

**The Quality of Life of Young People affected by Tourette  
Syndrome**

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## **Overview**

Tourette Syndrome (TS) represents a challenge for young people and their families to manage. Some feel that their TS has little negative effect in their lives whilst for others it is extremely distressing and represents a significant barrier to their aspirations. Understanding the ways in which TS can alter Quality of Life (QoL) can give insight into the factors which influence a young person's capacity to cope successfully with the symptoms of TS.

This thesis examines the influences on young people's QoL. QoL is influenced by the severity of the TS, and the number of associated difficulties. Other psychological and social factors, such as the family environment and self-esteem, may also play an important role. Interestingly, the qualitative research in this thesis suggests that the meaning that young people and those around them develop for their TS may be another important psychological factor which influences their QoL. These multiple influences on QoL suggest that psychological and social interventions could be effective ways of managing TS.

This thesis first examines the existing research, looking for associations between TS and indicators of physical, social and emotional well-being. It then presents the results of an empirical study which specifically investigated the QoL of a group of young people who had a diagnosis of TS. It concludes with reflections on the process of designing and conducting this research.

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## **Part 1: Literature Review**

### **The Impact of Tourette Syndrome and its Co-morbidities on Young People's Quality of Life**



## **Abstract**

This review considers how young people's Quality of Life (QoL) is affected by the symptoms of Tourette Syndrome (TS). The construct of QoL aims to capture how satisfied a person is with their level of functioning in life, rather than assuming that those with mild symptoms have a higher QoL than those with severe symptoms. This may be particularly relevant in TS, where a strong relationship between symptom severity and well-being has not been established. The research into QoL in TS is limited, but there is a wide body of evidence that suggests TS can impair functioning across many domains. This research reminds us that individuals with TS are coping with more than just tics. Rage attacks, attention and learning difficulties, obsessions and compulsions are also experienced by individuals with TS and can have an equally significant impact on their life.

## Introduction

Quality of Life (QoL) assessment enables researchers and clinicians to consider not just the level at which an individual is functioning, but also how happy they are with that level of functioning. QoL can be a useful way of understanding the impact of health difficulties, and acknowledging that this impact will be different for different individuals. This may be particularly useful in relation to TS:

'It is not uncommon, for example, to see children with severe tic symptoms who are in every discernable respect happy, confident, well related, popular, academically successful and comfortable with their families; it is also not uncommon, on the other hand, to see children whose tic symptoms are mild and that nevertheless contribute disproportionately to their dysphoria, lack of confidence, low self-esteem, poor peer relations, teasing, unsatisfying school performance and family discord' (Peterson and Cohen, 1998, p.63)

The review will begin by exploring the features commonly associated with TS. It will then briefly consider the construct of QoL and how this is defined in relation to health problems in paediatric populations. Finally, it will explore the existing evidence that TS has an impact on young people's QoL.

## **Tourette Syndrome**

Tourette Syndrome (TS) is a neurodevelopmental disorder. This is a medical disorder that affects a person's neurological system during the period in their life when they are experiencing rapid neurological development. A diagnosis of TS is given when multiple motor and vocal tics have been present for over a year, and the onset was before 18 years old. Tics are the clinical hallmark of TS, and are defined as sudden, rapid, non-rhythmic, stereotyped motor movements (motor tics) or vocalisations (vocal tics). Complex features such as involuntary swearing (coprolalia) are relatively uncommon in TS, occurring in less than a third of clinical cases, and in few children or mild cases (Robertson, 1994). Whilst TS is defined by tics, there are important associations between TS and other conditions such as Obsessive-Compulsive Disorder (OCD) and Attention Deficit / Hyperactivity Disorder (ADHD) (Robertson, 2000). Indeed, TS may be part of a spectrum of disorders with common causes and varying manifestations.

### *Characteristics of Tourette Syndrome*

Generally, tics begin in early childhood and then fluctuate in number, frequency, forcefulness and complexity, reaching a peak at around 10 or 11 years old. They often then decline gradually in severity throughout adolescence and in around 40% of cases may disappear entirely by the age of 18 (Leckman, Zang, Vitale, Lahnin, & Lynch, 1998; Bloch et al., 2006). In many other cases, tics are reported to decline to a minimal level by adulthood. It is of note that tics and TS are often at their worst during key developmental stages for young people and may potentially interfere with social and emotional development.

Tics are frequently preceded by a premonitory urge, and individuals with TS describe feeling a sense of relief when the tic reduces this urge. Such urges seem

to be common in adolescent and adult individuals with TS (Leckman, Walker, & Cohen, 1993), but may be less frequent in younger children (Banaschewski, Woerner, & Rothenberger, 2003).

It is generally difficult to predict the course of TS for any one person, as initial symptom severity has a weak relationship with adult symptom severity (Bloch et al., 2006). Superimposed on these general trends is a waxing and waning of each person's symptoms that occurs over weeks and months (Lin et al., 2002). This means that it is difficult for clinicians to provide families with information about the future course of the disorder at the time of diagnosis and this uncertainty can be difficult for families to manage.

The wide range of TS symptoms has made estimates of incidence difficult, especially as up to 20% of the population will experience transient tics. Recent estimates suggest that around 1% of schoolchildren between the ages of 5 and 17 may have some form of TS (Stern, Burza, & Robertson, 2005). However, this figure includes cases where the symptoms fulfil DSM-IV-TR criteria for TS but are so mild that they are likely to be of little concern to the individual.

The symptoms of TS have been shown to exist in many different cultures (Staley, Wand, & Shady, 1997), with similar associated conditions in addition to tics (e.g. impaired anger control and ADHD (Freeman et al., 2000)). However, culture may influence the expression of more complex tics in TS; for example coprolalia is less common in Japan than in the UK and USA. The vocalisations themselves can be influenced by culture and context. They may be derogatory comments about people nearby and or may be utterances which would be considered 'forbidden' in an individual's society (e.g. shouting "terrorist" on a crowded tube train). Such "non-obscene socially inappropriate" (NOSI) behaviours are present in a significant

minority of cases (Kurlan et al., 1996). In Kurlan et al.'s (1996) survey, 22% of TS patients reported insulting others and 30% reported having an urge to insult others which they often attempted to suppress. In order to produce such inappropriate behaviour, knowledge of the behaviours that are socially inappropriate must have some influence on the process of tic production. However, it is unclear how this interaction occurs.

### *Aetiology*

No single cause has been identified for TS. It is likely that it results from interactions between a number of risk factors. One current conceptualisation from developmental psychopathology is of a syndrome that develops under certain environmental circumstances (e.g. adverse perinatal events) in individuals who have specific genetic characteristics (Spessot & Peterson, 2006).

TS is linked to dysfunction in the basal ganglia structure of the brain, particularly the caudate nuclei which are critical for the initiation of movement and more complex behaviours. Some researchers suggest that the symptoms of TS result from a weakening of the moderating influence of the frontal cortex on the caudate nuclei (Goldberg, 2001; Olson, 2004). This results in disinhibition and difficulties with impulse control.

Extending this idea, it is possible that different subtypes of TS and the different co-morbidities associated with TS could reflect different patterns of interaction between the basal ganglia and the frontal lobes. Indeed, Goldberg (2001) suggests that when the left caudate nucleus is particularly involved, TS may be associated with OCD and repetitive behaviours (seen as a form of perseveration). In contrast, the right caudate nucleus's involvement may be responsible for the association with ADHD. However, there is as yet limited evidence to support this suggestion.

In this way, TS could be seen as a marker for neurological dysfunction, and the linkage with other disorders and symptoms such as rage attacks could sometimes result from dysfunction in the same regions of the brain. Importantly, it means that some co-morbid disorders which also link to problems with impulse control (e.g. OCD) may not be separate disorders but may be part of a spectrum of linked disorders.

### *Psychological Models*

TS has been described as “psychological factors acting on a biological substrate” (Mahler, 1949) and this gives some insight into the additional influences of the environment and of the individual on the exact expression of symptoms in TS. For example, there is some evidence to suggest that tics may worsen at times of stress (Walkup, 2001).

However, there are few current psychological models of TS. There is some work which considers the cognitive and behavioural aspects of tics (O'Connor, 2002; Azrin & Nunn, 1973). Both models include the idea of a cycle in which a tic is negatively reinforced as it relieves muscular tension. However, the muscle does not return to its fully relaxed state following the tic, and this increased tension increases the likelihood of future tics. The idea of a tension-relief cycle is useful in helping individuals with tics develop strategies to manage them. It may well explain how some tics are maintained. It contributes less to our knowledge about the aetiology of TS. The models also give little consideration to the impact of having tics on the individual, and neglect the role of the social context.

O'Connor (2002) adds cognitive elements onto this basic model of a tension-relief cycle. However, O'Connor's model concerns tics in general and is not specific to

TS. In addition, it relies on research in adult populations. Whilst a correlation is shown between a perfectionist style of planning action and the presence of tics (O'Connor, 2001), cause and effect are unclear as O'Connor's results only show association, not causation.

### *Co-morbidities*

Tics are only one of a number of associated features of TS. A survey of 446 patients (adults and children) with TS considered the frequency of associated conditions present (Wand, Matazow, Shady, Furer, & Staley, 1993). Children frequently experienced sleep problems (30%), mood swings (27%), problems with temper control (30%) and aggression (20%). Problems with mood swings, aggression and temper control declined in adulthood but were still present. The majority of surveys were completed by the parent or guardian of the individual with TS, with 28.4% completed by the individual with TS and 15.2% completed jointly by the person with TS and their parents.

All of the associated behavioural features present additional challenges for the child and his/her family to cope with. Spessot and Peterson (2006) note that the 'social, emotional and behavioural problems that occur in TS may be just as, if not more, disruptive in day-to-day functioning than the motor and phonic tics that define the disorder' (p.456).

There are well-established associations between TS and other conditions, such as OCD and ADHD. This co-morbidity is frequently observed in studies using clinic populations, although this could be due to referral bias. Clinic populations are made up of individuals who have sought help to cope with their TS and this often represents the more severe end of the spectrum of a disorder. People may seek help because of the extra difficulties they experience due to their co-morbidities. It

is therefore important to check that the same associations are present in the general population of individuals with TS, including those who do not seek medical help at clinics. The same associations between OCD and TS have now been shown in epidemiological studies of the general population. This suggests that the association observed in studies using clinical populations is not just due to referral bias (Robertson, 2000).

Studies of co-morbidity in TS use a predominantly medical, diagnostic perspective and frequently consider depression and anxiety as co-morbidities alongside ADHD and OCD. Such studies of incidence do not allow for consideration of the different possible causes of such associations. Stern, Burza and Robertson (2005) suggest that OCD is an integral part of TS and is genetically linked. They also suggest that ADHD may be genetically linked in some cases. They note that depression and generalised anxiety are more likely to be secondary to the experience of having TS. For example, a child who is teased about his/her tics may be depressed as a result. They also note that depression may sometimes be a side-effect of the medication used for TS.

For this reason, disorders such as ADHD and OCD are considered here as co-morbidities, given that it seems likely they are part of the spectrum of disinhibition in TS. Depression and anxiety are considered in later sections of this review, as they are considered to be secondary to the experience of TS and are more likely to reflect the impact of TS than a common underlying cause.

## **ADHD**

ADHD is a complex syndrome. The primary symptoms include impulsivity, distractibility and hyperactivity. It is likely that there are many different causes of ADHD and the diagnosis remains controversial (Timimi & Taylor, 2004). Numerous



studies have shown that ADHD occurs in a substantial proportion of TS clinic attendees, ranging from 21-90% (Robertson, 2000). This is unlikely to be entirely due to referral bias. ADHD-like symptoms do occur in mild TS cases identified in some epidemiological studies although at a lower rate and often below the threshold for ADHD diagnosis (Mason, Banerjee, Eapen, Zeitlin, & Robertson, 1998). However, this association is not always present (Peterson, Pine, Cohen, & Brook, 2001).

The factors behind the co-occurrence of ADHD and TS are unclear. Some authors have suggested a genetic or biological relationship, perhaps reflecting dysfunction in a common underlying biological substrate (Goldberg, 2001). Psychological mechanisms may also have a role. For example, it takes a considerable amount of attention to suppress tics and premonitory urges can also be distracting (Leckman, Peterson, King, Scahill, & Cohen, 2001). Premonitory urges may well contribute to the attention problems that accompany TS, as they can lead to an uneasy sensation and a resulting difficulty in sitting still or concentrating. In summary, it seems likely that symptoms labelled as ADHD in TS may reflect a number of underlying causes and may or may not be secondary to TS.

## **OCD**

There is a clear and strong association between TS and OCD in individuals with TS and their family members (Miguel, Shavitt, Ferrao, Brotto, & Diniz, 2003; Robertson, Banerjee, Eapen, & Fox-Hiley, 2002). It seems likely that TS and some cases of OCD are variable expressions of the same genetic vulnerabilities. It may be that tics and compulsions represent different points on a spectrum of similar behaviours (Cath et al., 2001).

However, it seems that the obsessive compulsive behaviour (OCB) and obsessive

compulsive symptoms (OCS) seen in TS are different to those seen in pure OCD (Robertson, 2000). The compulsions in TS are related to ordering, repeated touching, symmetry and getting things 'just right'. The obsessions may also have different themes. In pure OCD, the obsessions often relate to contamination and fear of something 'going wrong' and the compulsions are more likely to be preceded by autonomic anxiety and cognitions (Cath et al., 2001). This suggests that symptoms which we group under the label of OCD may have different aetiologies, some of which are related to those which cause TS.

### **Learning Disabilities**

General intellectual ability, as measured by intelligence quotients (IQ), does not seem to differ in individuals with TS compared to the general population when measured in epidemiological studies (Como, 2001). However, below average scores on tests of IQ have been reported in a clinical sample of children with TS when compared to population norms. There is therefore a discrepancy between clinical and non-clinical populations of individuals with TS and it seems that individuals with TS who attend clinics may be more likely to have lower scores on tests of IQ. The reasons for this discrepancy are unclear in the literature at present.

Burd, Freeman, Klug, and Kerbeshian (2005) compared individuals with TS with and without learning disabilities found that learning disabilities in TS were associated with five factors. These five factors were;

- Being seen for evaluation before the age of 18 years
- Being male
- Having fewer family members with tics or TS
- Having more perinatal problems
- Having more co-morbidities (especially ADHD).

The reasons for these associations are not yet established. In addition, the term

'learning disability' is used without a precise definition of the criteria for this term and this means that the study population is likely to be heterogeneous. This makes the results hard to interpret. One hypothesis might be that learning disabilities are more likely in more severe TS, but this is not a clear conclusion from the limited literature. Burd et al. (2005) also demonstrate that there is an increased prevalence of learning disabilities (LD) in individuals with TS but prominent learning and school problems are often highly correlated with the presence of ADHD.

### **Aggression and Rage Attacks**

Individuals with TS and their families also report aggressive behaviour, or sudden outbursts of rage. This can be seen as another feature of the disinhibition seen in TS. It could also be related to a child's frustration at not being able to manage their symptoms. Aggression is associated with comorbid ADHD or OCD (Budman, Bruun, Park, Lesser, & Olson, 2000; Stephens & Sandor, 1999), but is independent of tic severity or age. These studies were based on a clinic population, and so may not represent the population as a whole, as clinical populations tend to have more severe symptoms, which are more likely to encourage families to seek help.

### **Summary**

In summary, whilst it is clear that TS does co-occur with other disorders the reasons for this are complex. It may be that the associations have different causes in different individuals. It could be that a family environment that pre-disposes someone to TS or OCD also increases the chance of ADHD symptoms. Alternately, the linkage could represent dysfunction in the same areas of the brain, or genetic linkage.

Clinically, it is important to keep in mind that families are coping with more than just tics. Other features are likely to be present and may be equally or more problematic

than tics. For example, it may be that ADHD features have a much more significant impact on a child's QoL than tics. This association with QoL may not be related to symptom severity, as it is possible that some difficulties are just much more difficult for children and their families to manage. This review will now move on to briefly consider the construct of QoL before looking at the relationship between TS and QoL.

## **Quality of Life**

Quality of life (QoL) assessment measures seek to measure the well-being of individuals. The World Health Organisation (WHO) describes QoL as 'the individual's perception of their position in life, in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns' (WHOQOL Group, 1995). The inclusion of context is important, allowing for the way in which individual's expectations are influenced by their culture and environment. To understand whether a person is satisfied with their quality of life, it is necessary to compare a person's perception of their current situation to their ideas about the situation they aspire to. Prince and Prince (2001) noted that the consensus in the literature is that 'conceptual models should incorporate the relationship between the objective and the subjective dimensions of the human experience' (p.1008).

Criticisms of the QoL construct note that a rush towards measurement has meant that the underlying construct is not well tested or well-established (Hunt, 1997). This is evident in the diversity of measures of QoL, many of which rely on face validity and few of which specify the underlying construct they are trying to measure.

### *Subjective Quality of Life*

Subjective QoL constructs rely on the 'standards of the respondent to determine what is a good life' (Diener, 1984, p.534, cited in Prince & Prince, 2001). Subjective QoL measures consider individual difference and meaning. They also raise the question of who should determine what QoL is, for example, whether professionals or patients should determine the impact of an illness. Many existing outcome measures reflect what professionals and researchers see as important (objective QoL). For example, many clinical trials look at whether the symptom severity is reduced to judge whether a treatment is effective. Subjective QoL measures assess what the individual sees as important. For example, the Child Health-Related Quality of Life Questionnaire asks about functioning in a number of domains, satisfaction with this functioning and upset about this functioning (Graham, Stevenson, & Flynn, 1997). If school performance is not important to the child, he or she could score poorly on the level of functioning but not be upset by this and be satisfied with it.

It is likely that different measures of QoL are best suited for different purposes (Titman, Smith, & Graham, 1997). For example, the Pediatric Quality of Life Inventory (Varni, Seid, & Rode, 1999) provides a brief screen of a number of domains and so is useful for a brief assessment of functioning and QoL and for making comparisons. Semi-structured interviews and qualitative analysis would provide much more insight into an individual's own standards and their satisfaction with their quality of life, as subjective QoL can never be completely captured in a questionnaire. De Civita et al. (2005) note that '[m]ethodological triangulation, which is essentially the blending of qualitative and quantitative research approaches, shows promise in ensuring that the final product is meaningful from the child's perspective' (p. 667).

Therapeutically, subjective QoL measures usefully remind us that improving an individual's sense of well-being does not necessarily rely on removing their symptoms. Successful treatment could equally focus on psycho-education, or on helping the individual to develop a helpful meaning for their symptoms.

### *QoL and paediatric populations*

With younger populations, it is also important to consider developmental stage when assessing QoL (Eiser & Morse, 2001). The developmental stage is important in two ways. Firstly, the child needs to be able to reflect to some extent on their internal and external world and consider what distresses them and what makes them happy. Measures need to be simple enough to allow them to do this. Secondly, the stage of development may influence the child's priorities and aspirations. At different stages different domains become relevant. For example, concerns about physical appearance become more pronounced in adolescent girls (Hoare, Elton, Greer, & Kerley, 1993).

The QoL of the young person may also be assessed by asking informants such as parents and teachers to judge the child's QoL, using a proxy report. Their view may be different to that of the child, and it is important to bear this in mind when considering research that relies on parental report. It has been noted in previous research that the QoL rating of parents and clinicians often concern observable features such as the social skills of the child, whilst those of the child seem to represent their inner world, for example self-esteem (Bastiaansen, Koot, & Ferdinand, 2005). Parents and clinicians are not able to judge the child's subjective QoL. Only the child themselves can say which problems are most significant and distressing for them.

## *Summary*

In summary, a multitude of measures of QoL exists but the construct underlying some of them remains poorly defined. QoL measures need to strike a balance between capturing the individual's subjective view and allowing measurement of change and comparison by using more objective methods.

## **Impact of Tourette Syndrome on Quality of Life**

Few studies have considered QoL in TS and no studies to date have investigated QoL in young people with a diagnosis of TS. This section of the review will therefore begin by looking at the few studies which consider the impact of TS on QoL in adults. It will then move on to consider other relevant studies which look at the impact of TS on functioning in young people. Core domains of QoL in children have been identified as physical, emotional, and social (Seid, Varni, & Jacobs, 2000). Education is often added to these domains to reflect the importance of this setting in children's lives. The review will therefore consider the impact of TS in each of these domains. Many studies cover more than one domain and this will be noted where relevant.

## *Search Strategy*

A defined search strategy was used to identify literature that was relevant to the research question. Both PsycINFO and MEDLINE were searched for relevant articles prior to April 2007. Literature relevant to TS was identified using free text searches of 'tourette\*' <sup>1</sup>, 'tic' or 'tics' or the index term 'Tourette Syndrome'. When combined with search terms for QoL a small number of relevant articles were

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<sup>1</sup> The star represents truncation – Tourette\* finds Tourette OR Tourette's OR Tourettes

identified. The search was therefore expanded to consider emotional, behavioural and social functioning, using the search terms in

Table 1. Relevant review articles were also searched for references (Robertson, 2000; Peterson & Cohen, 1998; Spessot et al., 2006). Particularly relevant articles were also cross-referenced.

**Table 1: Search Terms used to Identify Relevant Literature in PsycINFO and MEDLINE.**

Domain of Functioning	Terms Used	
	PsycINFO	MEDLINE
QoL	"Quality of Life" or "LIFE SATISFACTION" or "QUALITY OF LIFE"	"Quality of Life" or "QUALITY OF LIFE"
Emotional	Psychopathology or "PSYCHOPATHOLOGY"	"Psychopathology" or "PSYCHOPATHOLOGY"
Emotional	"EMOTIONAL STATES" exploded or "STRESS" exploded	"EMOTIONS" exploded or "AFFECTIVE STATES" exploded
Emotional	"Self esteem" or "SELF CONCEPT" exploded	"Self-Esteem" or "Self Esteem" or "SELF CONCEPT"
Behavioural	"BEHAVIOR" exploded	"BEHAVIOR" exploded
Social	"FAMILY" or "FAMILY RELATIONS" exploded	"FAMILY" exploded or "FAMILY RELATIONS" exploded
Social	School*	School*

*Note.* Thesaurus or Index Terms are shown in capitals. Where search terms were exploded (i.e. all subordinate categories were also searched) this is noted.

## *Tourette Syndrome and Quality of Life*

### **TS and Quality of Life in Adults**

One UK study has shown that adults with TS have a significantly worse QoL than a general population sample but better QoL than patients with intractable epilepsy (Elstner, Selai, Trimble, & Robertson, 2001). QoL was measured in two ways; firstly, using the Quality of Life Assessment Schedule (QOLAS) (Selai, Trimble,



Rossor, & Harvey, 2001) and secondly, using the Medical Outcomes Study 36-Item Short-Form Health Survey (SF-36) (Ware, Snow, & Kosinski, 1993).

The QOLAS is a subjective measure of QoL and asks the patient to recount what is important for his/her QoL and then determine how TS affects this QoL. The QOLAS assesses the difference between expectations and reality for a number of distinct domains (physical, psychological, social, daily activities and cognitive functioning), which fits well with subjective constructs of QoL as defined by Cantril (1965). Whilst it is a subjective measure, comparison between groups is possible as the patients are asked to rate the size of the problem, providing quantitative data. It represents a good balance between research demands and a method of capturing clinically meaningful individual difference.

As can be seen in Table 2, people with TS were most likely to rate motor tics as a problem and mentioned that these can cause physical injury, exhaustion and interfere with daily activities. Adults also noted that concentration was a significant problem (e.g. “can’t listen attentively”), as was memory (e.g. “remembering appointments”). These are features which could be considered as ADHD-related symptoms. Alternately, they may reflect the impact of trying to control tics and worrying about TS. Depression and low mood were also reported frequently, however the reasons for this low mood were not established in this study.

The second measure of QOL in this study, the Short Form-36 (SF-36), found that patients with TS had worse scores on all subscales than the general population. The SF-36 is more functional in its assessment of QoL, looking at the impact of health difficulties on a number of activities (e.g. bathing and walking) and then asking individuals to assess how much their health problems have interfered with their life over the last two weeks. Increased TS symptom severity was associated

with poorer QoL as indicated by this measure, as was depression (assessed using the Beck Depression Inventory (BDI)). Other factors which influenced QoL were employment status, obsessive compulsive behaviours and anxiety.

**Table 2: Main problems reported by individuals with TS**

Dimension	Problem	Total (%)	Severe (%)
Physical	Motor tics	78	25
	Vocal tics	35	11
	Coprolalia	7	2
	Obsessive compulsive behaviour	19	9
	Drug side effects	9	5
Psychological	Depression	51	23
	Anger/aggression	31	15
	Anxiety	29	13
	Mood swings	19	8
	Low self-esteem	15	6
Social	Family	29	19
	Making friends	27	12
	Social life	20	9
	Self-conscious	15	8
Cognitive	Concentration	67	23
	Memory	50	17
	Short Attention Span	18	6
Economic	Interference with work/study	48	34
	Made redundant / unable to work/study	19	-

*Note.* Patients nominated two items of importance in each of the five domains and rated the severity in each. The percentage of respondents who listed each item is shown in the column Total, and the percentage who listed each problem as severe is shown in the column Severe. Taken from Elstner et al. (2001) (p. 55)

It is clear that the two methods of assessing QoL provide very different information. The functional SF-36 tells us that TS affects people's functioning across many domains and allows comparison with the general population. However, it does not tell us what causes these changes in functioning, or tell us whether people are distressed by these changes. In addition, the domains are pre-determined as the

measure was designed to be relevant for many different disorders. This means it may not capture those that are particularly important in TS. For example, the focus on physical activities does not capture the social aspects which are key to TS.

In contrast, the more qualitative QOLAS allows participants to describe the biggest problems for them. This information is perhaps more helpful clinically, particularly as there is little previous research telling us why QoL is adversely affected in TS. It measures a construct that fits with currently accepted definitions of QoL, and so has good face validity.

The study used a clinical population of adults and most of the patients in this study had long-standing TS. It is hard to generalise to a population of children with TS. Adults who are in contact with a TS clinic tend to represent the most severe end of the spectrum. Most children who have TS do not go on to attend adult clinics, as symptoms generally improve significantly with age. As TS commonly presents for the first time in childhood, children will have only recently been given a diagnosis of TS. In contrast, adults have known their diagnosis for many years and have been managing their symptoms for many years. This may alter the impact of the TS in children compared to adults.

### **TS and Quality of Life in Children**

There is almost no research which looks at QoL in children with TS. One study in Canada (Dooley, Brna, & Gordon, 1999) did not explicitly assess QoL but it did ask the families of children with TS to rate how “significant/bothersome” they found a number of symptoms. The results are shown in Table 3.

This is essentially subjective QoL. It is assessing not just the presence of symptoms but is also assessing how distressing families find these symptoms. The most

common features of TS, motor and vocal tics, were not the most bothersome for the families. Rage, attention deficit and learning difficulties were reported less frequently than tics but families believed these symptoms to be very significant/bothersome when they were present. It was the parents who completed the questionnaire in most cases and so these results represent the parent's perceptions of the most significant problems. Further research is needed to understand what the children's views would be.

**Table 3: Families' experiences of TS symptoms (Dooley et al., 1999)**

Symptom	Present at time questionnaire completed (%)	Rated as very significant when present (%)
Motor tics	98	9
Vocal tics	91	7
Attention deficit	85	29
Compulsions	79	14
Learning difficulties	68	24
Hyperactivity	67	16
Obsessions	64	21
Rage	59	36

In a large postal survey of 466 adults and children with TS in Canada, an open question asked them to identify the most disabling aspect of TS (Wand et al., 1993). The responses of adults and children were similar. The most disabling aspects of TS listed by both groups, beginning with the ones identified most frequently, were:

- Social isolation / embarrassment
- Severity of tics
- Lack of public and professional knowledge about TS
- Attention / learning problems
- Associated conditions – depression, anxiety, aggression, phobia, mood swings, and self-mutilative behaviour.

More children than adults identified attention and learning problems as a disabling feature. It would be interesting to find out whether this is because the educational context that they are in places more demands on their attention and their ability to learn is being explicitly evaluated at school. The classroom environment requires a child to be able to listen attentively and work independently, with little flexibility for breaks. In adulthood, individuals with TS may be able to find an occupation which fits with their abilities and so makes it easier for them to attend and concentrate.

The study provides limited information from one question, but having such an open question was a useful and simple way of allowing people to identify the features that are most important to them. The results of the study again highlight the social impact of TS, although it is not clear whether such social isolation leads to emotional difficulties.

## **Summary**

Studies looking at QoL in adults with TS have noted the social and emotional impact of the physical symptoms of TS and it seems to be in these areas that the individual's QoL may be affected most detrimentally. It is not clear that the research with adults can be generalised to child populations, although there may be some similarities as suggested by Wand et al's study (Wand et al., 1993).

The studies considered above either investigated QoL explicitly (Elstner et al., 2001) or assessed the distress caused by symptoms as part of a wider study (Dooley et al., 1999; Wand et al., 1993). As there is little research on QoL in young people with TS, studies of functioning may provide further clues as to which areas of their lives are most affected by TS. As noted earlier, functioning (e.g. performance at school, mood) is not the same as QoL, but there is some overlap between the constructs.

This review will now consider the impact of TS in three different domains of functioning; physical, emotional and social.

### *Physical impact of TS*

Few studies look at the physical impact of TS, other than to measure the frequency and severity of tics. Some authors have noted that although pain is not generally recognised as a symptom of tic disorders, there are some cases where pain is a prominent feature. For example, a repetitive tic such as neck movements may cause neck pain (Riley, 1989). It is not clear whether the lack of studies reflect researchers' views that this area is not important. It would be useful to ask individuals with TS whether it does have a physical effect which bothers them.

### *Emotional impact of TS*

As noted previously, most studies looking at the emotional impact of TS do so using standardised measures to identify the severity of symptoms of 'disorders', such as depression or anxiety. Conceptually, many studies consider depression and anxiety as co-morbidities rather than the emotional consequence of TS. This gives little insight into why TS has such an impact on individual's emotions and mood and why this impact differs between individuals. However, the results of the studies can help to establish whether TS does impact on mood. Given that the research uses the categories of depression and anxiety it is useful to explore the research using the same categories.

### **Depression**

Robertson provides a comprehensive review of the relationship between depression and TS (Robertson, 2006). Most studies of children use the Child Depression Inventory (CDI) (Kovacs, 1985) to assess symptoms and others use the relevant dimension of the Child Behaviour Checklist (CBCL) (Achenbach, 1991).

Studies using the CDI generally show that depressive symptoms are elevated in children with TS when compared to controls (Carter et al., 2000; Robertson et al., 2002) with the exception of one study which found no difference (Termine et al., 2006). The study by Termine et al. (2006) had only 17 participants with TS and so it is possible that the power of this study was not large enough to detect an effect. Studies using the CBCL have also found that individuals with TS score higher than controls on the anxious/depressed dimension of Internalising Symptoms (Carter et al., 2000; Cardona, Romano, Bollea, & Chiarotti, 2004; Termine et al., 2006; Nolan, Sverd, Gadow, Sprafkin, & Ezor, 1996; Hoekstra et al., 2004). Studies using different measures also find increased depression and anxiety (Wodrich, Benjamin, & Lachar, 1997).

A study in China of a group of children with TS found that the older children (15-19 years) reported more emotional distress and the younger children showed more somatisation and obsessive-compulsive symptoms (Chang, Tu, & Wand, 2004). The presence of emotional distress fits with the results from the above studies. Younger children may express distress in different ways and may find it harder to think about and verbalise their distress in order to fill in questionnaires.

There are inconclusive results as to whether increased tic severity correlates with increased depressive symptoms. Carter, O'Donnell, Schultz, Scahill, Leckman and Pauls (2000) found no relationship in a study involving 72 children, whilst Cardona, Romano, Bollea and Chiarotti (2004) found that internalising problems increased with the duration of the tic disorder. Hoekstra et al. (2004) found that scores on the anxious/depressive subscale of the CBCL correlated with ADHD symptom severity and with OCD severity but had no significant relationship with tic severity.

The mixed results may reflect the small numbers of participants in most studies, and

the different measures used in different research groups. However, it is also clear that hypothesising that depression is directly linked to tic severity will always oversimplify the relationship. Cognitive psychological approaches remind us that the meaning of the symptoms to the individual is critical. The impact of having tics on mood will be mediated by an individual's beliefs and thoughts about the tics. Correlational studies based on surveys are not able to take account of individual differences in beliefs.

In her review, Robertson (2006) notes that TS in itself is distressing, particularly with severe tics, but that depression could also result from the association with OCD or ADHD, or be a side effect of medication. It is likely that all these explanations are true in different cases and this highlights the difficulties associated with considering 'depression' to be a unitary disease entity rather than a mood state which can reflect a multitude of causes.

### **Anxiety**

Anxiety has received less attention in young people with TS. It is often considered alongside depression, as grouped in the CBCL. One study used the Test Anxiety Inventory but found no differences (Termine et al., 2006). However, this measure has poor content validity as a measure of general anxiety associated with TS. It looks specifically at anxiety related to test performance. In addition, as noted previously, the study may not have adequate power to detect an effect. Studies in adults using the Spielberger State-Trait Anxiety Inventory have found that adults with TS are more anxious than controls (Eapen, Fox-Hiley, Banerjee, & Robertson, 2004). This study suggested that increased levels of anxiety were not always related to OCD tendencies.



## **Self-Esteem**

A few studies have looked at self-esteem in TS, which could be considered as one mediating factor between TS and mood. One study which used the Self-Perception Profile for Children (Harter, 1985) found that children with TS were only different to controls in one domain, that of athletic self-competence (Bawden, Stokes, Camfield, Camfield, & Salisbury, 1998). There were approximately 25 children in the TS group and in the control group, so power was limited. Carter et al. (2000) also used the Self-Perception Profile for Children and found no significant differences between controls and children with TS on the global and social domains but did find a significant difference on the behavioural subscale. It seems that there is no general impact of TS on self-perception. However, the variability in the sample was not reported. Whilst there was no difference overall, there may be significant individual difference in the extent to which TS affects a child's perception of themselves.

A larger study by de Lange (de-Lange, 2000) found that adolescent boys with TS differ significantly from those without TS in terms of non-academic, academic and global self-concept. It is possible that TS could have more influence on self-esteem in adolescence, as self-concept is perhaps more fluid and vulnerable at this life stage. More longitudinal studies would be needed to investigate this.

## **Behaviour**

As noted in the study by Chang et al. (2004), younger children may express distress in terms of behaviour. This is recognised in the CBCL which divides symptoms into those which are internalising and those which are externalising. Some studies have found elevated scores on the externalising domains of the CBCL (Nolan et al., 1996; Termine et al., 2006; Wigley et al., 2000). Another study used the Connors scale and found a correlation between tic severity and the level of behavioural dysfunction (De Groot, Janus, & Bornstein, 1995).

There are many possible causes of difficult or disturbed behaviour. As noted earlier, rage attacks and mood swings can be a feature of the disinhibition associated with TS. These could elevate scores on measures such as the CBCL and Connors but reflect a part of the syndrome rather than a measure of its impact on the child. Equally, ADHD symptoms need to be carefully considered. Careful assessment is needed to consider the causes of the behaviour and distinguish behaviour that results directly from tics, ADHD and OCD and behaviour that is more reflective of the impact of TS. Parents may have difficulties making this distinction and it can be hard for the family to manage the child's behaviour when it is unclear if the behaviour is voluntary or involuntary.

Many studies use parental reports of behaviour and it is important to note that using other informants would possibly provide different results. Christie and Jassi (2002) found that teacher and parent reports on the CBCL were very different. Teachers tended to report fewer behaviours as concerning and rate those that were reported as less severe than parents did. This could reflect the different sensitivity of the reporters, or it may be that TS is experienced differently in different contexts.

## **Summary**

TS is associated with low mood and depression in young people, it seems likely that there are a number of different reasons for this association. The relationship to generalised anxiety is not yet established. While there is no general impact on self-perception, it is likely that individual differences in self-perception are masked in studies making group comparisons. It would therefore seem that TS has a negative impact on emotional state. It is unclear whether this impact reflects a poorer quality of life in TS.

## *Social impact of TS*

The impact of TS on the family, a key social context for young people, is considered here. The social impact of TS on peer relationships is also examined.

### **Family**

Several studies have used measures of family functioning in families where a child has been diagnosed with TS. Some view poor family functioning and caregiver stress as an outcome of the child's TS, indicating its impact on the family (Bawden et al., 1998). If TS impacts negatively on the family then this is likely to adversely affect the child's QoL. Other studies consider family functioning as a mediator of the impact of TS on the child's well-being. For example, a positive family environment could mean that TS has less impact on a child's QoL (Carter et al., 2000).

One study (Bawden et al., 1998) asked families to complete two measures of family functioning; the Family Assessment Measure (FAM-III) (Skinner, Steinhauer, & Santa-Barbara, 1984) and the Family Adaptability and Cohesion Scales (FACS) (Olson & Tiesel, 1991). The FAM-III assesses family members along seven key dimensions, as specified by the Process Model of Family Functioning. Bawden et al. (1998) compared families where a child had TS to families where a child had diabetes. There was evidence that mothers of children with TS had more difficulty solving family problems effectively than mothers of children with diabetes. There were no differences between the groups of fathers. The FACS showed a tendency by mothers and fathers of children with TS to rate their families as less cohesive than mothers and fathers of children with diabetes, although this difference did not reach significance. This reduced cohesion could reflect the effect of behaviour which is difficult to manage, or of the rage attacks experienced by some children with TS.

However, there was little overall difference between the groups. This may in part be due to high levels of variability in family functioning, which makes group comparisons difficult unless measures are specific and investigate clear hypotheses. Most significantly, neither group (TS or diabetes) was significantly different from the mean of the normative data for the measures. This may indicate that families of children with TS do not function in a way that is significantly different to other families.

TS may not alter family functioning, but it may challenge it and present additional difficulties for the family to manage which could mean that the well-being of the child within the family was altered. However, it is important to bear in mind that most families do successfully cope with the challenges presented; only 4% of families indicated that family problems were the most disabling aspect of TS (Wand et al., 1993), and when Elstner et al. (2001) asked individuals with TS to report any problems only 29% reported problems in their family. At the same time, TS can interfere with the family's daily activities, although they may not perceive this as a problem. In a survey of over two hundred Canadian families, many families indicated that TS often interfered with daily activities (Hubka, Fulton, Shady, Champion, & Wand, 1988).

Several studies have investigated the impact of TS on the child's carers. If the child's carers are struggling to cope, this will affect the child's care and could also affect their QoL. One study used the Family Impact Scale (FIS), a 34-item scale that consists of statements regarding the way in which an "ill" family member has affected the family, in terms of relationships, social life and finances (Wilkinson et al., 2001). This study found that TS had significant negative effects on the family. Greater symptom severity and increased co-morbidity (e.g. ADHD and OCD)

increased the impact on the family. The external validity of the study may be reduced, as all participants were members of the Tourette Syndrome Association. The sample could therefore be unrepresentative of the general population of individuals with TS. However, the results have been replicated in another study, which also found that the impact on the family was related to associated co-morbidities (Woods, Himle, & Osmon, 2005).

Another study investigating the impact of TS on the family (Cooper & Livingston, 2003) asked parents to complete the Child and Adolescent Impact Assessment (CAIA; (Messer, Angold, Costello, & Burns, 1996), a structured interview which looks at caregiver burden in four areas; finances, relationships, activities and well-being. The study also asked parents to complete the General Health Questionnaire which was used as an indication of their mental health, and asked an open question 'In what way does your child's illness impact most on your life?'.

The study used a comparison group of parents of children with asthma. The results showed that parents of children with TS were more likely to be 'psychologically unwell' (as indicated by a cut-off on the GHQ), and also experienced greater burden in the domains of relationships, well-being and activities. This suggests that the stress of TS is greater than the general stress of having a child with a chronic, severe illness. The main subjective causes of stress in parents of children with TS related to behaviour problems, and disagreements about how to manage the behaviour (e.g. distinguishing TS symptoms from misbehaviour). In the asthma group, concerns were more illness related. This highlights the difficulties for the family in coping with difficult behaviour over which the child has little control and in separating this from behaviour over which the child does have control.

Investigating this idea more fully, a qualitative study with mothers of children of TS,

looked at how they experienced and managed the aggression of their children (De Lange, 2000). The author noted that, when aggressive behaviour was present, the mothers had extreme difficulties in managing their child's behaviour and in coping with their own feelings. They questioned their parenting skills, were uncertain whether they were doing the 'right' thing and were anxious and concerned about the future of their child. This research suggests some reasons why caregiver burden may be increased in parents of children with TS, although it should be noted that not all children with TS exhibit aggressive behaviour. The study also highlighted that different mothers had different strategies for managing the child's behaviour – this raises the possibility that some families may be more successful at helping the child cope with the symptoms of TS than others.

Family characteristics may also act as mediating factors, altering the level of emotional or behavioural problems experienced by the child as a result of TS. Carter et al. (2000) noted that family functioning was associated with parental ratings of children's adaptive competence in the domain of socialization, even after controlling for diagnosis (TS and ADHD). Children in families where functioning was better were rated as more socially competent. This highlights the importance of assessing the child in the context of his or her family. It may be that emotional difficulties or behavioural problems are related to the family context rather than to the child's TS.

Finally, the family context may help the children cope with their TS and reduce the impact of TS on their QoL. One study found a significant, positive relationship between the self-concepts of children with TS and the self-concepts of those children's mothers (Edell-Fisher & Motta, 1990). This hints that a positive family environment could minimise the impact of TS on children's QoL.

In summary, it is clear that TS can present a challenge for families which can result in increased stress and impaired QoL. However, there may also be family characteristics that minimise the impact of TS. The causality may be bi-directional, with the TS symptoms affecting the family at the same time that family factors mediate the impact of the TS symptoms.

### **Peer Relationships**

Bawden et al. (1998) asked the classmates of children with TS to complete the Pupil Evaluation Inventory (PEI) (Pekarik, Printz, Liebert, Weintraub, & Neale, 1976). Each child in the class rated their classmates' social behaviour, and the results were grouped into factors labelled Aggression, Withdrawal and Likeability. Children with TS and ADHD were more likely to be seen as aggressive and withdrawn by their peers, and less likely to be nominated as likeable. Children with TS alone were more likely to be labelled as withdrawn, with no differences on the other two scales.

This study is interesting as it gives an indication of how children with TS are perceived by their peers. It seems that ADHD symptoms have a negative impact on peer relationships. However, TS alone does not affect peer relationships so negatively, although classmates reported that their peer with TS was withdrawn. The reasons for this withdrawal are less clear – it could reflect embarrassment, the effort of tic suppression, poor social skills or low mood. Children and adolescents can be embarrassed by their tics, and can fear losing control over tics in public which may affect their confidence in creating peer relationships (Rindner, 2005).

Another study (Hoekstra et al., 2004) assessed children's social behaviours using the Children's Social Behaviour Questionnaire (CSBQ), a parent-report measure (Luteijn, Luteijn, Jackson, Volkmar, & Minderaa, 2000). Their results indicated that poor social behaviours were associated with ADHD diagnosis. However, there may

be some circularity in these arguments, as it is possible that the CSBQ is measuring many of the same behaviours which caused the child to be given a diagnosis of ADHD. Whilst the CSBQ has questions which are beyond the defining symptoms of ADHD (e.g. social insight), they may still be associated with ADHD symptoms.

In conclusion, it seems possible that TS could affect the child's relationships with their peers in some cases, particular when associated with other externalising difficulties. It is not clear whether the children are distressed by this.

### *Impact of TS on Education*

Two studies of TS across whole school populations have noted that children with TS are more likely to be in non-mainstream classes, and are more likely to have impaired educational performance than other children in their school (Khalifa & von Knorring, 2003; Lanzi et al., 2004). A survey of parents of children with TS found that around a half of respondents reported moderate to severe academic impairment and peer problems. These difficulties may be related to ADHD (Abwender, 1996). There is limited research in this area, and the causes of the problems are unclear. It could be related to difficulties managing tics in the classroom. It is also unclear if these educational difficulties are distressing for the children and their families.

### *Impact of Stress on Tourette Syndrome*

The studies considered above have generally considered how TS causes distress or difficulties in functioning. However, all studies considered are correlational and show associations rather than causality. It is important to keep in mind that stress can affect the expression of TS. Individuals with TS may experience worsening of tic symptoms at times of stress (Walkup, 2001). In addition, individuals with TS may be at increased risk of experiencing higher levels of psychosocial stress and adversities throughout their life (Findley et al., 2003). Adverse circumstances could



therefore lead to a spiralling of symptoms due to positive feedback between stress caused by symptoms and symptoms escalated by stress.

### *ADHD, OCD and QoL*

Whilst there is no research on the impact of TS on children's QoL, there is research which considers the effect of ADHD on QoL. One study asked parents of children with ADHD referred to an ADHD clinic to fill in the 50-item parent version of the Child Health Questionnaire which measures psychosocial and physical health (Klassen, Miller, & Fine, 2004). When compared to norms for children without ADHD, the children with ADHD had comparable physical health, but deficits in all psychosocial domains, family activities and family cohesion. Greater symptom severity and greater co-morbidity resulted in a greater impairment of QoL. A later study by the same group (Klassen, Miller, & Fine, 2006) compared parent and child report of QoL and found more agreement on domains measuring observable features (e.g. physical function) and less agreement on domains which are not easily observed (e.g. self-esteem). Children tended to report their QoL more favourably than their parents.

Another study, using a more subjective measure of QoL, confirmed the finding that QoL is poorer in children with a diagnosis of ADHD (Lange et al., 2005). The result has also been replicated using other measures (Varni & Burwinkle, 2006; Escobar et al., 2005).

There are no studies of QoL in paediatric populations who have a diagnosis of OCD, but in adult populations QoL has been shown to be impaired (Sorensen, Kirkeby, & Thomsen, 2004; Masellis, Rector, & Richter, 2003).

Finally, Sukhodolsky et al. (2005) examined adaptive, emotional and family

functioning in children and adolescents with OCD and ADHD. Some of the children in the study also had TS, but this was found to have no effect on the dependent variables and so no separate TS group was used in the analysis. This suggests again that tics themselves may not have a significant impact on QoL in most cases, but that the other features associated with TS (ADHD and OCD) often do have a significant impact. It may well be that specific features present challenges in specific domains, which may impact QoL. For example, family dysfunction was associated with ADHD but not OCD (Sukhodolsky et al., 2005).

It is therefore well established that ADHD and OCD are linked to poorer QoL. This fits with the finding in the TS literature that co-morbidity results in greater impairment of functioning. The studies so far are descriptive and rely on quantitative methods. It will be interesting to see whether further research identifies the key difficulties that individuals experience, perhaps using more qualitative QoL measures.

### *Summary*

TS has been shown to have a significant impact on the QoL of adults. Whilst there are no studies which specifically examine subjective QoL in children with a diagnosis of TS, there is a great deal of relevant literature on social and emotional functioning. This literature highlights the association between TS and difficulties in physical, emotional, social and educational functioning. Generally, it seems that when associated difficulties such as ADHD and OCD are present the negative effect is worse. It is more than just tics which cause difficulties for children with TS.

At the same time, most studies show that less than half of individuals with TS perceive problems to be associated with their TS, and TS does not impact on all domains for every person. For example, only 40% of individuals with TS view social isolation and embarrassment as a major problem (Wand et al., 1993). It may be that

the majority of people with TS are coping well, and TS has little impact on their lives. The results of studies may also be biased by their use of clinical populations. Individuals who attend clinics, particularly the specialist clinics in tertiary care settings, are more likely to have problems with their TS, as they have had to seek specialist help for these problems. Population studies might show different results.

Most of the research is descriptive, pulling out associations between measures of emotion, behaviour and symptom severity. Nearly all of the research relies on questionnaire measurement of the constructs. This enables responses to be grouped, analysed statistically and compared to control groups. However, most of the research is not testing specific theories, and so the cause of associations remains unclear. It is easy to become lost in trying to understand which variables are linked, and forget that what we are trying to understand is how children are affected in their day to day lives by TS, and how they feel about this. Understanding how individuals cope with and perceive their symptoms would be useful as a way of understanding the reasons for associations, and would also be helpful clinically.

The relative rarity of TS means that many studies have limited numbers of participants, reflecting the difficulties of recruiting a clinical population. This means that not all studies in the literature are adequately powered, making relationships more difficult to detect reliably.

Finally, the studies vary in their use of controls. In some ways, the issue of a relevant control group is less important when considering QoL, as the construct by its very nature is subjective. Each individual has their own aspirations and hopes, and these will be affected differently by the symptoms of TS. We know that individuals without TS, or with asthma or diabetes, will not be affected by the symptoms of TS. It is therefore debatable whether such comparisons to control

groups are useful when investigating subjective QoL.

## Conclusions

It seems likely that future research will show that some young people's QoL is affected by TS. It also seems likely that research will show that there are some young people for whom TS has little impact on their QoL, particularly if all studies do not rely on research populations from tertiary care clinics. In some ways, the more interesting question seems to be not whether QoL is affected but how it is affected, and why differences exist between individuals who have similar levels of symptoms. There may well be a general trend, with more severe symptoms having a greater impact. The exceptions to this trend will be especially interesting to investigate, as they will provide insight into protective factors for young people and point to future directions for clinical interventions.

Few explicit links are made to psychological theory in the existing literature, and this reflects the relative lack of modern psychological perspectives on TS. Since medication has been shown to be effective in some cases, TS has been more frequently investigated by neurologists and psychiatrists, with less focus on the psychological and social aspects of the disorder.

TS is clearly a neurological disorder. However, it is one that has particularly strong interactions with psychological and social factors. For example, socially inappropriate behaviours reflect an interaction between an understanding of social rules and conventions and the unconscious production of tics. Stressful situations, or relaxing ones, can act as triggers for tics. People may be able to suppress the symptoms to some extent when they feel that they would be particularly inappropriate. Tics can be suggestible, being triggered by discussion of TS or by

environmental stimuli. Future research considering more psychological factors may help to understand the different ways in which TS presents, and the different impacts it can have. For example, perspectives from health psychology may help understand how people's beliefs about TS influence the way in which they cope with the disorder.

Given that TS is not treatable with medication in all cases, it seems that this would be a useful and valid direction for future research. It may be that the most useful treatment will be that which helps people learn to successfully live with their TS symptoms, increasing protective factors and reducing risk factors. For example, if it is shown that family relationships have a significant influence on the child's self-concept and quality of life then interventions could focus on shifting patterns of interaction in the family to ensure that they are the most helpful for the child and for the family. Alternately, interventions could focus on helping the child think more positively about themselves, and so feeling more able to cope with their tics.

Additionally, research which starts with few assumptions and asks the young people themselves about the parts of TS which are hard for them to manage will enable professionals to check their assumptions, and make sure that the areas they are focussing on are those which are most difficult for the family to manage. For example, if it is the co-morbidities which are most difficult, interventions offered should focus on these rather than tics.

In conclusion, this review has noted that there is relatively little research which investigates the views of children and their families about the impact of TS. It would suggest that this would be a useful area to begin further investigations into TS, as this will ensure that future research and interventions focus on the areas which are most relevant to individual's coping with TS.

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## **Part 2: Empirical Paper**

### **A Mixed Methods Study of the Quality of Life of Young People affected by Tourette Syndrome**

## **Abstract**

The purpose of this study was to consider the impact of Tourette Syndrome (TS) on young people's Quality of Life (QoL). The study used a mixed methods design, combining focus groups and questionnaire data. Questionnaires measured QoL, symptom severity and psychosocial variables (family functioning and self-perception). The study examined a UK sample of young people with TS.

The results showed that the QoL of children with TS was significantly worse than that of children in a UK normative sample. Poorer QoL was associated with increased symptom severity in the data reported by children. Analysis of transcripts from the groups identified four main themes; 'TS can be distressing and disabling', 'struggling to fit into society's expectations of normal behaviour', 'needing to control tics' and 'TS is one part of who I am'.

## Introduction

Tourette Syndrome (TS) is a neurodevelopmental disorder characterised by motor and vocal tics. Individuals with TS frequently have wider difficulties with impulse control in addition to tics. This disinhibition may be reflected in sudden outbursts of anger (Budman, Bruun, Park, Lesser, & Olson, 2000), or may result in hyperactivity and possibly a diagnosis of Attention Deficit / Hyperactivity Disorder (ADHD) alongside that of TS (Robertson, 2000). There is also a strong association with obsessive-compulsive behaviours and Obsessive Compulsive Disorder (OCD; (Robertson, Banerjee, Eapen, & Fox-Hiley, 2002) and some association with learning disabilities (Burd, Freeman, Klug, & Kerbeshian, 2005). Robertson (2000) suggests that some associated behaviours, such as obsessive-compulsive symptoms and some types of ADHD symptoms, are likely to be an integral part of the syndrome. Such behaviours are frequently present in individuals with TS, even if they do not reach the level required for a separate diagnosis of OCD or ADHD.

TS generally begins in middle childhood. Tics then increase in number, frequency, forcefulness and complexity, reaching a peak at around 10 or 11 years of age. They frequently decline in severity throughout adolescence, and will be negligible by early adulthood for at least 40% of individuals (Bloch et al., 2006; Leckman, Zang, Vitale, Lahnin, & Lynch, 1998). It is therefore important that studies of the impact of TS use paediatric populations, as it is during childhood and adolescence that the disorder is often at its most severe. Developmental theories note that risk and protective factors may be age-specific (Rutter, 2005), and this reinforces the importance of research with young people.

Subjective Quality of Life (QoL) considers an individual's sense of their own well-being (Prince & Prince, 2001). Subjective QoL can be measured to some extent

quantitatively, and captures both the level of functioning and the individual's satisfaction with that level of functioning. For example, the Paediatric Quality of Life Inventory (PedsQL; (Varni, Seid, & Rode, 1999) asks respondents to indicate whether they perceive aches and pains to be a problem for them (not just whether they have aches and pains). However, subjective QoL can perhaps be most fully understood using qualitative methods as these allow the individual experiences of young people to be captured (Smith & Osborn, 2003). Using a blend of qualitative and quantitative methods can ensure the final research captures the perspective of the young person more fully (De Civita et al., 2005).

There is a need to consider the elements of TS which have the most significant impact on young people's QoL, so that clinicians can manage these effectively. There are no studies to date that directly investigate the QoL of young people with TS. Parents report that symptoms such as rage attacks, attention deficits and learning difficulties are most difficult to manage in their children when they are present (Dooley, Brna, & Gordon, 1999). Young people rate social isolation and embarrassment as equally disabling as the symptoms themselves (Wand, Matazow, Shady, Furer, & Staley, 1993). It therefore seems that a broad psychosocial focus is needed, in addition to considering other difficulties such as rage attacks, hyperactivity and obsessive compulsive symptoms alongside tics. Research with adults has shown that motor tics and problems with concentration and working memory have significant negative effects on their QoL (Elstner, Selai, Trimble, & Robertson, 2001). However, this cannot be directly generalised to a paediatric population.

Existing research considering the impact of TS on social and emotional functioning has often examined the contribution of symptom severity, asking whether individuals with more tics show greater difficulties. In adults, greater symptom severity is

associated with reduced QoL (Elstner et al., 2001). The results in paediatric populations are mixed, perhaps in part because different studies use different measures to assess psychosocial functioning (Brand et al., 2002; Carter et al., 2000).

Another explanation for the mixed results is that the relationship between symptom severity and QoL is also influenced by other factors. Studies of the QoL of adolescents with epilepsy noted that many of the issues they experienced related to identity formation (McEwan, Espie, Metcalfe, Brodie, & Wilson, 2004), a key developmental task in adolescence (Erikson, 1968). Studies of self perception in TS have found minor differences between children with and without TS (Carter et al., 2000; Bawden, Stokes, Camfield, Camfield, & Salisbury, 1998), although these differences seem more marked in adolescence (De Lange, 2000). In addition, studies of children with ADHD show that they have a more negative self-perception than their peers without ADHD (Barber, Grubbs, & Cottrell, 2005). Existing studies in TS have not looked at the relationships between self-perception and QoL. This will be an important area to investigate, as positive self-perception can act as a protective factor against stress (Rutter, 1981).

TS presents a challenge for a family and can result in increased stress in the family environment and a reduced QoL, especially when there is co-morbidity with ADHD and OCD (Hubka, Fulton, Shady, Champion, & Wand, 1988; Wilkinson et al., 2001; Woods, Himle, & Osmon, 2005; Cooper & Livingston, 2003). However, a positive family context has been found to help children to manage their TS and can improve functioning (Carter et al., 2000; Edell-Fisher & Motta, 1990). This suggests that any association between family functioning and QoL could well represent bi-directional influences. Whilst a positive family environment could help to minimise the impact of TS on a child's QoL, having a child with TS has have a negative impact on the



family's functioning. This fits with broader psychological theories which view development as an ongoing interplay between the child's inherent predispositions, the family's characteristics and the wider environment (Brofenbrenner, 1986).

There are little data on the impact of TS on children at school, although a significant proportion of parents report academic impairment and peer problems (Abwender, 1996; Christie & Jassi, 2002). TS may have some effect on children's peer relationships. However, this effect may be largely due to associated ADHD symptoms, as children with TS and ADHD were shown to be at increased risk for poor peer relationships (Bawden et al., 1998).

In summary, the QoL of young people with TS seems likely to be influenced by symptom severity, self-perception and family functioning. It is also important to consider the impact of TS in other contexts, such as school.

### *Aims and Objectives*

This study used a mixed-method design to investigate the effect of TS on young people's QoL. The lack of previous research in the area meant that it was important to investigate general trends and make normative comparisons using quantitative methods. However, it was also seen as important to include a qualitative element to the research as QoL is inherently subjective and it was therefore necessary to have some understanding of the individual experiences of young people.

Quantitative methods were used to investigate the following hypotheses:

1. Young people with a diagnosis of TS have a lower QoL than young people without a diagnosis of TS
2. Greater TS symptom severity is related to decreased QoL.
3. Increased QoL is associated with positive self-perception and positive family

functioning.

Symptom severity measures in this study will not just include measures of tic severity but will also include measures of obsessive-compulsive behaviours and hyperactivity. These behaviours are strongly associated with TS and can be considered an integral part of the syndrome (Robertson, 2000) rather than distinct co-morbid condition with different causality.

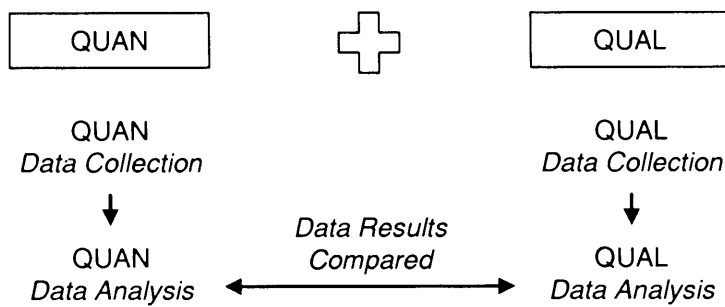
Qualitative methods were used to

1. describe the subjective experience of having TS as a young person;
2. explore which aspects of TS young people perceive to impact upon their quality of life.

## **Method**

### *Overview*

A concurrent triangulation strategy (Creswell, 2003) was used to combine qualitative and quantitative methods in this study. Separate quantitative and qualitative methods were used concurrently and the results were integrated at the interpretation phase. This is shown diagrammatically in Figure 1. Questionnaires were used to collect quantitative data and focus groups were used for the qualitative element of the study.

**Figure 1: Concurrent Triangulation Strategy (taken from Creswell (2003)).**

### *Setting*

Participants were recruited from a specialist Tourette Syndrome clinic at a national children's hospital. Most children in the UK with TS are managed by their local paediatric or child psychiatric services, but some children with challenging diagnostic or management issues are referred to this specialist Tier Four clinic. Whilst recruiting from such a clinic may have caused this sample to be biased towards complex and medication-resistant cases of TS, it also enabled an understanding of the aspects of TS which were challenging for families to manage. In addition, the rarity of TS meant that there would have been a number of practical difficulties recruiting from local services.

The clinic team consisted of a neurologist, neuropsychiatrist and neuropsychologist. Any diagnosis used in this study represented a consensus between members of this team, and was the result of a rigorous process.

### *Ethics*

The Institute of Child Health/Great Ormond Street Hospital Research Ethics Committee, in London, granted ethical approval for this work (Appendix One).

### *Procedure for Recruitment*

Inclusion criteria were a clear diagnosis of TS, and an age between 8 and 18 years. Exclusion criteria were severe neurological or physical impairments which would be hypothesized to have a significant effect on QoL. An introductory letter was sent to all families who had attended the clinic from January 2004 to April 2007 and who met the study criteria (Appendix Two). The notes of each child were reviewed to determine whether a child met the criteria for participation in the study before sending a letter. If the child did meet study criteria, their notes were reviewed in detail and the data from any previous questionnaires they had completed for past clinic appointments were collated.

The letter invited the parent and the child to participate in the study, with separate information sheets and consent forms for the parent (Appendix Three) and the child (Appendices Four and Five). The letter also included the questionnaires for the quantitative part of the study. Families were asked to indicate in their response whether the young person would be interested in participating in one of the focus groups. It was made clear in the letter that declining to participate would have no impact on the family's clinical care. Follow up phone calls were made to provide further information to families where no response was received. However, the researchers were careful not to coerce the families into participation.

### *Quantitative Measures*

The measures used in the study are summarized in Table 1. The PedsQL was selected as the quantitative QoL measure (Appendix Six). Appendix Seven includes examples of the other study questionnaires. The questionnaires sent out for the study were more extensive than those used prior to clinic appointments. Additional areas of interest (e.g. anger symptoms) were identified as important to include from the literature. Parental and child reports of QoL and of all symptoms were collected.

In addition, the study hypotheses also needed data on more psychological constructs such as self-perception and family functioning.

### *Focus Groups*

Focus groups are becoming an increasingly common method of exploring what individuals believe and feel in health psychology (Rabiee, 2004; Wilkinson, 2003). There is some suggestion that the group context may sometimes facilitate personal disclosures (Farquar, 1999), and groups can help people to explore and clarify views in a way that would be less easily accessible in a one-to-one interview (Kitzinger, 1995). In addition, focus groups acknowledge that the participants are the experts (Heary & Hennessy, 2002), and this can help young people to express their own view without feeling the need to give a 'correct' answer to a more powerful adult.

The groups lasted for an hour and a half, and were audio-recorded. They were facilitated by one researcher with the assistance of a colleague, who was the qualified psychologist who worked in the TS clinic<sup>1</sup>. This colleague also asked questions on occasions. The groups commenced with an explanation of their purpose, a discussion of confidentiality and instructions as to what the young people should do if they had any worries during or after the session. The young people were asked for their verbal consent to the recording at the beginning of the session, and a check was made at the end to obtain their consent for what they had said to remain on the recording. No individual participant withdrew their consent.

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<sup>1</sup> The psychologist who originally worked at the clinic, and had been involved as a supervisor in the research design, left in the course of this study. The pilot group was run with the first psychologist, and the other groups were run with her successor.

**Table 1: Measures Used in Quantitative Study**

Construct	Measure	Details	Respondents
QoL	Paediatric Quality of Life	Rates how much difficulty the child has had in four areas (Health & Activities, Feelings, Social,	<b>Child<sup>a</sup></b>
	Inventory (UK) (PedsQL; (Varni et al., 1999)	School) over the last month on a scale of 0-4. Relatively brief and easy to complete. Versions available for 5-18 years. Good reliability and discriminant validity in a large UK sample (Upton et al., 2005). Considered satisfactory in a review of measures of QoL (Eiser & Morse, 2001).	Parent
TS symptoms	Motor tic, Obsession and compulsion, and Vocal tic	Self-report scale which provides scores on five subscales: motor tics, vocal tics, obsessions, compulsions, and associated symptoms. Good sensitivity and specificity (Gaffney et al., 1994)	<b>Child</b>
	Evaluation Survey (MOVES; (Gaffney, Sieg, & Hellings, 1994)		Parent
Rage	Rage Attacks Questionnaire (Budman, Rockmore, Stokes, & Sossin, 2003)	Used to identify presence of episodes of rage in the participants -- brief version of scale used (four questions). Used as a screening questionnaire previously (Budman et al., 2003), reliability and validity not yet established.	Parent
ADHD features	Strengths and Difficulties Questionnaire (SDQ;(Goodman, 1997))	Measures strengths and difficulties on five scales: pro-social, hyperactivity, emotional problems, conduct (behavioural) problems, and peer problems. Satisfactory reliability and good validity in a large British sample (Goodman, 2001).	<b>Child</b>
			<b>Parent</b>

**Table 1 (continued): Measures Used in Quantitative Study**

Construct	Measure	Details	Respondents
OCD features	Short OCD scale (SOCS; (Lee, Jones, Goodman, & Heyman, 2005)	Narrow focus on obsessions and compulsions, based on the most discriminant items from the Leyton Obsessive Compulsive Inventory (Berg, Rapoport, & Flament, 1986). Eight items, rated on a 3- point scale of frequency. Good sensitivity to clinical change in symptoms (Lee et al., 2005).	<b>Child<sup>a</sup></b> Parent
Self-Esteem	Self-Perception Profile for children (Harter, 1985) or Adolescents (Harter, 1988)	Evaluates children's perceptions of competence in the domains of schools, peer, athletic, physical, behavioural and global functioning. Good internal consistency for subscales (Harter, 1985).	Child
Family Functioning	Short Version of Family Environment Scale (Moos & Moos, 1986)	A 30-item self-report instrument designed to evaluate family functioning. It provides three individual scales of family functioning; Cohesion, Expressiveness and Conflict. Acceptable test-retest reliability and internal consistency (Moos et al., 1986).	Parent

<sup>a</sup>Questionnaires which may have been completed prior to past clinic appointments, and which were sometimes present in the notes, are indicated in bold type.

A focus group schedule was used to guide the group discussion, and to ensure the discussion was relevant to the research questions (Appendix Eight). The guide was designed to fit with the information from the quantitative part of this study. For example, it specifically prompted the children to talk about the different domains covered in the Harter Self-Perception Profile. The group was semi-structured, and the guide was used flexibly. Many of the young people had attention difficulties, and so care was taken to use more than one modality, writing information on whiteboards or flip charts, and using visual cues such as a 'feelings thermometer' to indicate their level of distress. An example of this record of information is included in Appendix Nine.

The initial group was run as a pilot of the focus group schedule and the group size was intentionally small. No significant changes were made to the guide, and so the data from this pilot were included in the data for the study. Two focus groups were run as the main data collection for the study. The groups aimed to have 4-6 participants, and the sample was divided by age as far as possible. It is helpful to have smaller groups when working with young people than with adults, to enable the researcher to manage the group and support discussion (Hoppe, Wells, Morrison, Gillmore, & Wilsdone, 1995).

## *Participants*

### **Sample**

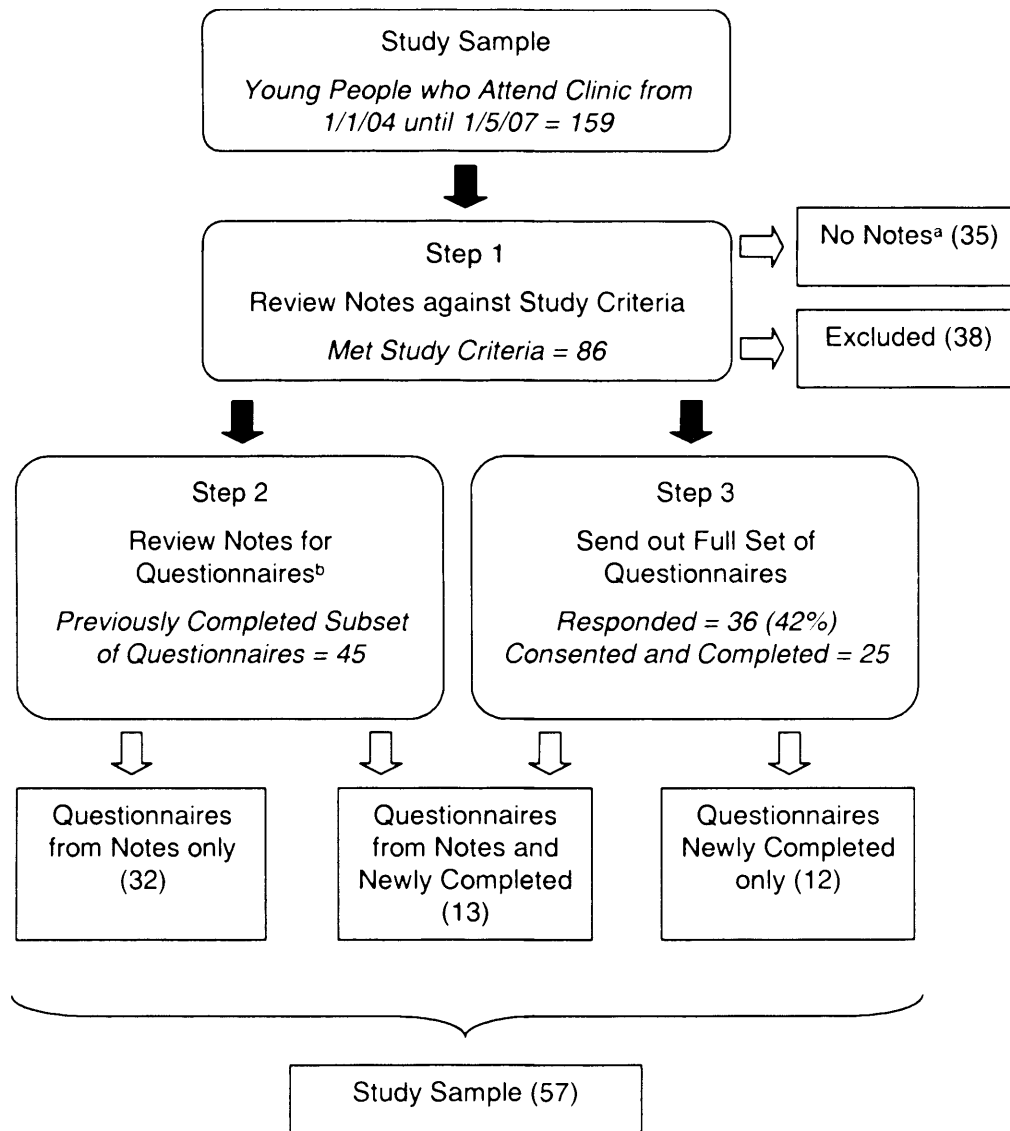
The sample population consisted of all children who attended the clinic from 1<sup>st</sup> January 2004 until 1<sup>st</sup> May 2007, a total of 159 children. 38 were excluded, 15 because they did not have a diagnosis of TS, 17 due to their age and 5 because they had complex neurological problems which may have influenced their QoL,



independently of having TS. One other participant was excluded due to a difficult home situation for reasons unrelated to TS. Unfortunately, the notes for 35 of the children who had attended the clinic were inaccessible and so it was not possible to include them in the study. Consent forms were therefore distributed to 86 of the 159 families with an introductory letter, as shown in Figure 2.

25 families returned their consent forms and the study questionnaires. 13 of these families had completed a subset of the study questionnaires in the past. The most recent data was used in all analyses, and the data set for each individual was collected at the same point in time. A limited subset of questionnaires (as indicated in Table 1) was available for an additional 32 families from notes review, bringing the total sample size to 57. This is shown diagrammatically in Figure 2. Eleven children agreed to participate in the focus groups, ten of whom had also completed the full set of questionnaires.

The average age of children in the sample was 11.4 years ( $SD$  2.36) with a range from 8 years to 17 years. 46 were male and 11 were female. The majority of parents who completed the study questionnaires described their children's ethnicity as white, with one exception (who indicated a mixed ethnic background). 18 children were diagnosed with co-morbid ADHD, and 10 were diagnosed with OCD alongside their TS.

**Figure 2: Flow Chart of Procedure for Recruitment indicating Sample Size**

<sup>a</sup> Medical records not available at the time of the study, due to re-organisation of historical notes by the medical records department; <sup>b</sup> Questionnaires are sent out prior to a clinic visit, and these are a subset of those used in the study, as shown in Table 1. This information had not previously been collated.

## Groups

The young people who participated in the groups were self-selected, as they had indicated that they would be prepared to participate on their consent forms. The final composition of the groups is shown in Table 2.

**Table 2: Characteristics of young people who participated in the focus groups**

Demographics				Diagnoses		Scores on questionnaires	
Gender	Ethnicity	Age (years)	OCD	ADHD	Child PedsQL	Child MOVES	
					Total <sup>a</sup>	Total <sup>b</sup>	
Pilot							
1	Male	White	11	No	No	52.2	42.0
2	Male	White	11	No	No	.	.
One							
3	Male	White	17	No	No	44.4	43.2
4	Male	White	17	No	No	63.0	20.0
5	Male	White	11	No	No	64.1	26.0
6	Male	Mixed	12	No	Yes	63.0	31.0
Two							
7	Female	White	8	Yes	No	41.3	40.0
8	Male	White	12	No	No	69.3	32.0
9	Male	White	11	No	Yes	82.6	13.0
10	Female	White	8	Yes	No	63.0	37.0
11	Female	White	7	No	No	77.2	24.0
<i>M</i> of group sample ( <i>N</i> = 10)			11.4			62.0	30.8
<i>M</i> of whole sample			11.4			62.6 <sup>c</sup>	25.8 <sup>d</sup>

Notes: A typically developing child would be expected to score around 80 on the PedsQL (Upton et al., 2005), below 10 on the MOVES (Gaffney et al., 1994) and below 6 on the Child SDQ Hyperactivity subscale .

a. Score from 0-100, higher values indicate better QoL; b. Score from 0-60, higher values indicate more TS symptoms; c. *N* = 57; d. *N* = 53.

## Analysis

### Quantitative Analysis

The data were analysed using SPSS for Windows, release 11.5.0. It began with exploratory data analysis, checking the frequency distributions of key variables, and

examining the correlations between measures.

To examine the second hypothesis of the study<sup>2</sup>, the association between measures of symptom severity and QoL was examined using a hierarchical regression. For the third hypothesis<sup>3</sup>, a second hierarchical regression was planned, adding in self-perception and family environment as independent variables.

A previous study looked at the relationship between psychopathology and QoL, and used the PedsQL as an outcome measure (Bastiaansen, Koot, & Ferdinand, 2005). The linear multiple regression in this study produced an R-squared of 0.29 for the child version of the PedsQL, and 0.47 for the parent version. With three predictors, this effect size would need a sample size of 17-31<sup>4</sup> participants to be detected at 80% power.

As noted in Table 1, some questionnaires were distributed prior to clinic appointments by the clinic. This information was added into the analysis to increase power. For some individuals, information was available at two time points as there was information from the clinic questionnaires and from the additional study questionnaires. The most recent data were used in cross-sectional analyses.

### **Qualitative Data Analysis**

The recordings were transcribed verbatim. Thematic Analysis (Joffe & Yardley, 2004) was used as the approach for the analysis. In accordance with this method, a

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<sup>2</sup> Greater TS symptom severity is related to decreased QoL

<sup>3</sup> Increased QoL will be associated with positive self-perception and positive family functioning

<sup>4</sup> Calculated using zumastat

series of steps were followed in order to identify the themes of the discussion, and examples of this analysis are shown in Appendix Ten.

1. The transcript of the first group was read through several times. The left-hand margin was used to annotate the text when interesting or significant points were made by the participants, producing an initial set of low level codes.
2. Once the whole transcript had been read in this way, the other margin was used to document emerging themes, drawing upon commonalities in the initial annotations.
3. The emerging themes were listed, and similar ideas were clustered together. Links to the original text were maintained with page and line number references.
4. The process was then repeated for another group's transcript, looking for themes which are both similar and different to those from the first group.
5. Finally, the three sets of themes were combined to produce the super-ordinate themes discussed here.

Three forms of credibility checks were made (Barker & Pistrang, 2005). Firstly, an audit trail was checked by an independent researcher who was not directly involved in the running of the groups or the data analysis. This researcher examined the transcripts and summaries of themes for each group to check that themes reflected the group discussion and that themes had not been lost in the process of summarising. Thematic qualitative analysis is a subjective process, and so another researcher analysing the raw data would produce a different account of the data. As a result, the audit was designed to ensure that this analysis was valid, rather than to confirm that this was the right way of analysing the data, as other accounts would be equally valid.

Secondly, the themes were sent to the participants, and they were asked to indicate

whether they felt the themes accurately represented their discussion. Thirdly, triangulation was inherent in the study design, and the results were compared to those from the questionnaires.

There was considerable overlap between the themes from each group, which suggested that some level of saturation had been reached. The themes which emerged reflected the items on the group schedule to some extent, but they tended to focus on certain areas which were clearly salient for the participants, for example school, friends, and the views of other people. Although the schedule did not explicitly ask the children about the meaning they attributed to TS, this emerged from their discussion.

### *The Researcher*

A single researcher (DC) ran the groups and completed the analysis. Having reviewed a literature dominated by difficulties and problems, I had some preconceptions that children would be distressed by their symptoms. I had attended clinics for children with TS, and I had previous experience of facilitating groups and of working with children.

## Results

### *Quantitative Results*

This section first presents the descriptive analysis of key variables, and then addresses each of the research questions in turn.

#### **Exploratory Data Analysis of Key Variables**

The dependent variable in this study was the overall score on the child and parent versions of the PedsQL. This variable was measured on an interval scale, and was normally distributed, and so met the assumptions for parametric tests. All of the independent variables also met these criteria, with the exception of the SDQ Hyperactivity scale. This was significantly negatively skewed, as both parents and children reported very high levels of symptoms, at the scale maximum. As a result, the validity and usefulness of this variable as a predictor in a regression are likely to be limited. ADHD diagnosis was therefore used as a binary predictor instead of the SDQ hyperactivity score (0 = No ADHD and 1 = ADHD diagnosis).

#### **Descriptive Analysis: Symptom Severity and Co-morbidity**

There were no significant differences between parent and child report of symptom severity on the MOVES Tic Scale or Short OCD Scale,  $t(24) = 0.21$ ,  $p = .84$ , and  $t(24) = 0.8$ ,  $p = .43$ . There was a significant difference between parent and child reports of hyperactivity on the SDQ<sup>5</sup>,  $T = 2.21$ ,  $p = .027$ , with parents reporting higher levels of hyperactivity.

Table 3 shows the scores on measures of symptom severity across three diagnostic groups. These diagnostic groups are separated in this analysis to allow sense

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<sup>5</sup> Calculated using the Wilcoxon Signed Ranks Test, a non-parametric test.

checking of the symptom severity data.

**Table 3: Symptom Severity Data from Parent and Child Report**

		Diagnostic Group			
		TS	TS + ADHD	TS + OCD	Total
Child Report Measures					
MOVES Tic	<i>M (SD)</i>	12.5 (5.1)	13.3 (5.4)	14.5 (4.4)	13.1 (5.0)
	<i>N</i>	27	16	10	53
Short OCD	<i>M (SD)</i>	5.5 (2.8)	5.4 (3.0)	7.6 (2.2)	5.9 (2.8)
	<i>N</i>	27	15	10	52
SDQ Hyperactivity	<i>M (SD)</i>	6.5 (2.4)	8.0 (1.6)	7.3 (2.0)	7.0 (2.2)
	<i>N</i>	29	13	10	52
Parent Report Measures					
MOVES Tic	<i>M (SD)</i>	11.8 (3.7)	14.2 (4.7)	11.6 (0.5)	12.6 (3.9)
	<i>N</i>	14	8	3	25
Short OCD	<i>M (SD)</i>	5.5 (2.7)	5 (2.4)	6.3 (1.1)	5.4 (2.5)
	<i>N</i>	14	8	3	25
SDQ Hyperactivity	<i>M (SD)</i>	7.2 (2.6)	9.5 (0.8)	7.2 (2.0)	8 (2.3)
	<i>N</i>	27	17	10	54

a. Scoring above a clinical cut-off of 10 is used as an indication of TS (Gaffney et al., 1994). b.  $M = 11$ ,  $SD = 3.1$  on this scale for children attending a specialist clinic for OCD, all of whom had a diagnosis of OCD (Lee et al., 2005). c. Scores of 7 or above are considered to indicate a high likelihood of a diagnosis of ADHD (Goodman, 2001)

There were no significant differences in scores on the MOVES across all the groups,  $F(2,50) = 0.51$ ,  $p = .60$  for the child versions and  $F(2,22) = 1.01$ ,  $p = .38$  for the parent version. This would suggest that there were no significant differences in tic



severity with a co-morbid diagnosis.

As expected, young people with a diagnosis of OCD scored significantly higher on the Child Report Short OCD scale ( $M = 7.6$ ,  $SD = 2.2$ ) than those without a diagnosis of OCD ( $M = 5.5$ ,  $SD = 2.9$ ),  $t(50) = 2.16$ ,  $p = .04$ . No comparison of parent report was made due to insufficient numbers in the OCD group. Child reports of hyperactivity on the SDQ were not significantly different for children with and without ADHD<sup>6</sup>,  $U = 168$ ,  $p = .07$ . Parent reports of hyperactivity were significantly greater in children who had a diagnosis of ADHD,  $U = 113$ ,  $p < .001$ .

**Table 4: Intercorrelations between measures of symptom severity (Pearson Correlation)**

	1 MOVES C	2 OCD C	3 SDQ C	4 MOVES P	5 OCD P	6 SDQ P
1 MOVES Tic Subscale Child	-	0.58**	0.51**	0.49*	-0.05	0.31*
2 Short OCD Scale Child		-	0.30*	0.15	0.54*	0.00
3 SDQ Hyperactivity Scale Child			-	0.34	-0.13	0.71**
4 MOVES Tic Subscale Parent				-	0.10	0.37
5 Short OCD Scale Parent					-	-0.08
6 SDQ Hyperactivity Scale Parent						-

*Note.* Non-parametric correlation coefficient (Spearman's rho) used for all correlations with SDQ

\* Significant ( $p < 0.05$ ); \*\*Significant ( $p < 0.01$ )

The correlations among the measures are summarised in Table 4. In general, the child report of TS symptom severity on the MOVES correlated strongly with all other

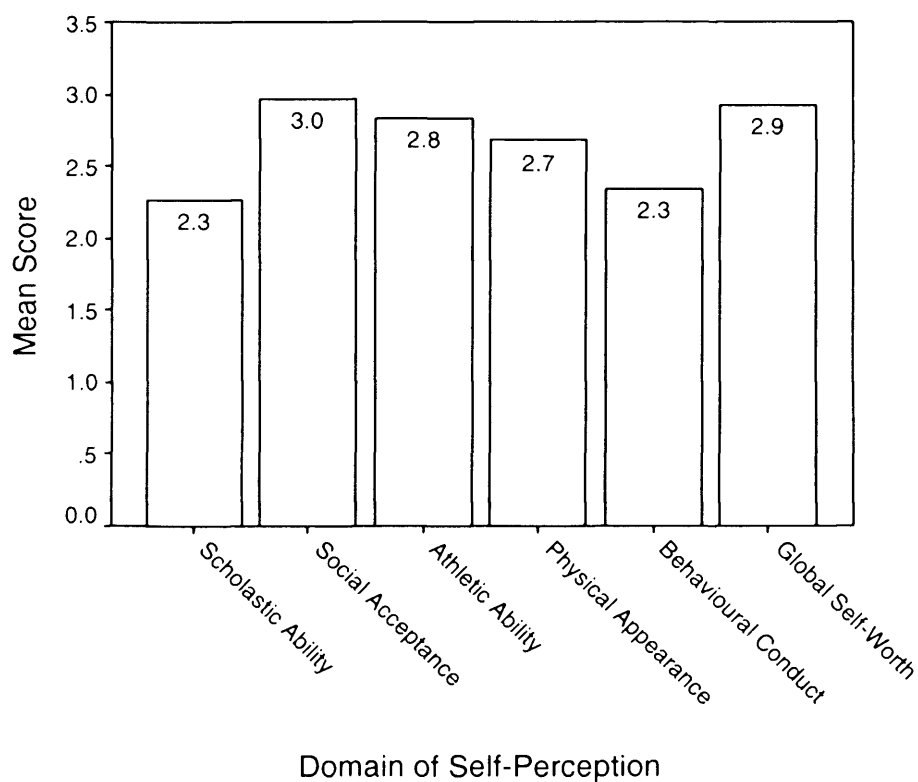
<sup>6</sup> Calculated using the Mann-Whitney Test, a non-parametric test.

child report measures, and less strongly with parental report of tic severity and hyperactivity. Parent and child report were significantly correlated for each of the scales.

### Descriptive Analysis: Self-Perception and Family Functioning

Children had positive self-concept (greater than the mid-point of 2.5; (Hoare, Elton, Greer, & Kerley, 1993) in all domains of the Harter Self-Perception Profile except scholastic ability and behavioural conduct, as shown in Figure 3. There were no significant differences between the domains of self-esteem,  $F(5,16) = 2.59, p = .07$ . However, the sample size in this analysis was small and as a result there may not be adequate power to detect significance.

**Figure 3: Mean scores of children (N=22) across all domain of the Harter Self-Perception Profile**



The information from the family environment scale is shown in Table 5. The study sample showed significantly more conflict and significantly less expression than a

previous sample of US families from a range of sources (newspaper adverts, church groups, school samples) which aimed to include families with a number of different structures and with different cultural backgrounds from across the US (Moos et al., 1986).

There were significant negative correlations between scores on the cohesion and expression scales and the severity of tics reported by children,  $r(24) = -.67$ ,  $p < .001$  and  $r(24) = -.44$ ,  $p = .03$  respectively. There were also significant correlations with the severity of tics reported by their parents,  $r(24) = -.53$   $p = .01$  for the cohesion scale and  $r(24) = -.57$   $p < 0.001$  for the expression scale. This suggests that increased tic severity is associated with less family cohesion and expression.

**Table 5: Comparison of scores on the Family Environment Scale in this sample to that in a normative sample (Moos et al., 1986)**

	Normative Sample <sup>a</sup>	Study Data	T
	N = 1,432	N = 24	
	<i>M (SD)</i>	<i>M (SD)</i>	
Cohesion	6.73 (1.47)	6.42 (2.48)	1.00
Expression	5.54 (1.61)	4.85 (1.79)	2.05*
Conflict	3.18 (1.91)	3.96 (2.14)	-2.00*

<sup>a</sup> Normative Sample of 1,432 US families (Moos et al., 1986)

\*Significant  $p < 0.05$

**Hypothesis 1: Young people with a diagnosis of TS will have a poorer QoL than young people without a diagnosis of TS**

Table 6 presents the means and standard deviations of the PedsQL scores from this

sample and compares them with those of a normative sample of UK school children from a published study (Upton et al., 2005), examining the first study hypothesis. The ages of the children in the normative sample ranged from 8-18 years ( $M$  for self report = 12.58,  $SD$  = 2.6;  $M$  for proxy-report = 11.86,  $SD$  = 2.3).

**Table 6: Comparison of study data on QoL to normative data collected from a sample of healthy UK children (Upton et al., 2005).**

Domain of PedsQL	Normative	Current Study	
	Sample mean ( $SD$ )	Data mean ( $SD$ )	$t$
Self Report			
$N$	1033	57	
Total Score	83.8 (11.8)	62.6 (14.9)	12.9*
Physical Health	88.5 (11.6)	73.6 (18.4)	9.02*
Psychosocial health	81.8 (13.2)	56.8 (16.0)	13.7*
Emotional Functioning	78.4 (17.9)	50.7 (20.4)	11.1*
Social Functioning	87.6 (16.4)	67.1 (20.3)	9.04*
School Functioning	78.8 (15.8)	51.8 (18.0)	12.2*
Parent Report			
$N$	665	25	
Total Score	84.6 (11.1)	55.8 (13.7)	12.4*
Physical Health	89.0 (12.2)	69.2 (23.0)	7.59*
Psychosocial health	82.2 (12.6)	48.8 (12.1)	12.9*
Emotional Functioning	78.2 (15.5)	38.4 (19.3)	12.4*
Social Functioning	86.8 (15.4)	63.4 (17.8)	7.41*
School Functioning	81.5 (16.0)	45.2 (18.3)	10.8*

\* $p < 0.001$

Without the raw data from the previous study, it was not possible to test for homogeneity of variance, which is an assumption of the  $t$  test calculation used.

However, the large sample size in the normative data makes the *t* test more robust to deviations from this assumption. The QoL reported by both children and parents in the study is significantly lower across all domains than that reported in the normative sample.

There were significant differences between parent and child report of QoL. Parents tended to report physical,  $t(24) = 2.33$ ,  $p=0.03$ , and emotional QoL,  $t(24) = 4.5$ ,  $p<0.00$ , QoL as significantly poorer than their children. This resulted in significantly poorer parental reports of psychosocial and total QoL, as emotional and physical QoL were summed in these totals.

A repeated measures ANOVA showed that there were significant differences between the domains of QoL in child report  $F(3,52) = 38.3$ ,  $p < 0.01$  and parental report  $F(3,21) = 14.2$ ,  $p < 0.01$ . Post hoc pairwise comparisons using the Bonferroni correction demonstrated that QoL in the emotional and school domains was significantly worse than that in the physical and social domains for both parental and child report. Child reports rated social QoL as significantly worse than physical QoL, there was no difference in parental reports.

### **Hypothesis Two: Greater TS symptom severity will show a relationship with decreased QoL.**

The second research question addressed the relationship between symptom severity and quality of life. Parental and child ratings of QoL were considered separately, as significant differences existed between them. Child ratings of symptom severity were used as predictors of child-rated QoL, and parent-reported symptom severity was used as a predictor of parent-rated QoL.

**Table 7: Summary of Multiple Regression Analysis for Symptom Severity Variables Predicting Overall QoL (Child-Report)**

Variable	Model 1			Model 2		
	<i>B</i>	<i>SE B</i>	$\beta$	<i>B</i>	<i>SE B</i>	$\beta$
MOVES Tics Score	-1.70	0.34	-0.58**	-1.03	0.38	-0.35*
ADHD Binary Label <sup>a</sup>				-7.22	3.49	-0.22*
Short OCD				-2.08	0.68	-0.40**
$R^2$	0.33			0.14		
F for change in $R^2$	24.6**			6.2*		

Notes: *N* = 49; \* Significant  $p < .05$  \*\* Significant  $p < .01$ ; <sup>a</sup> Entered as a binary variable where 0 = No ADHD diagnosis and 1 = ADHD diagnosis

Child report of QoL was negatively associated with children's reports of tic severity , as shown by the hierarchical regression in Table 7. Adding in a measure of obsessive compulsive symptom severity and an indicator of a clinical diagnosis of ADHD in a second block explained significantly more variance. The MOVES Tics score, ADHD diagnosis and the Short OCD Scale scores were all significant independent predictors of reported QoL. The regression showed a significant relationship between child-reported TS symptom severity and QoL.

The sample size for parental report of QoL was much smaller ( $N = 25$ ) than that for child report. In addition, one of the variables, parental report of hyperactivity on the SDQ, was significantly skewed. Given that the sample size was small and the assumptions of parametric tests were violated, non-parametric correlations were used to examine the relationships between the variables rather than a regression analysis. These are shown in Table 8. Only tic severity correlated significantly with parental report of QoL, although the correlation with the SDQ Hyperactivity score did

approach significance at  $p = .08$ .

**Table 8: Correlations between measures of symptom severity and parental report of QoL ( $N = 25$ )**

Measure	MOVES Tics	SDQ Hyperactivity	Short OCD
PedsQL Total	-0.41*	-0.36	-0.14

Note:  $N = 25$ , Spearman's rho used as correlation coefficient; \* Significant  $p < .05$

**Hypothesis Three: Increased QoL will be associated with positive self-perception and positive family functioning.**

Hierarchical multiple regressions were planned to determine whether adding in measures of self-perception and family functioning would increase the predictive power of the model. However, given the limited sample size, a multiple regression would not be valid. As previously, correlations were used as an alternative method of exploring the relationships between measures of QoL, symptom severity, self-perception and family functioning. There were no significant associations, as shown in Table 9, although the correlation between family cohesion and parental report of QoL did approach significance.

**Table 9: Correlations between measures of self-perception and family functioning and parental and child reports of QoL**

	<i>N</i>	Child QoL		Parent QoL	
		<i>r</i>	<i>p</i>	<i>r</i>	<i>p</i>
Harter Global Self Worth	21	-0.01	0.98	-0.11	0.63
Family Environment Scale					
Cohesion	24	0.32	0.13	0.39	0.06
Expression	24	0.26	0.23	0.33	0.12
Conflict	24	-0.10	0.64	-0.12	0.59

## *Focus Groups*

The analysis produced four main themes, and a number of sub-themes within each of these. These themes were similar across the groups. Firstly, there were times when the symptoms of TS had a direct negative impact on the young person's QoL, for example when tics caused pain. Secondly, additional difficulties were caused by young people's struggle to fit in with their peers, and behave in a way which was considered normal. This struggle to control tics was distracting for young people, especially at school. Finally, some young people were able to accept TS as one part of their personality, despite the fact that it had some elements which were distressing. It was also clear that young people were helped by other people accepting their TS, for example by schools who made allowances. The themes are summarised in



Table 10. The results are presented using extracts from the transcripts as illustrations.

There were some differences in participant's experiences of TS at different ages. It was noticeable that many of the younger children thought about the impact of the TS in a much more concrete way, and this fits with their developmental stage (Piaget, 2001). For example, the theme of acceptance, viewing TS as a "part of who I am" emerged particularly strongly in the older group, but some of the younger children had also begun to think in this way. The following text provides a summary of the content of the themes, alongside illustrative quotes.<sup>7</sup>

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<sup>7</sup> Square brackets enclose comments made by the transcriber, an ellipsis (...) indicates a pause or trailing off, a hyphen at the end of the word indicates that the speaker was cut off abruptly.

**Table 10: Themes from analysis of focus groups**

	Theme	Sub-themes
One	Tourette syndrome can be distressing and disabling	Tics get in the way and are hard to manage More than just tics
Two	Struggling to fit into society's expectations of normal behaviour	Others not understanding involuntary behaviours Being talked about and noticed Bullying and teasing Worrying about what others think
Three	Needing to control tics	Distracting and attention-consuming
Four	Tourette Syndrome is one part of who I am	

**Theme One: Tourette syndrome can be distressing and disabling**

There were a number difficulties directly related to the condition itself. All the young people acknowledged that there were times when they had been distressed by their TS, and it seemed that this distress was sometimes increased by the isolation they experienced.

There are still times when it does get to you and does upset you (G1, P3)<sup>8</sup>

I was the only one. I knew there were other people out there but I just hadn't met anyone, for years it was just like 'oh can I just see someone please' (G1, P5)

The symptoms often had a larger impact when they were more severe, or when the young person felt that they were out of their control.

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<sup>8</sup> The group number and participant number correspond to those in Table 2. (G1, P3) would refer to participant 3 in Group One.

Interviewer: What do you think would happen if you didn't try and control it?

P1: Everything would go wrong (Pilot, P1)

It was evident that families were also concerned about the impact of TS on their child's life.

Yeah, I wouldn't want a kid though that had Tourettes. I know how my mum and dad feel... bad. (G1, P5)

### ***Tics get in the way and are hard to manage***

The inherent nature of tics made them difficult to manage. For example, stressful situations could be made more stressful as they also triggered an increase in tics.

My dad didn't just tell me to stop, he used to shout at me and make me stop it, and that's what made me worse. (G1, P4)

Young people were bothered by pain caused by repeated movements, and by injuries resulting from tics which they were unable to control. For example, tics could get in the way of some sports, causing the children to flinch at inappropriate moments.

I go trampolining, and it is annoying. We practice doing a front flip and that and like I twitched in the air and I landed on my arm and hurt it [Pilot, P1]

At times the tics were physically disabling. The young people also noted that it was sometimes hard for them to sleep. Finally, there were times when impulses were dangerous.

Earlier I felt like going on the train track on the tube, I had to ask my mum to hold me, just squeeze me (G1, P5)

### ***More than just tics***

The young people also spoke about their difficulties concentrating and paying attention. When such problems were present, they had a significant negative impact on young people's lives. The young people linked these difficulties to their TS.

But in a way I sometimes wish I didn't have it, because, just so I could be able to concentrate and look at a book or a screen for more than like three seconds

or whatever, just to be able to sort of... (G1, P3)

Anger was frequently mentioned by the young people, even before explicit prompting by the interviewer. Getting angry was an unpleasant experience.

I hate getting angry, because it's just like, I don't want to get angry, it's really annoying when I get angry 'cause it makes me feel really sick and tired and it gives me a really bad headache and I go all red. [G2, P10]

## **Theme Two: Struggling to fit into society's expectations of normal behaviour**

Young people experienced difficulties fitting in, with their involuntary behaviours making it hard for them to behave in what general society considers to be a "normal" way of behaving. Some were explicitly aware of this; in Group One they discussed who had the right to determine normal behaviour.

### ***Others not understanding involuntary behaviours***

People naturally construe behaviours as intentional and controllable. Children with TS have many behaviours which they are unable to control. This means that they are sometimes perceived as naughty or annoying when they are unable to stop their tics.

He [Dad] used to like sort of if I ticked when I was umm... If I sort of ticked when he was watching TV or something he was like 'shhh be quiet', and I'm like 'I can't help it'. He never really understood that much, so... But he does quite a lot more now... (G1, P3)

This can be a particular problem at school, where children reported getting into trouble.

In reception, before I knew I had Tourettes, I used to squeak, and I lost my whole golden time, for squeaking (G2, P10)

They were very appreciative of understanding from school, when teachers did recognise that their behaviour was involuntary and make allowances.

We used to always have to stay still, and like I always used to have to move,

and like if you move you have to stay in for lunch, and like, but Miss didn't do it to me because like, she doesn't, and it's really hard for me to stay still. (G2, P8)

### ***Being talked about and noticed***

The young people experienced embarrassment when people stared at them and talked about them. Many wanted to be able to blend in with their peers but were unable to.

I told my mates that I've got Tourettes, but only a few, then they can tell like a brother or sister. Then the brother or sister if they are in high school can tell their mates, like 'don't tell anyone' and then they'll tell someone and then all through school would know that me, that I have got Tourettes. And it's really annoying. (Pilot, P2)

### ***Bullying and teasing***

Being different from their peers did mean that the young people were a target for bullying and teasing. In addition, many described their own aggressive and violent reactions to this bullying. These reactions could mean that they also got into trouble when they were bullied.

They like call me Tourettehead and stuff, I just end up kicking them or something, as I just get really angry and start kicking them and they start punching me, doing that back. And I end up falling out with them (G2, P7)

### ***Worrying about what others think***

The young people's experiences of other people's negative reactions to their Tourette Syndrome resulted in considerable anxiety about what others think of them.

If I do the noises they're like 'oh that's Tourettes isn't it' and I think [few words inaudible] and I'm scared they'll start thinking I'm weird and my relationships will go. I'm scared of that. (Pilot, P2)

Many young people tried to use secrecy as a defence against this anxiety, trying to hide their TS and avoid discussions about it.

My friends, knew I had Tourettes, and they know what it was, and then I said keep it a secret as I don't want loads of people knowing (G2, P8)

### **Theme Three: Needing to control tics**

Children felt a need to suppress their tics, in order to fit in with others and avoid attention from others which was experienced as negative.

Because when I look at people, they are really odd ones just like [demonstrates a facial tic] and when I look at people and I don't know them I try and keep them in, but if you do do them they like look at you and stare at you, it's really annoying. (G2, P10)

Well, it's when I'm outside, I tend to worry about what people see and what they think, so I'll stop it (G1, P4)

However, there were times when the children needed to let out their tics, particularly if they had been suppressing them for a while.

### ***Distracting and attention-consuming***

This struggle to control was attention consuming and meant that children were unable to give their full attention to the external world. This could be problematic at school, where there is a particular need to control tics but also a need to concentrate on the lesson's contents.

I think the reason that I can't remember is like sometimes I don't know what I'm doing because I'm thinking 'oh no the Tourettes is bad ohh got to control it control it control it' and I might ask just to go to the toilet so I can do it and then I've missed like 20 or 25 minutes of the lesson. (Pilot, P2)

Some young people reported measures put in place by their school to reduce the impact of their struggle to control tics. For example, some were allowed to leave lessons, or take their exams in separate rooms.

## **Theme Four: Tourette Syndrome is one part of who I am**

Some young people had been able to integrate their TS into their sense of self, even though it had some negative effects on their life.

Well, I suppose if I didn't have it I wouldn't be myself. So that's why, it's part of you and part of your personality, so if I didn't have it I wouldn't be me. So that's why I've just learnt to accept it. (G1, P4)

Well, I just don't think I would be the same [without Tourette Syndrome]. I can do different things now, and I can if I don't. But it's just I was born with it so I have to live with it so. (G2, P7)

This seemed to reflect a process over time, which was helped by diagnosis for many of the young people.

Yeah, I'm starting to enjoy it, well not necessarily enjoy like 'Yey I've got it whoopee look at me' but not like 'oh I've got it oh god'. So I just live with it and get used to it. (G1, P5)

It was helped by acceptance from friends and family, and it was evident from the children's descriptions that it was helpful when their family found out the cause of their behaviour.

So the ones I told a long time ago, think well [name] has got Tourettes but that doesn't mean he's a bad person, it just means that it's one of the things he's got wrong'. I'm not worried about them. (Pilot, P2)

My mum used to think I was being annoying, so in the end we sort of um went to our local CAMHS service and they sort of diagnosed it as Tourette Syndrome. (G1, P3)

## **Discussion**

This study reports on the quality of life of a small but well-characterised sample of young people who have TS. The mixed methodology used provided considerable

insight into the impact of TS on young people's QoL. The views of the young people themselves were clearly illustrated in both the qualitative and quantitative data, and this provided a valid description of their subjective QoL.

### *The QoL of Young People with TS*

The QoL in this sample of children with TS was lower than that of a large sample of UK school children without TS, fitting with the hypothesis that TS would have a negative impact on QoL. There was a differential impact across the domains of the PedsQL with emotions and school being affected most adversely. This fitted with the results from the qualitative data. The young people described how their TS sometimes upset them. They also explained how it was particularly difficult to manage TS at school, in terms of the distraction of trying to control tics and trying to behave in an acceptable way.

The young people in the focus groups generally reported that they had friends who were accepting of their TS. In support of this, scores on the Social Acceptance domain of the Harter Self-Perception Profile were high, indicating that the children in the quantitative study viewed themselves as having good friendships. However, young people also described bullying and teasing from a wider circle of peers, and were very worried about the views of others who they knew less well. The quantitative QoL results suggested that children's social QoL remained significantly poorer than that of UK school children without TS. These slightly mixed results are explained by considering the distinction between group acceptance (sociometric status) and friendship, which constitute two separate domains of the larger area of peer relationships (Gest, Graham-Bermann, & Hartup, 2001). It seems that TS may have little impact on a young person's ability to form and maintain friendships, but may impact more negatively on how they are perceived by their peers with whom they do not have close friendships (i.e. their sociometric status).



THE IMPACT OF TS ON QoL

The impact of TS on physical QoL was significant but limited. Sixty six percent of respondents indicated they were bothered by aches and pains<sup>9</sup>, and physical discomfort was also mentioned in the focus groups.

It is of note that parents tended to report QoL as significantly worse than their children did, highlighting the importance of asking the children themselves their view. Previous research has also shown this tendency for parents to under-report their child's QoL (Parsons, Barlow, Levy, Supran, & Kaplan, 1999). A mother's report of her child's QoL can be influenced by her own well-being (Eiser, Eiser, & Stride, 2005), suggesting that mothers may sometimes project their own feelings onto their child. Fitting with this idea, this study showed that parental report of QoL correlated with family cohesion, suggesting that parental reports of their child's QoL could also be influenced by the quality of a family's relationships.

### *Influences on the QoL of Young People with TS*

#### **Symptom Severity**

This study also explored some potential influences on the QoL of young people with TS, and these will now be considered in turn. Firstly, the quantitative results showed a relationship between child reported QoL and symptom severity (frequency and number of tics and OCD symptoms and ADHD diagnosis). The qualitative reports described how the symptoms could be both disabling and distressing at times.

The high frequency of symptoms of hyperactivity in this sample meant that it was not

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<sup>9</sup> Respondents who indicated they were bothered by aches and pains sometimes, often, or almost always.

possible to use a continuous measure of these symptoms in the study analyses. ADHD diagnosis was used as an alternative, and did account for a significant proportion of the variance in QoL scores. The qualitative results also highlighted that the young people were bothered by their difficulties in concentrating and in sitting still. Future research would benefit from a more comprehensive assessment of hyperactivity, attention and impulsivity difficulties.

Rage attacks were much more common than was expected, with 19 out of 25 children experiencing these outbursts. This meant that the group of children without rage attacks was too small to allow for the planned comparison of QoL for children with and without rage attacks. The ubiquity of rage attacks suggests that is an important area for clinicians to consider when assessing TS and determining treatment plans, especially as anger emerged as a feature of TS which distressed the children who participated in the focus groups. It is unclear whether such attacks of rage represent an expression of underlying frustration and distress, which builds up as a result of having to manage TS, or whether they reflect disinhibition and problems associated with the frontal lobes.

The relationship between parental report of QoL and symptom severity was less clear, in part due to the small sample size for this part of the analysis. As would be expected, a significant relationship was shown with tic symptom severity.

In summary, the child report quantitative and qualitative data highlighted that the symptoms of TS which had a negative impact on their QoL and which were difficult to manage were 'More than just Tics'. Symptom severity explained a significant but relatively small proportion of the variance in QoL. This influence may not be direct, it could be mediated by other factors. Other factors may also have a direct influence on QoL in TS.

## **Other factors**

In this study, family functioning and self perception were considered as additional possible factors which could affect QoL. No significant relationships were found between family functioning or self-perception and parent or child reported QoL. Previous studies (Hubka et al., 1988; Wilkinson et al., 2001; Woods et al., 2005; Cooper et al., 2003) have shown weak relationships between these psychosocial variables and QoL. The small sample size in this study may mean that it is not adequately powered to detect such a weak relationship. Fitting with this idea, it is of note that the relationship between family cohesion and parental QoL is close to significance. However, as a result of the study's limited power, no conclusions can be drawn about the influence of family functioning or self perception on QoL.

As noted in the introduction, theory and previous research would predict that a positive family environment and positive self-perception would act as protective factors for the child in minimising the impact of TS on his or her QoL (Rutter, 1981; Edell-Fisher et al., 1990; Carter et al., 2000). It was noticeable in the qualitative data that young people reported a positive impact when their families understood their TS, and also noted that their TS behaviours could be a source of conflict when they were not understood. However, further investigation is needed with larger sample sizes to establish whether this is a general trend or just a specific effect for some of the individuals in the focus groups of this study.

There was a strong negative association found between tic severity and family cohesion and expression as measured on the Family Environment Scale. This highlights that TS may have an impact not just on the child but on the whole family. This finding was reflected in the qualitative data. This negative association would also fit with the idea that a more positive family environment reduces the frequency and number of children's tics. This association, between tic severity and family

environment, could therefore represent bi-directional influences.

A comparison with a sample of US families where no individual had TS showed that the families in this study had more conflict and less expression. This could be because of cultural difference between the US and the UK. However, it seems likely that it does reflect the presence of TS to some extent, particularly given that within this study there is an association between TS symptoms and family expression.

### *Comparison of QoL in Adult and Paediatric Studies*

This results of this study closely match those of a study examining the QoL of adults with TS (Elstner et al., 2001). Both found that the QoL of individuals with TS was worse than that of individuals without TS and that this QoL was poorer when symptoms were more severe (both tics and obsessive compulsive symptoms). Young people and adults were distressed by the impact of TS on their family, education or work, and on their social interactions. These consistent results could reflect analogous clinic populations, as both studies were conducted in specialised UK TS clinics. However, not all young people with TS will attend clinics as adults as many will no longer seek help when their symptoms (or their ability to manage those symptoms) improve.

It is interesting to note that there may be continuity in individual's perceptions of TS from childhood to adulthood. This suggests that interventions in childhood could have a lasting effect on QoL in adulthood. Longitudinal studies would be interesting, as it would be useful to determine whether children with poor QoL become adults with poor QoL.

### *Beliefs about TS*

The qualitative study highlighted that young people have a variety of beliefs about

TS and that they are involved in a process of attributing meaning to their TS. The results suggested that acceptance of TS as “one part of who I am” could allow the young people to manage their symptoms more effectively and to minimise the emotional impact of TS. It could be that this acceptance allows them to stop battling with their TS symptoms and it may also reduce stress. The reduction in stress associated with acceptance could also lead to a reduction in symptom severity. Accepting TS in this way could therefore not only make the symptoms of TS less distressing but could also reduce the severity of the young person's symptoms.

Developmental psychology notes that stress is inevitable but that individuals learn to protect themselves in different ways (Rutter, 1981). It is useful to consider TS as a potential stressor, which could alter the developmental pathway of a child. One method of protection from such a stressor is to minimise the exposure to risk, for example by reducing the symptoms of TS, or by educating a child's peers about TS to prevent teasing and bullying (Marcks, Berlin, Woods, & Davies, 2007). An alternative method is to change the meaning of the risk, and this seems to be happening for some of the children who are able to change the way that they think about TS. This fits with other models from adult health psychology, which note that an individual's appraisals of whether a situation is harmful and of their own resources to manage that situation are key in determining the level of stress experienced (Lazarus & Folkman, 1984). TS can both act as a stressor and be exaggerated by stress, and so effective management of TS is particularly important.

Many factors could influence young people's ability to view TS in this way, for example symptom severity, family beliefs, individual beliefs or age. Future research could usefully explore the meanings that children give to their TS, and further investigate the influences on this process of acceptance. It would be interesting to investigate whether age and developmental stage influence their ability to

conceptualise their TS in this way. It would also be interesting to investigate quantitatively whether such acceptance corresponds to a better QoL.

### *Methodological Limitations*

The results of this study should be considered in light of its methodological limitations. There are several potential sources of bias which could reduce external validity. The participants were recruited from a specialist clinic and so may not reflect the full range of TS presentations. Such a population may exclude milder cases of TS. However, the results of this study would remain valid, as they are informative about the aspects of TS which can impair QoL and may contribute to an individual seeking medical help. In addition, the participants of this study were self-selected. This could mean that children whose QoL was particularly positive chose not to participate as they did not want to be reminded of their TS. Alternatively, children whose QoL was very poor may have been unwilling to think further about TS. This would imply that the study population was biased towards young people whose QoL was in the middle range of the distribution. However, it seems unlikely that this would be a distinct population, and so the results of this study could be generalised to individuals with higher or lower QoL.

In addition, the data is cross-sectional and so it is difficult to determine the directions of causality. However, theory suggests that many relationships in this study could be bi-directional, and such relationships would be difficult to entangle even in longitudinal studies.

The measures used had specific limitations, and meant that it was not possible to fully characterise all of the symptoms associated with TS. Future research could use a more objective assessment of symptom severity, to see whether this would show the same relationship with reported QoL. In addition, it is hard to interpret the

absence of relationships where they had been hypothesized due to the limited power of the study. Considering the qualitative and quantitative results alongside each other has helped to interpret the absence of relationships, as the qualitative data have sometimes suggested that such a relationship is present for some people, even if it does not reach significance in the group as a whole.

### *Clinical Implications*

The study has many clinical implications. It highlights the importance of considering 'more than just tics' when assessing and treating TS. Obsessive-compulsive behaviours and hyperactivity are often present alongside tics and need to be assessed and considered as targets for intervention. However, in this study, ratings of symptom severity were only able to account for around 47% of the variance in child related QoL. This suggests that other factors also have an influence on QoL, and these other factors could also be useful targets for intervention, especially given the side effects associated with the medications used to control tics.

One such potential target of intervention suggested by this study is children's beliefs about their TS. Further research is needed to demonstrate a clear relationship, but it seems possible that children who are able to accept their TS as 'one part of who I am' are more able to manage it and reduce the impact that their TS has on their QoL. This suggests a key role for psychological therapy in exploring the meaning that a child and their family give to the child's TS and considering whether this is helpful for that individual.

In conclusion, this study provides useful information for clinicians despite these limitations. It highlights the impact of co-morbidities and of wider impulse control difficulties in TS. It reminds us that it is the subjective experience of individuals that determines their QoL, and that this is not only influenced by symptom severity. As

noted above, the individual's beliefs about their life and their TS also influence their QoL and provide an alternative target for psychological interventions.



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## **Part 3: Critical Appraisal**

## Introduction

Waddell (2002) highlights a distinction made by Bion between learning “about” things and learning from experience of the-self-in-the-world. The empirical paper earlier in this thesis presents the learning this study provides “about things”. This critical appraisal summarises some of the learning from my own experiences as a researcher on this project, and from the process of research design.

## The Process of Research Design

### *Deciding on my Research Questions*

My experiences prior to training included working with adults with dementia and I developed an interest in QoL in neurological conditions. Some people were successful in continuing to live a life they felt was fulfilling, whereas others struggled. Neurological conditions can have a profound effect on an individual's sense of self and mental processes and this is clearly linked in part to changes in the brain. However, Kitwood (1993) began a school of thought that acknowledged dementia is not just a manifestation of a damaged brain, but also reflects the social environment and an individual's personality and life experiences. There has been a resulting shift away from a purely medical model in understanding the experience of neurological conditions.

I hoped that my research could provide evidence and information to help understand the social and psychological impact of other neurological conditions. I was particularly interested in working with young people, and when I met with my research supervisor I was caught up in her enthusiasm for her work with young people with Tourette Syndrome. Looking at the literature in this area, it was clear that psychological research could make a contribution to a largely medical body of

literature, focussed on symptoms and medication.

In the initial stages of the literature searches I found it very hard to keep a psychological perspective, given that the literature has few psychological models. In particular, when looking at quantitative questionnaire based studies it is difficult to pick up a sense of what the young people themselves are experiencing. A few personal accounts of and by adults with TS were very helpful in providing insight into the condition (Hollenbeck, 2001; Sacks, 1995) but there are no equivalent accounts by children. This suggested that this study might need to use more qualitative methods, perhaps alongside quantitative measures.

### *Choosing a Mixed Method Design*

QoL may be assessed using objective measures of symptom severity, for example the ability to attend school. QoL can also be assessed subjectively, by finding out an individual's own perception of their quality of life, for example looking at whether they are satisfied with their own level of school attendance. I did not feel that a purely objective assessment of QoL would add a great deal to the existing literature on functioning in young people with TS. For me, a motivating factor in choosing to study QoL was the potential to understand what children themselves perceived to be important in their own lives.

This led to a focus on the meaning of the illness to the young people. A qualitative approach to research was used to capture the individual experiences of young people. This fitted with a 'social constructionist' position which acknowledged that individuals develop subjective meanings for their experiences and that these meanings are varied and multiple. Meanings are formed through interactions with others. Socially constructed meaning turned out to be very relevant to this research, with young people's views of their TS often reflecting the reactions of other people.

In addition, given the relative lack of previous research at the time of the study, it was also important to understand whether young people with TS did have a QoL which was significantly different to that of the general population, and to investigate whether there were general trends in QoL. For example, if more tics always lead directly to worse QoL tic management would be an important focus for clinicians. A quantitative measure of QoL was included in this study to allow examination of such general trends.

This led to a mixed-method strategy for this research. Such methods are becoming increasingly recognised (Creswell, 2003). Creswell (2003) acknowledges that they do provide challenges for the researcher, including extensive data collection, the time-intensive process of analysing text and numerical data and the need to be familiar with quantitative and qualitative methods. All of these were challenges I experienced on this study.

However, the advantages of the mixed method design outweighed the practical difficulties. It allowed triangulation of the results, which was particularly important given that sample size was likely to be small in a tightly defined clinical population. In addition, the qualitative results could be used to make sense of the quantitative ones and vice versa (Burke Johnson & Onwuegbuzie, 2004). An example of such a process of sense-making was the consideration of the mixed quantitative results about the social impact of TS. Understanding that the children in the groups were happy with their friends but bullied by other children who were not their friends enabled the interpretation of the mixed results from the questionnaires.

### *Process of Recruitment*

Recruitment was always likely to be challenging in such a narrowly defined group, especially with a relatively rare disorder such as TS. However, research into TS



needs to continue, despite the difficulty of ensuring an adequate sample. Most studies in the literature are from the US (particularly the ones with larger sample sizes), and it is equally important to study a UK population as significant differences are likely to exist between the two.

I worked hard to obtain the sample that is included in the study, and the response rate was reasonable, given that this is a single centre study with a limited timescale. A nationwide survey would be another possibility, but members of nationwide organisations such as the Tourette Syndrome Association are often not representative of the population as a whole. Families who are motivated to join such organisations are already showing evidence of looking for information and thinking about how to live with TS. This may not be representative of the way in which all families cope with TS. Even with such a survey, the response rate would be likely to be low, resulting in a small sample size.

Families were very willing to engage with the research. Even when they did not wish to take part, many kindly wrote to let me know why they were declining to participate. Some were not taking part as their child's TS had improved dramatically and they did not wish to discuss something which was no longer relevant to them. Others felt that discussing TS could cause their child's symptoms or distress to increase. This highlighted the anxiety created in some families by the symptoms of TS and their fears that social factors could influence the level of their child's symptoms.

### **Phone contact with families**

I tried to strike a balance between allowing the families to make their own decision about whether to participate and encouraging as many as possible to participate. This was particularly important as I had not met many of the families face-to-face.

Many participants no longer attended the clinic and I knew little about their current situation. The need for a gentle approach was highlighted by one family whom I had initially been unable to contact. When I spoke to them a few months later it emerged that one family member had been diagnosed with a serious illness, but that they were still intending to participate now that the initial crisis was over. Had I been making repeated phone calls at the time of the illness this would have been very intrusive and distressing for them.

The phone calls to families were an important part of recruitment and the response rate would have been lower without them. They also provided additional information. Many families were very appreciative of the phone contact, as they felt isolated in their local area where there were no specialist centres and few other families dealing with the same difficulties. Families commented that they felt pleased someone was taking an interest in their experiences and wanted their experiences to be known more widely.

## *Questionnaires*

### **Selection**

The questionnaires that were selected generally had acceptable reliability and validity, as shown in Table 1. The choice of some questionnaires was restricted, as it was necessary to use the same questionnaires as those used in clinic to maximise the study power. None of the previous clinic questionnaire data had been collated, and collating this data was a significant and worthwhile task. Adding these clinic questionnaires into some sections of the analysis greatly increased the power of the study.

Table 1: Reliability and Validity of Measures used in empirical study

Measure	Reference	Internal Reliability <sup>a</sup>	Test-Retest Reliability	Validity	Notes
PedsQL	Upton et al. (2005)	Greater than 0.9 for the total PedsQL score		Higher QoL was reported for healthy children than for those with health problems (asthma, diabetes, inflammatory bowel disease, in remission from cancer)	Large UK sample Larger international samples also show good results (Varni, Limbers, & Burwinkie, 2007)
MOVES	Gaffney, Sieg, and Hellings (1994)	Split half reliability coefficient was 0.87	Over at least 2 weeks 0.54 for the tic subscale, and 0.69 for the Total MOVES score	<ul style="list-style-type: none"> <li>• Correlated significantly with other clinician rated measures of tic, obsession and compulsion symptom severity, (<math>r = 0.6 - 0.8</math>)</li> <li>• Sensitive to clinical change</li> <li>• Good sensitivity (87%) and specificity (94%) for diagnosis of TS when using the cut-off value of 10 or above on the total scale</li> </ul>	Clinical sample used to determine properties small but analogous to that in this study
SDQ	Goodman (2001)	Mean of internal reliabilities was 0.73 across all raters and subscale	Mean reliability over 4-6 months was 0.62	<ul style="list-style-type: none"> <li>• Good specificity when a cut-off is used to identify ADHD from the hyperactivity subscale score (92% for parent and 93% for child report)</li> <li>• Adequate sensitivity (74% for parental report and 68% for child report)</li> </ul>	Large survey of UK schoolchildren $N = 3,983$ and their parents, $N = 9,998$
Short OCD Scale	Lee, Jones, Goodman, and Heyman, (2005)	Not well established	Not well established	<ul style="list-style-type: none"> <li>• Shown to be very sensitive to clinical change</li> </ul>	11 item version shown has good psychometric properties (Bamber, Tamplin, Park, Kyte, & Goodyer, 2002)

Table 1: Reliability and Validity of Measures used in empirical study (continued)

Measure	Reference	Internal Reliability <sup>a</sup>	Test-Retest Reliability	Validity	Notes
Rage Attacks Screen	Budman, Rockmore, Stokes, and Sossin (2003)	Not well established	Not well established	<ul style="list-style-type: none"> <li>Screening questions used in the study are based on the DSM diagnostic criteria for Intermittent Explosive Disorder</li> </ul>	
FES	Moos and Moos (1986)	0.69 – 0.78 (N = 1,067)	2 Month r(47) = 0.73 – 0.86	<ul style="list-style-type: none"> <li>Case descriptions and family profiles in manual (Moos et al., 1986)</li> <li>Staff reports correlate with patients and spouses reports of cohesion, expressiveness and conflict (Spiegel &amp; Wissler, 1983)</li> </ul>	
Harter Self-Perception Profile	Harter, (1985)	0.71 – 0.84 (N = 1,543)	2 Year correlations from 0.35 – 0.47 (Muldoon, 2000)	<ul style="list-style-type: none"> <li>Factor analysis shows that the five factors are distinct in US schoolchildren, although possibly less so in UK children (Eiser, Eiser, &amp; Havermans, 1995)</li> <li>Concurrent validity with Rosenberg Self-Esteem Scale (Hagborg, 1993)</li> </ul>	

<sup>a</sup>Cronbach's Alpha unless otherwise stated

However, as discussed in the empirical paper, not all of the questionnaires provided an adequate measure of the construct required for this study, particularly the SDQ and Rage Questionnaires. In future research, it would be particularly important to fully screen for ADHD symptoms, perhaps using a scale such as the Conners' Rating Scale (Conners, 1997). In addition, the full version of the rage attacks questionnaire could be used (Budman et al., 2003).

### **Completion**

The questionnaires were completed by parents and children at home, and then sent back to the researchers. This does mean that parents may have been able to influence their child's view and did also mean that some were filled in incorrectly and so had to be excluded from the analysis. Every effort was made to obtain consent from young people and their parents, but it is possible that some young people were coerced into taking part by their parents. For example, one young person reported no symptoms or difficulties of any kind despite a recent diagnosis of TS and a very different report from his parents. Asking the young people themselves to complete questionnaires was extremely important to the research, but the disadvantage was that it made recruitment more difficult.

Young people sometimes had difficulties completing the Self-Perception Profile (Harter, 1985; Harter, 1988). This scale was chosen as it had previously been used in research with young people with TS (Carter et al., 2000; Bawden, Stokes, Camfield, Camfield, & Salisbury, 1998), but a simpler questionnaire may have been more useful.

### *Focus Groups*

#### **Choosing to use focus groups**

Having decided to include a qualitative element in the research there was then a

choice between individual interviews with young people or focus groups. My supervisor's previous experience of groups suggested that young people were able to talk about their TS in a group setting (Murphy & Heyman, 2007). The literature on focus groups acknowledges that the interaction process can stimulate memories and discussion in the group (Millward, 2006).

Focus groups represent a balance between processes of interaction and allowing the participants to express their individual thoughts, feelings and experiences. It could be that the interactions distort the expression of individual views. For example, a dominant group member who claims that 'tourettes is no problem' could make it difficult for other individuals for whom it is a problem to express their views.

In this study, the research aims were to understand individual perceptions, and so the group was facilitated with this in mind. Millward (2006) identifies two types of moderation that can be used by group facilitators. The style used in these focus groups more closely matched 'process facilitation' in which there is high process control to maximise involvement. This ensured that the discussion was productive in terms of specificity, range and depth of information. It also provided a structure to the group which was helpful for children with attention problems.

### **Recruitment for the groups**

The week prior to a group was often a peak time of anxiety for me. However, once all the participants were in a room together with the tape running the groups were extremely rewarding and enjoyable. They brought the subject to life for me and I hope that the qualitative results will be able to do the same for others.

Some children (and their families) were very anxious prior to the group. Anxiety, and the resulting increase in tics for some children, was a significant reason for drop

out from the groups. Children were generally able to overcome this anxiety once the group had begun. Illustrating this, one child who had been too anxious to participate in the pilot group went on to attend one of the later groups. Once the group had begun he was one of its most talkative members. After my experience of families needing to drop out of the pilot I over-recruited for all subsequent groups, as recommended by experienced focus group researchers (Wilkinson, 2003). This ensured that there were adequate numbers in each group.

Some children struggled to talk about their TS. This was particularly noticeable for one child in the pilot group, who said little without direct questioning throughout the group, when the subject matter was TS. In contrast, he became extremely animated discussing one of his hobbies in the problem-free talk at the end of the session. However, this was an exception and most children who chose to come to the groups were able to talk about their TS, possibly because they were allowed to self-select.

Most of the participants felt that they benefited from the groups, although the groups were not intended to have a therapeutic effect. This highlights the clinical value of support groups and of reducing the isolation of young people with rare disorders. The young people in Group One swapped contact details, as for some it was the first time they had met anyone else with TS.

### **Choosing a method of analysis**

It was also necessary to decide on the method of analysis for the data from the groups. I considered using Interpretative Phenomenological Analysis (IPA; (Smith & Osborn, 2003). Smith (2004) expressed some reservations about the use of IPA with focus group data, whilst acknowledging that is becoming increasingly common. His reservations stem from the possible masking of individual experiences by the group interaction, and he suggests that transcripts are checked for the effect of

group interactions.

In addition, some of the children in this sample were relatively young. When I began the analysis, it was clear that whilst the data was rich in many ways, the children were not always able to reflect upon their internal state (e.g. considering the meaning of their experiences). It was therefore decided to use thematic analysis rather than IPA, as Smith and Osborn (2003) note that 'the main currency for an IPA study is the meanings particular experiences, events, states hold for participants' (p.51). The steps followed in the thematic analysis corresponded to those that would have been followed in IPA. The difference in this study is that the end themes do not always relate to the meanings that particular experiences hold for the children.

This masking of individual experiences was prevented to some extent by the facilitators ensuring that all members of the group were able to answer each question and by an explicit statement that we were happy to hear different opinions from them all and that there were no right or wrong answers. The transcripts were also read through with group interactions in mind. Significant interactions were present, but did not prevent the expression of difference, as shown in the example below from Group One.

- P4: I'm fine with it [TS], I'd live with it for the rest of my life.  
 P6: I'm fine  
 INT: So you lot are fine, which bit would you get rid of?  
 P3: Well sometimes I, you know, I find it alright, it's just the...  
 P4: Awkward situations I suppose.  
 P3: And it's just the annoyance of trying to read or concentrate on something for me I think.

### **Validity of Qualitative Analysis**

The themes found in this study were specific to the accounts of the children in this study. It is likely that many would also emerge in the accounts of other children, but



there may be specific themes which are missing from the discourse in this study. In addition, another researcher with different characteristics may well have altered the discussion with the children, and made a different interpretation of the transcripts. It is important to acknowledge there are other accounts of this data which would also be valid, if they were also grounded in the data.

### *Feedback of Results*

Finally, I feel that feedback is an important part of the research process, particularly given the time and effort that the families and young people who participated invested in this project. As such, this research will be presented at a conference for teachers, professionals and families of children with TS in late June 2007. The work will also be fed back to the TS clinic, and will be presented at one of their clinic meetings.

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## **Appendices**

## **Appendix One: Ethical Approval**

**Institute of Child Health/Great Ormond Street Hospital Research Ethics  
Committee**

The Institute of Child Health

17 July 2006

Dr Tara Murphy  
Clinical Psychologist

Dear Dr Murphy

**Full title of study:**                      **Quality Of Life in Children diagnosed with Tourettes  
Syndrome**  
**REC reference number:**              **06/Q0508/23**

Thank you for your letter of 05 July 2006, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chairman.

**Confirmation of ethical opinion**

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised.

**Ethical review of research sites**

The Committee has designated this study as exempt from site-specific assessment (SSA). There is no requirement for [other] Local Research Ethics Committees to be informed or for site-specific assessment to be carried out at each site.

**Conditions of approval**

The favourable opinion is given provided that you comply with the conditions set out in the attached document. You are advised to study the conditions carefully.

**Approved documents**

The final list of documents reviewed and approved by the Committee is as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Application		23 February 2006
Investigator CV	CV for Tara Murphy, version 1	23 February 2006

Investigator CV	CV for Jane Gilmour, version 1	23 February 2006
Protocol	version 3.1	21 February 2006
Peer Review	peer review from	11 November 2005
Questionnaire	PedsQL, Paediatric Quality of Life Inventory (UK), 13-18	
Questionnaire	PedsQL Paediatric Quality of Life Inventory Version 4.0 Child Report (young person)	
Questionnaire	MOVE Survey & Short OCD scale (young person)	
Questionnaire	Strengths and Difficulties Questionnaire (young person)	
Questionnaire	"What I Am Like" (young person)	
Questionnaire	PedsQL Paediatric Q of L Inventory v4 Parent Report for Children (parent/guardian)	
Questionnaire	Rage Attacks Questionnaire, Budman 2003 (parent/guardian)	
Questionnaire	Strengths and Difficulties Questionnaire (parent/ guardian)	
Questionnaire	Family Environment Scale - Short version (parent/guardian)	
Questionnaire	PedsQL Paediatric Quality of Life Inventory Teacher Report (Teacher)	
Questionnaire	Strengths and Difficulties Questionnaire (teacher)	
Letter of invitation to participant	version 1	24 February 2006
Participant Information Sheet	Parent/ Guardian Information Sheet	superseded
Participant Information Sheet	Young Person's Information Sheet, version 1	24 February 2006
Participant Information Sheet	8-14 years old, version 1	24 February 2006 superseded
Participant Information Sheet: Parent Guardian Information Sheet with Consent Form		
Participant Information Sheet: 14+ Information Sheet with Consent Form	2	07 July 2006
Participant Information Sheet: 8 - 14 Information Sheet with Assent Form	2	07 July 2006
Participant Consent Form	Assent Form	superseded
Participant Consent Form	Participant Consent Form	superseded
Participant Consent Form		superseded
Response to Request for Further Information	Answers to REC questions	05 July 2006
Focus Group Agenda		07 July 2006
CV	for Dawn Cutler	

### Research governance approval

You should arrange for the R&D department at all relevant NHS care organisations to be notified that the research will be taking place, and provide a copy of the REC application, the protocol and this letter.

All researchers and research collaborators who will be participating in the research must obtain final research governance approval before commencing any research procedures. Where a substantive contract is not held with the care organisation, it may be necessary for an honorary contract to be issued before approval for the



research can be given.

### **Statement of compliance**

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees (July 2001) and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

**06/Q0508/23**

**Please quote this number on all correspondence**

With the Committee's best wishes for the success of this project

Yours sincerely

Chair

Email:

Enclosures: Standard approval conditions, SL-AC2 for studies other than Clinical Trials of Investigational Medicinal Products

Copy to: Great Ormond Street Hospital/ Institute of Child Health R&D Department

## **Appendix Two: Introductory Letter**

Parents of [CHILD] and [CHILD]  
[ADDRESS]  
[ADDRESS]  
[ADDRESS]  
[ADDRESS]

DATE

Dear parents of [CHILD] and [CHILD],

We are writing to ask if you would like to take part in a study we are carrying out in the Great Ormond Street Tourettes clinic. You do not have to take part. If you decide not to participate it will not affect your care in the clinic in any way.

The study is being carried out by Dawn Cutler, Trainee Clinical Psychologist, as part of her training. Dawn is working with Dr Jane Gilmour and Dr Tara Murphy, who you may know from the clinic.

We have enclosed some information about the study with this letter. There are information sheets for you and your child and two packs of questionnaires. Dawn will contact you to see if you have any questions about this study, or alternately she can be contacted on [PHONE NUMBER]. If you would like to take part, please fill in the consent forms and questionnaires and send them back to us in the stamped addressed envelope enclosed. The information sheets are for you to keep.

If you do not wish to take part, we would be grateful if you could send back the blank forms in the envelope provided so that we can re-use them.

Thank you for taking the time to read this.

Yours sincerely,

Dr Tara Murphy	Dr Jane Gilmour	Dawn Cutler
Clinical Psychologist	Clinical Psychologist	Trainee Clinical Psychologist

## **Appendix Three: Parental Information Sheet and Consent Form**

## **Quality of Life in Tourette Syndrome Parent or Guardian Information Sheet**

We would like to ask your child to take part in a research project. Before you decide, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. Ask us if there is anything that is not clear or if you would like more information, and take your time to decide. Thank you for reading this.

### **The aim of the project**

The aim of this project is to understand the way in which the different features associated with Tourette syndrome affect a young person's life. We will be looking at how satisfied an individual is with their quality of life, and whether this is related to certain symptoms associated with Tourette syndrome, e.g. tics. We also want to understand how this relates to a young person's self esteem and family life.

### **Why is the project being done?**

Understanding which symptoms of Tourette syndrome are seen as the most significant can help doctors decide what treatments to develop in the future, and how to best use current treatments. Importantly, understanding how people view their symptoms also teaches us how best to help people cope with and manage Tourette syndrome.

### **How will the project be done?**

People who attend the Great Ormond Street Hospital Tourette clinic are being asked if they would be happy to complete some questionnaires which tell us about the symptoms they experience and about their view of their life. Some of the questionnaires need to be completed by the young person, and others need to be completed by their parent or guardian. We are also asking whether you would be happy for us to send a form to your child's school, to be completed by their teacher or Head of Year.

The questionnaires will be sent to you in the post, and queries will be discussed over the telephone. There will be about four questionnaires for each of you, which will take about 30 minutes to fill in. You will be given a stamped addressed envelope to return them. Each person will only be asked to complete one set of questionnaires.

We are also asking your permission to look in your child's medical notes. This is because we would like to use the results of some questionnaires used by the clinic which are kept in your medical notes. In addition, we would like to see if your child has completed any tests in the past which look at their ability to solve problems and at their general knowledge, and to use the results of these tests in our study.

In addition to the questionnaires, we will be running one or two discussion groups in which we will be asking a group of young people to talk about how the symptoms Tourette syndrome makes them feel, and the effect this has on them. If you would be interested in knowing more about this part of the study, please tick the box on the consent form. This does not commit you to taking part in

such a group.

### **What are the risks and discomfort?**

It is possible that thinking about their life and the effect of Tourette syndrome upon them could be upsetting for you or your child. If the questionnaires do cause any kind of distress, we would ask you to let us know so that we can offer support, and think about whether further help is needed.

### **What are the potential benefits?**

Whilst there are no immediate benefits for those people participating in this study it is hoped that the results will help us understand more about the effects of Tourette syndrome. It will help us better understand how to help people cope with the features associated with Tourette syndrome.

### **Who will have access to the information collected?**

All information which is collected during the course of the research will be kept strictly confidential. Any information about your child which leaves the hospital will have your name and address removed so that you cannot be recognised from it. The consent forms will have the name and code number on, and are the only link between your child and the information collected about them. These will be stored securely at the Institute of Child Health.

The use of some types of personal information is safeguarded by the Data Protection Act 1998 (DPA). The DPA places an obligation on those who record or use personal information, but also gives rights to people about whom information is held. If you have any questions about data protection, contact the Data Protection officer via the switchboard on extension 1234.

### **What will happen to the results of the study?**

The results from the study will be written up by Dawn Cutler and published as part of her thesis, they may also be published as papers in medical journals. No names will be used in the publication. We will also write to you at the end of the study with a brief summary of what we have found out.

### **Do I have to take part in this project?**

No. If you decide not to take part in this project, this is entirely your right and will not in any way affect your child's present or future treatment. You can withdraw from the study at any point, even after you have sent back the questionnaires.

### **Who do I speak to if I have further questions or worries?**

Please contact Dawn Cutler, who is leading this project. You can contact her on extension 1234. If you have any concerns about the way in which the project is being or has been conducted, in the first instance please discuss them with Dawn. If the problems are not resolved, or you wish to comment in any other way please contact extension 1234, Head of R&D Office, Institute of Child Health, or, if urgent, by telephone on extension 1234.

Contact: Dawn Cutler, Trainee Clinical Psychologist  
Sub-Department of Clinical Health Psychology,  
University College London,

Identification Number: \_\_\_\_\_

## CONSENT FORM

**Title of Project:** **Quality of Life in Tourette Syndrome**  
**Name of Researchers:** Dr Tara Murphy, Clinical Psychologist  
 Dr Jane Gilmour, Clinical Psychologist  
 Dawn Cutler, Trainee Clinical Psychologist

Please  
 initial box

1. I confirm that I have read and understood the information sheet dated 28.07.2006 for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐
2. I understand that my child's participation is voluntary and that they are free to withdraw at any time, without giving any reason, without medical care or legal rights being affected. ☐
3. I understand that sections of my child's medical notes may be looked at by the researchers or by regulatory authorities where it is relevant to my taking part in research. I give permission for these individuals to have access to my child's records. ☐
4. I agree to taking part in the above study. ☐

\_\_\_\_\_  
 Name of Child

\_\_\_\_\_  
 Name of Parent or Guardian

\_\_\_\_\_  
 Date

\_\_\_\_\_  
 Signature

\_\_\_\_\_  
 Researcher

\_\_\_\_\_  
 Date

\_\_\_\_\_  
 Signature

☐ Please tick this box if you would be happy for us to contact you in the future to see if your child would like to participate in a discussion group. This does not commit your child to taking part.

☐ Please tick this box if it is OK for us to send some questionnaires to your child's school. Please fill in the school details overleaf. If you do not want us to contact the school, please do not tick this box.

Identification Number: \_\_\_\_\_

Name of Child \_\_\_\_\_

Your Name \_\_\_\_\_

Please fill in the school details if you are happy for us to send them some questionnaires, which look at how your child copes at school.

Person to Contact (e.g. Head of Year, Teacher) \_\_\_\_\_

Name of School \_\_\_\_\_

School Address \_\_\_\_\_

\_\_\_\_\_

\_\_\_\_\_

\_\_\_\_\_

\_\_\_\_\_

\_\_\_\_\_

School Telephone \_\_\_\_\_



## **Appendix Four: Young Child Information Sheet and Consent Form**

**(Ages 7-13 Years)**



## **How Tourette Syndrome Affects Your Life Young Person Information Sheet**

We work at the Tourette clinic at Great Ormond Street. We are asking lots of children who have Tourette Syndrome to help us find out more about what this is like for them.

Take time to decide if you want to take part or not. Please read, or ask someone to read this information for you. Don't worry if you don't understand it straight away. Your parents have also been told about this, and you can ask them to help you understand.

### **Why are we doing this?**

We would like to understand more about how things like tics affect a child's life, and so would like to ask you about this. This will help us understand how to help other children who have similar problems.

### **What will happen?**

If you agree to take part, we will ask you to answer some questions for us about you. These questions are written down, and you will write down your answers on the forms that we give you. It will take between 30 minutes and one hour for you to fill in the questionnaires. You can ask for help with this from your parents or from one of us. Your parents will also be asked to fill out some forms, and we will ask your teacher to fill out one form if this is OK with you and your parents.

We will also be asking some children to come to Great Ormond Street and be in a group with other children. They will talk together about what it is like to have tics, and how this affects them. If you would like to know more about this, please tick the box on the form to say yes or no. Ticking this box does not mean that you have to be in a group, it just means that you would like to know more.

### **What if taking part makes me worried or upset?**

Please speak to your parents to begin with. If you would like to speak to someone else your parents know how to contact Dawn Cutler, her address and her phone number are at the end of this sheet.

### **How will the information help people?**

The information will help us to understand how we can teach people to cope with tics, and how we can help them get on with their life. We will write to you after the study finishes to let you know what we have found out.

**Who will know what I have said on the forms?**

We will look at what you have said, and keep the forms in a safe place. We will put all the information that we collect from all the children and young people who take part together. We do not keep your name or address on the forms, so no one apart from us will know your name. We may write articles for professional magazines, or publish the results in a book. No names will be used.

**Do I have to take part?**

No. If you do not want to take part, you do not have to. You can decide to stop being in the study at any point.

**Who do I speak to if I want to know more about something?**

Your parents also have information about this project. You can ask them any questions. Alternately, you can ask Dawn Cutler who is running of this project. Your parents know how to contact her.

Contact: Dawn Cutler, Trainee Clinical Psychologist  
Sub-Department of Clinical Health Psychology,  
University College London,

Identification Number: \_\_\_\_

## ASSENT FORM

**Title of Project:**                      **How Tourette Syndrome Affects Your Life**

**Name of Researchers:**              Dr Tara Murphy, Clinical Psychologist  
    Dr Jane Gilmour, Clinical Psychologist  
    Dawn Cutler, Trainee Clinical Psychologist

***Please circle YES or NO***

Have you understood the information that you were given?                      YES                      NO

Have you been able to ask any questions that you have?                      YES                      NO

Would you like to take part?                      YES                      NO

\_\_\_\_\_  
 Name

\_\_\_\_\_  
 Date

\_\_\_\_\_  
 Signature

\_\_\_\_\_  
 Researcher

\_\_\_\_\_  
 Date

\_\_\_\_\_  
 Signature

Would you like to know more about being in a group to talk about what it is like to have tics? Saying yes does not mean that you have to be in a group.                      YES                      NO

Is it OK for us to send some questions to your school?                      YES                      NO

[1 for patient, 1 for researcher, 1 to be kept with hospital notes]

## **Appendix Five: Older Child Information Sheet and Consent Form**

**(14 Years and above)**

## **Quality of Life in Tourette Syndrome Young Person Information Sheet**

We would like to ask you and your parents for permission for you to take part in a research project about Tourette syndrome. Please just ask us if there is anything that is not clear or if you would like more information.

### **The aim of the project**

The aim of this project is to understand the way in which the different symptoms associated with Tourette syndrome affect a young person's life. We will be looking at how you feel about your life generally, and whether this is related to some symptoms associated with Tourette syndrome, e.g. tics. We also want to find out how this relates to your self-esteem and to your family.

### **Why is the project being done?**

Understanding how people view their symptoms teaches us how best to help people cope with and manage Tourette syndrome. It also helps us think about what treatments might be helpful, and what should be developed in the future.

### **Why have you been chosen?**

You have been chosen because you attend or have attended the Great Ormond Street Tourette clinic, and have been diagnosed as having Tourette syndrome.

### **How will the project be done?**

If you agree to take part, you will be given some questionnaires to fill in. These tell us about the symptoms you experience and also about your view of your life more generally. We will also ask your parent's to complete some forms. If you say it is OK, we will also send a questionnaire to your school.

The questionnaires will be sent to you in the post, and we can discuss any questions over the telephone. You will only be asked to complete one set of questionnaires. There will be about four questionnaires for you to fill in, which will take about 30 minutes. We are also asking your permission to look in your medical notes, and use some of the information kept in these notes.

In addition to the questionnaires, we will be running one or two discussion groups in which we will be asking a group of young people to talk about how having the symptoms of Tourette syndrome makes them feel, and the effect this has on them. If you would be interested in knowing more about this part of the study, please tick the box on the consent form. This does not commit you to taking part in such a group.

**Are there any disadvantages to taking part?**

If completing the questionnaires makes you feel upset or worried in any way, please either speak to your parents or contact Dawn Cutler so that she can help and think about whether further support is needed.

**What are the potential advantages?**

Whilst there are no direct advantages for those people taking part in this study it is hoped that the results will help us understand more about the effects of Tourette syndrome. It will help us better understand how to help people cope with the features associated with Tourette syndrome.

**Who will have access to the information collected?**

All information which is collected during the course of the research will be kept confidential. Any information about you that leaves the hospital will have your name, date of birth and other personal information removed so that you cannot be recognised from it. As a patient, you also have rights regarding medical information recorded about you. You may read your medical record if you wish to do so.

The results from the study will be written up by Dawn Cutler, they may also be published as articles in medical journals. Your name will not be used when the research results are published. We will write to you at the end of the study to let you know what we have found out.

**Do I have to take part in this project?**

No. If you decide not to take part in this project, this is entirely your right and will not in any way affect your present or future treatment. You can also decide to stop being in the study at any point, even after you have sent the questionnaires back to us.

**Who do I speak to if I have further questions or worries?**

Please contact Dawn Cutler who is running this project. You can contact her on 020 7608 2222. If you have any concerns about the way in which the project is being or has been conducted, in the first instance please discuss them with Dawn.

If the problems are not resolved, or you wish to comment in any other way please contact Miss Emma Pendleton, Head of R&D Office, Institute of Child Health, 8th Floor, 31 Great Ormond Street, London WC1N 3EH, or, if urgent, by telephone on 020 7608 2222.

Contact: Dawn Cutler, Trainee Clinical Psychologist  
Sub-Department of Clinical Health Psychology,  
University College London,

Identification Number: \_\_\_\_ \_\_\_\_ \_\_\_\_

## CONSENT FORM

**Title of Project:**                      **Quality of Life in Tourette Syndrome**

Name of Researcher: Dr Tara Murphy, Clinical Psychologist  
    Dr Jane Gilmour, Clinical Psychologist  
    Dawn Cutler, Trainee Clinical Psychologist

initial box Please

1. I confirm that I have read and understood the information sheet dated 24.02.2006 for the above study and have had the opportunity to ask questions. ☐
  
2. I understand that I am taking part as a volunteer and that I can pull out at any time, without giving any reason, without my medical care or legal rights being affected. ☐
  
3. I understand that sections of my medical notes may be looked at by Dawn Cutler or by regulatory authorities where relevant. I give permission for these people to have access to my records. ☐
  
4. I agree to taking part in the above study. ☐

\_\_\_\_\_  
 Name of Young Person

\_\_\_\_\_  
 Date

\_\_\_\_\_  
 Signature

\_\_\_\_\_  
 Researcher

\_\_\_\_\_  
 Date

\_\_\_\_\_  
 Signature

☐

Please tick this box if you would be happy for us to contact you in the future to see if you would like to participate in a discussion group. This does not mean that you have to take part.

☐

Please tick this box if it is OK for us to send some questionnaires to your school. If you do not want us to contact your school, please do not tick this box.



## **Appendix Six: Paediatric Quality of Life Inventory**

# PedsQL<sup>TM</sup>

## Paediatric Quality of Life Inventory

Version 4.0 – UK English

**CHILD REPORT** (ages 8-12)

### DIRECTIONS

On the following page is a list of things that might be a problem for you. Please tell us **how much of a problem** each one has been for you during the **PAST MONTH** by circling:

- 0 if it is **never** a 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 do not understand a question, please ask for help.

In the **PAST MONTH**, how much of a **problem** has this been for you ...

<b>ABOUT MY HEALTH AND ACTIVITIES (problems with...)</b>	<b>Never</b>	<b>Almost Never</b>	<b>Some-times</b>	<b>Often</b>	<b>Almost Always</b>
1. It is hard for me to walk more than a couple of streets (about 100 metres)	0	1	2	3	4
2. It is hard for me to run	0	1	2	3	4
3. It is hard for me to do sports activities or exercise	0	1	2	3	4
4. It is hard for me to lift heavy things	0	1	2	3	4
5. It is hard for me to have a bath or shower by myself	0	1	2	3	4
6. It is hard for me to do chores around the house	0	1	2	3	4
7. I have aches and pains	0	1	2	3	4
8. I feel tired	0	1	2	3	4

<b>ABOUT MY FEELINGS (problems with...)</b>	<b>Never</b>	<b>Almost Never</b>	<b>Some-times</b>	<b>Often</b>	<b>Almost Always</b>
1. I feel afraid or scared	0	1	2	3	4
2. I feel sad	0	1	2	3	4
3. I feel angry	0	1	2	3	4
4. I have trouble sleeping	0	1	2	3	4
5. I worry about what will happen to me	0	1	2	3	4

<b>HOW I GET ON WITH OTHERS (problems with...)</b>	<b>Never</b>	<b>Almost Never</b>	<b>Some-times</b>	<b>Often</b>	<b>Almost Always</b>
1. I have trouble getting on with other children	0	1	2	3	4
2. Other children do not want to be my friend	0	1	2	3	4
3. Other children tease me	0	1	2	3	4
4. I cannot do things that other children my age can do	0	1	2	3	4
5. It is hard to keep up when I play with other children	0	1	2	3	4

<b>ABOUT SCHOOL (problems with...)</b>	<b>Never</b>	<b>Almost Never</b>	<b>Some-times</b>	<b>Often</b>	<b>Almost Always</b>
1. It is hard to pay attention in class	0	1	2	3	4
2. I forget things	0	1	2	3	4
3. I have trouble keeping up with my schoolwork	0	1	2	3	4
4. I miss school because of not feeling well	0	1	2	3	4
5. I miss school to go to the doctor or hospital	0	1	2	3	4

## **Appendix Seven: Other key measures**

**MOVES Parent Report****INSTRUCTIONS**

Please tick ☒ the box (Never, Sometimes, Often or Always) that shows how much each of the following things has happened to your child over the last week.

In the <u>last week</u> it has happened....		<u>NEVER</u>	<u>SOMETIMES</u>	<u>OFTEN</u>	<u>ALWAYS</u>
1	Make noises or grunts that he/she can't stop	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2	Parts of their body jerk again and again, that they can't control	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3	They have bad ideas over and over, that they can't stop	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4	Does things in a certain order or ways (like touching things)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
5	Words come out that can't be stopped or controlled	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
6	At times has the same jerk or twitch over and over	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
7	Bad words or thoughts keep going through their mind	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
8	Does exactly the opposite of what they are told	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
9	Same unpleasant or silly thought or picture goes through their mind	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
10	Can't control all movements	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
11	Has to do several movements over and over again, in the same order	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
12	Bad words or swear words come out that they don't mean to say	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
13	Feels pressure to talk, shout or scream	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
14	Has ideas that bother them (like germs or cutting themselves)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
15	Does certain things (like jumping or clapping) over and over	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
16	Has habits or movements that come out more when they are nervous	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
17	Has to repeat things that they hear other people say	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
18	Has to do things that they see other people do	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
19	Has to make bad gestures (like the finger)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
20	Has to repeat words or phrases over and over	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

### Short OCD Scale Parent Report

Please answer each question by ticking ☒ the box that most applies to your child

	<u>NO</u>	<u>A BIT</u>	<u>A LOT</u>
Does your child's mind often make him/her do things – such as checking or touching things or counting things – even though he/she knows he/she don't really have to?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Is your child particularly fussy about keeping his/her hands clean?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Does your child ever have to do things over and over a certain number of times before they seem quite right?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Does your child ever have trouble finishing his/her school work or chores because he/she has to do something over and over again?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Does your child worry a lot if he/she has done something not exactly the way he/she likes?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Does your child have trouble making up his/her mind?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

If you have answered "**A LOT**" to any of these questions, please answer the next two questions as well (if not, please go onto the next section at the bottom of this page):

	<u>NO</u>	<u>A BIT</u>	<u>A LOT</u>
Do these things interfere with your child's life?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Do you try to stop them?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

### Rage Attacks Screening Questionnaire Parent Report

Please answer each question by ticking ☒ the box that most applies to your child

	<u>NO</u>	<u>YES</u>
Over the past month, has your child had explosive outbursts of anger during which he/she became intensely angry in a way that seemed grossly excessive or inappropriate to the situation and beyond his/her control?	<input type="checkbox"/>	<input type="checkbox"/>
Do you consider these rage attacks to be uncharacteristic of your child's baseline personality?	<input type="checkbox"/>	<input type="checkbox"/>
Have these behaviors included verbal attacks or abuse?	<input type="checkbox"/>	<input type="checkbox"/>
Have these behaviors included physical attacks to property and/or included physical attacks to yourself or other people?	<input type="checkbox"/>	<input type="checkbox"/>

## SDQ Parent Report

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of the child's behaviour over the last six months.

	Not True	Somewhat True	Certainly True
Considerate of other people's feelings	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Restless, overactive, cannot 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 (treats, toys, pencils etc.)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Often has temper tantrums or hot tempers	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Rather solitary, tends to play alone	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Generally obedient, usually does what adults request	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Many worries, 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 least 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, down-hearted 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 clings 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 on 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"/>
Sees tasks through to the end, good attention span	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Do you have any other comments or concerns?

Overall, do you think that your child has difficulties in one or more of the following areas: emotions, concentration, behaviour or being able to get on with other people?

No	Yes - minor difficulties	Yes - definite difficulties	Yes - severe difficulties
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

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
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

- Do the difficulties upset or distress your child?

Not at all	Only a little	Quite a lot	A great deal
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

- 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
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Signature ..... Date .....

Mother/Father/Other (please specify:)

**Thank you very much for your help**



## Harter Self-Perception Profile for Children Child Report

### INSTRUCTIONS

Please read these instructions carefully, and if you are not clear what you have to do ask your parents for help.

For each line, first decide what type of person you are, and draw a circle round that sentence. If you like to play outdoors in your spare time more than you like to watch TV, you would circle that sentence. For example,

Really True for me	Sort of True for me			Really True for me	Sort of True for me
<input type="checkbox"/>	<input checked="" type="checkbox"/>	Some kids would rather play outdoors in their spare time	BUT	Other kids would rather watch TV	<input type="checkbox"/>

Then, please tick ☒ the box to show whether each sentence is "really true for you" or "sort of true for you".

Really True for me	Sort of True for me			Really True for me	Sort of True for me
<input checked="" type="checkbox"/>	<input checked="" type="checkbox"/>	Some kids would rather play outdoors in their spare time	BUT	Other kids would rather watch TV	<input type="checkbox"/>

For each line, first chose one sentence to say what type of a person you are and then tick a box to say whether it is "Really True" or "Sort of True" for you.

### START HERE

	Really True for me	Sort of True for me			Really True for me	Sort of True for me
1	<input type="checkbox"/>	<input type="checkbox"/>	Some kids feel that they are very <i>good</i> at their school work	BUT	Other kids <i>worry</i> about whether they can do the school work assigned to them	<input type="checkbox"/>
2	<input type="checkbox"/>	<input type="checkbox"/>	Some kids find it <i>hard</i> to make friends	BUT	Other kids find it's pretty <i>easy</i> to make friends	<input type="checkbox"/>
3	<input type="checkbox"/>	<input type="checkbox"/>	Some kids do very <i>well</i> at all kinds of sports	BUT	Other kids <i>don't</i> feel that they are very good when it comes to sports	<input type="checkbox"/>

	Really True for me	Sort of True for me				Really True for me	Sort of True for me
4	<input type="checkbox"/>	<input type="checkbox"/>	Some kids are <i>happy</i> with the way they look	BUT	Other kids are <i>not</i> that happy with the way they look	<input type="checkbox"/>	<input type="checkbox"/>
5	<input type="checkbox"/>	<input type="checkbox"/>	Some kids often do <i>not</i> like the way they <i>behave</i>	BUT	Other kids usually <i>like</i> the way they behave	<input type="checkbox"/>	<input type="checkbox"/>
6	<input type="checkbox"/>	<input type="checkbox"/>	Some kids are often with <i>unhappy</i> themselves	BUT	Other kids are pretty <i>pleased</i> with themselves	<input type="checkbox"/>	<input type="checkbox"/>
7	<input type="checkbox"/>	<input type="checkbox"/>	Some kids feel like they are <i>just as smart</i> as other kids their age	BUT	Other kids aren't so sure and <i>wonder</i> if they are as smart	<input type="checkbox"/>	<input type="checkbox"/>
8	<input type="checkbox"/>	<input type="checkbox"/>	Some kids have a <i>lot</i> of friends	BUT	Other kids <i>don't</i> have very many friends	<input type="checkbox"/>	<input type="checkbox"/>
9	<input type="checkbox"/>	<input type="checkbox"/>	Some kids wish they could be a lot better at sports	BUT	Other kids feel they are good enough at sports	<input type="checkbox"/>	<input type="checkbox"/>
10	<input type="checkbox"/>	<input type="checkbox"/>	Some kids are <i>happy</i> with their height and weight	BUT	Other kids wish their height or weight were <i>different</i>	<input type="checkbox"/>	<input type="checkbox"/>
11	<input type="checkbox"/>	<input type="checkbox"/>	Some kids usually do the <i>right</i> thing	BUT	Other kids often <i>don't</i> do the right thing	<input type="checkbox"/>	<input type="checkbox"/>
12	<input type="checkbox"/>	<input type="checkbox"/>	Some kids <i>don't</i> like the way they are leading their life	BUT	Other kids <i>do</i> like the way they are leading their life	<input type="checkbox"/>	<input type="checkbox"/>
13	<input type="checkbox"/>	<input type="checkbox"/>	Some kids are pretty <i>slow</i> in finishing their school work	BUT	Other kids can do their school work <i>quickly</i>	<input type="checkbox"/>	<input type="checkbox"/>
14	<input type="checkbox"/>	<input type="checkbox"/>	Some kids would like to have a lot more friends	BUT	Other kids have as many friends as they want	<input type="checkbox"/>	<input type="checkbox"/>
15	<input type="checkbox"/>	<input type="checkbox"/>	Some kids think they could do well at just about any new sports activity they haven't tried before	BUT	Other kids are afraid they might <i>not</i> do as well at sports they haven't ever tried	<input type="checkbox"/>	<input type="checkbox"/>
16	<input type="checkbox"/>	<input type="checkbox"/>	Some kids wish their body was <i>different</i>	BUT	Other kids <i>like</i> their body the way it is	<input type="checkbox"/>	<input type="checkbox"/>
17	<input type="checkbox"/>	<input type="checkbox"/>	Some kids usually act the way they are <i>supposed</i> to	BUT	Other kids often <i>don't</i> act the way they are supposed to	<input type="checkbox"/>	<input type="checkbox"/>

	Really True for me	Sort of True for me				Really True for me	Sort of True for me
18	<input type="checkbox"/>	<input type="checkbox"/>	Some kids are <i>happy</i> with themselves as a person	BUT	Other kids are often <i>not</i> happy with themselves	<input type="checkbox"/>	<input type="checkbox"/>
19	<input type="checkbox"/>	<input type="checkbox"/>	Some kids often <i>forget</i> what they learn	BUT	Other kids can remember things <i>easily</i>	<input type="checkbox"/>	<input type="checkbox"/>
20	<input type="checkbox"/>	<input type="checkbox"/>	Some kids are always doing things with <i>a lot</i> of kids	BUT	Other kids usually do things by <i>themselves</i>	<input type="checkbox"/>	<input type="checkbox"/>
21	<input type="checkbox"/>	<input type="checkbox"/>	Some kids feel they are <i>better</i> than others their age at sports	BUT	Other kids <i>don't</i> feel they can play as well	<input type="checkbox"/>	<input type="checkbox"/>
22	<input type="checkbox"/>	<input type="checkbox"/>	Some kids wish their physical appearance (how they look) was <i>different</i>	BUT	Other kids <i>like</i> their physical appearance the way it is	<input type="checkbox"/>	<input type="checkbox"/>
23	<input type="checkbox"/>	<input type="checkbox"/>	Some kids usually get in <i>trouble</i> because of the things they do	BUT	Other kids usually <i>don't</i> do things that get them in trouble	<input type="checkbox"/>	<input type="checkbox"/>
24	<input type="checkbox"/>	<input type="checkbox"/>	Some kids <i>like</i> the kind of person they are	BUT	Other kids often wish they were someone else	<input type="checkbox"/>	<input type="checkbox"/>
25	<input type="checkbox"/>	<input type="checkbox"/>	Some kids do <i>very well</i> at their class work	BUT	Other kids <i>don't</i> do very well at their class work	<input type="checkbox"/>	<input type="checkbox"/>
26	<input type="checkbox"/>	<input type="checkbox"/>	Some kids wish that more people their age liked them	BUT	Other kids feel that most people their age <i>do</i> like them	<input type="checkbox"/>	<input type="checkbox"/>
27	<input type="checkbox"/>	<input type="checkbox"/>	In games and sports some kids usually <i>watch</i> instead of play	BUT	Other kids usually <i>play</i> rather than just watch	<input type="checkbox"/>	<input type="checkbox"/>
28	<input type="checkbox"/>	<input type="checkbox"/>	Some kids wish something about their face or hair looked <i>different</i>	BUT	Other kids <i>like</i> their face and hair the way they are	<input type="checkbox"/>	<input type="checkbox"/>
29	<input type="checkbox"/>	<input type="checkbox"/>	Some kids do things they know they <i>shouldn't</i> do	BUT	Other kids <i>hardly ever</i> do things they shouldn't do	<input type="checkbox"/>	<input type="checkbox"/>

	Really True for me	Sort of True for me				Really True for me	Sort of True for me
30	<input type="checkbox"/>	<input type="checkbox"/>	Some kids are very <i>happy</i> being the way they are	BUT	Other kids wish they were <i>different</i>	<input type="checkbox"/>	<input type="checkbox"/>
31	<input type="checkbox"/>	<input type="checkbox"/>	Some kids have <i>trouble</i> figuring out the answers in school	BUT	Other kids almost <i>always</i> can figure out the answers	<input type="checkbox"/>	<input type="checkbox"/>
32	<input type="checkbox"/>	<input type="checkbox"/>	Some kids are <i>popular</i> with others their age	BUT	Other kids are <i>not</i> very popular	<input type="checkbox"/>	<input type="checkbox"/>
33	<input type="checkbox"/>	<input type="checkbox"/>	Some kids <i>don't</i> do well at new outdoor games	BUT	Other kids are <i>good</i> at new games right away	<input type="checkbox"/>	<input type="checkbox"/>
34	<input type="checkbox"/>	<input type="checkbox"/>	Some kids think that they are good looking	BUT	Other kids think that they are not very good looking	<input type="checkbox"/>	<input type="checkbox"/>
35	<input type="checkbox"/>	<input type="checkbox"/>	Some kids behave themselves very well	BUT	Other kids often find it hard to behave themselves	<input type="checkbox"/>	<input type="checkbox"/>
36	<input type="checkbox"/>	<input type="checkbox"/>	Some kids <i>are</i> not very happy with the way they do a lot of things	BUT	Other kids think the way they do things is <i>fine</i>	<input type="checkbox"/>	<input type="checkbox"/>

Thank you!

### *Family Environment Scale – Parent Report*

There are statements about families in this scale. You are to decide which of these statements are true of your family and which are false. If you think the statement is TRUE or mostly TRUE of your family, make an X in the box labelled TRUE. If you think the statement is FALSE or mostly FALSE of your family, make an X in the box labelled FALSE.

		<u>TRUE</u>	<u>FALSE</u>
1	Family members really help and support one another.	<input type="checkbox"/>	<input type="checkbox"/>
2	Family members often keep their feelings to themselves.	<input type="checkbox"/>	<input type="checkbox"/>
3	We fight a lot in our family.	<input type="checkbox"/>	<input type="checkbox"/>
4	We often seem to be 'killing time' at home.	<input type="checkbox"/>	<input type="checkbox"/>
5	We say anything we want to at home.	<input type="checkbox"/>	<input type="checkbox"/>
6	Family members rarely become openly angry.	<input type="checkbox"/>	<input type="checkbox"/>
7	We put a lot of energy into what we do at home.	<input type="checkbox"/>	<input type="checkbox"/>
8	It's hard to 'blow off steam' at home without upsetting somebody.	<input type="checkbox"/>	<input type="checkbox"/>
9	Family members sometimes get so angry they throw things.	<input type="checkbox"/>	<input type="checkbox"/>
10	There is a feeling of togetherness in our family.	<input type="checkbox"/>	<input type="checkbox"/>
11	We tell each other about our personal problems.	<input type="checkbox"/>	<input type="checkbox"/>
12	Family members hardly ever lose their tempers.	<input type="checkbox"/>	<input type="checkbox"/>
13	We rarely volunteer when something has to be done at home.	<input type="checkbox"/>	<input type="checkbox"/>
14	If we feel like doing something on the spur of the moment we often just pick up and go.	<input type="checkbox"/>	<input type="checkbox"/>
15	Family members often criticise each other.	<input type="checkbox"/>	<input type="checkbox"/>
16	Family members rarely back each other up.	<input type="checkbox"/>	<input type="checkbox"/>
17	Someone usually gets upset if you complain in this family.	<input type="checkbox"/>	<input type="checkbox"/>
18	Family members sometimes hit each other.	<input type="checkbox"/>	<input type="checkbox"/>
19	There is very little group spirit in our family.	<input type="checkbox"/>	<input type="checkbox"/>
20	Money and paying bills is openly talked about in our family.	<input type="checkbox"/>	<input type="checkbox"/>
21	If there is a disagreement in our family, we try hard to smooth things over and keep the peace.	<input type="checkbox"/>	<input type="checkbox"/>
22	We really get along well with each other.	<input type="checkbox"/>	<input type="checkbox"/>
23	We are usually careful about what we say to each other.	<input type="checkbox"/>	<input type="checkbox"/>
24	Family members often try to one-up or out-do each other.	<input type="checkbox"/>	<input type="checkbox"/>
25	There is plenty of time and attention for everyone in our family.	<input type="checkbox"/>	<input type="checkbox"/>
26	There are a lot of spontaneous discussions in our family.	<input type="checkbox"/>	<input type="checkbox"/>
27	In our family we believe you don't ever get anywhere by raising your voice.	<input type="checkbox"/>	<input type="checkbox"/>

**Appendix Eight: Interview Schedule**

## Focus Group Agenda

Where possible, we will use more than one modality (i.e. written and spoken) to help young people with attention difficulties who may be easily distracted participate. Notes about visual materials and other equipment are in italics.

### Ground Rules and Confidentiality (10 minutes)

*Make name badges when come into room and during introduction*

Introductions: who we are (me and Patricia)

Ice breaker: Test recording equipment – go round group and get everyone to test by saying name and favourite sport / colour. *Rewind and check OK.*

Ground rules: confidentiality, don't need to answer if makes uncomfortable, let us/parents know afterwards if anything worrying  
(*Prepare sheet on flip chart to refer to which has two words on - confidentiality and worry*)

Record consent on audio-tape

### Symptoms and view of Tourette Syndrome (20 minutes)

What are the key things that Tourette syndrome makes you do?

- *Give everyone large post it notes to write things on. Stick on board. Check for similarities.*
- *Use participant's terms for symptoms throughout session.*
- Prompts:
  - Is there anything about your behaviour which is affected by Tourettes?
  - If not mentioned, ask specifically re. some symptoms e.g. tics, worries, being very active, hard to concentrate/sit still, angry at times

If a friend at school asked you what Tourette syndrome was, what would you say?

- *Get a volunteer to be "writer-upper" and put ideas on board*

Do any of these things upset or worry you? Which ones?

- *Have a separate sheet on wall titled "Things which worry me" and either move post-its across to this board or write up parts which worry participants*

It is OK to be worried about things sometimes, but we also have lots of ways to cope with our worries. What helps you to beat the worry?

- *Again have sheet titled "Things which help me cope" on wall and ask for volunteer to write up*

### Self Esteem (20 minutes)

Now we are going to work out which parts of your life Tourette syndrome affects, and whether it has a good or bad effect. We are going to start with sports. Does Tourette syndrome mean that sports are harder for you, does it have no effect, or does it make them easier?

- *Use thermometer to indicate whether hard or easy – each participant to have stickers with their initials on so that they can show how much affected. Use positioning to prompt discussion.*
- Prompts:

- Once stuck on, ask about position and find out why has that effect
- Why does Tourettes make it hard / easy?
- Which bit of Tourettes makes it harder?
- Does it bother or upset you that Tourette's affects these things?
- Repeat process for all domains covered on Harter Self-Esteem questionnaire:
  1. Sports and activities
  2. School work
  3. Making friends and being accepted by other people – generally and for close friendships
  4. Appearance – the way you look
  5. Relationships – having boyfriends or girlfriends
  6. Behaviour i.e. getting into trouble
  7. Working – getting a job (depending on ages of participants)

*Check covered impact on quality of life and return to question if necessary - Does it bother or upset you that Tourette's affects these things?*

*How do you cope? Likely to be discussed throughout but return to if needed*

### **Family (20 minutes)**

What does your mum / dad say about your Tourette syndrome?

- *Again write up on board (volunteer)*

Does it ever get you into trouble at home or cause arguments?

Who in your family is most worried about Tourette syndrome and why?

Is there anything they do which is helpful / unhelpful?

- *Put into two columns of helpful and unhelpful on flip chart*

### **Change Question (10 minutes)**

People often think about changing parts of themselves, and people do change as they grow up and go through school. If you didn't have Tourette syndrome what would be different in your life?

- Prompts:
  - School, friends, activities
  - Would it be better or worse?
  - Who else would it affect?
- *If finding it hard to say out loud, consider getting them to write down on paper and then put in pile in centre. Can then each pick one to read out..*

### **Summary of discussion and check for areas missed (15 minutes)**

Give summary of discussion and check whether participants agree

Give certificates for participation

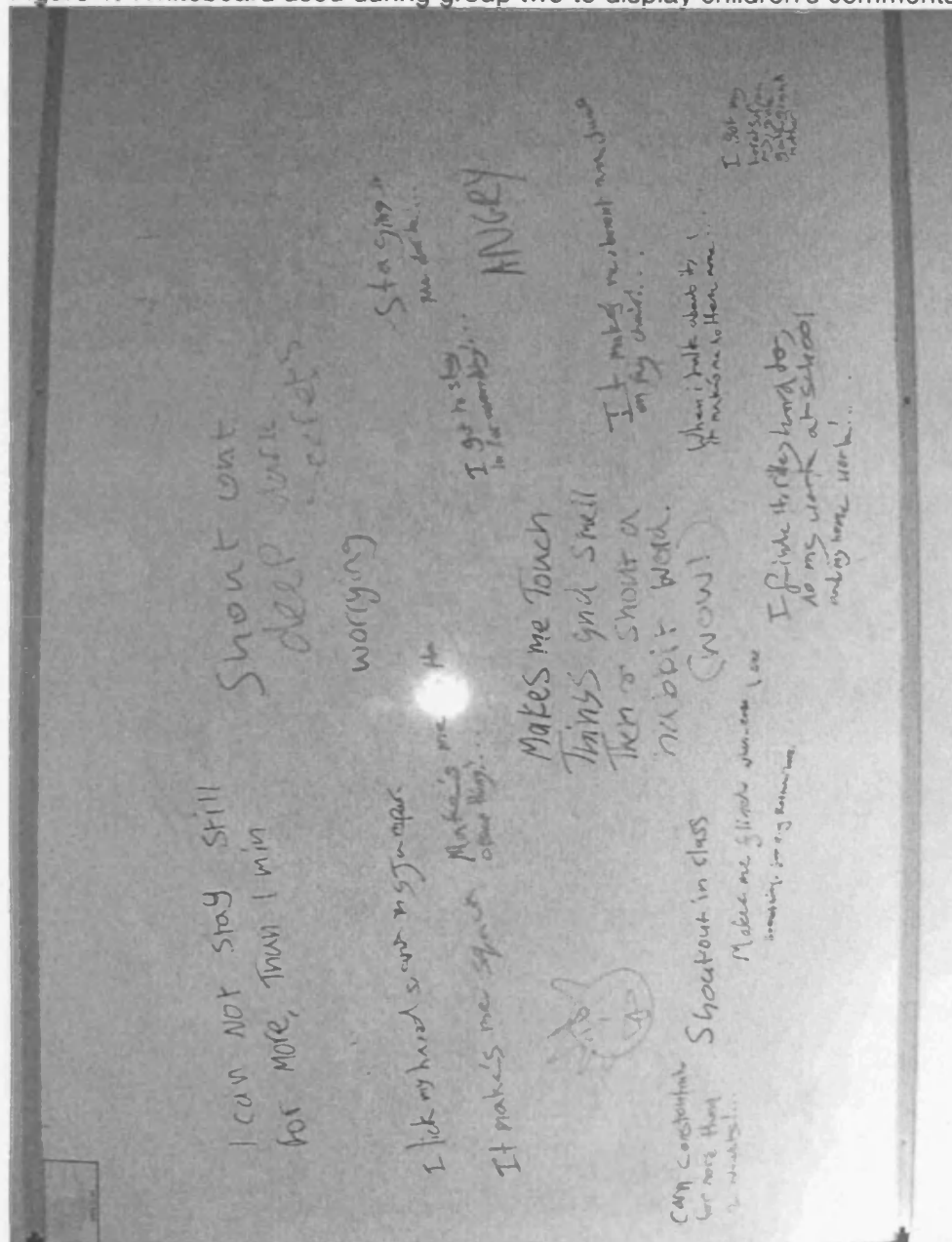
### **Positives and problem-free talk to finish (5 minutes)**

Tell us about an achievement you have managed in the last 6 months. What makes you feel proud about yourself?



## **Appendix Nine: Record of Information from Focus Group**

**Figure 1: Whiteboard used during group two to display children's comments**



## **Appendix Ten: Examples of Qualitative Analysis**

### *Step One of Analysis: Annotating interesting or significant comments*

**Table 1: Initial Coding of a Section of the Discussion in Group One (Left Margin)**

Initial Code	Original Transcript	
Hard to distinguish TS behaviour from other behaviour	P3	And my sister, she understands but I think sometimes she doesn't realise it's Tourettes, she thinks it's me being annoying, and is like 'oh stop it D', but I can't because its my Tourettes if you know what I mean.
	INT	What about for you, what does your family say?
Lack of understanding Stress worsens symptoms Hard to distinguish	P4	Well, me mum was the first person to notice it, and that was when I was younger, but I've only been diagnosed with Tourettes for two years, but when I used to tic when I was younger, as I said, my dad didn't just tell me to stop, he used to shout at me and make me stop it, and that's what made me worse. But me mum was like 'don't shout at him he can't help it' but other than that, everyone just, at first they though I was just being a pain in the butt but then they realised that I'd just got Tourettes.
	INT	So there are some things people can do that are helpful in your family, and there are some things people can do that aren't helpful?
Acceptance from family	P4	In other words, it was basically that my whole family was alright with it, but it was just my dad that tried to stop it.
	INT	Mmmm. And the trying to stop it made it worse for you?
Trying to control worsens Need to let out Acceptance	P4	Yes..
	P5	Yes, best to just let it out and be yourself.
	INT	What do your family say about it?
	P5	Well, my mum's sort of got it, so, she realises she had it when she was, talks to herself and goes [murmurs] but...
	INT	Does that make it easier or harder for you?
Understanding of others Isolated	P5	I sometimes say what are you doing, but then again I've got it, so I can't really, I don't make fun of it. That's the funny thing because I've never met anyone with Tourettes, apart from one person but...

## Step Two of Analysis: Document Emerging Theme Titles

**Table 2: Documenting the Emerging Theme Titles (Right Hand Margin)**

Initial Code		Original Transcript	Theme Titles
Hard to distinguish TS behaviour from other behaviour	P3	And my sister, she understands but I think sometimes she doesn't realise it's Tourettes, she thinks it's me being annoying, and is like 'oh stop it D', but I can't because its my Tourettes if you know what I mean.	Others not understanding
	INT	What about for you, what does your family say?	
Lack of understanding Stress worsens symptoms	P4	Well, me mum was the first person to notice it, and that was when I was younger, but I've only been diagnosed with Tourettes for two years, but when I used to tic when I was younger, as I said, my dad didn't just tell me to stop, he used to shout at me and make me stop it, and that's what made me worse. But me mum was like 'don't shout at him he can't help it' but other than that, everyone just, at first they though I was just being a pain in the butt but then they realised that I'd just got Tourettes.	Others not understanding Stress worsens symptoms
Hard to distinguish	INT	So there are some things people can do that are helpful in your family, and there are some things people can do that aren't helpful?	Others not understanding
Acceptance from family	P4	In other words, it was basically that my whole family was alright with it, but it was just my dad that tried to stop it.	Acceptance from family
	INT	Mmmm. And the trying to stop it made it worse for you?	
Trying to control worsens Need to let out Acceptance	P4	Yes..	Struggle to control
	P5	Yes, best to just let it out and be yourself.	Need to let out Acceptance
	INT	What do your family say about it?	
	P5	Well, my mum's sort of got it, so, she realises she had it when she was, talks to herself and goes [murmurs] but...	
Understanding of others	INT	Does that make it easier or harder for you?	Understanding of others
Isolated	P5	I sometimes say what are you doing, but then again I've got it, so I can't really, I don't make fun of it. That's the funny thing because I've never met anyone with Tourettes, apart from one person but...	Isolated

### *Step Three of Analysis: Listing and Clustering of Emerging Themes*

**Table 3: Examples of Clustering of Emerging Themes from Group One**

Theme	Page and Line Number	Sample Quotes
Cluster: Involuntary behaviours challenge other people's understanding and make it hard to fit in		
Others not understanding	2.44 5.44 7.4	'annoying' 'awkward to be around' 'look at you like you're mental'
Other people judge and stare	16.32	'kept on staring'
Not fitting in	3.1	'worry about what people see and think'
Secrecy as a defence	7.5	'don't know I have it'
Theme	Page and Line Number	Sample Quotes
Cluster : Balance between controlling symptoms and letting them out		
Struggle to control and keep inside	10.35 16.4	'try and control it more, and it gets worse' 'used to shy down and be quiet'
Need to let out	8.7 3.3 3.28	'it just lets out' 'let it out your system' 'when you get home you just [scream]'

### *Step Five of Analysis: Super-Ordinate Themes and Illustrative Quotes*

#### **Theme 2: Struggling to fit into society's expectations of normal behaviour**

Sub-theme: Others not understanding involuntary behaviours

Yes, cause as well, she [Teacher] told me off, and like I didn't want to say it cause she would send me to our head teacher, but I wanted to say 'well I can't help it', but she would have been really angry. [P10, Two]

Oh, once I had this teacher called Mr [name], and he, cause I done a tic he told me off for doing something with my eyes. [P7, Two]

Well, they [school] didn't understand until I was in Year 11, and it was only the last couple of months of me being there, they just thought I was being naughty. [P4, One]

I: So there is this thing that you might be being naughty, that you should be able to behave differently? P4: Yes, but people just think you can automatically do it. P6: They just think you are like other people [One]

He used to like sort of if I ticked when I was umm If I sort of ticked when he was watching TV or something he was like 'shhh be quiet', and I'm like 'I can't help it'. He never really understood that much, so... But he does quite a lot more now... [P3, One]

And my sister, she understands but I think sometimes she doesn't realise it's Tourettes, she thinks it's me being annoying, and is like 'oh stop it D', but I can't because its my Tourettes if you know what I mean. [P3, One]

I've just moved into my dad's because I was that bad at home. [P1, Pilot]

My mum used to think I was being annoying, so in the end we sort of um went to our local CAMHS service and they sort of diagnosed it as Tourette Syndrome. [P3, One]

#### **Theme 3: Needing to control tics**

I also do things which I can't control [P1, Pilot]

I: The noises bother you the most. Why are they the hardest for you? P: Because I can't really control them. Like sometimes I can control them like Tourettes, but other times like the Tourettes is out of control. [P1, Pilot]

Because when I look at people, they are really odd ones just like [demonstrates a facial tic] and when I look at people and I don't know them I try and keep them in, but if you do do them they like look at you and stare at you, it's really annoying. [P10, Two]

Well, its when I'm outside, I tend to worry about what people see and what they think, so I'll stop it [P4, One]

I think so, and knowing that if you concentrate and the tics come out and people around you are aware of it, you see people looking at me doing that [tics] and then you try and control it more, and it gets worse. [P3, One]