportunities for the design and implementation of psychologically orientated therapies to assist those with TS, and their caregivers.

Although rarely employed in a disorder specific context, the novel application of attachment theory in this research project provided a developmentally oriented theoretical framework within which to systematically explore the impact of TS on the close relationships and psychosocial functioning of those diagnosed. Furthermore, the robust and extensive evidence base of attachment theory provides support for detailed insights emerging from these exploratory studies.

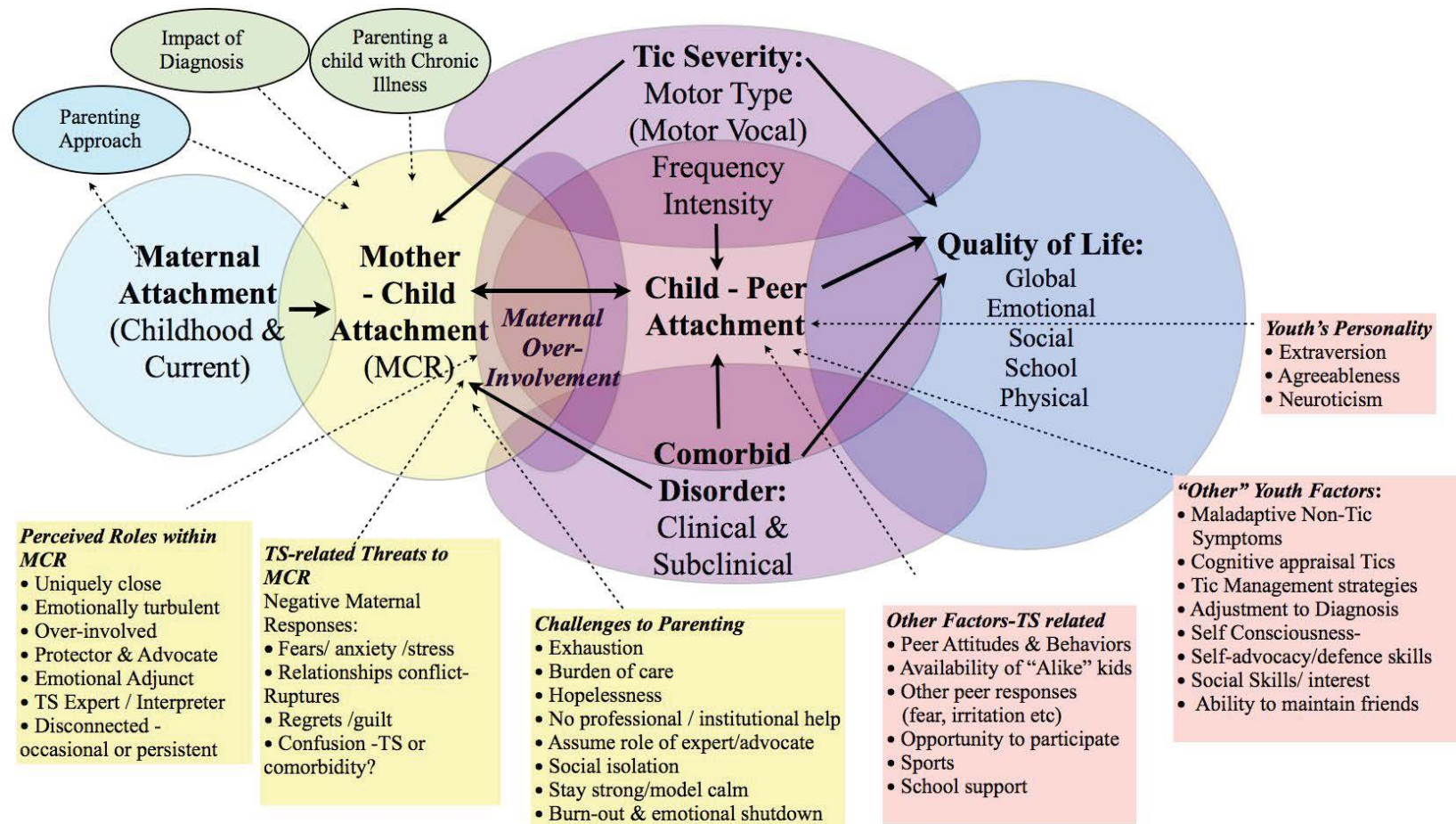
The employment of attachment theory also facilitated the development of a predictive quality of life model based on the integrated findings from the current research, which is graphically presented in Figure 6. Given the previously discussed challenges posed by TS to the clinician, this hypothetical model may provide a way to conceptualize the many difficulties confronting youth with TS and their caregivers, to prioritise treatment goals, and to plan interventions tailored to the unique requirements of the individual. Whilst the model encompasses the need to manage tic severity and comorbidity, it also highlights the importance of improving the attachment relationships of diagnosed youth as a key goal of treatment.

A diverse range of psychological interventions aimed at improving the psychosocial functioning and relationships of those diagnosed may be implemented in response, with the goal of improving quality of life and functional outcomes. Most



interventions rely on the existing skills of general, child and clinical psychologists, and do not require specialized training beyond psychoeducation regarding TS. With additional training, psychologists may also implement behavioural tic minimization strategies including habit reversal therapy (HRT) and competing response, massed practice and CBIT (Peterson, 2007; Woods et al., 2008; Woods et al., 2011). These help youth learn to how to change, camouflage, suppress and perhaps in the case of CBIT, extinguish tics when required. Other specialist services such as those offered by educational psychologists, family or relationship therapists may also be utilised.

The proposed model has the additional benefit of illustrating the impact of TS on the mother's relationship with the TS child or adolescent. As the current findings confirm, mothers play a pivotal, and often largely unsupported role, in the care of their TS diagnosed child. Developing a greater understanding of the dynamics of this relationship, the mother's attachment history, and the impact of TS on both the mother and youth with TS, may enhance the opportunity for clinicians to work more effectively with both. Although future studies will be required to strengthen and validate this model, the current findings may be employed to tentatively inform intervention.



**Figure 6.** Predictive Quality of Life Model for Youth with Tourette's Based on the Integrated Findings from Studies 1 & 2.

## Recommendations for Practice

### Therapeutic Approach

Many psychological approaches and modes of therapy may be employed to address the multidimensional difficulties presented by youth with TS and the caregivers. These range from supportive, psychodynamic, pragmatic solution-focused, behavioural therapies through to family systems therapy. With their popularity and proven efficacy in the context of pediatric and adult therapy, however, Cognitive Behavioural (CBT) (Beck, Rush, Shaw & Emery, 1979; Graham, 1998) and Interpersonal (IPT) approaches (NICE, 2005; Mufson et al., 1999), may be particularly helpful to interventions attempting to improve the relationships and social functioning of youth with TS and their mother-child relationships (MCR). These approaches also have the advantage of application across many modes of therapy including individual, conjoint, group or family therapies.

For children, CBT may be adapted to reflect the metacognitive ability and emotional fluency of the developing child, with the therapist focusing on behaviourally oriented interventions for younger children and gradually increasing cognitive techniques during middle childhood and adolescence. Whilst CBT can be successfully employed to change maladaptive thoughts and modify difficult behaviours, it may be particularly helpful for teaching social and interpersonal problem-solving skills, important goals in the context of the difficulties youth with TS experienced in this study.

Rooted in Bowlby's (1982) attachment theory, Interpersonal therapy (IPT) represents the approach most closely linked to the theoretical context of the current

study, and has been recommended for the treatment of many adult difficulties and psychiatric disorders, and pediatric disorders, particularly depression (NICE, 2005). Therapy begins by identifying the youth's problems, and then aims to decrease symptoms and improve social functioning within important relationships by developing effective problem-solving and communication skills. These skills are taught and rehearsed in session, and gradually extended to the real world setting.

### **Implications and Recommendations for Practice: Improving the Mother-Child Relationship**

#### **Assessment**

Whilst prior studies have reported the challenges shared by parents of youth with TS (Collins 2005; Dedmon, 1990, Einarsdottir, 2008; Shimberg, 1995, 2012; Stern et al., 2005; Walkup, 1999), the current findings (illustrated by the model presented in Figure 6) may be used to guide the clinician towards identifying and exploring those factors most relevant to individual dyads.

In addition to a comprehensive biopsychosocial intake interview, clinicians may seek to explore maternal attachment style. This may be achieved informally via interview or psychometrically assessed using standardized measures such as the self-reported Revised Adult Attachment Scale (Collins, 1996), the Experiences in Close Relationships Scale – Revised (Fraley, Waller & Brennan, 2000), or the Adult Attachment Interview (Kaplan & Main, 1985). The latter, however, requires substantial training to administer and interpret. The formal assessment of mother-child attachment is also challenging and complex, as it involves different methods for

different age groups, and varies in forms including standardised observational studies, to projective tests and questionnaires.

The mother's increased risk of psychopathology, attributable to the genetic links between TS and many comorbid disorders, should also be considered during assessment (Eapen & Črnčec, 2009). Finally, family and marital functioning may be considered, as the adverse impact of TS can extend to both (Ginsburg & Kingery, 2007; Hansen, 1992).

### **Psychological Interventions**

Present findings suggest that the most important early intervention for mothers of diagnosed youth is the provision of psychoeducation and comprehensive informational resources, in addition to referral to multidisciplinary services and peer supports (e.g. TSAA). Another important early intervention is assessing the mother's level of adjustment to the child's diagnosis of TS. Therapy may then be provided to resolve residual grief and trauma. Redressing the mother's guilt regarding pre-diagnostic parenting is also a priority, as this may mitigate or prevent maternal overcompensation within the MCR.

Supportive psychotherapy for mothers may also be an important early intervention that affords the opportunity for catharsis and debriefing. The therapeutic relationship also represents a source of empathy, acceptance and validation for mothers in the face of the low levels of understanding and acceptance of TS they frequently encounter. Indeed, an ongoing relationship with a therapist may become a valuable resource for the mother. By providing emotional support, and focusing on developing maternal coping skills, the therapist may help the mother as she confronts the various challenges accompanying different stages of her child's development

(particularly during puberty and adolescence), and dealing with the unpredictable and changing nature of the TS symptomatology and its unknowable prognosis.

Interventions may also target the specific threats posed by TS to the quality of the MCR, and the general difficulties accompanying parenting a child with a chronic, currently incurable disorder. Reducing maternal fear, anxiety and stress will assume a high priority, particularly for mothers with an insecure attachment history. Specific interventions may include cognitive behavioural therapy (CBT), formal stress management, and relaxation training. Mindfulness-based techniques and Acceptance and Commitment Therapy ACT (Harris, 2006) may be of particular benefit for those struggling with over-vigilant and catastrophic cognitions. The therapist may prioritise empowering mothers who have developed a sense of hopelessness in response to their child's disorder, in order to manage depression.

Minimising distress, conflict and ruptures within the MCR is also likely to be identified as a crucial treatment goal, particularly when it relates to the aggressive and disruptive behaviour of the youth with TS. Although challenging levels of aggressive behaviour are traditionally managed by medication, psychosocial and behavioural interventions involving both the child and the parent may be effective adjunctive or "first-line" therapies (Sukhodolsky & Scahill, 2007), although no studies have assessed their efficacy in the context of TS. In addition, behavioural strategies may be difficult for the already stressed and over-burdened mother to apply with consistency.

Application of behavioural interventions is further complicated by problems mothers experience in differentiating between behaviours that are, or are not, under the youth's control (Walkup, 1999). Discipline may also be difficult to enforce in the face of the mother's high level of empathy for her child's many difficulties (Ginsburg & Kingery, 2007). These factors highlight the need for the ongoing support of the

therapist, who can help the mother to both monitor, develop and respond consistently to her child's more challenging behaviours. For aggression and ruptures linked to adolescence, mothers may benefit from traditional parent training aimed at helping them to cope with developmental transitions, and changing roles within the previously close MCR. Finally, if aggressive, or impulsive behaviour represents a serious threat, the clinician may need to help the mother to develop a crisis plan.

The present study also identified the importance of reducing the burden of care the mother may experience. A pragmatic, solution-focused approach to therapy (De Shazer et al., 2007) might be employed to help address the multiple practical needs of the mother, to encourage the sharing and devolution of responsibilities where possible, and to encourage maternal self-care. Given the onerous, often-unavoidable tasks accompanying parenting a child with TS, the therapist may also assume a key role in helping to prevent maternal over-involvement, by helping the mother to maintain age and culturally appropriate boundaries with her child.

An important component of therapy aimed at minimising blurred emotional and behavioural boundaries within the MCR, is assisting the mother to manage her emotional response to and fears for both the child and herself. Once again this may be more difficult for mothers with an insecure attachment history. CBT, mindfulness and elements of Dialectical Behaviour Therapy (Linehan, 1993) may be effective in improving emotion regulation for mothers choosing to participate in individual therapy. Furthermore, the therapist may need to help the mother to maintain an identity beyond her role as caregiver.

Another target for therapy involves developing the mothers social, presentation, negotiation and assertiveness skills. These may minimise the stress and burden associated with the maternal role as child advocate and TS educator. These

skills will be of particular benefit in the context of the school, where effective communication and collaboration between parents and staff results in greatly improved academic and social outcomes for the child, and enhanced parental wellbeing (Kepley & Conners, 2007; Packer, 1997, 2005; Woods, 2002; Woods et al., 2003; Woods & Marks, 2005).

Interventions to improve family and marital functioning could include a range of problems identified by mothers in the current and prior studies. These include helping family members to tolerate and respond consistently to the youth's symptoms, resolving the neglect of siblings, intervening to reduce family and marital conflict, and helping parents to resolve differences in approaches to parenting. Finally, clinicians may be required to intervene to reduce the social isolation of mothers and the family. Offering psychoeducational services to extended family and friends may help increase acceptance and understanding. Linking mothers to organisations such as the TSAA, who can provide an alternative support network and social opportunities, may also be beneficial. This, however, must be cautiously considered in relation to the readiness of the mother, diagnosed youth and the family to encounter others from the TS community.

### **Implications and Recommendations for Practice: Improving Peer Relationships**

#### **Assessment**

As with the assessment of the security of the MCR, the clinician may gain informal insight regarding the youth's peer attachment security and level of social functioning by interviewing the youth and their caregivers. Peer attachment security



may also be formally assessed using the unidimensional AQC (Muris et al., 2001), or the multi-dimensional Inventory of Parent and Peers-Revised (Gullone & Robinson, 2005) (aged restricted to 9-15 years), both of which are available in self and parent-proxy versions.

### **Psychological Interventions**

As part of a comprehensive intake interview, the clinician may include an assessment of the youth's peer relationships. Where indicated, the process of designing interventions aimed at improving peer attachments for the individual child may be guided by the current findings. The major factors impacting the peer attachments of youth with TS are presented below, along with corresponding recommendations for intervention. Therapy, particularly for younger children, may be most effective if it involves parents or the family.

#### ***Improving Personality Functioning and Personality- Related Behaviours***

Current findings suggest the importance of determining the impact of the youth's personality traits and related behaviours on their peer relationships, as well as and the youth's ability to attract, recruit and maintain friends. Formal assessment of personality is not, however, recommended. An important therapeutic goal might be to facilitate those behaviours associated with "positive" traits such as Extraversion and Agreeableness, and minimize the "negative" behaviours associated with Neuroticism and low Agreeableness.

Current findings also indicate the need to monitor extraverted TS youth for a tendency to be too overbearing or "over the top", particularly if they have comorbid

ADHD or OCD, which may interact with their Extraversion to further alienate them from peers. Highly Extraverted youth with TS may also need to be closely observed, as their strong motivation for friendship places them at greater risk of adverse psychological consequences if they experience rejection and stigmatisation. The therapist may also need to closely assess, and address signs of neurosis, emotional dysregulation and depression, as symptoms of depression may be difficult to distinguish from trait neuroticism.

Therapists may choose from many methods to try to encourage behaviours that reflect “positive” personality traits. These include, but are not limited to CBT, IPT, contemporary personality-based IPT, and social skills training. It may also be necessary to facilitate opportunities for the child to participate socially and practice positive social behaviours.

### ***Facilitating Adjustment to Diagnosis of TS***

In this study, maladaptive emotional and behavioural responses to diagnosis of TS were found to impair both the social functioning and peer relationships of youth. Given this, and studies associating diagnosis of chronic disorder with a child’s increased risk of adjustment disorder (Wallander & Varni, 1998), evaluating the youth’s level of adjustment to their diagnosis in the early stages of assessment is recommended. If maladjustment is identified, supportive psychotherapy may facilitate the youth’s grieving process, augmented by psychoeducation and the therapist modelling acceptance and understanding of the disorder. A cognitive approach may enable the exploration of the “meaning” the youth ascribes to their diagnosis and symptoms, with the aim of changing maladaptive or unhelpful cognitions. The

therapist may need to be particularly alert to signs of “self-stigmatization” (Mukolo et al., 2010), and intervene to mitigate this, where present.

***“Coping with” Tics - Psychological Interventions.***

The current findings also suggest that psychological interventions such as CBIT and Habit Reversal, designed to help youth to “cope with” their tics, facilitate friendship and sociability. Although challenging to minimise, current findings highlight the need to prioritise the treatment of severe vocal tics, due to their disproportionately negative effects. Therapists, youth and caregivers however need a pragmatic, realistic approach to tic management. For reasons that are not well understood, many tics are highly resistant to change, so it is necessary to target those that are most likely to be successfully modified. Furthermore, the rapidly changing nature of tics, and the waxing and waning course of TS, mean that such interventions are likely to be ongoing, and require persistence on the part of all involved.

The youth’s age may also determine the choice of intervention. CBIT, for example, appears to require cognitive capacity or the presence of premonitory urge, which are not generally present in those under the age of 10 (Woods et al., 2008). Contingency management also offers the opportunity to reduce tics, by identifying triggers for tic exacerbation, factors associated with tic minimisation, and the development of strategies in response. For those whose tics are exacerbated by stress, anxiety, fatigue or change for example, stress management and relaxation training may help to reduce tic severity or frequency.

Current results however suggest that the goals of psychological interventions aimed at increasing the youth’s ability to “cope with” their TS symptoms, need to go beyond tic reduction. Whilst the increased severity of tics is likely to adversely affect

social functioning and relationships, the youth's level of "self-consciousness" regarding their tics may mediate these effects. As mentioned in the context of adjustment to diagnosis, a cognitive approach to therapy may be required to reduce the distress, negative self-appraisal and self-consciousness associated with tics. Other strategies may include encouraging optimal social activity during waning tic cycles, and developing alternative strategies for socialization during waxing phases of the disorder. The use of the Internet and social media, closely monitored for incidences of cyber bullying, may help youth at these difficult times, and reduce social anxiety.

The importance of being able to advocate, being open and defending themselves in the context of their diagnosis, were also identified as key to the youth's ability to "cope with" their TS. Teaching presentation and assertiveness skills may be useful to enhance these abilities, however, the decision to share the diagnosis, the amount of detail provided, and who to include in this process must be cautiously considered. Cost benefit analysis needs to be conducted on an individual basis and in a collaborative process with the child, caregivers and the school (Kepley & Connors, 2007). All youth with TS, however, may benefit from the development of strategies enabling them to defend themselves against negative peer behaviours such as bullying, teasing and rejection. Therapists also need to ensure that anti-bullying strategies are extended to the youth's environment, including home, school, sports clubs and other social settings.

### ***Minimisation of Problems Associated with "non-tic" TS Symptoms and Comorbid or Secondary Disorder***

The importance of providing concomitant treatment for the youth's "non-tic" TS symptoms and comorbid disorder also assumes importance in the context of

facilitating friendships and social functioning. A pattern of socially damaging “non-tic” behaviours attributed to TS identified in this study included aggressiveness; impulsivity; a tendency to dominate other children; to behave bizarrely and incongruently; or to withdraw from or fail to participate socially. The current study also identified the need to reduce obsessive symptoms, which were found to inhibit friendship by upsetting play and group activities. These included the youth’s cognitive rigidity, rule focus, dogmatic or controlling behaviour, obsessions and “just right” compulsive rituals.

Intervention to address internalising symptoms and behaviours associated with the youth’s anxiety and depression are also important, as these encourage the youth’s social withdrawal and contribute to their unpopularity with peers. Treatment of comorbid ADHD, ODD/CD or impulse control is also important to reduce the overtly damaging effects externalising symptoms (impulsivity, inattention, hyperactivity, anti-social behaviours) have upon all of the youth’s relationships.

### ***Managing Aggressiveness and Disruptive Behaviours***

As discussed in the context of the MCR, reducing the youth’s aggressive and disruptive behaviours assumes a high priority, based on the present findings. As mentioned, medication may be prescribed in the case of serious aggression, particularly when it is symptomatic of the youth’s TS, ODD or intermittent rage disorder. Various psychological interventions may also be employed. Therapy may include behaviour modification, formalised anger management programs for older children and teens, impulse control and frustration tolerance strategies. Contingency management may help to identify triggers and consequences of aggression, and inform behavioural interventions in response. For aggression linked to maladaptive

adjustment to diagnosis (youth lashing out in frustration, rage and denial), the therapist may provide psychotherapy to facilitate adjustment.

The therapist may also consider the role of emotion dysregulation in the youth's aggression. This may be improved by employing an age downward form of Dialectical Behaviour Therapy (Lineham, 1993), which may also address any underlying depression or anxiety. Understanding the role that the youths' social cognition, social anxiety and low self-esteem might play in encouraging aggressive behaviour could also be addressed. Finally, for aggression linked to adolescence, youth may be trained in ways to understand and respond to developmental transitions and their changing roles in peer and parental relationships.

### ***Treating Anxiety and Depression***

With the increased risk of anxiety and depression accompanying TS, clinicians may closely monitor and measure signs of emotional disorder. Evidence suggests that pediatric anxiety is effectively treated with CBT (Seligman & Ollendick, 2011) whilst CBT, IPT and short-term family therapy (NICE, 2005) are the most effective approaches for pediatric depression.

### ***Providing Social Skills Training***

As noted in prior TS studies, there is a need to address the lower general level of social skills reported for a significant percentage of youth with TS (Champion et al., 1989; Cohen & Leckman, 1994; Dykens et al., 1990; Marek, 2006). Social skills training may be provided in therapy and via formal social skills programs. Youth with TS may also be disadvantaged by limited opportunities to develop and rehearse social

skills, due to low levels of peer acceptance and social exclusion, and need assistance to improve opportunities for social interaction (see following section).

### ***Facilitating Opportunities for Friendship and Socialisation***

In collaboration with the youth, parents and relevant others, the therapist may help to identify opportunities to reduce social barriers and increase opportunities for socialisation. Both child-centered factors and those external to the youth impact these. Current findings suggest the positive impact of sports participation, shared activities and having access to “alike” youth, role models and mentors. The study also suggests the benefit of encouraging the child’s sense of humour, interests, talents and skills.

Therapists must, however, carefully consider participation at a level that the youth can accommodate in light of their symptoms, comorbid pathology, motivation for friendship and personality. The amount of time spent with peers may also need to be managed to avoid the exhaustion associated with tic suppression and, as mentioned earlier, limited during waxing phases of the disorder. In order to facilitate continuing friendship, the child’s peers may also require occasional respite and support.

Limitations and barriers, such as fine motor deficits that occasionally accompany TS (Leckman et al., 2006a), a lack of interest in sports, difficulty coping with the competitive pressures associated with team sports, and low acceptance of the child by teammates and their parents, must also be considered when encouraging participation.

In addition, the study identified an important role for parents in encouraging, teaching and modelling social behaviours, in addition to taking active steps to create opportunities for children to develop friendships and extend their social networks.

Such efforts must be undertaken carefully to avoid parental over-involvement and may be challenging for already overburdened parents and socially isolated families.

### ***Changing the Attitudes and Behaviours of Peers***

Finally, as regularly identified in the TS literature, this study demonstrated the importance of increasing peer acceptance and understanding, and reducing bullying and rejection of youth with TS (Collins, 2005; Conelea et al., 2011; Kurlan et al., 1996; Packer, 1997). Given the critical role a supportive and accepting school environment plays in the social outcomes for those with TS (Packer, 1997, 2005; Woods et al., 2003; Woods & Marks, 2005), interventions may be best delivered in the school environment. Programs aimed at changing the attitudes of classmates towards those with TS, by increasing awareness and understanding of the disorder, have been found to effectively increase compassion, lower ridicule and increase peer acceptance of youth with TS (Packer, 1997, 2005; Woods, 2002; Woods et al., 2003; Woods & Marks, 2005). Many resources are available at no cost for these purposes via the TSAA and other internationally based TS societies.

## **LIMITATIONS AND FUTURE STUDIES**

### **Limitations**

This section recaps the main limitations associated with this complex research project, which have been discussed in detail at the end of each research report presented in Section 3.

Firstly, employing the assistance of the national TS support organisations to recruit the TS sample may have introduced bias. In addition to possible ascertainment



bias, unmeasured differences in characteristics of those who do or do not join support groups may have influenced results, whilst membership may have conferred benefits to parents, such as knowledge, coping strategies and social support that influenced outcomes in the studies. Future epidemiological sampling, which was beyond the scope and resources of the current study, would be required to control for these risks.

The study also relied upon parental reports, and as two prior TS studies have found, there may be some discrepancies between youth self-reports and parental reports (Storch et al., 2007b; Conelea et al., 2011). Participation of youth in the study was, however, prohibited by the ethical and practical concerns of surveying children as young as age seven (in a remote mode- postal survey / telephone interview). The need to gain the youth's perspective, particularly in relation to their interpersonal experiences, is therefore a high priority for future research.

Thirdly, the use of survey methodology precluded clinical assessment of tic severity and disorder meeting DSM-V criteria. Tic severity was however assessed using a robust parent-rated instrument (PTQ) that has been found to correlate strongly with results from the gold standard clinician-rated Yale Global Tic Severity Scale (Leckman et al., 1989), and successfully employed in prior published TS research (Conelea et al., 2011). Parental reports of formal diagnoses were supplemented with the findings of the well-validated SDQ and whilst not a diagnostic tool, the SDQ is a highly reliable and valid screening instrument for major pediatric psychiatric disorder (Goodman, 2001). Furthermore, the SDQ correlates highly with measures such as the CBCL (Achenbach & Rescalora, 2000), which has been employed in several prior TS studies to identify comorbid symptoms (Storch et al., 2007). In light of the serious challenges associated with differential diagnosis in the context of TS, it is also arguable that clinician assessments can adequately control for comorbidity. Future

efforts to replicate current findings in clinic based studies, or those including more rigorous clinician rated assessments of symptom severity and comorbid disorder, would be recommended.

The homogeneity of the sample (Caucasian, biological mothers, married, middle income) may also limit the generalizability of results from Study 1, thus future studies could benefit from the inclusion of a more demographically diverse sample. Generalisability may also have been limited in Study 2 by the over-representation of mothers with difficult family of origin experiences. In addition, in order to maximise the sample for the study, a wide age range of youth was employed. The study therefore could not control for potential variability associated with different developmental stages. Both peer relationships and the MCR are likely to change during different developmental stages and these would be important to consider in future research. Furthermore, the study could not adequately control for gender due to the expected heavy bias towards male participants in the TS group.

The cross sectional design of the study also limited inferences relating to the nature and causality of relationships between TS, comorbid disorder, tic severity, security of peer attachment, and quality of life outcomes. Future prospective longitudinal studies are needed to further consider the directionality and relative influence of these variables, particularly in view of the waxing and waning nature and course of TS.

The assessment of peer attachment in Study 1 was also limited to the use of the single-item measure, due to inability to identify a multi-item instrument designed for middle childhood. The lack of sensitivity of the instrument may have resulted in under-reporting of insecure attachment (Muris et al., 2003; Roelofs, Meesters, Huurne, Bamelis & Muris 2006). This was however ameliorated by the rich detail

provided by the qualitative studies. The dichotomization of attachment in this initial study may also have restricted findings. Future studies with a much larger sample may include multiple categories of insecure attachment (e.g. anxious, avoidant, dismissive, disorganized) and as initial research suggests, these may generate more detailed findings (Rutter, Kreppner and Sonuga-Barke, 2009).

In Study 1, the effects of several measured comorbid disorders could not be evaluated due to low reported numbers. As is commonly encountered in TS research, the unequal size of groups of youth with specific comorbid disorders limited parametric analyses.

Furthermore, in order to facilitate comparison with undiagnosed peers, the study employed a general health related quality of life measure. This may have resulted in the underestimation of impairment in youth with TS. More accurate measurement may be achieved in future research using a disease specific QoL instrument such as the GTS- QOL (Robertson & Cavanna, 2008).

Study 2 was also limited by use of the unique methodology designed to explore and estimate attachment, and the lack of suitable available standardised measures meeting the current design requirements. The methodology developed for the study was, however, congruent with the exploratory goals of this novel qualitative research project. Furthermore, the reliability of the “estimates” of attachment resulting from the methodology was supported by the consistency of the results emerging from the triangulation of relevant findings from both studies.

Other limitations relating to qualitative components of the current research (Study 1 Part B & Study 2) include the possibility that the researcher may have introduced bias at any or all levels of this study, although multiple efforts to minimise this were undertaken. In addition, open-ended questions for the semi-structured

interview in Study 2 were designed to maximize individual expression, however the phrasing and order of delivery varied slightly between respondents, which may have contributed to variability in responses. Although consistency of response was examined and demonstrated across each interview, social desirability may also have biased participants' responses.

### **Future Studies**

In addition to those mentioned in the preceding discussions, a high priority for future research would be to replicate the current study based on youth self-reports. The inclusion of rigorous methods of assessing attachment would also benefit future studies. These may include pen and paper psychometric measures such as the Experiences in Close Relationships Questionnaire (Fraley, Waller & Brennan, 2000) and the Inventory of Parent and Peer Attachment (Armsden & Greenberg, 1987), which assess a more restricted range of ages than currently included, or semi-structured interviews including the Adult Attachment Interview and the Child Attachment Interview (Shmueli-Goetz, Tagert, Fonagy & Datta, 2008).

Future research might also be designed to test the study's proposed "Predictive Quality of Life for Youth with TS Model" using techniques such as structural equation modelling or path analysis. Further goals could also include extending research to additional attachment relationships. Preliminary research has demonstrated that secure paternal attachment has important but differing effects on child wellbeing and functioning to those associated with secure maternal attachment (Doyle, Markiewicz, Brendgen, Lieberman & Voss et al., 2000), and may mitigate the negative impact of insecure maternal attachment (Main & Weston, 1981).

Findings from the current study also suggest multiple unexamined factors impacting outcomes for those with TS and their families that may be included as

variables in future studies. In regard to the mother, these include the role of maternal over-involvement, parenting style, family functioning, maternal stress and coping skills and maternal psychiatric status. For youth, the role of subclinical levels of mood and behavioural disorder, cognitive style, stress and coping skills, social skills, and developmental stage may be included. The length of time since, and impact of diagnosis, and the level of social support and stigma experienced, may be important for all actors.

Current findings also strongly suggest the inclusion of personality in future studies regarding the wellbeing and functioning of youth with TS. Prospective longitudinal research that includes personality assessment may contribute to better understanding of the relationship between personality traits and functioning in childhood, the impact of TS and comorbid disorder, and the higher incidence of personality disorder noted in adults with TS recently reported by Robertson (2007).

The findings that youth with TS were vulnerable to mood disorder also indicates the need for a detailed exploration of the relative contribution of different mood and anxiety disorders, in addition to the impact of subclinical levels of these on the child's wellbeing and functioning. Identifying the strongest predictors of the exacerbation or alleviation of the youth's emotional distress could also be explored in future research, with the current study suggesting a diverse range of potential variables. These include the quality of the youth's attachment relationships, personality variables (especially neuroticism), the individual's cognitive style, vocal tic severity, comorbidity and the social consequences of living with TS. On the basis of these results, targeted psychological interventions could be designed and tested.

Results from the two qualitative studies could also be employed to develop a TS specific questionnaire for assessing the quality of peer relationships of youth with

TS, the youth's level of satisfaction with current peer relationships, and factors that may enhance or detract from the youth's ability to form secure peer relationships.

The efficacy of the mother's parenting strategies, particularly those employed in response to her TS child's distress, has also been under-research. This could be the subject of future study, with results leading to the development of improved guidelines and interventions to assist mothers. Future studies could also attempt to replicate the relationship between insecure maternal attachment history and maternal over-involvement that emerged from the current study. Results also imply a need for the development and testing of interventions aimed at assisting mothers to maintain age and culturally appropriate emotional and behavioural boundaries with her child with TS.

Finally, the results of the current study revealed the utility of including attachment as an independent variable in the study of individual differences that could be extended to other pediatric psychiatric or medical conditions.

### **Concluding Comments**

Many new findings emerged from this complex research project, which makes several original contributions to TS literature. It has advanced current understandings of TS by identifying an important role for secure attachment as a key psychological variable associated with individual differences in outcomes for those diagnosed. It also illustrates the many ways in which TS impacts the attachment relationships and functioning of diagnosed youth, and their primary caregivers. The findings also provided the basis for the development of an inclusive, predictive quality of life model for TS that may be tested in future studies and tentatively employed to inform assessment and intervention. Recommendations for psychological interventions

designed to improve the mother-child and peer relationships of youth with TS were also developed and presented, in response to the current findings.

With its novel application of attachment theory, this research also makes an original contribution to the attachment literature. Attachment theory provided an elegant framework, with a vast empirical base, within which to explore the impact of TS on the wellbeing and functioning of the developing child or adolescent. The successful application of attachment theory in the current study also suggests that it may be usefully applied to the study of individual differences in outcomes associated with a wide range of developmental, psychological and medical disorders, particularly in paediatric populations.

Results of this research also formed the basis of seven research reports. Several have been recently published, with the remaining articles currently under review or subject to recent submission (November, 2015). Two conference papers presenting findings from this project have been delivered at the recent national conference of the Australian Psychological Society (October, 2015). Published abstracts for the conference presentations, and online links to the digital poster are presented in Appendix E.

Finally, many recommendations for practice and policy were generated in response to the research findings. It is hoped that the results of this study will inform and encourage a greater role for psychologists in TS research, practice, and advocacy, and provide the impetus for improved services and supports for the Australian TS community.

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## Australian Tourette's Survey

**PART A**

Participant ID: (Anonymous)

Date:.....

**Dear Participant,****Please complete all questions, by placing a Tic the appropriate box, or as a written answer as indicated.**Example :  or please write your answer on dotted line .....**Your Details****1. Your Place of Residence:** (Please Tic appropriate box)Major city  Regional town   
Rural or remote **2. Your Relationship to the child / teen:** (Please Tic appropriate box)Biological mother  Stepmother   
Adoptive Mother  Other Biological father  Stepfather   
Adoptive Father  Other **3. Your Marital Status** (Please Tic appropriate box)Never married  Married /de facto   
Separated/divorced   
Widowed/widower **4. Living situation:** (Please Tic appropriate box)Living with a husband/partner   
Single parent  Other **5. Number of children living with you (full or shared custody)** (please write number here)

.....

**6. What best describes your single parent or family income per year?** (Please Tic appropriate box)Up to \$35,000  \$35,000 – 75,000   
Above \$75,000 **7. What race do you consider your child to be:** (Please Tic appropriate box)Caucasian   
Aboriginal /Torres Strait Islander **Your Child's Details****8.**

Age of child = ..... years.

Gender: Male  Female **9. Has your child been diagnosed with Tourette's Syndrome or Chronic Tic Disorder (CTD)?**

(Please Tic appropriate box)

Yes  No **10. Does your child have any brothers or sisters?**

(Please Tic appropriate box)

Yes  No 

If yes, how many siblings? (Please write number here) .....

**11. Do any of your child's brothers or sisters have Tourette's Syndrome or CTD**

(Please Tic appropriate box)

Yes  No **12. Has your child been diagnosed with any of the following disorders?** (Please Tic appropriate box)Attention/Hyperactivity disorder Obsessive/compulsive behaviour/disorder Anxiety disorder Conduct disorder Impulse control Autism Learning disorder Depression 

Other (please write here).....

Asian  Other **Please turn the page**

Australian Tourette's Survey

13. Do you think anyone else in your family may have or have had Tourette's or CTD?

(Please Tic appropriate box) Yes  No

14. Have any of your child's brothers or sisters been diagnosed by a professional with any of the following disorders:

(Please Tic appropriate box)

- Attention/hyperactivity disorder
- Obsessive/compulsive behaviour/disorder
- Anxiety disorder
- Conduct disorder
- Impulse control
- Autism
- Learning disorder
- Depression
- Other (please write here).....

15. Do you find it difficult to access medical and/or mental health services for your son / daughter with Tourette's or CTD?

(Please Tic appropriate box)

Yes  No

16. Do you feel that medical and mental health professionals know enough about Tourette's or CTD and its treatment?

(Please Tic appropriate box)

Yes  No

17. Do you feel the education system is supportive of those with Tourette's or CTD?

(Please Tic appropriate box)

Yes  No

18. Do you feel that your child is stigmatized as a result for having Tourette's or CTD?

(Please Tic appropriate box) Yes  No

19. Is your child receiving medication for his/her Tourette's or CTD?

(Please Tic appropriate box) Yes  No

20. Is your child receiving any treatment or support from any of the following professionals? (Please tic the appropriate box if Yes)

- Pediatrician
- Neurologist
- Psychologist (any type)
- Psychiatrist or child psychiatrist
- G.P.
- Counselor (school or other)
- Special education assistance
- Family therapist

21. Does your child have any trouble making friends?

(Please Tic appropriate box)

Yes  No

22. What do you feel makes it easy or difficult for your child to make friends?

(Please write here)

.....

.....

.....

.....

.....

.....

.....

.....

.....

.....

Now please turn the page and continue with PART B

**Part B:**

Please read the directions for each section of this survey carefully and answer every question as best you can. It is most important that you try to answer every question, even if you are not certain of your answer.

**PLEASE BEGIN.**

**Directions**

Below is a list of things that **might** be a problem for **your child**. Please tell us **how much of a problem** each one has been for **your child** during the **past ONE month** by circling

- 0 if it is **never** problem
- 1 if it is **almost never** a problem
- 2 if it is **sometimes** a problem
- 3 if it is **often** a problem
- 4 if it is **almost always** a problem.

There are no right or wrong answers.

If you are **not sure** of a response, **please give it your best estimate**.

It is **very important** that you please answer **all** items.

*Example:*

1. Walking more than one block	0	1	2	3	4
--------------------------------	---	---	---	---	---

**NEXT: Please answer ALL questions.**

In the **past ONE month**, how much of a **problem** has your child had with .....

Physical Functioning (problems with...)	<i>Never</i>	<i>Almost never</i>	<i>Sometimes</i>	<i>Often</i>	<i>Almost always</i>
<b>1. Walking more than one block</b>	0	1	2	3	4
<b>2. Running</b>	0	1	2	3	4
<b>3. Participating in sports activity or exercise</b>	0	1	2	3	4
<b>4. Lifting something heavy</b>	0	1	2	3	4
<b>5. Doing chores around the house</b>	0	1	2	3	4

Emotional Functioning (problems with...)	<i>Never</i>	<i>Almost never</i>	<i>Sometimes</i>	<i>Often</i>	<i>Almost always</i>
<b>1. Feeling afraid or scared</b>	0	1	2	3	4
<b>2. Feeling sad or blue</b>	0	1	2	3	4
<b>3. Feeling Angry</b>	0	1	2	3	4
<b>4. Worrying about what will happen to him or her</b>	0	1	2	3	4

Social Functioning (problems with...)	<i>Never</i>	<i>Almost never</i>	<i>Sometimes</i>	<i>Often</i>	<i>Almost always</i>
<b>1. Getting along with other kids</b>	0	1	2	3	4
<b>2. Other kids not wanting to be his or her friend</b>	0	1	2	3	4
<b>3. Getting teased by other kids</b>	0	1	2	3	4

School Functioning (problems with...)	<i>Never</i>	<i>Almost never</i>	<i>Some-times</i>	<i>Often</i>	<i>Almost always</i>
<b>1. Paying attention in class</b>	0	1	2	3	4
<b>2. Forgetting things</b>	0	1	2	3	4
<b>3. Keeping up with schoolwork</b>	0	1	2	3	4

**NEXT:**

For each item below, please mark the box for **Not True**, **Somewhat True** OR **Certainly True**. Please answer **ALL** questions as best you can even if you are not absolutely sure. Please give your answers *on the basis of your child's behaviour over the past six months*.

ITEMS <i>Over the past 6 months, my child....</i>	<b>Not True</b>	<b>Somewhat True</b>	<b>Certainly True</b>
Considerate of other people's feelings	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Restless, overactive, can not stay still for long	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often complains of headaches, stomach aches or sickness	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Shares readily with other children, for example toys, treats, pencils	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often losses temper	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Rather solitary, prefers to play alone	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Generally well behaved, usually does what adults request	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Many worries or often seems worried	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Helpful if someone is hurt, upset or feeling ill	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Constantly fidgeting or squirming	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Has at last one good friend	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often fights with other children or bullies them	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often unhappy, depressed or tearful	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Generally liked by other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Easily distracted, concentration wanders	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Nervous or clingy in new situations, easily loses confidence	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Kind to younger children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often lies or cheats	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Picked on or bullied by other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often volunteers to help others (parents, teachers, other children)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Thinks things out before acting	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Steals from home, school or elsewhere	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Gets along better with adults than with other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Many fears, easily scared	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Good attention span, sees chores or homework through to the end	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Overall, do you think your child has difficulties in one or more of the following areas: Emotions, concentration, behaviour or being able to get on with other people?  
 (Please tic appropriate box)

No      Yes- Minor difficulties      Yes- Definite difficulties      Yes- Severe difficulties  
                                                                 

If you have answered "Yes", please answer the following questions about these difficulties.

• **How long have these difficulties been present?**

Less than a month      1 - 5 months      6 - 12 months      Over a year  
                                                                 

• **Do the difficulties upset or distress your child?**

Not at all                      Only a Little                      Quite a lot                      A great deal  
                                                                 

• **Do the difficulties interfere with your child's everyday life in the following areas?**

	Not at all	Only a little	Quite a lot	A great deal
HOME LIFE	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
FRIENDSHIPS	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
CLASSROOM LEARNING	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
LEISURE ACTIVITIES	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

• **Do the difficulties put a burden on you or the family as a whole?**

Not at all                      Only a Little                      Quite a lot                      A great deal  
                                                                 

**NEXT:**

Please choose **One** description from the **three options** below (**Tic One box only**) that **best describes your child**, even if it is not a totally accurate description.

**Description 1.**                     

*My son / daughter finds it easy to become close friends with other kids. My son/daughter trusts them and is comfortable depending on them. He / she does not worry about being abandoned or about another kid becoming too close friends with them.*

**Description 2.**                     

*My son / daughter is uncomfortable to be close friends with other kids. He /she finds it difficult to trust them completely, and it is difficult for him / her to depend on them. My son / daughter gets nervous when another kid wants to become close friends with him / her. Friends often come more close to my son / daughter than he / she wants them to.*

*(Please turn over for Description 3)*



**Description 3.**      □

*My son / daughter often finds that other children do not want to get as close as he / she would like them to be. My son / daughter is often worried that his / her friend doesn't really like him / her, and that they may want to end their friendship. My son / daughter prefers to do everything together with his / her best friend. However, this desire sometimes scares other kids away.*

**NEXT:**

*Please Note: If your child has never had any tics, you are not required to complete the following section of this survey*

**“Tic Questionnaire”**

**Step 1.** For each of the tics listed below, please mark “Yes” or “No” as to **WHETHER OR NOT your child has had the tic in the PAST MONTH**

**Step 2.** For each tic you mark as “Yes”, please **circle** how **FREQUENTLY** the tic occurred over the past week according to the following:

- Constantly**, almost all the time during the day
- Hourly**, at least once per hour
- Daily**, at least several times per day
- Weekly**, just a few times or less

**Step 3.** Under **INTENSITY**, rate how intense you believe the tic **FELT** to your child over the past week. For example, if it **was very mild**, like a weak twitch, that would be a “1”. A much **more forceful** tic that would be **very noticeable to others** and **may even be painful** would be rated as a “3” or higher. Any tic that would be **obviously noticeable to others** should be rated as at least a “2”.

*Example:*

Eye blinking	Yes	No	Constantly	Daily	1	2	3	4
			Hourly	Weekly				
Eye rolling/ darting	Yes	No	Constantly	Daily	1	2	3	4
			Hourly	Weekly				

*We will begin with your Child's Motor Tics (that is, tics that involve some part of the body moving). Please answer every question.*

MOTOR TICS	PRESENT Yes or No		FREQUENCY		INTENSITY 0 – 4			
Eye blinking	Yes	No	Constantly	Daily	1	2	3	4
			Hourly	Weekly				
Eye rolling / darting	Yes	No	Constantly	Daily	1	2	3	4
			Hourly	Weekly				
Head jerk	Yes	No	Constantly	Daily	1	2	3	4
			Hourly	Weekly				
Facial Grimace	Yes	No	Constantly	Daily	1	2	3	4
			Hourly	Weekly				
Mouth/ tongue movements	Yes	No	Constantly	Daily	1	2	3	4
			Hourly	Weekly				

Shoulder Shrugs	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Chest / Stomach Tightening	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Pelvic tensing movements	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Leg/ feet movements	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Arm/hand movements	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Copying others gestures	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Obscene gestures	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Other motor tics	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Complex motor combinations (multiple tics at once)	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4

*Now let's look at your child's VOCAL TICS (any noises words or sounds made)*

**Please answer every question.**

<b>VOCAL TICS</b>	<b>PRESENT</b> <i>Yes or No</i>		<b>FREQUENCY</b>		<b>INTENSITY</b> <i>0-4</i>			
Grunting	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Sniffing	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Snorting	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Coughing	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Animal noises	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Syllables	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Words	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Phrases (a few words together)	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Repeating the words or sounds of others	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Obscene or offensive words	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Blocking/stuttering	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Other	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Other vocal tics	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4
Complex vocal combinations (multiple sounds /words together)	Yes	No	Constantly Hourly	Daily Weekly	1	2	3	4

*The End*

- **Thank you so much for your participation**
- **Please check that EVERY question, on EVERY page has been answered, as best you can**
- **Please place the *completed questionnaire and your consent form* in the addressed envelop provided and mail it as soon as possible, or return your completed forms directly to the researcher. NO POSTAGE REQUIRED**

## INFORMATION SHEET

**“The Lives of Australian Children and Teens with Tourette’s Syndrome: A parent’s perspective”**

You are invited to take part in a research project that aims to help psychologists to learn more about the lives of Australian children and teens with Tourette’s Syndrome (TS) or chronic tic disorder, conditions that have been understudied, particularly in Australia, as well as children who have neither of these experiences. As parents and primary caregivers, your knowledge and understanding of your child is of great value and importance. Parents and caregivers are being asked to share their understanding about their children to enable professionals to develop appropriate and improved services.

This study will be an important step towards building local knowledge about the Australian TS community and its needs. Deirdre O’Hare is conducting this research in completion of her Doctor of Clinical Psychology, at James Cook University, Queensland.

If you agree to be involved in this study, you will be invited to complete a questionnaire that will ask for your observations of your child or teenager’s behaviours, emotions and strengths. This questionnaire should take approximately 20 - 30 minutes to complete. Once completed, you may return your questionnaire to the researcher in the stamped, addressed envelope provided. You will also be asked to indicate whether you are willing to participate in a follow-up interview scheduled at your convenience to discuss your views on these issues in more detail.

Taking part in this study is completely voluntary and you can stop taking part in the study at any time without explanation or prejudice. You may also withdraw any unprocessed data from the study.

Although no distress is anticipated from participating in this research, occasionally, people find certain items a little upsetting, If for any reason you become concerned whilst you are completing the questionnaire, or have any questions in regard to the questionnaire or the study, please advise Deirdre and you will be referred to someone who can help you. Deirdre’s phone and email details are provided below. In addition, 24 hour counselling assistance is available via LIFELINE by calling 13 11 14.

If you know of others that might be interested in this study, please pass on this information sheet to them so they may contact Deirdre to volunteer for the study. The more parents that participate, the more we can learn.

Your responses and contact details will be strictly confidential and your participation anonymous. The data from the study may be used in research publications. However, at no stage over the course of the research, or in subsequent publications, will either you or your son / daughter be identified.

If you have any questions about the study, please contact **Deirdre O’Hare** or her supervisors, **Dr Kerry Anne McBain** or **Professor Edward Helmes**.

Thank you so very much for your time,

*Deirdre*

**Deirdre O’Hare**  
**Doctor of Clinical Psychology Candidate (James Cook University)**

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**Kerry.mcbain@jcu.edu.au; Beryl.buckby@jcu.edu.au**  
**Edward.helmes@jcu.edu.au;**

*If you have any concerns regarding the ethical conduct of the study, please contact:*  
**Sophie Thompson, Human Ethics and Grants Administrator, Research Office**  
**James Cook University, Townsville, Qld, 4811**  
**Phone: (07) 4781 6575 (Sophie.Thompson@jcu.edu.au)**

This administrative form  
has been removed

*Semi Structured Interview: Study Two*

*Interview Questions*

Welcome

Confidentiality and Consent to participate and record (Audio)

Interview (One and a half hour long individual interviews conducted by principal investigator)

Introduction & Point to cover. *Thank you , for agreeing to talk to me about yourself and your child.*

*I have a number of questions that I'll be asking you, and all the other mothers participating in this study.*

*These questions will be about your thoughts in regard to aspects of your child's life that may be affected by Tourette's. Please be assured that everything we talk about today will be completely confidential and if at any time you are not comfortable to respond to any questions, we can stop.*

*We will be speaking for about an hour, and we do have a few questions to get through, so I will try to keep us on track. If you would like to speak to me further, we can arrange that following this part of the interview.*

*Are you OK to start?"*

Let's start with you telling me a little about your *child* (Use name)

*(General Impact)*

- How would you describe your child?
- What was it like when your child was diagnosed with TS?
- How do you feel TS affects your child? (*global well being and specific concerns*) (*prompt as necessary...like family, school, social etc...mother's implicit theory..*)

*(Relationships /attachment/ mother as secure base safe haven)*

- How would you describe your relationship with your child or / tell me about the quality of your relationship? (How does it compare with your relationship with your other child/ren (if applicable)?)
- What, if anything, has changed in any way since the diagnosis?
- How does your relationship with your child differ from /or is the same as your relationship with your own parent? (*mum's attachment history-overview*)
- How does your child act when he is upset?

***If you have any concerns regarding the ethical conduct of the study, please contact:  
Sophie Thompson, Human Ethics and Grants Administrator, Research Office  
James Cook University, Townsville, Qld, 4811  
Phone: (07) 4781 6575 (Sophie.Thompson@jcu.edu.au)***

## APPENDIX A

- What do you do when he is upset?
- How do you feel when he is upset?
- How did you handle your upset when you were a child?
- As a parent, what do you do now when you are upset?
- How does your child get along with other kids? (prompts if necessary:  
*type of friendships (few, lots, close, distant etc.), ability to make and keep friends; other children feel about and interact with your child?*)
- How is your child's social life the same as / or different to other kids?
- How do casual acquaintances act towards your child /and your family?
- How do you think TS affects his social life or your family's social life?(*Invitations, sports, activities, participation, holidays, limited opportunities etc*).

*(Close)*

- What do you feel parents and kids with TS need the most?
- Would you like to add anything?

*Thank you. Check-in and close with details re follow-up.*

**DATA DICTIONARY  
For the Coding Proforma**

**STUDY TWO**

<b>PART ONE - MOTHER CHILD ATTACHMENT</b>	
<b>1. Mother–Child Relationship. MCR</b>	
<i>How mothers described their relationship and role with their TS diagnosed child.</i>	
<ul style="list-style-type: none"> <li>• <b>MCR Close</b> “Unique, close and special” relationship based upon empathy, acceptance and unique understanding of their child, and the perception that the child may have greater needs for mother’s close involvement than non-TS children, often differs from that of relationships with child’s siblings.</li> </ul>	
<ul style="list-style-type: none"> <li>• <b>MCROverinvolved/ Enmeshed</b> Mother may be overinvolved with her child and /or possibly emotionally enmeshed. Mother’s sometimes aware of this, but not necessarily. Characterized by high levels of maternal involvement beyond what might usually be considered age “appropriate” for the child. Enmeshed behaviours and emotions may also be noted. Seems to go a little beyond simply empathizing with child.</li> </ul>	
<ul style="list-style-type: none"> <li>• <b>MCR Lioness</b> Mum as “lioness” – Mum is the child’s protector, advocate, defender etc.</li> </ul>	
<ul style="list-style-type: none"> <li>• <b>MCR Regulate</b> Mum as child’s emotional adjunct-“partner” in emotion regulation. Mum helps the child to manage their emotions</li> </ul>	
<ul style="list-style-type: none"> <li>• <b>MCR Interpreter</b> Mum as “Interpreter and facilitator”, trying to help others to understand and cope with the effects of TS and comorbidity</li> </ul>	
<ul style="list-style-type: none"> <li>• <b>MCRU andme</b> “You and me against the world”- both mother and child are united by feeling isolated, judged, misunderstood and unsupported, either consistently or at various times and across contexts.</li> </ul>	
<ul style="list-style-type: none"> <li>• <b>MCR Rollercoaster</b> The emotional roller coaster experienced within the relationship, with incidences of rage, aggression and depression in the child posing the most difficult emotional challenges for mothers. There is individual variation in mother’s ability to cope with this. Mothers noted particular difficulty associated with aggressiveness during adolescence – a period of developmental transition</li> </ul>	
<ul style="list-style-type: none"> <li>• <b>MCR Witsend</b> A step up in terms of severity of distress experienced by the mother from Rollercoaster. “At wits end” – feeling overwhelmed and/or exhausted and/ or disconnected from the child (Occasional or persistent)</li> </ul>	
<ul style="list-style-type: none"> <li>• <b>MCR Normal</b> Mother striving to have, or to view as “normal” the mother-child relationship and / or the child and / or their lives in general</li> </ul>	
<ul style="list-style-type: none"> <li>• <b>MCR TSGift</b> Mother views Tourette’s (in part) as “a gift” that creates or strengthens interpersonal bonds, or strengthens ‘character’ (of the child, mother or family members) in some way.</li> </ul>	
<ul style="list-style-type: none"> <li>• <b>MCR Atypical</b> Mother describes her mother-child relationship as being unlike the “usual’ mother child relationship.</li> </ul>	



<p><b>MCRChangeTS</b> Does the mother perceive any change in her relationship with her child since the diagnosis of TS? Some may find that the mother-child relationship is closer and or more intense. Change may be driven by several factors including but not limited to the perception (or the reality) of increased “needs” of the child, evoked sense of compassion for the child, mother’s increased anxiety and fear, trauma and grief post diagnosis, guilt and remorse regarding pre-diagnostic parenting behaviour and attitudes to the child, and a sense of injustice that this has happened/ loss of their “ideal” child.</p>
<ul style="list-style-type: none"> <li>• <b>Yes</b> <b>N/A</b></li> </ul>
<ul style="list-style-type: none"> <li>• <b>No</b> <b>Recent change</b></li> </ul>
<p><b>2. Mother’s (mother –child) Relationship Concerns</b> <b>MCC - Specific Concerns</b> <i>expressed by the mothers that impacted the mother –child relationship</i></p>
<ul style="list-style-type: none"> <li>• <b>MCCDependence.</b> Being too close – Blurred boundaries, balance needs for independence /dependence. Possibly overinvolved/enmeshed. Concerns regarding the child’s autonomy and overdependence on mother / caregivers</li> </ul>
<ul style="list-style-type: none"> <li>• <b>MCCFuturechild</b> The mother’s concerns for her child’s future</li> </ul>
<ul style="list-style-type: none"> <li>• <b>MCCFutureRel</b> Mother’s concern regarding how to appropriately “transition” the mother-relationship as the child approaches adulthood</li> </ul>
<ul style="list-style-type: none"> <li>• <b>MCCMumsfuture</b> Mother’s concern for of own future”; <i>what will be my role? Lack of “own” life, “martyred” to TS, lack of identify once child has grown away, no idea what to do when her child has grown.</i></li> </ul>
<ul style="list-style-type: none"> <li>• <b>MCCRuptures</b> The mother-child relationship may be stressed by difficulties, conflicts or distress prompted by:</li> </ul>
<ul style="list-style-type: none"> <li>○ <i>Unanticipated or difficult to understand changes in the child’s behaviour</i></li> </ul>
<ul style="list-style-type: none"> <li>○ <i>Aggressive behaviour and attitudes of the child</i></li> </ul>
<ul style="list-style-type: none"> <li>○ <i>“Normal” developmental transitions (eg. Puberty and Adolescence).</i></li> </ul>
<ul style="list-style-type: none"> <li>• <b>MCCTSConfused ;</b> TS or Not TS? Mother is at times confused and has difficulty disentangling “normal” developmentally appropriate behaviour from unacceptable or maladaptive behaviours and attitudes that may be symptoms of TS and / or co-morbid disorder. Can be particularly problematic for discipline .</li> </ul>
<ul style="list-style-type: none"> <li>• <b>MCCBadmum</b> “Bad Mother” issues ; <i>Mother’s self-criticism</i></li> </ul>
<ul style="list-style-type: none"> <li>○ “Neglect” of siblings- Mother’s concerns regarding the amount of time, attention or the quality of her parenting of her other children.</li> </ul>

<ul style="list-style-type: none"> <li>○ Mum’s self doubt regarding her abilities to mother a child with TS. <i>Can I handle this? Am I equipped? “Out of my depth” etc.</i></li> </ul>
<ul style="list-style-type: none"> <li>○ Guilt, shame and remorse regarding pre-diagnosis parenting of her TS child - trying to make up for it now. It appears to be an enduring feeling, and can be hard for mothers to forgive themselves.</li> </ul>
<ul style="list-style-type: none"> <li>• <b>MCCPressure</b> Maternal pressures (<i>Specific pressures felt by the mothers in their parenting role of their TS child</i>)</li> </ul>
<ul style="list-style-type: none"> <li>○ Need to stay strong/calm for their child</li> </ul>
<ul style="list-style-type: none"> <li>○ Being Overly vigilant- Anticipating disaster</li> </ul>
<ul style="list-style-type: none"> <li>○ Being too exhausted, stressed, anxious and overwhelmed to function &amp; struggling to maintain optimism and hope (occasional or persistent)</li> </ul>
<ul style="list-style-type: none"> <li>○ Having to work everything out / do everything by themselves in the face of ignorance and lack of support</li> </ul>
<ul style="list-style-type: none"> <li>○ Assuming the burden of care – most caring duties born by mum</li> </ul>
<ul style="list-style-type: none"> <li>• <b>MCCAlone</b> The loss of other relationships – Mother’s perceive that they have lost previously important relationships or that level of social support from significant others such as spouse, family and/ or friends has significantly reduced since diagnosis.</li> </ul>
<ul style="list-style-type: none"> <li>• <b>MCCDisconnected</b> Mother feels disconnected / Burnt –out / or a loss or absence of sense of maternal love for their child (transient or pervasive)</li> </ul>

<p><b>3. Mother’s Emotional Response to her Child <b>MERC</b></b> The main emotions the mother expressed (conveyed in words and by tone of voice) regarding her child <i>in general</i>.</p>
<p><b>a. <b>MERC Positive</b></b> (emotions classified as being positive)</p>
<p>Positive: <b>Love</b> and <b>Joy</b></p>
<ul style="list-style-type: none"> <li>• “Maternal Love”- <b>acceptance, understanding, compassion, empathy etc</b></li> </ul>
<ul style="list-style-type: none"> <li>• <b>Pride</b></li> </ul>
<ul style="list-style-type: none"> <li>• <b>Humour and delight</b> and relief</li> </ul>
<p><b>b. <b>MERC Negative</b></b> (emotions classified as being negative)</p>
<p>Negative: <b>Anger, Sadness</b> and <b>Fear</b> plus (Emergent &amp; Shutdown)</p>
<ul style="list-style-type: none"> <li>• (<b>SADNESS</b>) <b>grief, loss, depression, hopelessness, sadness, unhappiness</b></li> </ul>

• (SYMPATHY) Pity, sympathy
• (NEGLECT) Rejection, embarrassment,
• (DISSAPOINTMENT) disappointment, dismay
• (SUFFERING) hurt, anguish
• (SHAME) Guilt, shame, remorse, regret
• (HORROR) Fear, panic, alarm
• (NERVOUSENESS) anxiety, concern, worry, apprehension, distress
• (Emergent) Confused, bewildered, lost, stressed, helpless
• (RAGE) Anger, bitterness, hostility
• (IRRITATION EXASPERATION) Frustration, irritation, exasperation, agitation
• (SHUTDOWN) numb /blunted/ utterly exhausted

<b>Summary</b>
<b>Individuals Mostly Positive :</b>
<b>Mixed Positive and Negatives:</b>
<b>Individuals Mostly Negative:</b>

<p><b>4. Mother’s representations of their child</b></p> <p><i>The classification of the mother’s representation of their child - is based upon the mother’s general description of their child and their behaviours, and the emotions mothers expressed towards the child. This is assessed as being either generally positive, ambivalent or generally negative.</i></p> <p><b>Generally Positive:</b> <i>more positive than negative generally</i>  <b>Ambivalent</b> – <i>balance of both positive and negative (may also seem confused or contradictory)</i>  <b>Generally Negative:</b> <i>more negative than positive generally</i></p>
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<b>5. Mother as Secure Base MSBase</b>
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<p><b>A MERCDistress</b> <i>Mother’s emotional response to child’s distress - The main emotions the mother expressed (conveyed in words and by tone of voice) in response to her child’s distress</i></p>
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- |   |
|---|
| <b>1. MERCDistressPositive</b>  |
| • Love- Empathy, compassion, pity, care, sympathy                               |
| <b>2. MERCDistressNegative</b>  |
| • Sadness- grief, tearfulness, helplessness, hopeless, guilt, despair, sympathy |
| • Fear- Concern, anxiety, stress  |

• Anger- frustration, irritation, sense of injustice, self pity
• Restrained or Effort to suppress
• Overwhelmed, paralyzed, numb, blunted
<b>SUMMARY (negative only)</b>
<b>NEGATIVE</b> (Mostly Negative and/or Overwhelmed/Numb etc):

**B MBR Mother’s Behavioural response to child’s distress** – The actions mother’s describe taking in response to their child’s distress

• <b>MBRTalk</b> Talking it through (mum uses cognitive and emotional strategies to process feelings and solve problems etc.)
• <b>MBRPhysical</b> Mother provides physical comfort
• <b>MBRStrength/calm/control</b> Mother provides and models “calm” and “strength” by controlling her own emotions and behaviours, to help the child
• <b>MBRDistraction</b> Using distraction
• <b>MBREmpathise</b> Empathizing with the child
• <b>MBRRescue</b> “Mum to the rescue” - mother very actively responds, often using multiple means to help the child, depending upon the situation. Mum’s can sometimes make a considerable effort to be innovative or to find best ways to help.
• <b>MBRLogicalDetach</b> Mother remains logical and tends towards being detached
• <b>MBRShutdown</b> Mother is Immobile, overwhelmed or experiences helplessness – she can not or will not respond to the child’s distress (transient or persistent)

<b>SUMMARY</b>
<b>Active and Positive:</b>
<b>Active positive + sometimes overwhelmed /immobilized /disconnected:</b>
<b>Immobilized and Negative:</b>
<b>MOTHER - CHILD ATTACHMENT (SUGGESTED ONLY)</b>
<b>SECURE</b>
<b>SECURE + signs of overinvolvement /enmeshment</b>
<b>INSECURE + signs of overinvolvement/enmeshment</b>
<b>INSECURE- Ambivalent</b>

<b>PART TWO (A) - MOTHER’S ATTACHMENT</b>
<b>Mother’s Attachment Style</b>

<b>1. MAP Mother's Childhood Attachment (Parents)</b>	
• <b>MAPSecure.</b> Mother describes a positive affective bond (in her childhood) with her primary carer (usually her mother)	
• <b>MAPInsecure</b> Mother reports the lack of a positive affective bond (in her childhood) with her primary. Poor quality parenting, neglect or abuse may be reported.	
• <b>N/A: Information not available</b>	
<b>2. MP Mother's presentation as she discussed her attachment history (<i>Insecure only - determined by tone of voice and text</i>)</b>	
• Matter of fact	
• Distressed	
• Saddened	
• Angry	
• Resigned	
• Presenting a "brave face"	
• Reconciled and forgiving	
• Shutdown	N/A:
<b>3. Mother's reported experience of secure base (own parents secure base availability) <b>MABase</b> <i>Did the mother report having access to her primary caregiver as a source of emotional support?</i></b>	
<b>A. MABase Present</b>	
<b>B. MABase Absent</b>	N/A:
<b>4. Mother's Mode of comforting self as a child <b>MAS3comfort</b> <i>What did the mother report doing as child when she was distressed?</i></b>	
<b>A. MAS Secure</b> (use parental figure for comfort)	
<b>B. MAS Insecure</b> (autonomous- take care of self)	N/A:
<b>5. Mother's Mode of comforting self as an adult <b>MAC</b> <i>What did the mother report doing as an adult (now) when she is distressed?</i></b>	
<b>A. MAC Secure-</b> (seek comfort from partner/significant other)	
<b>B. MAC Insecure-</b> (no reference to comfort from others)	N/A:
<b>PART TWO (B) - MOTHER'S PARENTING STYLE</b>	
<b>Mother's self-reported parenting style <b>MPS</b> <i>Did mothers report that their parenting style was the same as or different to the way they were parented?</i></b>	
• <b>MPSame</b>	
• <b>MPDifferent</b>	N/A:

***Mother's rationale of differences: Mothers reports of what the primary differences were.***

- **Generational** (different approaches /methods to parenting across generations)
- **Talk** (*Mother talks to her child more*)
- **Involved** (*Mother is more involved*)
- **Knowledge** (*Mother has more or different knowledge re parenting*)
- **Impact** (*Mother knows the impact that bad parenting can have from experience*)
- **Loving/open** (*Mother is more open and loving*)
- **Acceptance /understanding** (*Mother is more accepting and understanding*)
- **Situations too different to compare :** (*Mother couldn't compare her parenting situation with that of her own childhood*)

**PART THREE - CHILD'S ATTACHMENT TO PEERS**

**Mother's reports of aspects of her child's attachment to peers CAP including:**

**1. Sociability CAPSoc** *Mother reports her child's current level of friendships.*

- Has no friends
- Close relationship only with parents
- Close relationships only with siblings or other family members
- Gets on better with those younger or older
- Has friends but no close friendships
- Has a few good close same aged friends
- Has a few close same aged friends from early childhood days
- Has a few close friends from school
- Has a few close friends with (primarily) peers with difficulties (eg. LD etc)
- Has a normal social life with some close friends, acquaintances and gets on well with class-mates
- Is very gregarious and popular, has many peer friends and acquaintances
- Has a twin

**2. Desire for friendships CAPDes** *Mother reports her child's current desire and satisfaction with current level of friendships*

- Prefers own company / Not interested in making friends
- Interested in close relationship with immediate and /or extended family members

• Interested in friends who are older or younger than him
• Prefers adult company
• Is nervous of making friends
• Would like to have more friends
• Has difficulty maintaining relationships with any children, including same-aged peers
• Enjoys being with same aged friends, but for limited periods of time e.g. at school but not weekends
• Friendships prevented / impaired by bullying, teasing or social rejection by same-aged peers
• Enjoys having very small group of close friends (a few), but not interested in classmates etc.
• Enjoys being with groups of peers through activities such as sport
• Enjoys a “normal” social life, with several close friends, normal classroom relationships and has acquaintances
• Is highly sociable, charismatic and popular with same aged peers and others
• Number and quality and desire for friendships improved by late adolescence. Has some friends but none are close
• Wants (2) / has (1) a romantic relationship

<b>4. Mother’s perception of Factors effecting their child’s socialization with peers:</b>
<b>1. Positive:</b> ( <i>Mother reports on what she feels makes it easier for her child to make and maintain friendships</i> )

1. Accepted understood (the child is)
2. Age (of the child)
3. Availability of “Like” kids
4. Can defend self (the child)
5. Has friends but are not his Peers (the child)
6. Likes school (the child)
7. Longstanding friends (the child has)
8. Parental support/facilitation (of the child)
9. Positive Behaviours (the child’s)
10. Positive Personality (the child’s)
11. Sense of humor (the child’s)
12. Shared interests (the child shares interests with others)
13. Shy but OK (the child)
14. Social skills (the child’s)
15. Sporty (the child’s)
16. Supportive school environment (the child’s)
17. Talents/ability (not sport) (the child’s)
18. Unselfconscious re TS (the child is)

19. Kids that defend/ support the TS child/teen
20. Having less severe tics (the child's)
<b>2. Negative</b> ( <i>Mother reports on what she feels makes it <b>more difficult</b> for her child to make and maintain friendships</i> )
1. Bullying/teasing (of the child by others)
2. Co-morbid factors (the child's)
3. Denial of TS (the child's)
4. Being Different to peers (the child)
5. Low acceptance/understanding (by others)
6. Low interest (the child's)
7. Negative behaviours (the child's)
8. Negative personality (the child's)
9. No like kids available (to the child)
10. Not sporty (the child)
11. Other peer reactions (to the child)
12. Poor social skills (the child's)
13. School aversion (the child's)
14. Self conscious re TS (the child is)
15. Social exclusion/stigma (of the child)
16. Having Tics (the child)
17. Unsupportive schools
18. Behaviour/Tics too hard for peers to cope with (the child's)
19. Can't defend self (the child)
<b>Overall Assessment of Attachment to Peers CAPOverall</b>
1. <b>Overall Peer Secure</b> (Having peer friends, being satisfied with the level of friendships and not having difficulty making or maintaining friendships with peers)
2. <b>Overall Peer Insecure</b> (Not having any peer friends, being dissatisfied with the current level of friendships and/or having difficulty making or maintaining friendships with peers)
3. <b>Moving from insecure to secure (during mid-adolescence)</b> Level and quality of peer friendships, desire for friendships and ability to make and maintain friendships improving as the teen reaches mid to late adolescence.
<b>PART FOUR - GENERAL FINDING'S RELATING TO THE MOTHER'S EXPERIENCES OF PARENTING A CHILD WITH TOURETTE'S MPE</b>
<ul style="list-style-type: none"> <li>• <b>Mothers assumes BURDEN of care</b> * Mother's do the lions share of the caring. This may involve self-sacrifice / Determination to do the best by their child etc.</li> <li>• <b>Mother forced to TAKECHARGE/EXPERT/ADVOCATE</b> * Mother's need to "take charge" (becoming the experts, educators and advocates for</li> </ul>



<p>their child and their families) in response to generally high levels of ignorance re TS within the medical, academic and community contexts</p>
<ul style="list-style-type: none"> <li>• <b>BRIDGE to the outside world</b> * the importance of connecting with other’s who “get it” e.g. Peer support (TSAA, TSAV, TS Mothers support groups), understanding medical professional or teachers- to counter the isolation, distress and difficulty associated with the lack of acceptance and understanding of TS by others (including extended family and friends in some cases family)</li> </ul>
<ul style="list-style-type: none"> <li>• <b>FAMILY FIRST- (Home as Sanctuary)</b> * Focus on and “surviving as a nuclear family” (closeness, pulling together, balancing siblings needs, support of siblings, holidays, normal life, home as sanctuary. Can be forced by isolation and lack of support from family and friends)</li> </ul>
<ul style="list-style-type: none"> <li>• <b>LIVING WORRIED (stress, anxiety, coping difficulties etc.) &amp; STRUGGLE to maintain optimism and effort</b> * Living “worried” ...riding the emotional roller coaster life, as tics wax and wane; as crises of confidence in parenting abilities arise in the face of the TS child’s needs; Frustration and despair over lack of treatment options, concern over medication side effects, lack of prognosis re course of TS etc. “Staying in the struggle” (battling exhaustion and despair, fear of the future against hope and optimism)</li> </ul>
<ul style="list-style-type: none"> <li>• <b>CRITICAL TIMES (Diagnosis; Adolescence; new school year, commencing high school etc.)</b> * Commonly experienced increases in problems and difficulties that associated with specific times or events.</li> </ul>
<ul style="list-style-type: none"> <li>• <b>SERIOUS/CREDIBLE (Battling ignorance, fighting for understanding and Acceptance)</b> Mother’s identify the widespread lack of understanding and acceptance of their children, themselves and TS as the critical contributor to the adversity that they experience (including the negative effects of stereotypes, misinformation and stigma).</li> </ul>
<ul style="list-style-type: none"> <li>• <b>SOCIAL ISOLATION (child / mother/ family)</b> Mothers, their children and their families may feel socially isolated. They may report loss of relationships and lower levels of social support. They can experience difficulty maintaining ‘normal’ social lives by having to restrict some activities or by being social excluded.</li> </ul>
<ul style="list-style-type: none"> <li>• <b>SCHOOL as a key player</b> Mother’s identify the critical role that the school plays in their child/teens lives. The positive contributions made by supportive schools, teachers, assistance and policies are a key factor in maximizing the child’s development, in addition to the wellbeing of both the child and the mother.</li> </ul>
<ul style="list-style-type: none"> <li>• <b>DIAGNOSIS</b> – Getting diagnosed (difficult process and traumatic experience). Highly charged emotional process; often very traumatic. Often requiring mother to drive a very difficult process (primarily attributed to low levels of awareness of TS within the medical and academic communities, the tendency not to take mother’s observations and intuitions seriously, and the difficulty gaining access to specialists etc.)</li> </ul>
<ul style="list-style-type: none"> <li>• <b>Difficult process</b>    Yes            No</li> </ul>
<ul style="list-style-type: none"> <li>• <b>DIAGNOSIS - Emotionally traumatic</b> -Yes            Relief- Yes</li> </ul>

- **GRIEF/LOSS of IDEAL CHILD** Mother's specifically refer to their grief regarding the loss of their "ideal" child (as a result of diagnosis with a chronic condition) - The loss of the hopes and dreams they had for themselves as parents and their children. This can be compounded by the diagnosis of a neurodevelopmental disorder at an unexpected time (i.e. early / middle childhood or teens), when for many the onset of symptoms and diagnosis follows a 'normal' infancy and early childhood.
- **TANGLED (problems re TS, comorbid vs "normative" behavioural issues)** Difficulty disentangling TS and Comorbid symptoms from 'normative' unacceptable or maladaptive childhood or adolescent behaviour,

## DATA CODING PROFORMA: STUDY TWO

<b>MOTHER CHILD ATTACHMENT</b>	
<b>1. Mother–Child Relationship. <u>MCR</u></b>	
<i>How mothers described their relationship and role with their TS diagnosed child.</i>	
• <b>MCR Close</b>	
• <b>MCREnmeshed</b>	
• <b>MCRLioness</b>	
• <b>MCRRegulate</b>	
• <b>MCRInterpreter</b>	
• <b>MCRUandme</b>	
• <b>MCCRollercoaster</b>	
• <b>MCRWitsend</b>	
• <b>MCRNormal</b>	
• <b>MCRTSGift</b>	
• <b>MCRAtypical</b>	
<i>Did MCR relationship change with diagnosis? <b>MCRChangeTS</b></i>	
• <b>Yes</b>	<b>N/A</b>
• <b>No</b>	<b>Recent change</b>
<b>2. Mother’s (mother –child) Relationship Concerns <u>MCC</u></b>	
• <b>MCCDependence</b>	
• <b>MCCFuturechild</b>	
• <b>MCCFutureRel</b>	
• <b>MCCMumsfuture</b>	
• <b>MCCRuptures</b>	
○ <i>Unanticipated or difficult to understand changes in behaviour</i>	
○ <i>Aggressive behaviour and attitudes</i>	
○ <i>“Normal” developmental transitions (eg. adolescence).</i>	
• <b>MCCTSConfused</b>	
• <b>MCCBadmum</b> “Bad Mother” issues	
○ “Neglect” of siblings	
○ Mum’s self doubt regarding her abilities to mother a child with TS.. Can I handle this? Am I equipped? “Out of my depth”	

<ul style="list-style-type: none"> <li>○ Guilt, shame and remorse regarding pre-diagnosis parenting- trying to make up for it now. It appears to be an enduring feeling, hard to forgive themselves.</li> </ul>
<ul style="list-style-type: none"> <li>• <b>MCCPressure</b> Maternal pressures</li> </ul>
<ul style="list-style-type: none"> <li>○ Need to stay strong</li> </ul>
<ul style="list-style-type: none"> <li>○ Overly vigilant- Anticipating disaster</li> </ul>
<ul style="list-style-type: none"> <li>○ Being too exhausted, stressed, anxious and overwhelmed to be the mother the child needs &amp; difficulty “Staying in the struggle”- maintaining optimism and hope (occasional or persistent)</li> </ul>
<ul style="list-style-type: none"> <li>○ Having to work everything out / do everything by themselves in the face of ignorance and lack of support</li> </ul>
<ul style="list-style-type: none"> <li>○ Assuming the burden of care</li> </ul>
<ul style="list-style-type: none"> <li>• <b>MCCAlone</b></li> </ul>
<ul style="list-style-type: none"> <li>• <b>MCCDisconnected</b></li> </ul>

<b>3. Mother’s Emotional Response to her Child <u>MERC</u></b>
<b>a. MERC Positive</b>
Positive: <b>Love</b> and <b>Joy</b>
<ul style="list-style-type: none"> <li>• “Maternal Love”- <b>acceptance, understanding, compassion, empathy etc</b> <b>Pride</b> <b>humour and delight</b> and relief</li> </ul>
<b>b. MERC Negative</b>
Negative: <b>Anger, Sadness</b> and <b>Fear</b> plus (emergent- Shutdown)
<ul style="list-style-type: none"> <li>• (SADNESS) grief, loss, depression, hopelessness, sadness, unhappiness</li> <li>• (SYMPATHY) Pity, sympathy</li> <li>• (NEGLECT) Rejection, embarrassment,</li> <li>• (DISSAPOINTMENT) disappointment, dismay</li> <li>• (SUFFERING) hurt, anguish</li> <li>• (SHAME) Guilt, shame, remorse, regret</li> <li>• (HORROR) Fear, panic, alarm</li> <li>• (NERVOUSENESS) anxiety, concern, worry, apprehension, distress</li> <li>• (Emergent) Confused, bewildered, lost, stressed, helpless</li> <li>• (RAGE) Anger, bitterness, hostility</li> <li>• (IRRITATION EXASPERATION) Frustration, irritation, exasperation, agitation</li> <li>• (SHUTDOWN) numb /blunted/ utterly exhausted</li> </ul>

<b>Summary</b>
<b>Positive-</b> Love and Joy <b>Negative –</b> Sadness Fear <b>Anger</b>
<b>Individuals Mostly Positive or Mixed Positive and Negatives:</b>
<b>Individuals Mostly Negative (including Shutdown):</b>
<b>N/A:</b>

#### **4. Mother's Narrative representations of their child (SUGGESTED)**

*Based on a short written narrative summarizing each mother's description of their child's characteristics and behaviours. Mother's words used where possible. Then the overall impression of the narrative is assessed as being either generally positive, ambivalent or generally negative*

**Generally Positive:**

**Ambivalent/confused or contradictory:**

**Generally Negative:**

#### **5. Mother as Secure Base MSBase**

**A MERCDistress** *Mother's emotional response to child's distress*

##### **1. MERCDistressPositive**

- Love- Empathy, compassion, pity, care, sympathy

##### **2. MERCDistressNegative**

- Sadness- grief, tearfulness, helplessness, hopeless, guilt, despair, sympathy
- Fear- Concern, anxiety, stress
- Anger- frustration, irritation, sense of injustice, self pity
- Restrained or Effort to suppress
- Overwhelmed, paralyzed, numb, blunted

#### **SUMMARY**

**Mostly Positive**

**Mixed Positives and Negatives**

**NEGATIVE** (Mostly Negative and/or Overwhelmed/Numb etc):

#### **B MBR Mother's Behavioural response to child's distress**

- **MBRTalk** Talking it through (cognitive and emotional strategies)
- **MBRPhysical** mother provides physical comfort
- **MBRStrength/calm/control** mother Providing "strength", modelling "strength"

<ul style="list-style-type: none"> <li>• <b>MBRDistract</b>ion Using distraction</li> </ul>
<ul style="list-style-type: none"> <li>• <b>MBREmpathise</b> Empathizing with the child</li> </ul>
<ul style="list-style-type: none"> <li>• <b>MBRRescue</b> “Mum to the rescue”</li> </ul>
<ul style="list-style-type: none"> <li>• <b>MBRLogicalDetach</b> mother remains logical and tends towards being detached</li> </ul>
<ul style="list-style-type: none"> <li>• <b>MBRShutdown</b> mother is Immobile, or experiences helplessness</li> </ul>
<b>SUMMARY</b>
<b>Active and Positive:</b>
<b>Active positive + sometimes overwhelmed /immobilized /disconnected:</b>
<b>Immobilized and/or Mostly Negative:</b>
<b>MOTHER - CHILD ATTACHMENT (SUGGESTED)</b>
<b>SECURE</b>
<b>SECURE + signs of over-involvement/enmeshment</b>
<b>INSECURE + signs of over-involvement /enmeshment</b>
<b>INSECURE- Ambivalent</b>

<b><u>MOTHER’S ATTACHMENT</u></b>	
<b>Mother’s Attachment Style (<i>SUGGESTED</i>)</b>	
<b>1. MAP Mother’s Childhood Attachment (Parents)</b>	
<ul style="list-style-type: none"> <li>• <b>MAPSecure.</b></li> </ul>	
<ul style="list-style-type: none"> <li>• <b>MAPInsecure</b></li> </ul>	N/A:
<b>2. MP Mother’s presentation as she discussed her attachment history (<i>Insecure only</i>)</b>	
<ul style="list-style-type: none"> <li>• Matter of fact</li> </ul>	
<ul style="list-style-type: none"> <li>• Distressed</li> </ul>	
<ul style="list-style-type: none"> <li>• Saddened</li> </ul>	
<ul style="list-style-type: none"> <li>• Angry</li> </ul>	
<ul style="list-style-type: none"> <li>• Resigned</li> </ul>	
<ul style="list-style-type: none"> <li>• Presenting a “brave face”</li> </ul>	
<ul style="list-style-type: none"> <li>• Reconciled and forgiving</li> </ul>	
<ul style="list-style-type: none"> <li>• Shutdown</li> </ul>	N/A:
<b>3. Mother’s reported experience of secure base (own parents secure base availability) <b>MABase</b></b>	
<b>C. MABase Present</b>	

<b>D. MABase Absent</b>	<b>N/A:</b>
<b>4. Mother's Mode of comforting self as a child MAS3comfort</b>	
<b>C. MAS Secure</b> (parental figure for comfort)	
<b>D. MAS Insecure</b> (autonomous)	<b>N/A:</b>
<b>5. Mother's Mode of comforting self as an adult MAC</b>	
<b>C. MAC Secure-</b> seek comfort from partner/significant other	
<b>D. MAC Insecure-</b> no reference to comfort from others	<b>N/A:</b>

<b><u>MOTHER'S PARENTING STYLE</u></b>	
<b>Mother's self-reported parenting style MPS</b>	
<ul style="list-style-type: none"> <li>• <b>MPSame</b></li> <li>• <b>MPDifferent</b> <b>N/A:</b></li> </ul>	
<b><i>Mother's rationale of differences:</i></b>	
<ul style="list-style-type: none"> <li>• <b>Generational</b></li> <li>• <b>Talk</b></li> <li>• <b>Involved</b></li> <li>• <b>Knowledge</b></li> <li>• <b>Impact</b></li> <li>• <b>Loving/open</b></li> <li>• <b>Acceptance /understanding</b></li> <li>• <b>Situations too different to compare</b></li> </ul>	

<b><u>CHILD'S ATTACHMENT TO PEERS</u></b>	
<b>Mother's reports of aspects of her child's attachment to peers CAP including:</b>	
<b>1. Sociability CAPSoc</b>	
<ul style="list-style-type: none"> <li>• Has no friends</li> <li>• Close relationship (only) with parents</li> <li>• Close relationships only with siblings or other family members</li> <li>• Gets on better with those younger or older</li> <li>• Has friends but no close friendships</li> <li>• Has a few good close same aged friends</li> <li>• Has a few close same aged friends from early childhood days</li> <li>• Has a few close friends from school</li> </ul>	

• Has a few close friends with (primarily) peers with difficulties (eg. LD etc)
• Has a normal social life with some close friends, acquaintances and gets on well with class-mates
• Is very gregarious and popular, has many peer friends and acquaintances
• Has a twin

## 2. Desire for friendships CAPDes

• Prefers own company / Not interested in making friends
• Interested in close relationship with immediate and /or extended family members
• Interested in friends who are older or younger than him
• Prefers adult company
• Is nervous of making friends
• Would like to have more friends
• Has difficulty maintaining relationships with any children, including same-aged peers
• Enjoys being with same aged friends, but for limited periods of time e.g. at school but not weekends
• Friendships prevented / impaired by bullying, teasing or social rejection by same-aged peers
• Enjoys having very small group of close friends (2-4), but not interested in classmates etc.
• Enjoys being with groups of peers through activities such as sport
• Enjoys a “normal” social life, with several close friends, normal classroom relationships and has acquaintances
• Is highly sociable, charismatic and popular with same aged peers and others
• Number and quality and desire for friendships improved by late adolescence. Has some friends but none are close
• Wants (2) / has (1) a romantic relationship

## 4. Mother’s perception of Factors effecting their child’s socialization with peers:

### 1. Positive:

21. Accepted understood
22. Age
23. Availability of “Like” kids
24. Can defend self
25. Has friends but are not his Peers
26. Likes school
27. Longstanding friends
28. Parental support/facilitation
29. Positive Behaviours



30. Positive Personality
31. Sense of humor
32. Shared interests
33. Shy but OK
34. Social skills
35. Sporty
36. Supportive school environment
37. Talents/ability (not sport)
38. Unselfconscious re TS
39. Kids that defend/ support the TS child/teen
40. Having less severe tics

## 2. Negative

20. Bullying/teasing
21. Co-morbid factors
22. Denial of TS
23. Different to peers
24. Low acceptance/understanding
25. Low interest
26. Negative behaviours (OTT etc.)
27. Negative personality
28. No like kids available
29. Not sporty
30. Other peer reactions
31. Poor social skills
32. School aversion
33. Self conscious re TS
34. Social exclusion/stigma
35. Having Tics
36. Unsupportive schools
37. Behaviour/Tics too hard for peers to cope with
38. Can't defend self

### Overall Assessment of Attachment to Peers **CAPO**Overall (SUGGESTED)

4. <b>Overall Peer Secure</b>
5. <b>Overall Peer Insecure</b>
6. <b>Move from insecure to secure (during mid-adolescence)</b>

### GENERAL FINDING'S RELATING TO THE MOTHER'S EXPERIENCES OF PARENTING A CHILD WITH TOURETTE'S **MPE**

- **Mothers assumes BURDEN of care \***

• <b>Mother forced to TAKECHARGE/EXPERT/ADVOCATE *</b>
• <b>BRIDGE to the outside world *</b>
• <b>FAMILY FIRST- (Home as Sanctuary) *</b>
• <b>LIVING WORRIED (stress, anxiety, coping difficulties etc) * &amp; STRUGGLE to maintain optimism and effort *</b>
• <b>CRITICAL TIMES - Diagnosis; Adolescence- puberty; New school year, commencing high school etc *</b>
• <b>SERIOUS/CREDIBLE (Battling ignorance, fighting for understanding and Acceptance)</b>
• <b>SOCIAL ISOLATION (child and/or mother and or family)</b>
• <b>SCHOOL as a key player</b>
• <b>DIAGNOSIS - Difficult process</b> Yes <span style="float: right;">No</span>
• <b>DIAGNOSIS - Emotionally traumatic -Yes</b> <span style="float: right;">Relief-</span> Yes <span style="float: right;">No</span>
• <b>GRIEF/LOSS of IDEAL CHILD (compounded by diagnosis at an unexpected time – i.e. early /middle childhood or teens)</b>
• <b>TANGLED (problems disentangling behaviours/symptoms- re TS, comorbidity, and “normative”)</b>

**Data Dictionary for “Big-Five” Personality factors****(Goldberg, 1993; Costa & McCrae, 1992)****STUDY ONE PART B**

**1. Extraversion**- *Personality and behavioural traits characterized by the tendency towards being outgoing, energetic and sociable versus solitary, reserved and less active (introversion). Includes (but is not restricted to) energy, assertiveness, positive emotions, talkativeness, sociability and seeking stimulation in the company of others.*

**a. High Extraversion** “Friendly, sociable, happy, fun, enjoys (likes) the company of others (other kids), being talkative, chatty, likeable, popular, assertive, energetic, active, over-the-top, etc”.

**b. Low Extraversion** (introversion)- “shy, low(or no) interest in others, prefers for own company, prefers to be one on one”.

**2. Neuroticism**- *Personality and behavioural traits characterized by the tendency to be Sensitive and Anxious vs confident and secure. To experience negative emotions (such as fear, anger and depression) and being more emotionally unstable, moody, and impulsive.*

**c. High Neuroticism** –reference to child’s Neuroticism most noted were “Anxious, fearful, self-consciousness, low confidence, low self esteem, worry, emotionally unstable, moody, aggressive, angry, embarrassed, paranoid, etc”

**d. Low Neuroticism** - “Confident, optimistic, easygoing, happy go lucky, cheerful, emotionally stable, unselfconscious, high self-esteem etc”.

**3. Agreeableness**- *Personality and behavioural traits characterized by the tendency towards being compassionate, warm and interested in others versus hostile, unkind and cool. A tendency towards being compassionate, understanding and co-operative as opposed to being suspicious, antagonistic or disinterested regarding others.*

**e. High Agreeableness** Being “sensitive to others (needs), empathic, warm, caring, helpful, able to listen, personable, nice etc”.

**f. Low Agreeableness-** “aggressive , bossy, “over the top”, controlling etc”  
“Wanting games to be played on his terms, according to complicated rules”

**4. Openness** – *Personality and behavioural traits characterized by the tendency towards being open to experience versus being closed minded and cautious. A tendency towards being able to examine and know one’s own feelings, intellectual curiosity, creativity, appreciation of the different and the unusual versus closed mindedness, lack of curiosity and lack of appreciation of novelty, preference for consistency and the status quo.*

**g. High Openness** - being “open” with others, open to own feelings-self accepting, able to advocate for self, being “open” re tics and TS (oh its TS; , being open-minded, curious, “intelligent, creative, enthusiastic, nonjudgmental.”  
Bright, musical,

**h. Low Openness** - associated with being dogmatic, closed-minded- “he bosses children around when playing with them”, “won’t talk about his TS..”

**5. Conscientiousness-** *Personality and behavioural traits characterized by the tendency towards being efficient and organized versus disorganized and impulsive. Being highly organized rather than spontaneous can in the extreme appear close to obsessive, compulsive and ritualistic/repetitive behaviours.*

**i. High Conscientiousness** - being overly rule-focused (has trouble playing ”or obsessive- compulsive eg. “Wanting games to be played on his terms, according to complicated rules”

**j. Low Conscientiousness** - can be associated with being “inattentive” and “impulsive”

**CODING Key for Survey Data****STUDY ONE PART B.****FACTORS**

*Text in BLUE = Factors associated with **POSTIVE IMPACT** on Child's ability to make friends*

*Text in RED = Factors associated with **NEGATIVE IMPACT** on Child's ability to make friends*

1. Characteristics and behaviours indicating the **“Big-Five” Personality factors**

E	Extraversion	hE (high)	lE (low)
N	Neuroticism	hN	l.N
O	Openness	hO	lO
A	Agreeableness	hA	lO
C	Conscientiousness	hC	lC

2. **“OTHER”** (non –personality) Factors

1. Social/communication skills (aside from personality characteristics)- “High SS” or “Low SS”
2. Specific reference to low Social Interest “SI” (High SI or Low SI)
3. Specific reference to sports participation “Sport”
4. Specific reference to parental support, facilitation or modelling “P”
5. Specific reference to activities and interests “A/I”
6. Specific reference to School (context/environment etc) “PosSchool” & Specific reference to Negative School “Neg School”
7. Specific reference to opportunity to develop social skills or make friendships (not via sport or shared interest- activity) “Opport”.
8. Context (contextual factors such as geographic location) “Context”
9. Sibling factors “SIB”
10. Specific reference to sense of humour/fun “Fun”
11. Specific reference to friendships with similar children “Like”
12. Specific reference to Negative response from others (bullying, teasing, social exclusion etc) “NegOthers” & Specific reference to Supportive or Positive behaviour of others “PosOthers”
13. Specific reference to TICS (mild, Control Waning, open, advocate etc) “TicOK” & Specific reference to Tics (severity, type, self consciousness – negative impacts) “TicNeg”
14. Specific reference to child being or feeling different “DIFF”
15. Specific reference to child's maladaptive and co–morbid behaviours /symptoms “MAL.”
16. Specific reference to child having Long-term friends LONG
17. Specific reference to Understanding and Acceptance (UA)
18. Specific reference to Age or passage of Time (makes things easier-improves or young so not a problem) “Age/time”
19. Specific reference to a preference for older or younger friends. OYOK
20. Specific reference to child having trouble maintaining friends MAINT

21. Specific reference to preference for I – small group of friends *text in in italics*

**APPENDIX C.****STUDY TWO****Maternal Narrative Descriptions of Her Child**

The following short narratives were constructed from the mother's own words (*indicated in italics*) and the findings from the data analyses regarding the mother's general emotional response to her child (As reported in the Results Section). Some mothers were more forthcoming and articulate regarding their children than others; therefore there is notable variability in the level of detail and amount of content demonstrated between narratives.

An approximation (indication) of the quality of the Mother's Representation of her Child was made:

- A. **Generally Positive** (mother appeared to describe and express more positive than negative child characteristics and general emotional responses towards her child)
- B. **Ambivalent or Contradictory MIXED–Positives and negatives** (mother appeared to describe and express a fairly even distribution of positive and negative child characteristics and general emotional responses towards her child)
- C. **Generally negative** (mother appeared to describe and express more negative than positive child characteristics and general emotional responses towards her child)

**A & J*****Summary.***

- **Representation- MIXED.** Always some positives, deep compassion, but high level of needs provoked strong stress and anxiety regarding her son's dependence, "neediness" and aggressiveness. These problems are improving with maturity, and mother holds a slightly more positive and optimistic view of him and his future as a result.
- (Mother's emotional response to child in General): **Mixed.** Positives & lots of sad, fear and a little anger

### **Representation**

MIXED but *becoming a little more positive now that her son is getting older* and mother is seeing positive and hopeful change. In childhood, mother and son were *loving and close* but she made lots of reference to negative representations of him as a *needy, tearful, quiet, dependent child- shy, worrisome* (and who provoked a lot of emotional response from mother), *so different from his brother*. Then his *aggressive* adolescent behaviours and peaking *irritating* tics made it even harder for her, heightening her ambivalent feelings towards him. It appeared to be difficult for mother to discuss the negative aspects but she was clear relieved as she proudly discussed his current progress and the hope that this brings. Her tone of voice reflected these variations in her feelings as she described her son.

*J. is Compassionate, good, always was quiet, everyone loves him, so different to his brother, J. is very caring, wimpy.....but don't mean that in a bad way...soft, big young man now...used to be clumsy, if something bad was to happen (like falling over) it would happen to Josh...reserved. Not keen to do things with us like going to a beach or somewhere....getting more confidence now as he is getting older and doing better.....has always had a few close friends, but never been the one to initiate social stuff...happy enough to do things by himself.....agro as a young teenager,.... temper and not his usual self....He was a very "needy" child. Had to do everything for him...he stuck to his dad and I all the time.*

### **B & D.**

#### **Summary.**

- **Representation- MIXED** Positive and negative, reflecting recent adolescent behavioural changes (autonomy)
- (Mother's emotional response to child in General): **Mixed:** Positive and negatives sadness, fear and anger (part related to frustration, hurt and anger re adolescent behavioural changes and resulting mother- child conflict)

### **Representation**



Mother describes both the positive and negative characteristics of her son. Most difficulties were associated with changes accompanying adolescence. Conflict and ruptures are very difficult for them both. He is striving for autonomy and mother acknowledged her *over-protectiveness* as a problem. There seems to be a transfer of attachment (developmentally appropriate) but this is creating difficulty in mother-child relationship, with noted changes over past 12 months in increased aggressiveness, conflict and anger. Her tone of voice reflected these variations in her feelings as she described her son.

*“He gets on well with those older (adults) and especially well with younger kids... “got lots of young kids around him and he gets along fantastically with them all... I think he’s look he’s one of those kids that um is really helpful and friendly. I don’t think he’s got a nasty bone in his body at all. BUT...Agro since his teens....we have big blues...he frustrates the hell out of me. Has OCD traits and can’t give up on things. (These lead to distressing arguments with this mother, then afterwards he is very remorseful... “sorry mum”...). And I think it’s made even worse because his outside, outward appearance to everybody else is just you know loveable child who’d do you know go out of his way to help you and he is generally like that, so he doesn’t have that sort of behaviour for anybody else it seems to be reserved specifically for me. So people sort of look at me like I’m the fruit loop”* . (Mother feeling judged by others and isolated. Only she is privy to range of negative behaviour).

*“And he yeah he’s not afraid to sort of get out there. ....I think that D ...he’s insecure in some ways he’s quite confident in others. His self-esteem has been up and down the last few years. .... probably had enough positive people around him to sort of you know have some more ups than downs but he does have and especially like when we’ve had arguments together .... he has real concerns at school he doesn’t mix a lot with them (his small group of friends) on the weekend.....generally he has a fairly good group of friends and they’ve been pretty supportive but he has the odd one or two that like as I try to tell him they’re not really your friends. I think D’s going to have to be multi millionaire to have all the things he says he’s going to have in his life! D. has become a lot more aggressive I suppose in the last oh 12 months I would say but then like you know how much... I have often wondered how much of that is normal teenage behaviour and how much of it is the Tourette’s.*

**D & T*****Summary.***

- ***Representation- Generally Positive*** : Very positive, loving and proud. Mother is intolerant of those who don't understand his TS, and slightly disturbed by recent uncharacteristic but increasingly aggro behaviour (coinciding with puberty)
- (Mother's emotional response to child in General): ***Positive mostly***, (love and joy) with a little negative (guilt, anxiety, irritation)

***Representation.***

Mother's tone of voice is strong and proud as she speaks of her son. Annoyed and frustrated tone when she discussed the failure of others to cope with her son and his TS. A little hesitant in tone when discussing his aggressiveness... the only time her responses wavered at all. Otherwise her *representation of him is wholly positive*, and mother was proud and confident in her responses. *Very small amount of negativity* re his aggressiveness and her fear of what that may mean in the future if not well controlled. Lots of fondness and devotion in her tone of voice too.... "*very close since birth. As he was premmie.*"

There was some indication of over-involvement-enmeshment, particularly when she discussing her son's developmentally appropriate transference of attachment to others (father, friends) as he is in early adolescence. Mother noted that ... "*I will need to find myself...*" as her children "*grow away*" "*He's (very open), not scared in coming to me about issues as well which is really good because I generally know about things as soon as they happen, problems with his reading and writing but every single report I have is that he applies himself and he tries his hardest he's actually performing to the best of his ability.*" *He (can get very anxious), but at the same time no, no he is very confident, very, very confident outgoing, with adults and kids, um person you know it's only when his tics are really, really playing up that he tends to (withdraw). Sometimes in certain situations he's pretty much out there he's a very um forward type of kid. He's a very social person*

*He's very forward with his Tourette's to so he tells anyone you. Has problems relating to Obsessiveness (cognitive...completion. Just right etc).. and anxiety hand in glove with Obsessiveness... handles his tics well (suppression) , but it is it is very, very hard for him.*

## **D & H**

### ***Summary.***

- **Representation- Generally Positive** Very positive, mentions some negatives but high level of understanding and acceptance of these
- (Mothers emotional response to child in General): **Mixed.** Some, sad, fear and irritation (tics)

### ***Representation***

Mother's representation of her daughter is overwhelmingly positive: Full of pride, understanding, love, empathy, insight, with tone of voice to match. She described her efforts to empower her daughter. Lets her make decisions. Focuses on listening to her needs but is not afraid to make decisions when needed. Mother is only fearful of the future regarding behavioural changes and pressures associated with (immanent) adolescence and the impact peer pressure may have on her daughter's body image/ her self-esteem etc., noting some early signs already-increased need for privacy, anger frustration etc. ). She is sensitive to her daughter's difficulties, and spoke compassionate regarding the impact that the recent death of her husband and H's father has had on them both. She stressed that neither she nor H. wanted TS to "define her".

*"Oh she's awesome is one word to describe H. She's kind and sweet um she's just the nicest kid. If I you know had a classroom full of H's I would be just so happy.*

*I mean she's incredibly clever.....she's incredibly private at times and has no interest in letting anyone know you know about what's going on ....you know she calmed herself down and she got back up again. ... You know and I just go hats off to you H. and I've, you know I'm hoping that I'm instilling in her you know you can do anything*

*You know she just feels like “I’m normal I just want to be normal I don’t want special attention I just want to be... she just wants to just blend in you know and that’s it.. There are little thing (that she does sic.) ..she’s a bit quirky at times and so are her friends so that works, but she doesn’t even want to worry about her tics you ... normal...and that’s how I see her...*

*We’ve always just gotten along. She’s very easy going I mean, she’s also very independent I should add that about her um and um and as long as I give her the space that she needs sometimes to make decisions.... However...H. can get very frustrated... I try so hard not to get cross or anything back at her if she’s snappy because she’s got enough to deal with....Yep and I just think there’s hormones there as well, frustration. H.’s always been an independent kid you know and she’s you know she can be incredibly stubborn at time I just think her tics has just made it worse for her*

*She’s extremely capable um and she’s just she doesn’t let things get her down.*

*... I think she’s able to reflect a lot um and you know and work out what she’s doing With her friends she’s a very kind caring person, I think she’s very considerate um, I mean she’s got a wicked sense of humour*

## **C & M**

### ***Summary.***

(Mother is not very articulate -and is a little embarrassed by this- so the interview was not “pushed”.)

- Representation: **Positive:** Despite his considerable difficulties (TS and moderate Autism) mother is totally accepting, loving, understanding and compassionate of her high needs son.
- (Mother’s emotional response to child in General): **Mixed Positives and Negatives (fear and sad in part. With some frustration)**

### ***Representation***

Despite his considerable difficulties (TS and moderate Autism) mother is totally accepting, loving, understanding and compassionate of her son. She describes him as a ‘normal’ boy, even though he has significant behavioural and emotional

problems associated with his pronounced Autism. Her tone of voice throughout the interview was full of love and acceptance when talking about her son. The only time her tone of voice changed was when she expressed her fears and concerns regarding his tics or other issues not related to her son.

Mother sees her son as *a happy 'normal' boy* who has some difficulties and requires a *lot of day-to-day care* and her *total support*.

*“Overall pretty happy really, normal; he’s got to learn to control his temper (giggle), but that’s an autism thing, He’s pretty happy, normal yeah (sounds bright and loving when describing him).*

*He is sort of like my little baby sort of thing; we get along really well. I understand him pretty well and yeah I just love him; He understands more than people think...can’t express himself...too autistic.. I understand what he is thinking, feeling..*

*When he gets his tics - Oh he can get a bit aggressive sometimes um.....Yeah because when he gets really angry sometimes he’ll pinch his sister or me or.... oh it’s hard to explain but it’s just the way he lets out his anger, but it’s not like he’s doing it to actually hurt me or sister , it’s just that’s how he (handles it) ..... But that’s the way he does it .....he attacks. He loves jumping on his trampoline, ..loves his sister .... It just breaks my heart (when he is upset by his tics)*

## **D & J**

### ***Summary.***

- **Representation** – MIXED; Multiple positives but also many freely disclosed negatives. His high need for her attention and puberty have created stress and frustration at times. Stretched by OCB and mild Asperger’s
- (Mother’s emotional response to child in General): Mixed. Very positive; love and joy (compassion, understanding acceptance, pride, humour), with negative feelings featuring Sadness (grief, loss, hurt, dismay etc.) fear and some anger (frustration, irritation, exasperation)

### ***Representation***

Mother describes many positives, but also multiple freely discussed negatives. His long-term high need for her attention and the advent of puberty have created stress and frustration in their relationship and altered her view of him at times. Mother speaks with love, affection, empathy, compassion, pride and delight countered by irritation, frustration, anger, fear, exhaustion and self-pity alternating with annoyance at self. She pulls herself together in the interview - actively rationalizes her ambivalent feelings and frequently uses humor as a coping strategy.

They have a very loving but volatile relationship that is stretched by his OCB and mild Asperger's. *One child wasn't what we planned but that's how it happened and you sort of have hopes and dreams that your kid's going to be happy and successful and their lives are going to be good and it makes their life harder but it's also made him quite a special person and that's...yeah sometimes I actually have to stop and remind myself of that....*

*Even though he yells and screams and swears at me he's very respectful of his parents and he knows the difference between what's right and what's wrong and I think that's the best we could possibly do with this kid you know I mean yeah... And I think at the end of the day honestly that I think we've done a pretty good job.. ...another good thing with Asperger's actually these kids are um very, very honest and even though it can get them into trouble at least you know they're not going to lie to you and when they do something bad they know it's bad ....they'll do it but they'll think about it and know why they shouldn't have done it and they won't do it again*

Mother describes a healthy physically large, not handsome boy, who displays lots of mild Asperger's or OCB characteristics and behaviours: Obsessive ruminative thoughts, little rituals, great talents in some areas of interest, no interest or ability in others. His tics minimize his school functioning, exhaust, frustrate and physically cause him pain, now effectively medicated . *"Fantastic" musician, mixed academic skills, very bright as a child -Asperger's pattern.*

*He is a very, very caring emotional child,.....but has a tendency to ruminate on the negatives aspects of people's behaviour or injustices that he sees in the world- Emotionally naïve. Extremely shy as a child but "coming out of his shell now" that he is older, perhaps helped by his musicianship and the positive attention this has created But still "can not make eye contact, even (struggles to do so) with me"(mother.).... Social skills have progressed from being used to be "extremely shy"*

and unwilling to converse “*but is happily joining in now, although won’t start a conversation*”. ...and his confidence is growing...*fantastically and funnily!.. he’s quite popular at school the girls think he’s really cute 1023+* (“the cool guitar guy”), ..

“Loves to talk, especially with adults who generally find him very interesting” due to his savant like knowledge. He is “*fantastic and patient with younger kids*”, and will seek them out in preference to peers, and “*is very comfortable*” in adult company. Mother feels he avoid peers because he fears their judgment and the social pressure this creates. “*He is a deep thinker..... worries about what other people say,(he is) like his mother*”. Emotionally mother still feels that he is very “*immature*” and requires a lot of her assistance; “*I know he’s not because he’s emotionally this kid of mine is um still a little boy in some ways, you know he needs mother around. This creates fear of future as he approaches adulthood I hate that I had to make him understand that he needed to say sorry because he’s too big for that now. He’s nearly an adult and um (his dad) and I have talked about that he’s got to start accepting that he’s responsible for what he does and um... and we can’t be there to you know look after him all the time ...often I say to my friends you know we worry about Joel I said to them I stop and I say but you know what he’s a really good person he doesn’t drink, he doesn’t do drugs, he doesn’t go out with a bunch of boys and hang around the streets.*

His aggressiveness increased as a teenager creates relationship ruptures.

## **J & S.**

### ***Summary.***

- **Representation** Generally positive; initially reluctant to disclose negatives
- (Mother’s emotional response to child in General): **Mixed**: Positives and negatives fear and some irritation /exasperation (anger)

### ***Representation***

J. has a *very positive view* of her son. She took a while to disclose anything negative about him, and these negatives related primarily to her concerns over her own hurt regarding his “*pulling away*”. This (“pulling away”) mother is attributing, but not with great certainty, to adolescence. She clearly finds these difficult and

hopes to “*get him back later*”. The relationship however seems to be very close and positive. There are elements of enmeshment. “*Unconditional love*” between them when he was younger she misses. Her tone of voice suggests that she is feeling a little hurt and rejected by him, but she does process this. Slight blurring of boundaries, as she for example discloses her inability to cope with his emotional distress until she can fix it “*rescue mother*”. “*Spoils her kids*”..... “*over compensates*”. She has devoted herself entirely to finding ways to manage his TS. *Devastated* by the diagnosis, she utterly rejected the notion that TS was “*incurable*” and chronic, she launched a very comprehensive and proactive search for treatments etc.

She is proud, loving, totally accepting of him, empathic, compassionate and very involved in helping him to be the best he can. Mother is very pro-active and works hard to keep a positive and optimistic frame of mind for both of them.

*“S. is extremely confident and enthusiastic he’s a bundle of energy um not necessarily physical energy um not necessarily physical energy he would be jittery but he, he’s not really into sport, he although he does play sport um but his...the energy is all mind energy and you know talking and thinking and conversing and very positive, very high self esteem. Very happy child, most of the time.*

*From the moment he was born, from the moment he was pulled out of my tummy with those forceps his eyes were open, I was fascinated his eyes were open and he was looking around the room. He didn’t cry he rarely ever cried as a baby and it turns out he is going through a bit of a naughty phase but it’s you know and he said look I thought I was funny that’s why I was shouting and it wasn’t like an issue, it wasn’t Tourette’s at all . He’s happy and enthusiastic and a ball of energy....yeah, yeah so he was just born that way you know.*

*I’d say he’s hyper but not naughty so I don’t know how that falls in the ADHD yeah I guess I’d know if he had it (ADHD).. he’s always excited about everything you know in an almost permanent excited state. And he likes having Tourette’s by the way well we told him it was special a really cool thing happy enthusiastic popular...not “Mr Popular”*

*Good at articulating his feelings...Very sensitive ... smell. Tastes..etc like other kids with TS are heightened. You know recently at the parent teacher interviews, that the teachers were concerned and S. has impeccable behaviour at*



*school always has had so we're really very lucky aren't we, but um the teacher said we're a bit concerned S.'s been naughty and he's answering back, he's shouted out a few times and the girl sitting next to him said he'd been swearing under his breath and she said I think it was his Tourette's...He has very high integrity...He wouldn't let anybody else change his mind about what he wants.*

*He's affectionate (though not so much as he used to be)...so confident and resilient he...whereas my daughter lacks self esteem, she has no self esteem whereas S. has high self esteem, but I've tackled that and I think I've conquered that too. I did everything. He can't say no- he is like me.*

*he's yeah he's lazy, he's a bundle of energy mentally but no, no he never has tantrums. I don't think he had one single tantrum in his li um if he's upset now he tries not to cry because he used to cry a lot he was extremely emotional and he still is very emotional, tries really hard not to cry Sometimes now he's probably very annoying. You know like he will maybe say something and he'll have to say it over, over and over again you know what I mean you know he'll be very irritating...that...*

## **J & S**

### ***Summary.***

- **Representation- MIXED** /Positive and negative (big and difficult for mother to manage and accept negative changes accompanying adolescence; change again with diagnosis of TS (mother feels more compassionate and understanding)- very late at age 16- beginning to be more optimistic and positive as he is happy and succeeding at work)
- (Mother's emotional response to child in General): **Mixed**; Positive (love and Joy) Negative. Sad, Anger, Fear

### ***Representation***

Mother's representation of her son is both positive and negative. He was not diagnosed until recently at 16, which mother feels increased her understanding and compassion for him.

She expresses love, pride, empathy and understanding for him but is distressed, disappointed, angered and feels rejected by him as well at times. The

ambivalence mother reports is related to her feelings regarding his difficult adolescence, significant accompanying personality and behavioural change, with his increasingly aggressive behaviour over past three year. In addition she feels loss and rejection as he becomes more autonomous. Adolescence, increasingly severe tics and problems dealing with this impacted adversely on everything, particularly school and academic performance.

She understands her sons negative behaviours though she finds it it's difficult to differentiate between TS related behaviours and adolescence - drive for autonomy. He is "*her pet*", some signs of over involvement / enmeshment with mother having trouble letting go. They are close and she feels that she is more lenient with him than her girls. She is proud of his popularity and ability to cope with tics etc. but frustrated by his underachievement at school. She is struggling with him blocking her from his new adult work life. The relationship can be quite volatile: they have *flare ups* but are resolved well. She mentions that he has a very close relationship with his twin sister. His twin sister is accepting, supportive and permissive with him, "*like me*". Father struggles more with the TS. He is a more permissive than mother. Mother has considerable guilt regarding his late diagnosis, which came as the result of bad drunken fight and her son's final admission that he drinks to reduce his tics. *I understand him really well...can calm him down etc...we are very close .. oh yeah, yeah he was always with mother, he loved his mother, always wanted a cuddle and all stuff like that hop into bed with us and yeah*

His withdrawal from parents has hurt mother a little however she is proud and happy that he is enjoying his job so much. *..It's a different situation now because he's older and you know they don't want to be around their mothers. yeah listen S., I can't say it too loud <in a soft voice> is my pet you know he's the one that I worry about the most and you know I let him get away with a lot more than I should a lot more than what I would let the girls get away with different to relationship with the girls...*

*oh that's right and B.'s (his twin sister) like me she lets him get away with a little bit let's him get away with murder. I suppose he is her twin yeah and also because the Tourettes she's very sympathetic towards S., very sympathetic. She was so upset when he came home that night that he got beaten up (twin sister) He's always been a happy kid, outgoing lots of friends...*

*In High school things changed a little..years 7 and 8 (the feral years)  
OK..lots of friends, happy, played sports.....all the normal things...year 9, 10 and  
11...became very aggro, uncommunicative,..... Isolated himself in his room...  
He can't sit still!*

*When he did year 11, that was um last year he was shocking he was just  
horrible child to be around. Really angry all the time, come home from school so  
angry because he had been trying to hold his tics in. He'd be sitting in class trying  
to hold his tics in.. He passed year 11 just ..He was disruptive in class because he  
couldn't concentrate because he was trying to hold his tics in and stuff like that  
yeah. This year he's still very <pause> sort of angry but he's not as bad as what he  
used to be like I'm talking punching walls, yeah and walls and thinking and he's an  
idiot and stuff like that. You know this has all just come out in the last say 12  
months that's the way he actually thinks about himself... so his self esteem has been  
really hammered by this Yeah that's right..... because S.'s go real potential you  
know. Like he did really well in Primary School um he was doing quite well in year  
7 and 8 and then it just went completely downhill after that yeah but that's when I  
think the full on tics started coming into it at year 9 so that makes him 15 or  
something like that.*

*Well.. our is ...it's just typical family that you know they don't want to talk to  
their parents yet they will be really nice to a visitor when they come in, that's  
double standards, but that's alright .....house devils that's fine by me.*

## **J & C**

### ***Summary***

- **Representation - Generally Negative**- Compassionate and understanding and proud at times re C, but multiple negatives expressed. She describes herself as his “ally in everything” but her positive affective bond with his twin his much stronger and easier.
- (Mothers emotional response to child in General): **Mostly negatives** Positive and a lot of negatives sad, fear and anger

### ***Representation***

Mother's description of her son is generally negative. She is very open about her need to tell the truth regarding his negative behaviours/traits (impulsivity, ODD in particular). She speaks with pride, frustration, concern, exhaustion, love and empathy regarding him. She describes herself as his ally. She stays very firm and micromanages his life (Her regulatory function, her protectiveness) and appears to repress many of her feelings - makes a big effort to stay calm and focused on her job with him, though her empathy for him and fear for his future is revealed towards the end of the interview where she recounts her contact with the adult with TS in K-Mart. His ODD, his impulse control difficulties (dangerous-self harm), his emotional and behavioural excitability and his social skills deficits create the most angst and challenge for her.

***In contrast-***

***Twin B. (Fraternal Twin Brother- Mild TS only- Not included in this study).***

Her relationship with his twin is much easier. She describes a more positive child, a more positive and easier relationship. Easier for her to be loving, no need to be as worried about him, he is not exhausting. Does not have to be "hard" on him. No negative qualities described, she identifies more with H. (sense of humor shared and ability to interact easily) and he proves to be less of a parenting challenge. "Opposite to C. in every way!"

**Representation**

*How to describe him? Enthusiastic ..he's um he can be very loving and very warm he's um I'd say a bit of a "mummy's boy" but I think that's because I'm his ally in a lot of these things, do you know what I mean...he um he's aah growing confidence and things but then the flip side of the coin he can be wild and um nasty and you know and this is my boy and I love him to death, ....*

*(Her need to discuss the negative aspects of kids with TS - negative aspects of it then what will end of happening is that... more research like your own will just be kind of reflecting the 'everything's honky dory in Tourettes land'...and it's not it's um it's not it's really hard sometimes. There are times when um I just think I can't be near him right now he's just draining and unlikeable aggressiveness...scary and unpleasant..yeah it can..like ok all of his impulse control fits into that and all of his you know flare ups of anger fit into that and even if trouble with some of his um functioning at school and stuff..... Understand it now*

*since TS diagnosis. From the age of one...I knew something not normal.. was just that thing where his reactions were extreme he um he just wasn't .. I understand about the TS's.now...the anxiety...etc...because it's just that thing where like at school they're like we don't see any of that you know and even now you know he comes homes and starts ticking and has a huge tantrum and you know takes everything out on the world and yet he's this lovely pleasant child at school.*

*That GP saw.....this kid climb all over his mother...you know can be completely interruptive and you know being a brat really. Can get wound up..over excited and cant calm himself.*

*He doesn't and he has um elements of different things he's definitely got some oppositional defiance going on/ He's impulse control is getting better but it's pretty low but because a lot of these things are kind of tied in with Tourette's itself know he's not having huge problems at school or in any other area ...we went down to the local shops um there was a clown during school holiday entertainment, all of the kids there were maybe 3 to . Goes straight in there front row loving it.... unself-conscious...*

*Cognitive rigidity, rules based black and white thinking etc. poor social skills...lack of social consciousness/self consciousness. Both boys quite driven..(as demo'd by their judo success, mothers observation). Re his Martial arts success...C is now more as self-accepting*

## **K & C.**

### ***Summary.***

- ***Representation*** - **Generally positive** but sometimes bewildered by his behaviour
- (Mothers emotional response to child in General): **Mixed** positive and Negative Positive and some sadness and fear

### ***Representation***

Mother speaks with love and affection, pride, some concern, understanding, acceptance but bewilderment at times regarding her son. Her description of him is *generally positive but she sometimes struggles to understand him.* She is very

concerned for his struggle with his social side, and inability *to fit in* and to *be what he wants to be (the cool dude, not the nerdy kid)*. She does however describe a devoted relationship with her son, but her own experience with epilepsy makes it hard for her to empathize with his very different response to his TS. *He is very focused upon his TS* (adjustment problems perhaps) whilst she is very non-plussed by her epilepsy. She is proud that he has qualities that she feels she lacks (*being "out there" etc.*)

Their relationship is cerebral (*we are both thinkers..... and talk deeply*). That's how she feels they best connect. She is not a natural caring person..... *not my way* (attributed to her family of origin experience) but she does express empathy, if bewilderment and uncertainty/lack of confidence occasionally in her parenting ability. Increasingly over last year *he has lashed out at her..(maybe related to TS)*, but he is very quick to apologize. Recent big change in this following *seeing me (mother) have an epileptic fit for the first time*. Now mother is concerned that there is a *role reversal* with him being hyper-vigilant for her.. *being very caring. A big turnaround* and one she is trying to prevent from cementing itself. Some signs of over-involvement -enmeshment and mother is aware of this.

She expresses a lack confidence in her parenting at times, and relates this to her own experience as a child (insecure). Feels that she lacks the usual maternal caring quality that most mothers have. Mother sees her husband as the primary source of comfort and authority with their son. She does however describe a devoted relationship with her son.

*He's got a good sense of humour, he's very out there person, he's a strong character, if something is um if he's being told off for doing something wrong he has no qualms in telling you, you know, what he thinks. He's like he very much sort of wants to be sort of a stand up person, standing up for people's rights.*

Social skills/ social rules *maybe not great - He likes to get his opinion across um he wants to be friends with children but he's not <sigh> he hasn't got the correct skills I feel um <hesitant tone> sometimes he might go too big I suppose being my opinion is my opinion.*

Concern- *he's not fitting in with the mold of what he would like to fit in with. He's got one or two sort of close friends but he's not fitting in with the group that he wants to fit in with... it's a hard thing to sort of say because he's a social person... um he will like we go camping so he'll take his football and he'll go out and kick it*

*and you know the kids they come, they do come to him..... it could be different characters which he's not that ...a personality thing.....maybe it's um a hard one but yeah he doesn't have a big range of friends*

*He's very much I feel <sigh> a thinker ...like as in deep he will take things on and he will really think them through.. and I feel that's probably a bit more than what most kids would. He really listens too....he'll ask the questions if he doesn't understand as well...When he is sort of older he'll have a lot of skills which um you know he might not have some skills but he will have other skills.*

*School....um so he's sort of like medium as in like you know his rate of work um and a lot of the times he'll sort of get, behaviour might need to be improved sort of type thing. He's also doing piano this year which he's thoroughly enjoying and I would say he is a bit theatrical too.*

*Can lash out at me...oh yeah he can get angry ...I'm finding that a lot this year um just in general um I'm finding a lot of explosion towards me and it comes out big time but then he will be five minutes later "oh sorry mother"*

*Seeing mother having a fit for the first time...Yeah it's been quite a turnaround for him um so I suppose in one way it's a good thing but now he even is taking on the worrying side, a role reversal.*

## **L & L.**

### ***Summary.***

- **Representation- MIXED:** Largely positive but as the interview progressed her over-vigilance and concern for her sons anxiety emerged
- (Mother's emotional response to child in General): **Mixed:** Positive (love and joy) and negative – sadness and fear

### ***Representation***

Mother began by speaking in very positive terms about her son. She speaks with love, empathy, pride, understanding but as the interview unfolded her high levels of concern/ worry / anxiety for her son emerged. She is aware of her need to let go of some of her hyper vigilance and worry, particularly as her son has just turned 14 and as she feels, needs to be and deserves to be more independent.

She describes a *chatty, talkative* boy who has long struggled with *anxiety*. She reports that he is *generally pretty happy go lucky and resilient*. He has *learning difficulties so is constantly playing catch-up at school*, but has been able to manage with her assistance. He *didn't even notice his tics until* they became very noticeable last year, prompting diagnosis. He is much more comfortable in the company of those *older* (including adults) and *younger* than himself. He seems to be more *anxious around his peers*, and particularly so *since he tics got worse* this last year. Mother expresses concern that he *avoids close peer friendships* and *is a bit standoffish*. He does make friends at school but doesn't make an effort to maintain them. But mother's not sure why right now. She posits that it may just be adolescence, *a phase he's going through-* being a bit less communicative and isolating himself a little. His sister lets mother know that he is socializing *at school and laughing in groups*. Mother withdrew him from team sports recently as the atmosphere was *too competitive and the stress was not helping him or his tics*. He has taken up fishing and other individual sports and activities. He enjoys a good relationship with his sister.

Proud of his resilience *I'm really impressed by him. I mean and despite the fact that he struggles at school he still keeps going back and he's pretty happy go lucky most of the time. He is ..Very social, talks constantly, very, very happy, but quite anxious; anxiety is definitely actually there is one thing I can definitely say apart from that his anxiety has always been a big problem, a bit of separation anxiety in the earlier years I forgot to mention that. Actually his sister I know we shouldn't go she has been diagnosed with separation anxiety disorder quite severe but no ticks but I'm wondering if it's all related..*

*He loves fishing, playing at he beach swimming. Had to stop team sports recently as it was so competitive and harsh.. ...too hard. If you keep pushing the competitive sport I don't think, I think it just makes it worse because they really it just emphasizes I don't really fit in any way or if there's just no enjoyment in it what's the point.*

*Well we definitely talk a lot more about things, he's not afraid to approach me about anything, yeah it is because I'm always trying to think simple ways to explain things. Still affectionate- he quickly gets out of the car in the morning but he still wants the odd hug at home when no one's looking but he's always been a very affectionate child...yeah a bit of a mother's boy.*



So it's very open (mothers relationship with her son)?...Yeah it is, it is and I hope it remains that way because um you don't want him to try and deal with it on his own...but he does sometimes (adolescence). We definitely talk a lot more about things, he's not afraid to approach me about anything, when he's upset um yeah look at some when he was young very cuddly very affectionate relationship, um he's not wanting that as much naturally

How much do you really know about your adolescents social life?...Yeah it is and you know I've got to stop worrying because I know gosh when I was young parents didn't know anything I wouldn't have told I just did my own thing. Mother reports being anxious...since becoming a mother (family history husbands side of separation anxiety and school refusal) and does want her kids to be dependent and for her to not worry so much. Aware.

## L & A.

### *Summary*

- **Representation- Generally positive:** Very positive and compassionate description of her son, tempered by recent increase in aggressive behaviours since puberty (early developer)
- (Mother's emotional response to child in General): **Mixed:** Love and joy. (proud compassionate accepting, delight) with some sadness and fear (strong at times)

### *Representation*

Mother has a very positive representation and relationship with her son, but she is also aware of changes in his behaviour since puberty - increased *back-chat*, *frustration* and *aggressiveness* particularly in response to his quite severe ticking, which he finds very distressing and exhausting. She finds this slight rupture a little distressing (hurt and rejected and under-appreciated, reflected in her tone of voice) but generally understands this as a part of normal adolescence. Mother is very empathic, understanding, loving, proud of his achievements, committed to her role as her son's advocate and partner. She is very distressed when she sees him upset by his tics. He can however be very demanding of her, and his tics can be both frightening and irritating to herself and others. Her own OCD traits give her even

greater insight into his plight and motivate her to control them (in order to remain calm and strong for his benefit). His lack of close friends deeply saddens her, but she holds hope that he will find friendships in the future. She seems to view her son as a partner and an individual so although very close, and to a great extent unsupported by many others, they don't appear to be enmeshed.

*So A. isn't silly, he's a smart boy. His a big boy....bigger than me.... very developed for his age and already into extra-large men's size at 12. He is friendly shy, has low self esteem but it is improving. Loves fishing and raising animals of all types. He loves talking to adults, loves talking to other people about anything. However he doesn't relate well to kids his own age: he craves deeper friendships than most 12 year olds, can't really join in their games (playing chasings) due to his physical size, can't play football as it was too vigorous for him, and finds the interests of kids his age tedious and irritating. He has also missed so much school that mother feels the other kids friendships have moved on without him.*

He also also very "busy with tics", which are moderate to severe. They sometimes make him physically ill, irritate others (compulsive touching for example) and can be very frightening. The tics and his dyslexia impact adversely on his schooling and school refusal is a problem that mother has to address (monetary incentives etc).

*He can be very demanding of my (mother's) time, and his compulsive tics hard to live with (eg. jumping etc can be hard (new carpet to block the noise!))* Mother is very innovative and self-directed re making life better.

He had normal social relationships up until age 7 when compulsions began (touching /hitting other kids), and when teacher moved him away from the others kids, his anxiety began. As he enters adolescence, he tries not to cry when frustrated or upset (particularly by tics) and instead reacts with frustration and aggression. He is beginning to challenge his mother as well, which she interprets as normative teenage behaviour.

*Aggression/ frustration Close yeah...but he s getting to the back chatting putting me down stage,..age.....but still wants me there..but that will happen! Probably when he gets to high school.... When upset...He mumbles under his breath and he swears a lot, and negative self talk, or he mumbles and gets really grumpy and or does get quite aggressive as well*

*Well I think frustration, frustration and anger I should say like when, one time I just cooked him poached eggs, he loves poached eggs and he was carrying the plate over to the table he did a massive tick and it all ended up on the floor... and he burst into tears and he was so frustrated.....How do you feel when that happens? Terrible (very tearful...)*

## **N & J**

### **Summary**

- **Representation- Very negative** but still empathic and compassionate (change with adolescence)
- (Mothers emotional response to child in General): **Mostly negative** Love but multiple negatives (sad, fear and anger)

### **Representation**

Mother's presentation was initially brittle. Her words and tone of voice conveyed restraint, pent-up anger and intense frustration, hopelessness as she spoke of her concerns for her son. She was also quite shutdown, blunted, numbed by the relationship. "Over it all" and "wants to escape it"...get respite.

Her description of him is *highly negative* however her empathy and compassion for his plight is also apparent at times. She understands his situation and expresses despair, fear and desolation in response but is exhausted by the entire process of mothering him. Since diagnosis and adolescence his behaviour has radically deteriorated. In the past she has been a highly protective and pro-active partner...goes into combat for him. Tough love approach...tries to help by getting him to confront his TS and see that its not the end of the world... and to accept treatment but all is utterly rejected by her son.

*Mother-<nervous giggle>J. is a living nightmare... no he's an angry boy, he's um he's aggressive, he's um intelligent but he's um yeah he's quite aggressive now quite nasty aggressive and he doesn't really have any like he doesn't care if he makes me cry or his sister cry he doesn't care less, you know what I mean, that's a new thing isn't it? big, big, big difference ? Probably yeah that's probably been in*

*the last 12 months ...So you think you can sort of see a shift in his personality as things have gotten tougher? Oh yeah definitely yeah definitely in the last 12 to 18 probably 18 months.... (since diagnosis and onset of puberty).*

She feels his personality *has completely changed*. She describes a boy full of rage, lashing out, social withdrawing, *sabotaging himself, poor self esteem, immature but very bright ( IQ 145), no social skills (social inept), extremely stressed* and not coping at all. Behaviour so alarming that he had on *6 week hospitalization* last year...*no long lasting benefit* but gave family some *respite*.

And he is expressing this violently, particularly toward his family. He is now aggressive, *violent, nasty, actively sabotaging all of his relationships* and the relationships of anyone else. He has rage episodes and shows no remorse for his actions, *makes his mother and sister cry* and is *violent* towards them (*punching*). He is isolating himself in his room. Some elements of *paranoia* perhaps.

He *refuses to accept that he has TS*. He *refuses medication....* convinced that it will *destroy his very high intellect..* which is the only thing he feels he has of value.

She expresses grief for his lost potential.. She has reached a point where she feels totally overwhelmed and exhausted and has little energy to continue but feels trapped. Her only current hope is that he will finally accept his TS, accept treatment and that his tics will ease over the next few years. Meanwhile she describes a week by week struggle....*survival mode*.

## N & S

### **Summary.**

- **Representation-Very positive.** No current difficulties noted (newly diagnosed)
- (Mothers emotional response to child in General): **Mostly Positive** (only 1 neg. irritation re. Tics)

### **Representation**

Mother speaks with love, acceptance understanding, no stress, has coped with difficulties positively and did not sound overly distressed or concerned about

any particular issue or problem. She describes a *caring, active* little 7 year old, who can be *as annoying as any 7 year old boy at times, Caring, ..can be a bit “over the top”.. Very active.. Restless...always needs to be moving*

*A little introverted...A bit shy at first... but gets on well then (overcomes this and becomes) friendly and chatty,..Very honest”. He is a little more needy eg. Needs constant playmates...not good at entertaining himself.*

Has had some problems with bullying in the past, which he seems to have largely overcome at this point (home schooling for 18mths..now back and *doing better* school). *We took him out of school..home schooled...He is growing in confidence*, and has a tendency to prefer relationships with *older* and *younger* friends.

As mother is very familiar with TS, the diagnostic process was not traumatic and she felt prepared to cope with his symptoms, which are also very mild.

She describes a close relationship with her son, but can not attribute it to the TS....maybe its *personality* driven or *the fact that he is* (her only) boy.

## **R & T**

### ***Summary.***

- ***Representation - Generally Positive;*** Positive -Love and joy (proud, accepting etc. Acceptance of slight agro emerging as he hits puberty and occasional rage /anxiety episodes)
- (Mother’s emotional response to child in General): **Mixed;** Very positive (love and joy ,pride delight etc and relief) and negative. Sadness (Rejection on son’s behalf, hurt, grief) and fear (anxiety and stress)

### ***Representation***

Mother speaks with great affection, love, delight and pride for her son. She is totally accepting and understanding of all of his behaviours, and expresses few current concerns for him. However she becomes very emotional when discussing his problems in earlier childhood before his diagnosis. She was deeply distressed and intensely fearful for and protective of him when no-one understood his difficulties.

She was devastated when he was rejected by a family member and deeply grieved when he was diagnosed.

Mother describes a *very bright, eccentric, outgoing, well-rounded multi skilled child*, who is *popular and charismatic*, and *has a "normal" social life*. He has a *very strong ego and positive self-esteem* that mother feels enables him to cope well with his TS. His *is very witty*, with a great sense of humour. Mother describes him as *gushy, very affectionate boy who loves physical affection*. He was a *demanding baby who didn't sleep well*, had *very high energy levels* and encountered difficulty in pre-school, where his high energy-externalizing behaviours (rage and aggressiveness) and other OCB related tendencies were misinterpreted as *bad behaviour* at times. Problem behaviours relating to his OCB (sense of justice; back and white thinking; "just right" obsessiveness) still create occasional difficulties (frustration) and tensions particularly when engaging with *other strong-minded children*. He also *has high energy and ADD like tendencies when his tics wax*. He does well in school, and *is at the top of his class* (has a classroom aid).

He is a *natural leader* who is able to defend himself, has good insight into his TS and is learning to cope with some of his problem behaviours, which are for the most part quiet minimal. He is *confident and articulate and open* regarding his TS, which is in the mild to moderate range and has developed an understanding of and empathy for *those who are "different"*. She anticipates some challenges to his *self-esteem* in particular, when he enters *adolescence*.

Mother describes an *'incredibly' strong bond* her son. She feels their very close mother-child relationship with her son developed in part as a result of *his neediness as a baby* (he had to sleep in her arms for the first three years). They remain *physically affectionate*, even as he approaches adolescence. Their relationship is *very open* and they *share everything*.

*"...it's a really hard one to put actually like it's T. and I had an incredibly strong bond and still do as a mother son relationship but at the same time um I think because of all that physically, he was so physically needy (As a baby...slept in mother's arms until aged 3) when he was younger there was a different sort of bond there ( to his sister.)"*

**Summary.**

- **Representation** : **MIXED**- Positive, loving, proud and compassionate and understanding. His Impulsive (OCB) - ODD behaviours can be very challenging and difficult to manage at times and create ruptures in their relationship. However she is very forgiving, compassionate and understanding so can quickly reconcile these problems.
- (Mothers emotional response to child in General): **Mostly positives**; (love and joy) and balanced by some negatives (Sadness, fear and anger-exasperation)

**Representation**

*G is a very sensitive, outgoing, caring personality, very soft on the inside but tries to come hard on the outside with strong language....< sigh> he is a happy boy, I would say in general .... he forgives very quickly and like he's not a resentful person, he has a lot of humour. He's very focused on what he wants um to the extent that he's not giving up on anything, which is sometimes hard if he wants something he's just like very, very determined ...which is good and sometimes can be a little bit over the top and he's very diligent as well. He has goals and he is going towards those goals so he's um...he wants something he's really doing something about it... that's something very positive about G.*

*G. is very intelligent and we already knew.. we felt that there was something going on with him since age 5... he had a lot of problems in childcare we .... I thought he would have ADD ..an IQ test 135 IQ and the psychologist said that I have to take him out of childcare that's the reason why he would be difficult is that he would be bored he's very intelligent.*

*I like that he is not an aggressive person... but unfortunately he is not defending himself, so really his self esteem is very low as he is a target of being bullied quite a lot. He can be also be very annoying for his little brother, he is not respecting his space or other people's space, but its not intentional, it's because he is not able to self-control himself. He's very impulsive, (and compulsive) ie. he talks non stop and not able to contain himself which results in people being very upset and annoyed of him. He also lacks social skills ie can't read body language well, social cues, can't follow social rules very well and this causes terrible difficulty for*

him. He annoys other children so they either bully or ostracize him. *they just can't cope with him.* This is very upsetting for G. and for his mother. Mother believes that his loneliness and inability to find a close circle of friends who understand him is the most serious problem for him. *..Not being able to control himself and all the years of not knowing why has left him very insecure and anxious. He's even talking more, which is like making everything a little bit more complicated and so that's why children start to bully him. As a mother makes me very sad to be honest and I feel that's the biggest impact for him socially; not that he is not social but it's hard for him to behave to be socially accepted.*

His ODD (lack of respect, constant challenging, temper tantrums / frustration), his annoyance and apparent lack of insight or empathy for his brother's OCD, and his compulsive use of bad language (not diagnosed with coprolalia) cause problems in their usually happy, secure home and challenge mother and dad the most. His bad language is particularly challenging for his father, and creates trouble at school etc. *it's not coprolalia because he says it when he is angry so it's not just coming out in conjunction when he is angry about something but he does choose extremely bad language which we never use at home and it's very upsetting for us and he is in a lot of trouble but he says he can't help it he says I can't help it I don't want to really think bad but I just can't help it so I don't know (mother thinks its to make him look tough). .... I don't know, this is the main point I don't understand and this leaves a lot of tension also between my husband because my husband gets very upset with G.'s language um he's more than I do and I just don't understand I just can't explain it why. ODD is difficult for us because he is not respecting authorities especially not at home that's where he feels secure that's where everything we say is no, no, no everything has to be discussed.*

Mother describes her relationship with her son as *very intense, very close,* because she feels the *need to protect* him so powerfully. She has concerns about enmeshment and dependence and actively fights against *her tendency to 'over-mother'* her two boys (both have diagnoses) but finds this very difficult. She clearly loves and deeply empathizes with her son, but even she sometimes cannot cope particularly with his uncharacteristic aggressiveness outbursts and provocative behaviour. This creates transient ruptures in their relationship (She *sometimes loses it* and then *feels extremely guilty* as she *tries so hard to remain calm* for her sons to prevent *escalation*). However she is very forgiving and understanding so can



reconcile these problems quickly. She is *heartbroken* by his social situation and finds this very difficult to observe.

## S & J.

### *Summary.*

- **Representation** **Positive**; Realistic, loving narrative of her deeply disturbed child
- (Mothers emotional response to child in General): **Mostly positives** :Positive ; love and joy (huge compassion, as well as love, pride and delight) and powerful negatives (extreme fear, and sadness with a little anger-injustice)

### **Representation**

Mother speaks with profound love, empathy, understanding, compassion, fear, terror, horror, and resignation bordering on hopelessness at times. Mother describes a boy who was a *happy typical* child until the sudden onset of symptoms around the age of 5 when he became “*very unusual, self-absorbed, anxious and emotional*”. Shocked by this change, she pursued diagnosis and was devastated by the result. *He’s got incredible energy and love and compassion, he’s so intuitive and insightful it’s quite amazing. He is caring and giving and people love him. His energy is incredible and he can focus on the here and now and the beauty of the minutest of things, like nobody I’ve ever known in my whole life ..it’s quite a gift.* However his deteriorating mental health and behaviour presents terrible challenges for them both. Her son’s extremely serious problems include *self-harm, suicidality, sexually inappropriate* behaviours, *impulsivity, episodes of psychosis (command hallucinations), and rage attacks*, which can be *extremely violent*. These episodes are highly unpredictable and characterized by extremely rapid escalation. He has been both homicidal and suicidal.

He has *very low tolerance of sensory stimulation* and requires a very high level of routine and *a calm, unchanging environment*. He attends a *local school* but needs a teachers aid and has benefited from having an *excellent well-informed teacher over the past two years*. He does however have great social difficulties.

Although *people love his energy and humor, his friends (who still care for him) have been increasingly pulling away from him at the same time because they are incredibly drained, burnt out by his neediness and they call him weird and crazy.*

His friends (all school based) *still care about him* but can't cope with protecting him, *being his carers*. And he is too intense to play with so they have been withdrawing from him and not socially including him (birthday parties etc..).so sad and hurtful for them both, but mother understands. His social world has been shrinking rapidly, as has his mother's.

Mother is totally dedicated to doing whatever she needs to do to make her son's very difficult life as easy and as independent as possible. She clearly loves him and deeply empathizes with him, her empathy heightened by her own experience of mild TS. Their relationship may be described as over involved-enmeshed but his extremely high level of needs demands almost all of her energy and time, mental and physical resources. She needs to be able to monitor him constantly for any sign of deterioration of his mental or physical status and in the process has had to relinquish a large portion of herself and where own life, even to the point where she is prepared to die at his hands in the worst but possible case scenario. She believes her role as his carer was *fated* and that as his mother, there was absolutely no choice in the matter. They are both at very high level of risk from his homicidal and suicidal, violent behaviour yet mother feels she has no alternative other than to remain with him.

She does also appreciate that this may not be sustainable, but again cannot tolerate the alternative. She has intense fear of the future for both herself and her son, and is aware that she minimizes and denies the risks at this stage. She also knows that she has totally sublimated her own needs and wellbeing to the care of her son but makes this decision openly and expresses no regret, just enormous sadness. They are trapped in an extremely isolated and increasingly small world, and although she has good support from friends within the mental health services sector who understand, she has little other support that she can rely on. (*see prior coding for multiple relevant quotes*)

## **T & T**

### *Summary.*

- **Representation - Generally positive;** Very positive and compassionate even in the face of his considerable difficulties (anxiety and sensitivity). A normal boy, just “extra needy and precious”
- (Mothers emotional response to child in General): **Mixed:** Positive; Love and joy (love and compassion, pride and delight) with multiple negatives as well, fear sadness, and some irritation /exasperation

### **Representation**

Mother speaks with great compassion, love, care, pity, concern, worry, as well as fear of future /apprehension (He has only recently been diagnosed with TS). Her representation of her son is positive yet she openly acknowledges his difficulties. Mother describes a *fragile, pathologically anxious, overly-sensitive* little boy. “*He is a lovely and caring boy*”, whose only negative behaviours relate to the exhaustion and frustration that his ticking creates (*when he can become grumpy and moody*) and negative behaviours caused by his *anxiety and OCD* over which he has little control. He has a low tolerance for *change*, and will react extremely (*panic attacks* etc), thereby limiting both his own and the family members lives. He has *slight learning difficulties* (reading and spelling).

He has a few close long standing friendships with *boys who are protective and accepting of him*, friendships mother ascribes to *his caring nature*, but his anxiety and acute *shyness* prevents him from approaching other children. He is *too anxious* to endure *team sports*, has some limits to his *social skills* (eg. *Difficult to make eye contact*), and cannot manage to be in groups, even at family gathering. Once he overcomes his anxiety though *he can open up to others and get to know them* (and visa versa).

Mother chooses to view him as a *normal child*, just as precious as his siblings, but with tics and other issues that make him “*unique and special*”, but at the same time a little bit *extra precious and extra needy*.

*I think it is in a way that I suppose I mean they're all precious but he obviously feels that little bit extra precious um not in I don't feel any more different not different love but you have to really um the whole family we feel like we have to tip-toe around him a lot because of the way he is. So I suppose a little bit extra*

*needy and special, extra care in certain areas...try very hard to keep things the same..*

Mother obviously loves, cares for deeply, empathizes with powerfully and works hard to make life and their mother child relationship as normal as possible; and comparable to her relationship with her two other children. This is difficult however as she acknowledges his “neediness and specialness”, that require extra care in certain areas : “*the family have to tip-toe around him*”. Her experience of her own mother’s mental illness (bipolar disorder) heightens her sensitively to and anxiety regarding his mental health issues. This creates an intense fear for his future, leaving her hyper- vigilant and committed to doing whatever she can to help him. Although she and the whole family help make his life easier, she is also trying to give him skills to manage his own difficulties, thereby encouraging his autonomy. She does however find his distress at times almost too much to bear, expressing intense sorrow and compassion and desire to rescue. This, along with fatigue and a sense of being overwhelmed at times she combats by rallying herself with positive thinking and skill and knowledge acquisition to try to maintain hope, and a sense of balance and control. It is still early days in the diagnosis and they are both still adjusting to and learning about the TS.

## **T.O & J**

### ***Summary.***

- ***Representation*** – **MIXED** : Positive but with multiple contradictions (eg. Multiple difficulties relating to his TS, OCD, ADHD , Anxiety, LD and depression
- (Mothers emotional response to child in General): **Mixed**; Positive- (Love and pride, and relief) and negative- sadness and fear

### ***Representation***

Mother’s representation of son is positive, however many contradictions are also evident. She speaks with acceptance, love and pride, believes that he is a “*normal*” boy with a few problems however she also expresses great anxiety regarding her son and tends to be hyper-vigilant. Although she has always felt that there was something wrong for her son, she has consistently failed to notice his

signs and symptoms (until pointed out by others) and this is both highly bewildering and distressing for her. She describes a very close, possibly over-involved enmeshed relationship with him that is characterized by love (*he is her baby*), acceptance and mutual empathy but is also characterized by occasional serious disconnects with mother demonstrating poor insight (both regarding her own feelings and behaviours and her sons). His recent depression-suicidality, and the way her behaviour may have contributed to his depression (as suggested by his counselor) were a profound shock to her, and have heightened her protectiveness and vigilance. She describes “*feeling what he feels*” in times of distress (boundary blurring), feels that their relationship is *closer* than her other maternal-child relationships, and is anticipating more difficulties with him as he pulls away from her in adolescence, though she intellectualizes this as being normative (he is however her baby).

She reports her own high anxiety eg. when she or her husband is not with him, and does not often leave him alone. She is aware of her ‘*clinging*’, as colleagues have brought it up with her, but she hopes that she is not a “neurotic” mother/ to “*Cut the apron strings.*”

Mother describes a *quiet homebody* who *loves helping people*, and has *been able to make friends and fit in now* that he has found “like” children. *Anxious since he was a young child*, he can lack confidence and *his self-esteem is not strong*, however his ability to defend and assert himself is improving. He has been bullied and teased a little in the past and is still occasionally. His biggest problems she feels relate to his anxiety and obsessive behaviours (rituals, obsessional thinking) not helped by his TS. He is *very creative*, but *not sporty*, *gets on well with his sister* and *is very close to his father (preferring his company to hers when his is home)*. He is *never aggressive* but *withdraws* when angry. He ruminates over negatives for a long time, and this is difficult to counter. He has *learning disabilities*, some obsessive behaviours and was school averse in the past, but with the right help is now coming along well at school, and is only a little behind.

Self esteem and anxiety is a big issue too.....*head gets big, body small.....* looking back can see it from kindy (Anxiety). *He’s more of a homely type, but does have his friends. Creative, loves computers and art. Not really sporty., Loves helping people, loves the oldies..* “*He is quiet...not sensitive...confidence and self esteem a bit low sometimes? Yeah, yeah../ We are close.. he’s my baby... Prefers to do things with his dad....close to him as well (but dad is regularly away from home*

*for work).He talks a lot about troubles (to mother).. can't let it go". "He can be obsessive...withdraws when he is anxious...not angry...just withdraws..."*

She responds to his distress powerfully but sometimes with poor insight. (it was hard for her to find the words to describe). She has regularly failed to notice his symptoms and changes in mood etc. ...An example of this: *and like he said like take him home he'll be fine he said it may go away it may never ever come back he said or it could come back worse and I left there thinking well ok it's going to go away.... because it already was starting to go and I thought so you know I just thought it's just one of those things that it won't come back and but when it did come back I was the last person to see it...* Mother feels bewildered and guilty re her observational, insight failures.

*I am more in tune, more switched on.. with her son than with her other children...but when I say that like with him he also with me like he I suppose because I have that with him he's got to read me too and he knows.* Yes that's right. It can be quite intense can't it? *Yeah like he'll say to me oh 'you've had a bad day mum or your tired mum'..*

## **V & D**

### ***Summary.***

- ***Representation- Generally Negative*** with very slight positive (compassion and faint hope)
- (Mother's emotional response to child in General): ***Mostly negatives:*** Some Love but mostly negatives, fear, and anxiety and some anger.

### ***Representation***

Mother expresses a range of emotions over the course of the interview when describing her own and her daughter's difficulties. These included feeling trapped, defensive, overwhelmed, sad, fearful and numbed with a small glimmer of hope expressed towards the end of the interview in addition to relief resulting from having had the opportunity to discuss her concerns. Her representation of her daughter was generally negative, however she admired her daughter's ability to face such a difficult life.

*(No transcript available for direct quotes; consent for interview only)*

Mother's description of her 17-year old daughter is defined by her daughter's depression. A withdrawn, quiet but loving young child who gradually became more troubled. She never really had friends. Was close to her parents but never a "cuddly" child. Became increasingly withdrawn as she approached adolescence and with the increase in her tic severity. Mother very distressed by her daughter's aggressiveness, rage and impulsive outbursts, which peaked in mid-adolescence. Describes her daughter's behaviour and state of mind in this time as being very depressed, unpredictable, frightening and overwhelming. Finds that this has stabilized but daughter remains very withdrawn and socially isolated (prefers her own room).

Leaving school has helped her mood, which mother attributes to less pressure.

Reports no interest in any activities or socializing with anyone aside from her immediate nuclear family. She is intensely private, will not discuss her TS with anyone and rejects the idea of meeting others with TS. She now has a cat and this proving to be very helpful as she talks to and takes responsibly for the care of her pet. Mother expresses pride and admiration for her daughter's willingness to "keep going" in the face of her terrible difficulties. This is the only positive feeling she has expressed for her daughter in the current context. She has intense fears for her daughter's future, and for her own, as she does not see that her daughter will ever be able to gain any level of independence, and will require her long term, intensive care and support.

## Y & H.

### *Summary.*

- **Representation** - **Highly positive** with a tendency to minimize negative aspects of son's behaviour or personality (eg, rage, self destructive, impulsive behaviours, OCB tendencies)
- (Mothers emotional response to child in General): **Mixed**: Positive (love and joy) and Negative (sadness, fear (worry, confusion, distress)and anger (irritation frustration )

### *Representation*

Positive with a tendency to minimize negatives aspects / behaviours- high degree of empathy and mother adopts of positive psychological approach (her son sees a positive psychologist and this may also influence her representational style). Mother also has three sons with TS or OCD or both so has a high level of acceptance and understanding of TS and comorbid disorders.

Mother does not hesitate to launch into a very positive comprehensive description of her son when asked. She speaks with love, pride, affection, empathy, optimism, but by contrast does not provide a lot of detail regarding the more negative or concerning aspects of his behaviour, which become more prominent later in the interview. She treasures him as a special person and provides third party back up regarding his exceptional nature (kindergarten teacher). She describes *an optimistic, spiritual, happy, relaxed little boy who was always and still is, the peacemaker*, though his *frustration and self directed aggression* is increasing with age. He is *creative, empathic, highly intelligent, and is always looking after someone else, to his own detriment* at times (placing his needs last, like his father does). He is also *confident, charismatic and very popular with other children*, and at this stage feels that his *TS is a bonus that makes him unique*, though his self-consciousness regarding the way he presents is increasing. He *tries very hard at school* and gets on well, but his difficulties with *the structure of school* and lack of an adequate level of support are beginning to create difficulties.

Negative aspects also include her concern that *he undervalues all that he achieves* if he has required even the smallest level of assistance- and the huge blow to his self-esteem this entails and subsequent belief in his worthlessness. This tendency towards self-sacrifice, perfectionism, the extreme fatigue that results from his efforts to control his TS and OCD and a tendency to be the entertainer even at school when not appropriate are amongst his problems.

She also is inclined to glance over some fairly severe and *self-destructive impulsive behaviours*, and increasing episodes of *self-directed rage* and frustration associated with his impulsivity, Tics and OCD, even mentioning one episode of *self-harm*. He has some serious difficulties arising from his OCD, such as executive functioning difficulties that create emotional havoc and problems at home and school, (planning and organizing in particular), in addition to (OCB type) ruminative negative thought patterns, that negatively impact his mood and self-esteem. Mother acknowledges a steady rise in problems for him (*frustration, self-*



*esteem, self consciousness, decreased school performance etc)* but also tries to view this optimistically as his self management skills are increasing slightly (eg. His occasional ability to self-soothe when he recognizes an escalation in his OCD anxiety). Like a few of the other mothers, it is possible that mother must continue to minimize her sons difficulties in order to remain grounded, calm and able to cope. She has three sons with disorders and a touch of OCB herself. She admits to “*loosing the plot*” on occasions when he has “*meltdowns*”.

*As a little boy...Right well he was always happy, optimistic, relaxed um when he was really little he was easy going, the peace-maker and he still is, always is the peace maker. As he's gotten older well he was always the entertainer which then became a problem because he didn't quite get when to stop at school, but he's always the one who looks after everyone else. He's empathetic, he's creative, he's just a beautiful boy, he's very spiritual- he's into crystals and Asian things. He's just a beautiful child but as he's getting older he's getting a he gets angry very quickly,.. he's irritable, he just gets frustrated easily. I'm discovering that the peacemaker in him seems to be unfortunately he puts himself last and so he'll always say no you have it, but you know at the same time he's actually building that up, he's quietly seething or being anxious about it yeah but he and my husband's exactly the same he will always do no, no whatever you want but you know that at some point he'll take it out on himself he won't do it to anyone else he'll just get really angry about something and it's such a build up of stress because they always want everyone else to be happy. He looks after people; at kindy I used to call him the Messiah. Kids just flock they flocked to him and it's really strange he's just always had that affect on other kids. He is charismatic and outgoing. He's the one who is looking for fun things to do.*

*Has the Tourette's changed any of that for him? No they're all there because when he was diagnosed we always said to him it's not that there's something wrong with you but now we've found out why you are like you are and he always says there's nothing wrong with me it's just something extra I've got and he absolutely loves it. He loves that it's given him all those good qualities so he's amazing.. wow he's an optimist that's wonderful isn't it? Yeah is he's beautiful- he always says, we went to the psychologist and she I think she misunderstood why we were there and she was saying well you know we can make this go away and we can make you feel and he was so terrified that she was going to take away his Tourette's and I say no*

*she's only going to take away! Only take away the things that make your life difficult and help you and he was going.... no I don't want it I don't want to lose it. So he values his difference? Yeah he knows that it's what makes in him special..Oh he's just I think he's lived somewhere before I think he's just a quite advanced soul really.*

*Another problem... his perfectionist tendencies, maybe related to OCD?...and he doesn't feel like unless he's done it himself he thinks oh well you've done it all and so I'm not very good I'm really quite stupid (this baffles mother somewhat) (I think )... he's going to be fine I think he's going to be fine I think he's got um he's got it all sorted in his head he's going to be fine once he gets out of school and he can make his own structure I think he's going to be much better.*

## APPENDIX D.

## Summary of Estimated Attachment of Maternal Attachment, Mother-Child Attachment and Child-Peer Attachment for Each Dyad: Study Two

Code ID. Child's age & diagnoses	Mother's Childhood attachment	M's Safe Haven child	M's self-comfort child	M's Adult Comfort	Mother's parenting style	Mother's Representation of child (narrative)	Mothers Description of MC relationship / role	Relationship Concerns	Mum's emotion response to child	Mum's emotional response Child's distress	Mothers behavioural response child's distress	Suggested M-C attachment"	Child attachment to Peers
A 18 TS only	<i>Secure</i>	Present (Father)	Secure (father)	n/a	Different/more loving and open	MIXED ; Positive and negative (big and difficult for mum to manage and accept negative changes accompanying adolescence; change again with diagnosis of TS (more compassion and understanding)- very late at age 16- beginning to be more optimism and positive as he is happy and succeeding at work)	Changed with diagnosis, Close, Possibly Enmeshed, Regulate	Ruptures- (adolescence, aggressiveness, unanticipated), TS Confused, Badmum(Guilt)	Mixed; Positive (love and Joy) Negative. Sad, Anger, Fear	Mostly Negative and/or Overwhelmed/Numbed etc): guilt sadness, overwhelmed, plus compassion /empathy. m. Emotional response to the aggression is mostly negative	Active and positive:Talk, comfort empathy	<i>Secure</i> , but highly challenged by recent ruptures	<i>Secure</i> - 'normal' social life. Close to twin sister
B 7 TS only	<i>Insecure</i> (inconsistent/neglect) matter of fact	absent	Insecure	Secure	Different. Opposites.	Positive. No current difficulties noted (newly diagnosed)	Not changed by diagnosis, Close (perhaps because he is her only boy or the TS)	Futurechild (alleviated by husband having TS) Badmum-neglect of siblings only	Mostly Positive (only 1 neg. irritation re. Tics)	Positive; 'normal' as v. little distress	Active and positive: normal" physical , rescue, strength /calm	<i>Secure</i>	<i>Slightly insecure</i> (history of bullying). Beginning to make some friends at school
C 13.5 TS, OCD, Gifted (IQ 148)	<i>Secure</i>	present	secure	Insecure no ref. to others	Different needs/can't compare	Negative but still empathic and compassionate (change with adolescence)	Changing now (after diagnosis). Close, lioness, regulate, Interpreter, rollercoaster, Witsend	Futurechild, Ruptures (++ central issue of interview- adolescence, aggressiveness, unanticipated.),	Mostly negative Love but multiple negatives (sad, fear and anger)	Mostly Negative; Positive (love) but multiple negatives (including	Negative (currently shutdown/overwhelmed)	<i>Previously secure, now a little insecure</i> (ruptured)	<i>Insecure</i> Alienating all peers with his agro. Isolating himself as tics worsen. Better with odder and

								Badmum(sibling neglect ), Pressures( Exhaust/ Workout/burden/s truggle)		shutdown)			younger.
D 10 TS only	<i>Insecure</i> (dad died, mum unavailable); Resigned .	<i>Absent</i>	<i>Insecure</i>	n/a	<i>Different</i> (all except generational)	Generally positive but sometimes bewildered by his behaviour	Changed on diagnosis; lioness, regulate, rollercoaster UNUSUAL v. close to dad.	Ruptures (Aggressiveness), Confused, Badmum (self doubt, guilt)	Mixed positive and Negative Positive and some sadness and fear	Mixed; Love empathy but also sad, fear,	Active and Positive + and sometimes (Disconnected ) talk, physical, rescue and logical.	<i>Slight insecurity. And early signs of over-involved/ enmeshed</i> (but also sometimes mum bewildered and lacks confidence Closer to dad emotionally.)	<i>Insecure</i> (but has a couple of same aged friends). Would like more.
E 20 TS and Mild LD	<i>N/A</i> (Resigned reconciled)	<i>Absent</i>	<i>Insecure</i>	<i>Secure</i>	<i>Different</i> generational, talk, involved knowledge loving – open)	MIXED . Always some positives, but high level of needs provoked stress and anxiety Improving with maturity, and more positive/hopeful	Changed post diagnosis ; Close, Enmeshed but improving, lioness, interpreter, rollercoaster (in-adolescence)	Dependence (past) future child, Ruptures (x 3), Confused; Badmum (guilt), Pressure (strong, burden, struggle)	Mixed: Positives & lots of sad, fear and a little anger	Mixed: Positive and negatives (sad fear and anger & restraint)	Active and positive: talk, physical, strength/calm.	<i>Secure</i> ..but tested mum’s tolerance and coping ability Perhaps over-involved – enmeshed earlier age, as he was a very “needy” child	<i>Moving from insecure to secure</i> as he matures
F 11 TS Anxiety (Panic Attacks)	<i>Secure</i> (mum..dad not) Matter of fact, angry , resigned	<i>Present</i>	<i>Secure</i>	<i>Recently Widowed.</i>	<i>Different</i> to father involved, impact	Positive, mentions some negatives but high level of understanding and acceptance of these	No change, Close, lioness, regulate, interpreter, Normal,	Future child, Ruptures (very mild-adolescence), Pressures(strong. Work out, burden, struggle), Alone	Generally positive. Some, sad, fear and irritation (tics)	Mixed: Positive, with some sad, fear, and anger (injustice) and restraint	Active and Positive: Talk, physical, strength/calm distract/empathy/rescue	<i>Secure</i>	<i>Secure.</i> Small group close friends and some classmates.
G 11 TS Only	<i>Insecure</i> saddened resigned	<i>Absent</i>	<i>Insecure</i>	<i>Insecure</i>	<i>Different</i> (all except generational)	Generally positive; initially reluctant to disclose negatives.	Changed, even more protective and close (preemie baby). Close, enmeshed, lioness, regulate,	Future child, Ruptures(adolescence +), Badmum(siblings neglect), Guilt), Pressures(work	Mixed : lots of positives but negatives as well.... sad, fear and some anger	Mixed; Positive and negative (sad/fear)	Active and positive. Talk and rescue.	<i>Secure</i>	<i>Secure</i> Small group close friends, sports pals.

								out, burden, struggle)) Alone					
H 13 TS, OCD and Anxiety Blackouts? Eplispsy	<i>Insecure</i> , matter of fact, resigned	<i>Absent</i>	<i>Insecure</i>	<i>Insecure</i>	<i>Different</i> involved, talk, , acceptance, loving	<i>Positive</i> , accepting, realistic	Changed since diagnosis; Close, enmeshed, lioness, regulate, interpreter, Gift.	Dependence, future child, Confused (blackouts), badmum (siblings neglect. Self doubt, guilt) Pressures(strong, over-vigilant, work out, burden, struggle)	<i>Mixed</i> : Positives and negatives fear and some irritation /exasperation (anger)	<i>Mixed</i> ; Positives and fear/anger and restraint	<i>Active and Positive</i> : Positive, talk, physical, strength/rescue	<i>Secure...concerns re dependence/over-involvement/enmeshment</i> due to mums fear of his 'seizures'	<i>Secure</i> Several close friends, "nerdy"- like kids
I Frat.Twins 11 C TS ODD, Impulse Control Disorder H (twin) TS only	<i>Insecure</i> distressed, angry	<i>Absent</i>	<i>Insecure</i>	n/a	<i>Different</i> , impact..in every way different	<i>Generally Negative</i> - Compassionate and understanding and proud at times re C, but multiple negatives expressed. She describes herself as his "ally in everything" but her positive affective bond with his twin his much stronger and easier.	Changed since diagnosis; Lioness, Regulate, Interpreter, UandMe, Rollercoaster	Dependence, future child, Ruptures (aggro-ODD;) Badmum (self doubt), confused, Pressures (all!). Alone, Disconnect (transient)	Positive and a lot of negatives sad, fear and anger <i>Mostly negatives</i>	Positives and multiple negatives-sad, anger, restraint <i>Mostly negatives</i>	<i>Active and Positive but sometimes disconnect</i> etc. Logical/detach h/rescue/calm / (Clancy)	A challenging relationship with C. (TS +ODD) <i>Insecure-Ambivalent</i> . Secure attachment with H. (twin with mild TS only)	<i>Insecure</i> (Clancy), has a few but not close friends, poor social skills. Has twin bro.
J 8 TS and ASD (mod to severe)	<i>N/A</i> . "Cant remember"	n/a	n/a	<i>No partner</i>	<i>Same</i> for daughter, not for TS son	<i>Positive</i> ; despite his considerable difficulties and accepting and understanding	No change with diagnosis (already ASD). Close, Enmeshed, Lioness, Regulate, interpreter, UandMe, Witsend	FutureMum, Confused, BADmum( self doubt++)Pressures (all!) Alone,	<i>Mixed</i> : Positives and negatives (fear and sad in part. With some frustration)	<i>Mostly negative</i> : Positive but also sad, overwhelmed /numb/paralyzed at times	<i>Active and Positive but sometimes overwhelmed/helpless-immobile-distract/physical/ calm-strong</i>	<i>Secure but V. high level of dependence—over-involvement/enmeshment perhaps</i> . plus mum isolated	<i>Insecure</i> Mod to severe ASD precludes. Its is a source of relief to mum in some ways that he does not care how other children view him and his Tics
K. 17 TS, Depression, Rage and Impulse Control	n/a	n/a	n/a	n/a	n/a	<i>Generally Negative</i> with very slight positive (compassion and faint hope)	Closer on diagnosis; Close (past), Lioness, rollercoaster, witsend,	Dependence, future child, future mum, future rel, Ruptures (mid teens part. all three) Confused (depression)	<i>Mostly negatives</i> : Some Love but mostly negatives, fear, and anxiety and some anger	<i>Mostly negatives</i> ; Positive but generally negative. Sad, dear, anger, overwhelm	Talk and rescue but more often <i>recently overwhelmed and shutdown and detached</i>	<i>Relationship very challenged at present</i> . Hard to determine due to level of distress rendering	<i>Insecure</i> Enjoys her cat. Never really managed to be sociable-depression-pathology.

Disorder								Badmum ( self doubt, guilt) Pressures (over-vigilant, work out, burden, struggle) Alone, Disconnect (a little better of late)		ed-& numb/blunted		mother unavailable at the moment	
L 16 TS ADD, LD, OCB Traits	<i>Insecure</i> Reconciled and forgiving	<i>Absent</i>	<i>Insecure</i>	<i>Insecure</i> -Autonomous but does talk to mother a little	<i>Different</i> , more talk and understanding/acceptance	<i>MIXED</i> ; Positive and negative, reflecting recent adolescent behavioural changes (autonomy)	Changed with diagnosis; Close, Enmeshed, Lioness, UandMe, Gift	Dependence, futurechild, Ruptures), adolescence, aggro and unanticipated) Badmum (self doubt, Guilt), Pressure-work out, burden, struggle.	<i>Mixed</i> : Positive and negatives sadness, fear and anger (part related to frustration, hurt and anger re adolescent behavioural changes and resulting MC conflict)	<i>Mostly Negative</i> ; Positive compassionate with sadness anger and occasionally overwhelmed.	<i>Active and positive and occasionally disconnected</i> ; Talking (and at a bit of a loss re conflict of late)	<i>Secure</i> , however there appears to be a developmentally appropriate transfer of attachment to peers that is distressing mum	Attachment to peers improving as he gets older. <i>Moving from insecure to secure</i> . Small group of friends
M 9 TS OCD and Anxiety	<i>Insecure</i> (bipolar mother) Mater of fact, reconciled	<i>Absent</i>	<i>Insecure</i>	<i>Secure</i> (husband)	<i>Different</i> (In all ways) Being able to cope where mother couldn't.	<i>Generally positive</i> ; Very positive and compassionate even in the face of his considerable difficulties (anxiety and sensitively). A normal boy , just "extra needy and precious"	No change. had issues before (OCD, Anxiety). Close, Lioness, Regulate, Interpreter, Normal,	FutureChild; Confused; Pressures( Strong, Over-vigilant, exhausted, burden, struggle) Alone,	<i>Mixed</i> : Positive; Love and joy (love and compassion, pride and delight) with multiple negatives as well, fear sadness, and some irritation /exasperation.	<i>Mixed</i> : Positive (compassionate) with some negative, sadness, fear and an effort to suppress, retrain)	<i>Active and positive</i> ; Talk, Physical, Strength, Distraction, empathizing, Rescue	<i>Secure</i> .	<i>Secure</i> . has a small group of close , protective and accepting friends
N 12 TS, OCD Traits, ODD, Gifted	<i>Insecure</i> Matter of fact	<i>Absent</i>	<i>Insecure</i>	n/a positive relationship, with	<i>Different</i> in every way. Opposite to her childhood	<i>MIXED</i> ; Positive loving, proud and compassionate and understanding. His OCB and ODD behaviours however	Did not really change with diagnosis. Close, Enmeshed, Lioness, Regualte, Interpreter,	Dependence, Future Child, (Ruptures- Aggressive behaviour) BADmum (self	<i>Mostly positives</i> ; (love and joy) and balanced by some negatives	<i>Mixed</i> Positive (compassionate and understanding) but	<i>V. Active and positive + Occasionally overwhelmed</i> Talk, strength,	<i>Secure..with issues of over-involvement / enmeshment</i> : mum aware	<i>Insecure</i> . Can't defend himself, does not knowhow to behave socially. Lack of friends

(IQW 135) Younger bro. 10 TS and OCD.				husband		can be very challenging and difficult to manage at times and create ruptures in their relationship. However she is very forgiving, compassionate and understanding so quickly reconciles these problems. She is also determinedly positive in her approach to her son.	UandMe, Witsend (with ODD) occasionally	doubt) Pressures (all!), Alone	(Sadness, fear and anger-exasperation)	occasionally sad (helpless etc) and anger (frustration etc) and effort to restrain/suppress	rescue..	of the intensity and closeness of her relationship with her three boys (all have diagnosis),	mother's gravest concern for him
O 9 Full Blown (Super) TS, Epilepsy, Bipolar 1, Psychotic Episodes, Anxiety, OCB, provisional Atypical Autism	n/a	n/a	n/a	n/a Marital Break-down	n/a	Positive; Realistic, loving narrative of her deeply disturbed child.	Changed since diagnosis; Close, enmeshed, lioness, regulate, interpreter, UandMe, Rollercoaster, Witsend	Dependence, Futurechild, Future mum, Ruptures (Aggro and Diff behaviours), Pressures(all), Alone,	Mostly positives :Positive ; love and joy (huge compassion, as well as love, pride and delight) and powerful negatives (extreme fear, and sadness with a little anger-injustice)	Mostly Negative Positive (Deeply compassionate), but a high level of Negatives-sadness (devastation etc) and fear(intense and realistic) and occasional overwhelmed /paralyzed/ numb.	Active and positive; but also sometimes overwhelmed by his extremely high needs; Physical, calm, Distraction, rescue. Mum takes charge of everything.. she has to.	Secure..to the point of over-involvement/enmeshment but this is difficult to determine as his needs are so genuine and so high, mum does not have any real alternative.	Insecure Children can no longer cope with him. Did have friends before but they are burning out.
P 12 TS mod to severe, OCD, Anxiety, Dyslexia, Dysgraph	Insecure Implied. History of EYOH and mental health problems	n/a	n/a	Insecure - autonomous	n/a	Generally Positive: Very positive and compassionate description of her son, tempered by recent increase in aggressive behaviours since puberty (early	n/a change. Close, lioness, regulate, interpreter, UandMe (very much) Rollercoaster (just starting), Witsend occasionally	FutureChild, Ruptures (aggro, adolescence), Pressures (strong .very much so; exhausted, work out, burden, struggle, Alone	Mixed: Love and joy. (proud compassionate accepting, delight) with some sadness and fear	Mixed: Positive - Compassionate and Negative-sadness and restraint.	Active and positive. Talk, distraction and rescue	Secure, although very close they don't appear to be overinvolved-enmeshed	Insecure, his quite severe tics and multiple days off school alienate him from other kids, who reject him.

ia	in family of origin, distressed					developer)	(remote location may contribute to the closeness of the MC relationship)		(strong at times),				
Q16 TS, Mild Asperger's, OCB, Auditory processing disorder	<i>Insecure</i> Reconciled and forgiving	<i>Absent</i>	<i>Insecure</i>	<i>Secure</i>	Different mum really wanted the child. Own parents immature.	<i>MIXED</i> ; Lots of positives, but multiple difficulties were easily discussed. His high need for her attention and puberty have created stress and frustration at times. Stretched by OCB and mild Asperger's.	Changed with diagnosis; Very close, lioness, regulate, rollercoaster, witsend occasionally	Dependence, future child, future mum, Ruptures (all three), Confused, Pressure (exhausted, work out, burden, struggle), Alone	<i>Mixed</i> : Very positive; love and joy (compassion, understanding acceptance, pride, humour), with negative feelings featuring Sadness (grief, loss, hurt, dismay etc) fear and some anger (frustration, irritation, exasperation)	<i>Mixed</i> ; Positive: compassionate and negative fear, sadness and some anger (injustice, self pity)	<i>Active and positive + sometimes disconnected</i> . Now that he is 16, talk is primary mode but sometimes this leads to conflict.	<i>Secure. Very close but sometimes volatile</i> relationship, which may <i>feel a little overinvolved/emmeshed at times as she can identify closely with him</i> . Her grief over the loss of her ideal child may also impact MC attachment	<i>Insecure</i> but maybe moving towards more secure peer relationships as he gets older.
R13 undiagnosed anxiety, ADHD traits (not meet criteria) Mild LD	<i>Insecure</i> implied distressed	n/a	n/a	<i>Secure</i> (husband)	<i>Different</i> .	<i>MIXED</i> : Largely positive but as the interview progressed her over-vigilance and concern for her son's anxiety emerged.	No change with diagnosis. Close, some signs of enmeshment, lioness, regulate, rollercoaster	Dependence, mumsfuture, rupture (just a little withdrawn post puberty) Badmum (self doubt) Pressures (all!) Alone	<i>Mixed</i> : Positive (love and joy) and negative – sadness and fear	<i>Mixed</i> ; Positive (love) and Negative (	<i>Active and positive</i> : talk, physical, strength, empathizing, rescue (magic wand).	<i>Secure attachment with signs of over-involvement/emmeshment</i> that mum acknowledges needing to let go as he enters adolescence	<i>Insecure</i> with peers. He appears to be avoiding peers at this time as his tics have exacerbated.
S13 TS, ADD, OCD< Anxiety,	<i>Insecure</i> Resigned, brave face.	<i>Absent</i>	<i>Insecure</i> (implied)	n/a	<i>Different</i> . Involved Impact	<i>MIXED</i> : Positive but with multiple contradictions (eg. Multiple difficulties relating to his TS, OCD, ADHD,	No change with diagnosis; (already had OCD). Mum describes very Close, some signs	Dependence, Ruptures (Unanticipated behaviour- eg suicidality), Confused,	<i>Mixed</i> ; Positive- (Love and pride, and relief) and negative-	<i>Mostly negative</i> : Positive-compassion but more negative	<i>Active and positive but sometimes does not detect distress!</i> (disc	<i>Attachment a little uncertain</i> . Possibly <i>Secure</i> but Very close	<i>Secure</i> .. has a small group of close, "like" pals.



Auditory Processing Disorder						Anxiety, LD and depression	of enmeshment, Lioness, Regulate, Rollercoaster.. even though there is evidence of disconnection., she is “clinging”	Badmum ( Self doubt; Guilt shame re failure to recognize sons depression etc) Pressures(strong, over-vig, struggle, exhausted)	sadness and fear	fear (confusion, panicky and concerned etc) and anger (irritation)	connected) Talk, distraction, rescue.	and signs of some degree of over-involvement/enmeshed but with insight failures (or denial as ego defence?) on part of mum.	
T 14 TS, OCD Traits, Anxiety, Premature. Sister, Mild undiagnosed TS.	Insecure (with mother but did have good stepfather later) Saddened , brave face angry	Absent	Secure-stepfather (eventually) never with mother	Secure (husband)	Different Impact	Generally Positive : Very positive, loving and proud. Intolerant of those who don't understand his TS, and slightly disturbed by recent uncharacteristic but increasingly aggro behaviour (coinciding with puberty)	No change. Always very close. Possible tendency towards enmeshment (unrecognized) lioness, regulate, Interpreter,	Dependence, futurechild, future relationship, future mum, Ruptures (all three) Badmum (neglect siblings, self doubt, guilt), Pressures (strong, exhausted, burden, struggle)	Positive mostly, (love and joy) with a little negative (guilt, anxiety, irritation)	Mixed. Not a lot of distress reported	Active and positive. Talk, physical, strength	Secure attachment but mum may be slightly over-involved/enmeshed.... as evidenced by her distress at his developmentally appropriate transference of attachment to father and peers.	Secure attachment to peers “Normal”social life and some pals who defend him
U 12 TS, ADHD traits, OCB Traits, & A- 10 F. TS OCB traits, Precursor Bipolar1	Secure	Present	Insecure	Secure (husband)	Different Generational differences Openness priority.	Generally Positive; Overwhelmingly positive. Love and joy (proud, accepting etc. Acceptance of slight aggro emerging as he hits puberty and occasional rage /anxiety episodes	N/A re change; Closer Bond with Son.:V. close, Lioness, regulate, Interpreter, rollercoaster, Gift. (adversity makes him , and all, a better person)	Dependence, Confused, Pressures (strong, work out, burden, struggle, sometimes exhausted/anxious .	Mixed; Very positive (love and joy ,pride delight etc and relief) and negative. Sadness (Rejection on son's behalf, hurt, grief) and fear (anxiety and stress)	Mixed: Positive - Compassion, understanding, Negative: Fear Suppress/re train	Active and Positive. Talk, Physical, Strong, Rescue,	Secure. Incredibly close, open and sharing relationship but balanced by encouragement to learn self mgt. skills and independent.	Secure. Popular and charismatic with great group of close friends and acquaintances.
V. 11, TS and	Secure	n/a	n/a	n/a good relation	Same in general. More talk	Generally Positive with a tendency to minimize negative	n/a re change V close, signs of enmeshment,	Dependence, futurechild, Ruptures (all	Mixed: Positive (love and joy) and	Mixed: Positive-compassion	Active and Positive Talk,	Secure attachment. Deep	Secure peer attachments. Popular, with



**APPENDIX E.****Conference Abstracts: Papers Presented**

Conference Papers: October 2015 Conference: Australian Psychological Society  
(APS50) Golden Jubilee Conference,  
Gold Coast Exhibition Centre, Queensland, Australia

**“Advancing Psychological Interventions for Tourette’s Syndrome: A novel application of Attachment theory.”**

**Deirdre O’Hare**<sup>1</sup> E Helmes<sup>1</sup>, V Eapen<sup>2</sup>

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**Aim:** Tourette Syndrome (TS) is a surprisingly common, under-recognised neurodevelopmental disorder with adverse psychological, behavioural and social consequences, yet psychologists are under-represented in both the provision of care and the advancement of research in its regard. In a novel application of attachment theory, the current research tested the hypotheses that a psychological variable - the security of attachment - would account for individual differences in the quality of life and functioning of diagnosed youth and that TS would uniquely threaten the attachment relationships of diagnosed youth.

**Method:** This research consisted of a controlled, survey-based quantitative and qualitative study (Study One) of parents of young Australians with TS (n=86) and control group peers (n=108) and a qualitative interview-based study of mothers of TS youth (Study Two, n =22). A national sample was recruited from the TS support group database and multiple sites. Quantitative assessments of quality of life,

functioning, attachment and tic severity were conducted using the Paediatric Quality of Life (PedsQL) inventory, Strengths and Difficulties Questionnaire (SDQ), Attachment Questionnaire for Children (AQC) and the Parent Tic questionnaire. The security of peer, mother-child relationships (MCR) and maternal history were estimated using methodology designed for the qualitative analyses.

**Results:** As hypothesised, multivariate analyses revealed that insecure peer attachment strongly and consistently predicted impaired quality of life, higher rates of psychopathology, behavioural and social dysfunction ( $p < 0.01 - p < .001$ ).

Qualitative analyses revealed the complex impact of TS on attachment relationships.

Multiple factors effected peer attachment including personality, TS and comorbid symptom severity, the youth's psychological and behavioural adjustment to their disorder, coping strategies and the behaviour and attitudes of peers. TS determined uniquely close Mother-child relationships (MCR), shaped maternal roles and functions and multiple threats posed by TS to the MCR were identified, including high level parenting stress and maternal over-involvement.

### **“Recognising and Treating Tourette’s Syndrome in Young Australians”**

**Deirdre O’Hare**<sup>1</sup> James Cook University

**Background:** A recent Australia-wide survey suggests that psychologists are under-represented in the provision of care of young Australians with Tourette Syndrome.

With a surprisingly high prevalence (1/1,000 in paediatric populations), this complex neurodevelopmental disorder is often accompanied by comorbid disorders including OCD and ADHD and is associated with adverse psychological, behavioural and social

consequences. With no known cure and treatment greatly dependent upon psychopharmacotherapy, the need for psychologists to become involved in the diagnosis and treatment of this clinical population is evident.

**Aim:** The aim of the present session is to help interested clinicians to recognize, diagnose and treat those with this highly complex and challenging disorder. This will include providing information regarding recent developments in research regarding the aetiology of TS, the clinical phenomenology of TS in young Australians, medical and psychological management of TS, and the multiple roles for psychologists as part of multidimensional team.

**Method:** This participatory information session will focus on diagnosis, case conceptualisation and the design of interventions to suit the needs of the individuals.

Resources will include up to date psychoeducational material regarding TS, recommendations for psychometric assessment to facilitate diagnoses of TS, comorbid disorders and ongoing monitoring of symptoms, psychological and behavioural status; and information regarding existing psychological interventions such as CBIT.

**Conclusion:** The session aims to encourage and equip clinicians to work with this in need clinical population. For those with no prior knowledge or experience of TS, this session should provide a solid foundation that allows them to begin to use existing clinical skills to work with those diagnosed as part of a multidisciplinary team. The session will also provide more experienced clinicians with an opportunity to share insights and receive the most up to date information regarding the disorder, thereby enhancing their practice

**The End**