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# Volume I:

# Literature Review and Empirical Paper

**Caroline Formby**

**School of Psychology**
**Faculty of Psychology**
**University of Birmingham**

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# Volume I: Literature Review and Empirical Paper

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# Literature Review:

## Is There Any Evidence For Psychologically Mediated Psychopathology In Tourette Syndrome: A review and hypothesis of process mechanisms involved

Prepared for Neuroscience and Biobehavioural Reviews

## Abstract

A literature review sought to determine whether there is any evidence for psychologically mediated psychopathology in Tourette Syndrome (TS). A description of TS is followed by review of literature investigating the occurrence of a variety of psychopathological co-morbidities encountered in the disorder. This is followed by a review of empirical studies examining the impact of TS in children; peer relationships, family function, self-concept and adjustment and behavioural difficulties. Empirical studies investigating the impact of TS in adults are then reviewed. A hypothetical psychological model provides a framework by which the reciprocal relationships between TS and co-morbid OCD and ADHD as neurological vulnerabilities, peer and family relationships, internalising and externalising behavioural difficulties might be mediated by self-representation and self-efficacy to result in the development of psychopathology. Further discussion of the modes by which these factors may influence psychopathology in an adult TS population is provided. Implications for treatment and further research are discussed.

## Introduction

The aim of this paper is to review recent, currently published literature about psychopathology in Tourette Syndrome (TS). This is in order to determine if there is any evidence for psychologically mediated psychopathology in Tourette Syndrome. If this is the case, then a hypothetical psychological framework will be suggested highlighting the process by which these factors may be mediated.

The first part of this paper aims to summarise research about the co-occurrence of psychopathology in individuals with Tourette Syndrome (TS). Secondly, to review a number of studies that have purported to investigate the psychosocial and perceived psychological worlds of children and adults with TS. Thirdly to suggest a psychological framework for the processes observed through empirical research. This framework could conceivably be used in future research as a basis for hypothesis testing in order to clarify the possible mechanisms involved. Finally, a short discussion will reflect on the clinical implications of these findings and suggestions for further research.

## Description of Tourette Syndrome

Tourette Syndrome is defined as a neuropsychiatric disorder, with a mean age of onset of 5-7 years old (Leckman, Zhang, Vitale, Lahnin, Lynch, Bondi, Kim & Peterson, 1998; Baron-Cohen, Mortimore, Moriarty, Izaguirre, & Robertson, 1999). It is characterised by multiple motor tics and at least one vocal tic, which have lasted for at least one year, although they need not occur simultaneously (American Psychiatric Association, 2000; World Health Organisation, 1992). A tic is described as a sudden, rapid, recurrent, non-rhythmic, stereotyped motor movement or vocalisation (APA,

2000), which can be simple or complex. Simple tics involve only one group of muscles and a simple motor tic might appear as a brief jerk-like movement or twitch, whereas a simple vocal tic might be a meaningless sound such as a sniff, throat clearing, grunt, squeak, snort etc (Jankovic, 2001). Complex tics are defined as those which mimic a co-ordinated sequenced movement or gesture that might otherwise be interpreted as an everyday movement. These might include touching, jumping, skipping, hitting, gesturing towards others or copying others' gestures. Complex vocal tics might be meaningful utterances, such as saying or shouting obscenities or profanities, repetition and imitation of others' phrases or of their own phrases or words (Jankovic, 1997). Jankovic (1997) explains that most tics can be considered to be semi-voluntary or involuntary but suppressible and are often preceded by a sensory experience known as a premonitory urge (Leckman, Walker & Cohen, 1993).

The lifetime course of TS is typically episodic, although it has been found that between half and two thirds of diagnosed cases resolve completely or that tic symptoms are significantly reduced by early adulthood (Hedren, 2002; Leckman et al., 1998).

**Prevalence Rates for Tourette Syndrome**

Prevalence rates for Tourette Syndrome have been estimated from both clinical and community samples in different countries (Apter, Pauls, Bleich, Zohar, Kron & Ratzoni, 1993; Hornsey, Banjeree, Zeitlin & Robertson, 2001; Kadesjo & Gillberg, 2000). Overall agreement seems to have been reached that the accepted rate is around 5 in 10 000 children (Robertson, 2000). However, there is some debate as to whether this represents an underestimate, given that a number of studies have found rates

much higher, when looking at specific populations (Mason, Banjeree, Eapen, Zeitlin & Robertson, 1998). For example, Eapen, Robertson, Zeitlin, & Kurlan, (1997*a*) reported that out of 81 children in special education schools in the UK, 65% had a definite tic disorder, and 11.55% were diagnosed with definite TS. Furthermore, Comings, Hines & Comings (1990) found a prevalence rate of 63 in 10 000, in children referred to a school psychologist. However, different researchers have used different criteria to ascertain diagnostic categories and many are thought to have yielded inflated estimates, e.g. Kurlan, Como, Miller, Palumbo, Deeley, Andresen, Eapen, & McDermott (2002) who reported a 27% rate of tics as opposed to "no tics". See Scahill, Tanner & Dure (2001) for a review of the epidemiology of TS and tic disorders.

**Aetiology of Tourette Syndrome**

Tourette Syndrome (TS) is thought to be a predominantly inherited disorder transmitted by an autosomal dominant gene, not yet identified, which represents a susceptibility to the disorder (Pauls, 2001). This finding has been corroborated by twin and family studies and by genetic segregation analysis investigating TS and chronic tic disorders with and without Obsessive Compulsive Disorder (OCD) (Pauls, 2001). The marked incidence of OCD or OC symptoms co-morbid with TS and tic disorders and the pattern of family occurrence of both, have led researchers to hypothesise a spectrum of tic disorders of which TS and OCD are represented by the same genetic trait (Swerdlow, Zinner, Farber, Seacrist & Hartston, 1999; Pauls, 2001; Kurlan et. al. 2002; Miguel, Rosario-Campos, Shavitt, Hounie, & Mercadante, 2001; Alsobrook & Pauls, 1997). However, it is also postulated that numerous other factors such as infections, pre- and perinatal stressors may also contribute to the occurrence

of TS (Bessen & Lombroso, 2001; Hallett & Kiessling, 1997; Walkup, 2001). Robertson (2000) provides an overview of these different hypotheses, which are beyond the current scope of review.

Similar genetic investigations of the marked incidence of co-morbid Attention Deficit Disorder (with or without hyperactivity) (ADHD) in TS have reportedly not supported the hypothesis that ADHD is genetically linked in the same way as OCD to TS. Although many researchers argue that this is the case, the precise link has not yet been identified (Pauls & Leckman, 1986).

In terms of neuroanatomical and neurochemical correlates, TS is broadly hypothesised to involve the cortical-striatal-thalamo-cortical circuits (Peterson, 2001). Further investigations have proposed that similar neuroanatomic localisation involving the caudate nuclei and serotonin neurotransmission might explain the association between TS and OCD, and furthermore abnormal dopamine levels have also been implicated (Kurlan et. al., 2002; Singer & Wendlandt, 2001; Castellanos, 2001).

**Psychopathology and Tourette Syndrome**

Even though there has been criticism levelled at studies based on clinical populations because of the possibility of over estimates due to ascertainment bias, there is still a postulated link between OCD and ADHD to TS, as outlined above. It seems that co-occurring mechanisms of psychopathology may in fact take two forms, the first being the neurobehaviourally linked disorders and the second being other psychopathologies co-occurring with TS that may be linked by psychological and/or organic factors. The

following section describes studies that have investigated the frequency and nature of OCD, ADHD, anxiety, depression and a limited number of other psychopathologies, co-occurring in children and adults with TS. It is important to highlight that many of the following studies have assessed children or adults or both and that this distinction is not specified for some studies. It was therefore not feasible to separate these studies by a predominantly child or adult focus.

*Obsessive Compulsive Disorder*

Robertson (2000) reports that suggestion has been made of three types of OCD, the first being a familial one related to tic disorders, the second a familial type unrelated to tics and a third being a non-familial type. Even though research methods have used different assessment tools and have often failed to differentiate between OCD itself, obsessive or compulsive symptoms and traits, this hypothesis still appears to be favoured (Robertson, 2000).

In the UK, OCD is estimated to be prevalent in approximately 1.5% of the adult population (Jenkins, Lewis, Bebbington, Brugha, Farrell, Gill & Meltzer, 2003) and around 0.25% of the child population (Heyman, Fombonne, Simmons, Ford, Meltzer & Goodman, 2001). Similar figures have been reported for adults in the USA at a rate of 2.1% prevalence (Narrow, Rae, Robins, & Regier, 2002).

The diagnostic categories used to define OCD in those with TS have varied to include Obsessive Compulsive Behaviour and Obsessive Compulsive Symptoms, which represent a possible sub-threshold to OCD itself. However, as Robertson (2000) points out, the high co-morbidity of OC phenomenology in TS is remarkable,

regardless of whether this distinction is made. In a Japanese study, Kano, Ohta & Nagai (1998) found a co-morbidity rate of 62.5% in a clinical population of people with TS with Obsessive Compulsive Symptoms (OCS) and Comings & Comings (1987*d*) found rates of between 27.9% and 68.9% with OC symptom frequency increasing with tic severity. Coffey & Park (1997) reported the prevalence of sub-threshold OCD to be 52% in their TS clinic population, with 25% meeting the full diagnostic criteria for OCD. In a survey of 431 children and adults with TS, Stefl (1984) found that almost three-quarters of respondents reported OC behaviours (OCB), often or sometimes. Assessed by standardised rating scales for obsessive and/or compulsive behaviours, Robertson & Trimble (1988) found that over one third of TS participants reported OC problems. Comings & Comings (1987*d*) conducted clinical interviews with 246 patients diagnosed with TS and compared results with non-TS controls, those with TS and ADHD and those with ADHD alone. They concluded that on overall obsessive-compulsive scores, participants with TS were significantly more likely than the general population to experience such symptoms and that they were more likely to experience multiple OC symptoms. More recent studies have made similar findings with both children and adults with TS (Robertson & Gourdie, 1990; Robertson, Channon, Baker, & Flynn, 1993; Carter, Pauls, Leckman & Cohen, 1994; Kadesjo & Gillberg, 2000). Nolan, Sverd, Gadow, Sprafkin & Ezor, (1996) found that in children and adolescents with TS, a total of 20% were rated above the cut-off for OC behaviours as rated on the Child Behaviour Checklist (Achenbach, 1991) and that those rated as having more severe tics were significantly more likely to experience OC symptoms.

Further studies have sought to determine whether people with TS are more likely to experience different sorts of obsessions and/or compulsions that might distinguish them from pure OCD types. Miguel, Baer, Coffey, Rauch, Savage, O'Sullivan, Phillips, Moretti, Leckman, & Jenike (1997), Swerdlow et. al. (1999) and Miguel et. al. (2001) all describe the phenomenology of co-morbid OCD with TS as having a different quality in that obsessions and compulsive behaviours are less reliant on cognitive processes, are less likely to result in autonomic anxiety and are more likely be accompanied by sensory phenomena. For example, people with TS plus OCD have been reported to experience more obsessions about violent images, fear of illness and other intrusive images or sounds than those with pure OCD (Swerdlow et. al., 1999). Compulsions not accompanied by cognitions and autonomic anxiety like touching, 'evening-up' and counting were also reported more frequently in those with TS and OCD than pure OCD. Those with pure OCD reported more obsessions and compulsions about contamination, cleanliness and checking that were also accompanied by cognitive rumination and autonomic anxiety (Miguel et. al., 1997; Swerdlow et. al., 1999). Furthermore, there is also an overlap between complex tics and compulsions in that complex tics often mimic compulsive behaviours and that 'mental tics' might intersect with intrusive and obsessional thoughts making them difficult to distinguish too (Miguel, 2001).

Even though there are methodological differences across studies, the fact that OCD occurs in both adults and children with TS at a markedly higher rate than has been found in the general populations, of at least the UK and US, and by control group comparison, suggests that TS may be mediated by an underlying factor other than the psychological consequence of TS itself. In addition, studies outlining the

phenomenological differences between TS with OCD and OCD alone lend further support for the hypothesis that OCD co-morbid with TS is more likely to be linked by genetic and organic factors, in terms of neurobiology, than by the psychological or psychosocial consequences of TS itself (Miguel et al., 2001; George, Trimble, Ring, Sallee & Robertson, 1993).

*Attention Deficit Hyperactivity Disorder*

For the purpose of this paper, Attention Deficit Disorder with or without hyperactivity will be referred to as ADHD as a distinction between these clinical presentations is beyond the scope of this review. ADHD has also been noted to be markedly co-morbid with TS (Freeman, 1997). Spencer, Biederman, Coffey, Geller, Faraone & Wilens (2001) discuss the very many studies that have investigated TS co-morbid with ADHD and report between 35% and 90% co-morbidity, with an average of around 52%. Conversely, Spencer et. al. (2001*b*) found a co-morbid rate of only 12% of children with ADHD identified as having a tic disorder, when studied retrospectively. However, this difference may reflect ascertainment bias due to those with dual diagnoses being more likely to be referred to a specialist treatment clinic.

It has become widely accepted that ADHD occurs in a large number of children diagnosed with TS (Spencer et. al., 2001; Robertson, 2000; Comings & Comings, 1988). Comings & Comings (1987*b*) suggested that ADHD might be genetically related to TS, although this hypothesis has been disputed by others (e.g. Pauls et. al., 1988, Eapen & Robertson, 1996). In reviewing the existing evidence Robertson (2000) suggests three further hypotheses. Firstly, that there may be two types of co-morbid ADHD in TS; one which is secondary to TS and one which is independent of

TS. Secondly, that ADHD represents a different disorder to TS with ADHD. Thirdly, that ADHD co-morbid with TS may represent a sub-threshold sub-type of attention problems, resulting from decreased concentration and attentional capacity necessitated by TS itself (Robertson, 2000; Freeman, 1997). This would mean that prevalence estimates of co-morbid ADHD with TS depend upon the rigor of diagnostic criteria used in individual studies (Budman & Feirman, 2001). These hypotheses have been further confounded by the notion that medication used to treat ADHD (often identified at a younger age than TS, APA, 2000; Budman & Feirman, 2001; Spencer et. al., 2001*b*), may precipitate TS in individuals who might not otherwise have experienced the disorder (Spencer et. al., 2001; Kurlan et. al., 2002). Furthermore, when ADHD follows the onset of TS, there may be a greater likelihood of a genetic relationship between the two (Kurlan et. al., 2002). This latter notion might be supported by Spencer et.al's (2001*b*) suggestion that TS and ADHD have higher rates of co-morbidity when investigated through those identified as having been previously diagnosed TS rather than ADHD.

As yet, even though there are complex cases to be presented for and against the genetic mediation of organicity in ADHD with TS, the precise nature of this relationship remains unclear (Kurlan et. al., 2002). It is clear though that over half of many clinic samples of children with TS have also been diagnosed with ADHD. However, whether neurobiologically based or not, it is likely that the experience of OCD or ADHD in addition to TS might result in further psychological difficulties for example anxiety, depression or behavioural problems.

**Further Findings of Co-morbidity in Tourette Syndrome**

Evidence that Tourette Syndrome may have other psychopathological associations that cannot be explained wholly by the presence of OCD, ADHD or by genetically mediated organic factors, is important in terms of clinical treatment implications. Findings from numerous studies have identified increased prevalence rates of other co-morbidly occurring difficulties in both children and adults with TS (e.g. Kulisevsky, Berthier, Avila, Gironell, & Escartin, 1998; Coffey & Park, 1997; King & Scahill, 2001). These have been assessed by numerous different methods including clinical interview, self/parent/teacher ratings, using a variety of measures for both children and adults. Whilst it is true that these may be related to an individual's experience of TS, their associated family or psychosocial circumstances, it has been suggested that they may also be associated with co-morbidly occurring OCD or ADHD symptomatology (Cohen, Ort, Leckman, Riddle & Hardin, 1988; Sukhodolsky, Scahill, Zhang, Peterson, King, Lombroso, Katsovich, Findley & Leckman, 2003). In order to attempt to disentangle some of these factors, it is necessary to briefly examine the literature on the occurrence of other psychological difficulties associated with TS, although it was not feasible to separate them in terms child or adult studies because of the way they are reported.

*Anxiety and Depression*

A recent epidemiologic study in the US estimated prevalence rates of 'any anxiety' disorder in the population for all ages over 18 years, to be 11.8%, with generalised anxiety disorder at 2.8% (ages 18-54), panic disorder at 1.4%, social phobia at 3.2% and 'any phobia' at 7.8% (Narrow et. al., 2002). The same study estimated 'any mood disorder' to be prevalent amongst 5.1% of the population at any age and major

depressive episode in 4.5% of the population. In the UK, rates of neurotic and mood disorders were measured on a national scale by clinical interview. Generalised anxiety disorder was found to be prevalent among 3.4% of those surveyed, with all phobias, panic disorder and OCD prevalent amongst less than 2% each of those surveyed and a depressive episode was reported in 2.1% (Jenkins et. al., 2003). Briggs-Gowan, Horowitz, Schwab-Stone, Leventhal & Leaf, (2000) found rates of 0.7% of major depression or dysthymia, 0.5% of overanxious disorder and 6.1% for any anxiety disorder in a paediatric sample.

Anxiety and depression have been widely reported to co-exist in both children and adults with Tourette Syndrome and there is little dispute that this is the case (Robertson, 2000; Kulisevsky et. al., 1998; Coffey & Park, 1997; King & Scahill, 2001; Comings & Comings, 1987*c*; Comings & Comings, 1987*e*). In a survey of 431 members of a US TS Association, of both children and adults, a rating of "often" was given for extreme anxiety and mood swings in 31.6% and 32.7% of respondents respectively (Stefl, 1984). Further analysis of her data yielded no significant differences between respondents when divided into four age categories. This suggests that people with TS experience extreme anxiety or depression, regardless of their age (Robertson et. al., 1993). Robertson & Gourdie (1990) however, did not find increased incidence of psychopathologies including depression and anxiety, in a study examining one large family pedigree, compared to that which might be expected in the general population, or when comparing 'cases' of TS with 'non-cases'. However, they did find an increase in OCB and self-injurious behaviour. This study might be criticised though, because the authors failed to use well-defined or standard measures in determining the presence of TS or probable TS, using a short undefined interview

and it is unlikely that a sample of a single family pedigree would yield generalisable results.

In a large scale (n=1596) community based study to identify tics and psychopathology in school children, using standardised clinical interviews and rating scales, Kurlan et. al. (2002) found a range of between 8.9% and 29% frequency for different anxiety disorders in children identified as having tics. Furthermore, all of these were at a significantly higher frequency than in children not identified as having tics. Kurlan et al., (2002) found major depression and dysthymia to be less common, occurring in between 1.2% and 9.7% of those identified, not significantly different from those with 'no tics'. Again, this may be due their categories of 'tics' and 'no tics' rather than following diagnostic guidelines as set out by DSM-IV-TR (APA, 2000) or ICD-10 (WHO, 1992). However, the markedly high rate of anxiety encountered suggests that this criticism may only account for part of these findings and that anxiety at least, is more likely to occur in individuals with tics.

Although investigating a relatively small sample size of children with TS (n=47), Nolan et. al. (1996) reported 32% receiving an additional diagnosis of overanxious disorder, with 21% also receiving a diagnosis of simple phobia and/or separation anxiety, based on parent and teacher reports. In addition, 29% received a diagnosis of dysthymic disorder or major depression. They also found that when groups were divided by tic severity, those with more severe tics were significantly more likely to experience anxiety problems although this relationship was only noted as a tendency for dysthymia. Coffey, Biederman, Smoller, Geller, Sarin, Schwartz & Kim (2000*a*) also found that non-OCD anxiety disorders were highly associated with tic severity,

as did Comings & Comings (1987*c*) with phobias and panic attacks. Other researchers however, have failed to find a relationship between tic severity and psychopathology, although their findings support the high incidence of anxiety and mood problems in TS (Dian, Stoddard, Cates, Hartman, Bobenrieth, Hill, & Hill, 1995; Carter et. al., 2000; Sukholodsky et. al., 2003). Given the inconclusive evidence of there being a direct relationship between anxiety and depression and tic severity, this suggests that an individual's appraisal or perception of their tic disorder and its impact, is at least as important in the development of anxiety and possibly depression, as are the severity of the tics themselves.

According to Coffey et. al. (2000*b*), neither OCD nor ADHD were major predictors of psychiatric hospitalisation in children and adolescents with TS. Furthermore, Nolan et. al. (1996) found no significant difference between any co-morbid psychopathology and a diagnosis of ADHD, indicating that additional difficulties associated with ADHD could not account for these differences in children with TS. However, a number of authors have found the opposite, that children with TS and ADHD are more likely to exhibit behavioural difficulties and have problems with social adaptation and competence (e.g. Spencer, Beiderman, Harding, O'Donnell, Wilens, Faraone, Coffey, Geller, 1998; Stephens & Sandor, 1999; Carter et. al., 2000; Sukhodolsky, 2003).

In children identified as having a first-degree relative with TS who had not been previously diagnosed with TS, Carter et. al. (1994) found an increased incidence of tics and TS. Amongst those identified, 24% were diagnosed as having additional anxiety disorders other than OCD, with 4% having a major depressive disorder. It

appears that many studies have not controlled for OCD in TS, particularly with other psychopathologies as ADHD has been. This might be an important area for further research, given the marked prevalence of OCD in TS.

Further hypotheses have been presented that anxiety other than OCD might also be genetically and organically linked due to the implication of the basal ganglia and arousal systems in TS (Cummings & Frankel, 1985; King & Scahill, 2001; Robertson, Trimble & Lees, 1988). However these findings have been controversial and inconclusive and it has also been suggested that adverse environmental influences are likely to increase psychopathology (Edell & Motta, 1989; Silva, Munoz, Barickman, & Friedoff, 1995).

*Other Psychopathology in Tourette Syndrome*

A smaller number of studies have investigated the prevalence of other psychiatric and psychological disorders in people with TS. Comings & Comings (1987*d*) reported a significantly higher rate of schizoid symptoms including auditory hallucinations and paranoid ideation in a sample of 246 patients with TS than was found in a control group (auditory hallucinations; 14.6% versus 2.1% respectively). In addition, the rate of schizoid behaviours was seen to increase significantly with tic severity. There have been a few anecdotal reports of psychotic phenomena occurring in people with TS but as yet these remain inconclusive (Berthier, Kulisevsky & Campos, 1998).

A small number of studies have also investigated the incidence of bipolar disorder in people with TS (Berthier, Kulisevsky & Campos, 1998). Comings & Comings (1987*e*) found a significant difference between the numbers of TS and control

participants reporting mania (19.1% versus 0%). In examining the factors associated with illness morbidity in young people with TS, Coffey et. al. (2000*b*) found that 12% of 156 children and adolescents with TS required psychiatric hospitalisation and that along with major depression, bipolar disorder was the most robust predictor of that hospitalisation. Kerbeshian, Burd & Klug (1995) also found in a sample of 205 patients with TS that there was a prevalence of bipolar disorder at a rate above four times that of chance, although this was not found to be statistically significant. Again, because little empirical evidence exists, it is difficult to draw any conclusions about a possible association between TS and bipolar disorder.

One clinic study by Robertson, Banjeree, Fox-Hiley & Tannock (1997) has investigated the association between personality disorder and TS. Significantly more TS patients were found to meet the criteria for a personality disorder than a control group (64% and 6% respectively). The highest was for borderline personality disorders at 53% of those identified with depressive, obsessive-compulsive, paranoid and passive aggressive personality disorders, all second highest at 23%. There are however, methodological questions to be raised about this study, with relation to the relatively small sample, ascertainment bias and the particular control group employed.

*Summary*

There appears to be little doubt that a number of psychopathologies exist in conjunction with TS and that there is evidence to suggest OCD and ADHD are more likely to be related in terms of genetic and neurobiological factors. It seems clear that children and adults with TS experience a range of anxiety disorders at a markedly increased frequency than the general population or when compared to non-TS

controls, especially when sampled through clinic populations. This appears to sometimes be true even when co-morbid diagnoses of OCD or ADHD are controlled for. Rates of depression in people with TS appear to be elevated in comparison to the general population. There also remains debate about whether tic severity and anxiety and depression are related. It has been suggested that this relationship might be best understood in terms of the individual's perception of the impact of their TS. Findings from studies investigating other psychopathologies are inconclusive due to the small number of research studies. However, it is noted that the one study examining personality disorder found such a remarkable occurrence of this in adults with TS that this issue would warrant further research.

**Empirical Studies Examining the Impact of Tourette Syndrome in Children**

Many authors have hypothesised about psychological processes that might underlie co-morbid difficulties associated with TS. However, few have sought to empirically investigate these process mechanisms. A series of studies have examined other variables in children diagnosed with TS. These have investigated peer relationships, self-esteem, self-concept, adjustment, family function and childhood behavioural difficulties.

*Peer relationships*

Stokes, Bawden, Camfield, Backman & Dooley (1991) attempted to quantify peer relationship problems that might be encountered in children with TS. Twenty-nine children with a mean age of 11.4, with mild to moderate TS were selected from a clinic sample. They completed a series of self-esteem scales, whilst their parents and teachers completed a series of behaviour rating scales. Classmates of the selected

children completed the Pupil Evaluation Inventory, which gives overall measures of aggression, likeability and withdrawal. Although the self-reported self-esteem of children with TS fell within the average range and through these self-reports children were not deemed to have either internal or external locus of control, they were reported by their parents as being over or close to the cut-off on externalising behaviour, internalising behaviour, aggressive, hyperactive, somatic complaints and on OC factors. So there was found to be some discrepancy between the child with TS report of their difficulties and that of their parents and teachers. Compared with their same sex classmates, children with TS were rated as being significantly more withdrawn, aggressive and less popular. Thirty five percent of the children with TS obtained the poorest rating in their class on one or more of the three factors of withdrawal, aggression or likeability. Peer ratings of aggression were correlated with teacher ratings of school performance and total social competence. Peer ratings of withdrawal were also correlated with teacher ratings of internalising and on the unpopular factor. Finally, peer ratings of likeability were correlated with teacher ratings of school performance and total social competence. These correlations may represent cross-validation of the children's (peer) responses, strengthening these findings. Teachers rated the children with TS as more withdrawn and aggressive but did not rate them significantly differently on measures of likeability. A further finding was that children with TS and additional psychiatric diagnoses, particularly ADHD, obtained ratings that were as poor as those without additional diagnoses. This study also highlights a possible discrepancy between children's self-esteem and negative perceptions of them by their peers.

In an attempt to replicate the above findings, Bawden, Stokes, Camfield, & Salisbury, (1998) conducted a similar study but compared their sample of 25 younger children with TS (mild to moderate severity) with 25 children with another chronic condition, diabetes mellitus (mean ages 5.5 and 5.3, respectively). Again they found that children with TS were rated as significantly more aggressive, withdrawn and less likeable than their classmates, although when those with ADHD were excluded, the aggression factor became non-significant. Although children with TS were rated more poorly on all three scales than children with diabetes mellitus, this was not statistically significant. However, children with TS received the worst scores in their class on one or more of the aggression, withdrawal or likeability factors, significantly more often than children with diabetes mellitus. However, an important point to note in Bawden et. al.'s (1998) study is the relatively young age of the children. Given that the mean age of onset for TS is 5-7 years old, it may be that any indirect psychosocial effects of TS had not been apparent at the time of the study.

Another study was conducted by Friedrich, Morgan & Devine (1996) to assess the efficacy of providing educational information about TS to classmates of a proposed TS child (an actor), in terms of peer ratings of attitudes and behavioural intentions toward that child. Whilst it was found that on a measure of attitude, children rated this supposed peer with TS less positively, additional information about the nature of TS did not affect these ratings. However, on a measure of behavioural intention (e.g. preferred behaviour with the TS child on items like 'watch TV together; invite to my house'), no difference was seen between the no TS, TS and TS with information conditions. In an attempt to account for bias in terms of responses being more 'socially desirable', the children also completed a further questionnaire rating the

supposed behavioural intention of their classmates. Friedrich, Morgan & Devine (1996) further maintained that these scores were indeed answers perceived to be more 'socially desirable' and that these more negatively rated scores might more accurately reflect the children's truer, even less positive intentions toward a peer with TS. Although employing a sophisticated design, the authors criticise their work for making use of questionnaires versus actual behavioural measurements, the use of the same child actor in each condition and the fairly limited and short video footage the children were shown. In addition, Friedrich, Morgan & Devine (1996) criticise the experimental nature of their study as decreasing the "realness" of the situation for the children. Nevertheless, these findings substantiate those of Stokes et. al. (1991) and Bawden et. al. (1998) in terms of negative peer perceptions of children with TS. Furthermore, they also suggest that children without TS have more negative behavioural intentions towards children with TS than they perceive to be 'socially desirable'. One way to overcome the design difficulties here might be to conduct an observational study in a naturalistic setting.

*Family Functioning, Self Concept and Adjustment*

Another series of studies have attempted to clarify the relationship between family functioning, self-concept and adjustment in children with TS.

Dian et al. (1995) designed a study to predict self-concept in children with TS on the basis of their reported symptom frequency. Thirty-five children with TS, with a mean age of 12.5 years completed the Piers-Harris Children's Self-Concept Scale and a TS symptom frequency measure. They found that the most robust predictor of global self-concept was that of internalised symptoms, followed by simple motor and simple

phonic tics. Internalised symptoms were composed of a cluster of somatic symptoms such as severe headaches, parasomnias and dyssomnias. Whilst Dian et al. (1995) point out that the internal consistency between these factors was low, undermining the validity of their findings, they conclude that perhaps it is not tic severity that is important in terms of detriment to self-concept, but rather additional somatic complaints which may not routinely be assessed in clinic. A further suggestion might be that these somatic complaints may represent secondary symptoms as a result of internalised anxiety. However, Frank, Sieg & Gaffney (1991) maintain that their findings of significantly increased somatic complaints over a control group, in children with TS, were comparable to the frequency of reported unexplained physical symptoms in children with other psychiatric diagnoses.

Edell & Motta (1989) investigated factors that might affect the emotional adjustment of children with TS. Taking a systemic perspective of chronic illness within the family, they hypothesised that mother's self-concept, child perceptions of parental behaviour, family adaptability and severity of TS would be predictors of emotional adjustment particularly with self-concept and anxiety. Edell & Motta (1989) included 60 children with a mean age of 10.9 years from a clinic population. Overall, of the four variables tested, only the child's perception of parental behaviour significantly correlated with child self-concept and anxiety. Edell & Motta (1989) explain these findings in relation to literature regarding child perceptions of parental behaviour, in particular psychological control over the child, supported by their finding that children's perceptions of their parents' behaviour as controlling was a significant predictor of trait anxiety in these children. Edell & Motta (1989) comment on the finding that parental self-concept did not explain a significant amount of the variance

to account for their children's self-concept, by conceding that this was not consistent with other findings at the time.

In an attempt to replicate these findings, Edell-Fisher & Motta (1990) compared the self-concepts of 30 children with TS and their mother's with 30 control participants and their mother's. They found that the self-concept of children with TS did not differ from that of children without TS. A further finding was that severity of TS was positively correlated with the child's own self-report of behaviour disturbance and dysphoric mood. Furthermore, the mothers of children with TS were found to have lower self-concepts than mothers of children without TS.

In another clinic study, Dykens et. al. (1990) investigated the intellectual, academic and adaptive functioning of children with TS, with and without ADHD. They found that socialisation skills emerged as a relative weakness in adaptive functioning and that this was true regardless of ADHD status. In particular, this item related to interpersonal relationships, use of play and leisure time and coping skills. Dykens et. al. concluded that all of their sample of children with TS were at a level of social functioning considerably below that expected for their chronological age. These children did show significant strengths in domestic skills and personal daily living skills, suggesting a higher level of self-sufficiency than social skills. These results indicate that children with TS do experience difficulty with social skills and this may be related to peer relationship difficulties.

In a Canadian study, 210 children and adults with TS (70% were children) were surveyed and 42% of respondents reported experiencing difficulty forming and

maintaining friendships in childhood and later in adult life (Champion, Fulton & Shady, 1988).

*Childhood Behavioural Difficulties*

A recent sophisticated study (Carter et. al., 2000) found that children with TS and ADHD were significantly more likely to experience externalising behaviour problems and have problems with social adaptation than children with TS only or unaffected controls. However, children with TS were found to exhibit more internalising symptoms than unaffected children, being reported by parents as more anxious / depressed, as having more somatic complaints and as being more withdrawn than control children. Carter et. al. (2000) also noted that, with the exception of withdrawal, children with TS and ADHD were significantly more likely to experience these problems than both control children and children with TS alone. Carter et. al. (2000) go on to suggest a possible continuum of risk of further problems for children with TS, increasing with ADHD as a co-morbid factor. Furthermore, they did not find significant associations between tic severity or attention regulation and social, emotional and behavioural functioning as other researchers have (e.g. Nolan et. al., 1996). Carter et al. (2000) did however conclude that lower family functioning was associated with poorer social and emotional adjustment, even after controlling for ADHD status.

Another study set out to further investigate the hypothesis that children with TS and co-morbid ADHD are likely to have poorer behavioural outcomes than those without ADHD. Sukhodolsky et. al. (2003) examined disruptive behaviour along with social adaptive and family functioning in 207 children with diagnoses of TS and ADHD, TS

without ADHD, ADHD alone and an unaffected control sample. They used standardised parent and teacher rating scales and a measure of tic severity. Whilst there was found to be no significant difference on the tic severity measures for both TS groups, the authors also included an OCD diagnosis in their analysis and found no significant differences on any of their dependent variables for any of the participants with TS, regardless of ADHD status. In terms of disruptive behaviour, children with TS only did not differ significantly on measures of aggression, delinquency or conduct from children in the control group. However, the children with TS and ADHD obtained significantly higher scores than control children but they were not significantly different to children with ADHD alone on the same measures. All children with TS, TS and ADHD and ADHD alone were found to differ significantly on measures of social competence and daily living skills, from unaffected children. On measures of family functioning, the group of children with TS and ADHD were found to have significantly poorer family function in terms of conflict and cohesion, than TS alone, ADHD alone and unaffected children. This suggests a continuum of difficulties associated with TS and TS with co-morbidity, increasing with ADHD. Indeed, Spencer et. al. (1998) found that whilst tic severity was not associated with additional psychological or psychiatric difficulties, severity of ADHD was found to be significantly associated with these variables. They concluded that ADHD might be more related to other co-morbid psychopathologies in TS, than TS in its own right. However, there are criticisms to be levelled at this study in terms of ascertainment bias and small number of participants with TS only (Spencer et. al., 1998). Sukhodolsky et. al. (2003) also concluded that tic severity could not be firmly associated with any of these factors.

Further studies have also noted the increased rate of behavioural difficulties in children with TS, as reported by teachers and parents, increasing further with additional diagnoses of ADHD and OCD (Kadesjo & Gillberg, 2000; Stephens & Sandor, 1999). Abwender, Como, Kurlan, Parry, Fett, Plumb, & Deeley (1996) assessed the effect of co-morbidity, tic severity and social and / or family problems, in TS on academic difficulties in children without learning disabilities. They found that ADHD was the only significant predictor of school problems other than learning disabilities in children with TS.

It is perhaps not surprising given the likely internal and external distractibility posed by a disorder such as TS, that there is seen to be something of a continuum of associated behavioural difficulties with attentional problems. However, in terms of treatment implications, it is important to consider whether a child has attentional difficulties resulting from their experience of TS that if untreated may result in conduct problems or oppositional defiance escalating because of unhelpful inconsistent, reciprocal or rejecting behaviour of others. Or, whether the child has a truer form of ADHD with TS, likely to require medical treatment, in order to minimise the impact of the associated behavioural difficulties on childhood development. The former being optimally treated with psycho-education and behavioural management techniques (Overmeyer & Taylor, 1999).

*Summary*

Studies investigating peer relationships in children with TS, highlight a number of possible factors that may effect the psychological functioning of these children. Self-reported self-esteem and self-concept have been reported to be equivalent in children

who have TS to children without TS of the same age. This is whilst being frequently rated as the most or one of the most unpopular children in the class by their peers, in addition to negative perceptions of them overall. This reflects something of a discrepancy between children's self-esteem and negative perceptions of them by their peers. In addition, educational intervention to children about the nature of TS did not change their supposed behavioural intentions toward that child nor their negative attitudes towards them.

When mothers of children with TS were compared to mothers of children without TS, they were found to have lower levels of self-concept. However, this was not found to be related to the self-concept of their children with TS. Internalised symptoms were found however, to be a predictor of global self-concept in TS children. The TS child's perception of parenting behaviour has been found to be related to the child's self-concept.

It appears that children with TS alone are more likely to exhibit internalising problems, whether these are via somatic complaints or by social withdrawal, anxiety or depression. It also appears that this may lead to difficulties with social competence and possibly in the formation of good interpersonal relationships with peers. Children with co-morbid TS and ADHD appear to exhibit more externalising difficulties and again experience problems in the area of social competence and also with forming interpersonal relationships with peers. There appears to be little conclusive evidence of the effect of co-morbid OCD in these children with TS and it appears that this is an important area of undeveloped understanding, given the high rates of OCD found in children with TS. Furthermore, at present, there appears to have been no empirical

research on the changing nature of self-concept, adjustment, self-efficacy and personality development from early childhood years, through the years of adolescence in those with TS. If the psychological effects of TS in childhood and in later adulthood are to be better understood, this is an important gap in the existing literature.

**Empirical Studies Investigating the Impact of Tourette Syndrome in Adults**

There appears to be an increased risk of the mothers of children with TS experiencing lower self-concepts, and children with TS alone and TS co-morbid with ADHD and or OCD having more family dysfunction, and adjustment and social skills difficulties in addition to peer relationship problems and increased rate of behavioural difficulties than children without TS. If this is so, how might this affect development into adulthood? It can be seen from studies discussed earlier that the numbers of adults with TS and co-occurring psychopathologies are very much higher than would be found in the general population. A small number of empirical studies have attempted to explain why this should be so for an adult population with TS. These studies have also attempted to understand to some extent, the experience of the world as an adult with TS. The following section describes the few studies investigating the impact of TS in adults.

O'Conner, Gareau & Blowers (1994) attempted to elicit personal constructs associated with tics and different risk situations for tic-ing. These participants all met the criteria for chronic motor tics. Participants were required to keep a diary of their tics and then with the researchers, identify high-, medium- and low- risk tic situations. Low risk situations tended to be passive states, habitual automatic actions and

interactions with familiar people. Medium risk situations tended to be ones that required more effort and were unexpected or were difficult or demanding situations. High-risk situations were those that involved working under pressure, multi-tasking, being over-stimulated and waiting or worrying. Using a repertory grid analysis O'Connor, Gareau & Blowers (1994) elicited personal constructs relating to the different risk situations. These could be grouped into three principle categories. The first was relating to self-image e.g. fear of judgement versus feeling valued / fear of rejection versus reassurance; the second, to degree of involvement in a given task e.g. occupied versus losing or wasting time; and finally the type of task demand e.g. rushed versus relaxed or complex versus routine. All of these categories of constructs could be actual or anticipated. The most consistently reported emotion experienced in high-risk tic situations was that of frustration or impatience rather than anxiety, although anxiety was related to cognitive appraisal of anticipated situations where tic-ing might be a concern.

Even though this study stands alone and employed a small sample of people with chronic tic disorder rather than TS, it is perhaps important to note that, from three main constructs derived, one was centred around a fear of being judged and rejected.

Elstner et. al. (2001) rated the quality of life (QOL) of 90 adults with TS identified through clinic populations in the UK and compared them with normative data and with data obtained from sufferers of intractable epilepsy. The QOLAS (Kendrick & Trimble, 1994) is a qualitative measure using the repertory grid technique, which allows the participant to nominate their own items of importance in terms of quality of life and then rate them. A medical outcomes questionnaire consisting of eight

domains including physical and social functioning, role limitation, emotional problems and mental health was also employed. Compared with a previously assessed sample of patients with intractable epilepsy on the QOLAS, people with TS scored significantly better on factors relating to physical, psychological, social and cognitive functioning and daily activities but scored equally poorly in other domains. Although no normative data is yet available for this measure, the authors maintain that the TS participants reported relatively higher percentages of difficulties in each of the domains than might be expected from a normal population. Seventy-eight percent of participants reported motor tics to be problematic in the physical domain. In the psychological domain, 51% reported depression, 31% reported anger / aggression and 29% reported anxiety as problematic. In the social domain 29% reported problems with family, 27% and 20% reported problems with making friends and with their social lives respectively. A further 15% reported self-consciousness as a problem including feelings of embarrassment and social stigma due to their TS. With respect to cognitive problems participants reported problems with concentration, memory and having a short attention span. Finally, a further 48% and 19% reported interference with work or study or said that TS meant that they were unable to work / study or that they had been made redundant. On the medical outcomes measure, TS participants obtained scores significantly different from controls on all but one domain, measuring vitality. In addition, they also obtained scores that were not significantly different from those from the intractable epilepsy clinical population, except on measures of role limitation due to physical problems and social functioning. Elstner et. al. (2001) concluded that that QOL in patients with TS is impaired.

There do not appear to be any other studies specifically assessing quality of life in people with TS, which is surprising given the chronic nature of TS and also given the extensive research in this area on other chronic conditions. Champion, Fulton & Shady (1988) found that 42% of their TS respondents conveyed problems with forming and maintaining friendships and almost half of their adult respondents reported difficulties with dating. However, one study has addressed the effect of TS on a number of employment variables. Shady, Broder, Staley, Furer, & Papadopolous (1995) obtained information from 167 adults with TS indicating that almost half felt their TS had influenced their choice of job. Over 20% claimed to have been dismissed or fired from a job because of their TS and 16.8% reported having been denied a job for the same reason, with a further 11.8% believing they had not received promotion due to their TS. Further analysis of difficulties associated with TS, were factored down to four domains; problems with hyperarousal, sleep problems, childhood behavioural problems and vocalisations. These factors were then regressed on to four employment variables. Few problems relating to hyperarousal and few childhood behavioural problems were found to be the greatest predictors of job satisfaction. In addition, current interpersonal and behavioural problems as well as hyperarousal and sleep difficulties were the most important predictors of the perceived impact of TS on job choice. Shady et. al. (1995) also maintain that their results reflect the possibility that adults with TS may be 'underemployed', given their educational attainment.

It is interesting to note that the enormous self-reported rate of mood, aggression / anger, anxiety, social and interpersonal problems are comparable if not higher than clinically diagnosed rates of psychopathology. However, an important area frequently

omitted from clinical prevalence studies is that of interpersonal and social difficulties. In order to understand the difficulties that people with TS may experience, even though the intensity of these difficulties might be sub-threshold diagnoses, this seems to be another important area for further research.

In a self-selected population of adults with TS, Thibert, Day & Sandor (1995) surveyed self-concept and self-consciousness. Dividing their sample by self-reported high or low OCD symptoms, Thibert, Day & Sandor maintained that those with TS without OCD obtained scores comparable to normative data. Those with co-morbid OCD were reported to have impaired self-concept, higher levels of self-consciousness and social anxiety. Even so, the authors comment on qualitative data obtained from participants detailing current feelings of embarrassment, being teased or even ostracised when they were children, resulting in low self-esteem and feeling disadvantaged in a number of social and academic spheres including career aspirations. The empirical findings of this study seem to be at odds with those of Elstner et. al. (2001) regarding quality of life in adults with TS. One explanation for this might be the methodological issues encountered in Thibert, Day & Sandor's (1995) study in that a self-selected sample may be biased and a postal questionnaire means diagnostic certainty is difficult or that Elster et. al. (2001) failed to account for co-morbidity. Furthermore, the psychometric properties and theoretical constructs underlying the Tennessee Self-Concept Scale used in Thilbert, Day & Sandor's study have been questioned (Bishop, Walling & Walker, 1997).

In their survey of Canadian people with TS, Champion, Fulton & Shady (1988) reported that over a third of 210 respondents experienced moderate to significant

problems in coping and a further 47% reported some mild difficulties with coping. In a study to investigate the relationship between coping strategies and styles in adults with TS and the occurrence of anxiety and depression, Hickey & Wilson (2000) found that coping style was relatively independent of tic severity although anxiety, depression and stress were negatively correlated with planful problem solving (a form of problem-focused coping). Furthermore, those with TS who were anxious or depressed tended to use at confrontative style of coping.

Overall, it has been highlighted that adults with TS experience a relatively poor quality of life compared with a normal control population and they may be compared in some respects to adults suffering from intractable epilepsy (Elstner et. al., 2001). They have also reported difficulties in a number of life domains, for example, in interpersonal relationships, maintaining and forging friendships as well as study and employment factors (Shady et. al., 1995). However, one study has reported that adults with TS were comparable to those without TS on measures of self-concept and self-consciousness, although co-morbidity with OCD was associated with increased difficulties in these areas (Thilbert, Day & Sandor, 1995). Furthermore, constructs relating to feeling judged or rejected by others have been elicited by adults with TS with relation to their tics (O'Connor, Gareau & Blowers, 1994). Moreover, the coping styles of adults with TS who experience co-morbid anxiety and depression have been associated more with emotion-focused rather than problem-focused approaches (Hickey & Wilson, 2000).

**Summary of Findings From Studies Investigating the Impact of TS**

Findings from studies that have highlighted difficulties associated with TS in both children and adults that may be beyond neurobiological vulnerability, have been outlined above. Unfortunately, relatively few studies have been undertaken in this area and not many of those have been reliably replicated. In addition, methodological and design variation, and differences with operational definitions, make it hard to draw firm conclusions from the existing evidence. As such, a relative dearth of empirical psychological study has been highlighted, in comparison to the vast body of literature investigating the neurobiological, psychiatric, genetic and pharmacological treatment aspects of TS. Nevertheless, clinical researchers have acknowledged their observations of psychological distress and psychosocial difficulties in people with TS in their experience as clinicians and researchers time and again (e.g. Robertson & Trimble, 1988; Trimble, 1989; Carter et. al., 1994; King & Scahill, 2001).

It seems clear from the literature that children and adults with TS experience a range of anxiety disorders at a markedly increased frequency than the general population or when compared to non-TS controls, especially when sampled through clinic populations (e.g. Kurlan, 2000). This appears to sometimes hold true even when co-morbid diagnoses of OCD or ADHD are controlled (Nolan et. al., 1996).

The findings relating to mood disorders in TS seem less consistent than for anxiety disorders. This may be because the diagnostic categories used by some studies to distinguish dysthymia from a depressive episode or major depression, are unclear (e.g. Kurlan, 2002; Nolan et. al., 1996). They may reflect the characteristics of particular samples studied, or they may reflect the lesser likelihood of depressive disorders in

people with TS. Depression has been hypothesised by Coffey et. al. (2000*b*) to indicate a syndromic co-morbidity of major depression and bipolar disorder with TS rather than a reactive one, although the evidence for this is inconclusive. Nevertheless, rates of depression in children and adults with TS appear to be elevated compared with the general population and have even been compared to similar rates found in general psychiatric populations (e.g. Frank, Sieg & Gaffney, 1991). There also remains debate about whether tic severity and anxiety and depression are associated. It has been suggested that this relationship might be best understood in terms of the individual's perception of the impact of their TS (regardless of severity) rather than the actual measure of severity itself (Rosenberg, Brown & Singer, 1995).

Findings from studies investigating other psychopathologies are inconclusive due to the small number of empirical research studies published. However, it is noted that the one study examining personality disorder (Robertson et. al., 1997) found a remarkable occurrence of this in adults with TS. Personality disorder in TS is an important issue that would warrant further research.

There appears to be little doubt that a number of psychopathologies exist in conjunction with Tourette Syndrome and that there is evidence to suggest OCD and ADHD are more likely to be related in terms of genetic and neurobiological factors. There is little empirical evidence regarding the additional consequences of OCD in TS, however it might be reasonably assumed that obsessional thoughts and compulsive behaviours might be disruptive to the child and in the wider social sphere. ADHD is a disruptive and often socially disabling disorder. Whether ADHD with TS represents a sub-threshold form of the disorder or not, there is evidence that the

associated behavioural consequences may have greater implications for the child with respect to development of social skills and competence (e.g. Carter et. al., 2000; Sukhodolsky et. al., 2003). Ascertainment bias is important to remember because many of these findings relate specifically to clinical populations and may not be generalisable to a TS population who never seek neuropsychiatric treatment. Nevertheless, this does not detract from the findings that there are high rates of co-morbidity in TS clinic populations in whom a psychological approach to treatment could be beneficial.

Many authors have contemplated psychological processes that might underlie other co-morbid difficulties associated with TS, although relatively few studies have sought to empirically investigate the possible process mechanisms involved. This is especially important, given that anxiety, depression and personality disorder in particular, are frequently related to psychosocial development, learned behaviour, and the use of unhelpful coping strategies in reaction to adversity, often in early life (see Rutter & Maughan, 1997). There appears to be no longitudinal research to date that can determine whether the children who experience co-morbid problems with TS in childhood are the same people who experience these difficulties in adulthood. Research in this area might allow treatment programmes to be more effectively targeted across the lifespan.

Other factors relate to parents; that of parental self-concept, how parents adjust to the diagnosis of a disorder like TS in their children and in turn how their relationship with their child may change because of this. The strength of the parent-child relationship and the child's perception of this change in relationship may be important protective

factors, buffering against the development of psychological distress. Studies investigating peer relationships in children with TS, highlight a number of possible factors that may affect the psychological functioning of these children. Self-reported self-esteem and self-concept have been reported to be equivalent in children who have TS to children without TS of the same age. This is whilst being frequently rated as the most or one of the most unpopular children in the class by their peers, in addition to negative perceptions of them overall. This reflects something of a discrepancy between children's self-esteem and negative perceptions of them by their peers. It might also be supposed that peer rejection could be a consequence of negative peer perceptions of children with TS, which can have lasting psychological impact (see Rutter & Maughan, 1997).

Childhood behavioural difficulties associated with TS have been found in the domains of both externalising (e.g. aggression) and internalising problems (e.g. withdrawal) (e.g. Carter et. al., 2000). In addition, it has been consistently found that children with TS have a lower level of social competence than their peers and which might be predicted for their chronological age. These difficulties have also been related to co-morbidity with ADHD and less so with OCD, with a continuum of behavioural difficulties occurring, increasing with severity of the co-morbid disorders (e.g. Sukhodolsky et al., 2003). Additional difficulties associated with childhood ADHD and OCD without TS are also documented in the literature (e.g. Woodward, Dowdney & Taylor, 1997; see Samuels & Nestadt, 1997).

Adults with TS have reported a relatively poor quality of life compared with a normal control population (see Elstner et. al., 2001) and have reported difficulties in a number

of life domains, for example, in interpersonal relationships, maintaining and forging friendships as well as study and employment factors (see Shady et. al., 1995). Even so, one study has reported that adults with TS were comparable to those without TS on measures of self-concept and self-consciousness, although co-morbidity with OCD was associated with increased difficulties in these areas (Thilbert, Day & Sandor, 1995). In addition, constructs relating to feeling judged or rejected by others have been elicited by adults with TS with relation to their tics (O'Connor, Gareau & Blowers, 1994). Furthermore, the coping styles of those with co-morbid anxiety and depression have been associated more with emotion-focused rather than problem-focused approaches (Hickey & Wilson, 2000).

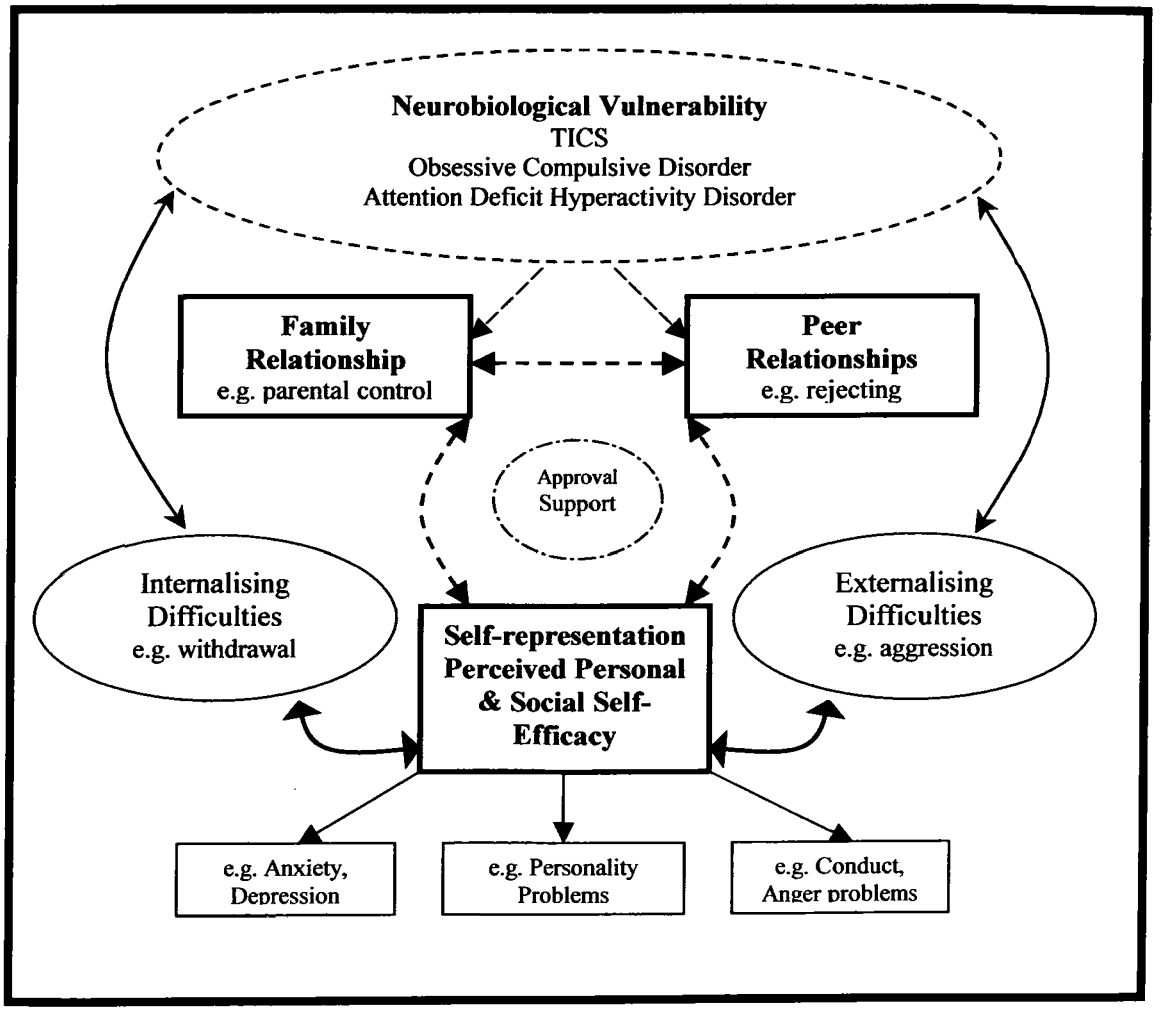
**Psychological Framework for Psychopathology in TS**

The following section intends to amalgamate the preceding evidence in order to suggest a psychological framework, which may provide additional insight for professionals working with people who have TS, presenting to neuropsychiatric clinics with more complex presentations. In particular, psychosocial relationships in childhood will be focused upon. Due to existing evidence being limited, the purpose of the following section is to provide a possible psychological framework for some of the findings that have been made as well as a hypothetical model (see **Figure 1**) illustrating how difficulties associated with TS in childhood may be linked by reciprocity. Research from other areas of childhood disability and chronic illness will also be drawn upon in order to support some of the findings from the TS literature. A number of broad and specific theoretical dimensions will be alluded to for example, Bandura's (1997, 1994) model of self-efficacy and Harter's (1999) model of parental

and peer relationships as pathways to childhood and adolescent psychological difficulties.

If it is accepted that an underlying neurobiological vulnerability exists with relation to ADHD and OCD in children with TS, then there may be other factors specifically relating to the experience of the disorder that may form the basis of psychological difficulties in these children, which may persist into adulthood. A number of observations have been made regarding the nature of tic severity and the experience of additional problems.

**Figure 1: Hypothetical Model of Psychological Factors in Children with TS**



Rosenberg, Brown & Singer (1995); Dian et. al. (1995); Carter et. al. (2000) and Sukhodolsky (2003) all found no relationship between tic severity and the occurrence of co-morbid anxiety and mood disorders, although Coffey et. al. (2000*a*) found a relationship between tic severity and non-OCD anxiety disorders. It is hypothesised in the model outlined in **Figure 1**, that perception of difficulties arising from tic symptoms and perception of the appraisal of these symptoms by others is more salient in the development of psychopathology, than the actual tic symptoms themselves. Thus, someone experiencing relatively mild tic symptomatology may suffer from severe social anxiety, whereas an individual with severe tic symptoms may experience little in the way of co-morbid problems. There is a wealth of literature suggesting that an individual's cognitive appraisal and perception of the impact of their disorder is more important than the actual severity and functional impairment resulting from the symptoms of the disorder itself. Thompson & Kent (2001) review the literature on adjustment to disfigurement in children with acquired and congenital conditions. They argue that in these children, there is an interaction between interpretation of their disfigurement, their own sense of self and their perceptions of encounters with others, which mediates this adjustment process. Although TS is different to the congenital, dermatological, and traumatically acquired disfigurements alluded to by Thompson & Kent, the perceived 'oddness' or 'bizarreness' and sometimes distracting nature of tic behaviours might be seen as similarly socially stigmatising and disabling. In discussing the impact of psychosocial adversities in childhood on the development of psychopathology, Rutter & Maughan (1997) also argue that it is the perceived effect of adversity rather than the nature of the adversity itself that is of primary importance.

The model presented in **Figure 1** maintains that a sense of self-representation and personal and social self-efficacy are learned through interactions with parents and peers. Instead of the term self-concept, Harter (1999) argues that 'self-representation' is a more accurate reflection of this construct. Self-representation is therefore defined as the description of the self i.e. 'what I am' (Harter, 1999). Self-efficacy has been defined as the belief in one's capability and capacity to perform within the context of a given environment (Bandura, 1994). Personal and social self-efficacy are argued to be mediated through the social modelling of parents and peers. The internalisation of these experiences, coupled with the opportunity to reciprocate in social interactions, allows a child to gain personal skills and social proficiency, resulting in a sense of mastery or competence (Bandura, 1997, 1994). Self-efficacy may be conceived of as one mechanism by which individuals appraise their own competence, i.e. 'how good I am' (Harter, 1999). Bandura (1994) also maintains that evidence suggests a low sense of self-efficacy may result in unfulfilled aspirations giving rise to withdrawal and resulting in the longer term in depression and anxiety.

Some studies of children with TS have found similar levels of self-concept (likened here to self-representation) in these children compared with their peers (e.g. Edell-Fisher & Motta, 1990). Even though the aforementioned study found reduced levels of self-concept in the mothers of these children compared with mothers of non-TS children, the current model proposes that self-representation may be developed in earlier years, through the attachment relationship with parents (see Bowlby (1969) for a detailed discussion of attachment relationships). The onset of TS is usually between the ages of 5-7 years old. With this in mind, it is hypothesised that a child's self-

representation might be well developed by the time of symptom onset, in the sense that it might be mediated by positive parenting experience prior to this. In particular, approval and support might be mechanisms through which this positive parenting is experienced. This factor may buffer children with TS who have a positive sense of self-representation at this stage against the adverse consequences of TS itself or the adverse effects of negative peer relationship experiences.

One explanation for the finding of mothers' reduced self-concept may be a reflection of the reaction to the onset of symptoms or diagnosis of TS in their children. However, it is postulated here that this self-representation is also open to alteration as children enter middle and late childhood (5-11 years). This has been broadly suggested by Bandura (1991*b*) to be when children's cognition becomes less influenced by the external control of significant others, more internalised and more related to the social environment, including an awareness of others' critical appraisal. Routledge, Mani, Pence & Hoskins (2001) conducted a qualitative longitudinal study of children, following them into early adulthood and reported findings in support of this shift from self-representation being primarily mediated by parental relationships in younger children, to becoming increasingly mediated by peer relationships in middle childhood and adolescence. This is also supported by the findings of Hay & Ashman (2003) who found that adolescents transfer their emotional attachment from parents to peers. Routledge et. al. (2001) argue that their results represent an interconnectedness of parental and peer relationships in the continuing development of self-representation. Hanson & Onikul-Ross (1990) also point out that as a result of this increased awareness, middle childhood is often the time when children begin to tease and bully others. Rigby (1999) found that peer victimisation was significantly

correlated with reduced physical and mental health in school children, in particular with internalising problems.

The current model suggests that the acquisition of approval from both parents and peers is important in the achievement of a strong sense of self-efficacy. Furthermore, if this support and approval is unavailable in either domain, then children will seek it from the other (Steinberg & Morris 2001). Routledge et. al. (2001) reported that when adolescents' relationships with parents were difficult, they turned to peers as a source of support and approval. This process was also observed in the opposite direction, that parents became a source of support and approval during times of difficult relationships with peers. An observation has been made in the literature regarding the occurrence of anxiety in children with TS and their perception of parents as 'controlling' (Edell & Motta, 1989). Whitebeck, Simons, Conger, Wickrama, Ackley, & Elder (1997) and Steinberg & Morris (2001) found that controlling and authoritarian parenting styles were linked to poor self-efficacy in children. Family adjustment and coping with chronic illness has also been reviewed by Rolland (1989). He argues that families coping with disorders with an episodic course, must remain flexible and that strain on the family is determined by coping with transitions between crisis and non-crisis. Crisis begins at the time of symptom onset and diagnosis. It includes a period of readjustment and the development of coping strategies and may be used differently depending on the family style (Rolland, 1989).

Harter & Whitesell (1996) and Harter, Waters & Whitesell (1998) propose a series of processes by which depression or adjustment difficulties might occur in adolescence.

They emphasise the importance of both parental and peer approval and support in the appraisal of their abilities and the development of subsequent depression. Harter et. al. (1996, 1998) propose that scholastic competence and behavioural conduct are factors which influence parental approval and support. They further propose that peer approval and support is determined in part by physical appearance, likeability and athletic competence. Even though this model was developed to explain depression and adjustment in adolescence, it appears to explain some of the findings from younger children with TS. Paralleling part of Harter's model, Bandura, Pastorelli, Barbaranelli & Capara (1999) also proposed a structural series of causal paths that influence childhood depression. These were found to be mediated by personal self-efficacy beliefs about social-efficacy, academic-efficacy and the problem behaviour. In a comprehensive review examining the role of childhood peer relationships, Deater-Deckard (2001) maintains that there is evidence to suggest children who are rejected by their peers, are more prone to developing internalising disorders like depression and anxiety. Furthermore, it is suggested that it is the perception of rejection that is the most important factor in determining later psychopathology. In addition, Deater-Deckard (2001) finds evidence to suggest that children who are more withdrawn, aggressive, have difficulty with emotional regulation or who have attentional problems, are more likely also to be rejected by their peers, although they were also more likely to have inflated perceptions of their status within their peer groups. These children, with more inaccurate peer ratings, tend to be lonelier and have higher levels of internalising and externalising problems.

Three studies of children with TS have investigated peer relationships (Stokes et. al., 1991; Bawden et. al., 1998; Freidrich, Morgan & Devine, 1996) and found that these

children are rated more negatively, particularly in terms of likeability, aggression and withdrawal than non-TS peers or than children with diabetes mellitus. Realmuto, August & Hektner (2000) found that in school children, aggressive-disruptive, sensitive-isolated and social etiquette were the best predictors of later externalising and internalising problems as well as with adaptive skills, respectively. It has also been suggested that more negative peer perceptions can result in peer rejection, which can in turn become a self-fulfilling prophecy as children learn to predict their rejection. In turn, they may opt out of social activities, resulting in the longer term, in reduced social competence or a lack of the appropriate social skills for the situation (Erwin 1993). Bandura (1994) also emphasises that perception or anticipation of imagined ability in whatever context, is as important a determinant of self-efficacy, as is actual ability itself. Self-efficacy can be thought of as a mechanism by which self-esteem is developed and maintained (Harter, 1999). However, the finding that peer rejection might exist concurrently with a level of self-esteem in children with TS that is comparable with their peers is difficult to explain. One suggestion is that these children may have an inflated sense of self as a self-process which serves a protective function. Harter (1999) and Harter, Marold, Whitesell & Cobbs (1996) explain that this 'false belief' behaviour can take the form of suppressing a 'true self' with the goal of obtaining approval of others. Thus, a sense of personal and social self-efficacy could be maintained by 'false self' beliefs in children with TS. From their findings, Harter et. al. (1996) argue that adolescents' subjective reaction (i.e. perception) to their lack of support or unconditional support was directly related to the motivation of false-belief behaviour. It is debateable whether these findings can be generalised to younger children, although Hanson & Onikul-Ross (1990) highlight literature for and against the positive adjustment seen in the literature about chronic

illness in children as being a protective factor, or as representing good psychological adjustment.

The model presented here in **Figure 1** suggests a hypothetical circularity of influence which has two main cycles of reciprocal relationships, each channelled through the child's perception which may in turn lead to the development of psychopathology. The outermost circle represents the relationship between neurobiological vulnerability, which may be experienced as tics occurring alone or co-morbidly with ADHD or OCD and internalising and externalising behavioural difficulties in childhood. These difficulties may affect the child's sense of self and their perceived social and personal self-efficacy, in turn exacerbating the effect of behavioural difficulties that may impact directly on the exacerbation of tic, ADHD or OCD symptoms. It is also hypothesised that neurobiological vulnerability can also affect family and peer relationships directly, particularly at the time of symptom onset and diagnosis. Furthermore, it is possible that OCD as a neurobiological vulnerability may temporally develop to become a defensive function due to over-compensated self-monitoring as a way of coping with the social adversity implicated by the tics themselves. This hypothesis offers an additional psychological explanation concerning the maintenance of obsessional thoughts, and how they may often become indistinguishable from tics.

The second inner cycle hypothesises a reciprocal relationship between the child with TS seeking or gaining approval and support through family and or peer relationships and the extent to which this determines the child's perceived personal and social self-efficacy. This perceived efficacy in turn determines the nature of relationships sought

by the child in family and peer contexts. Hence, a self-fulfilling prophecy in the reciprocity of relationships may develop. If a sense of personal and social self-efficacy is not awarded through parental or peer relationships, it is therefore proposed that low self-efficacy may ensue. This reduced sense of competence of both social and personal achievement may lead to actual personal and social incompetence and to the setting of inappropriate or unhelpful goals. This may be a pathway to psychopathology in the longer term. However, a strong self-representation is hypothesised to buffer against this negative dynamic and may be used to seek the necessary approval and support in parents and peers. This could explain why some children develop psychological difficulties beyond those directly caused by neurobiological vulnerability and others do not.

The purpose of this model has been to suggest possible pathways to psychopathology in children with TS, which may be tested in the future. It has also been highlighted that the process by which adults with TS may develop psychological difficulties or may carry such problems from childhood is unclear. A longitudinal research design of children with TS, following them into adulthood may clarify a number of issues arising here. However, in their review of recently published studies Rutter & Maughan (1997) present compelling evidence that psychopathology in childhood, negative parenting experiences, chronicity of adverse experience, poor attachment to parents or peers and antisocial behaviour all increase the risk for the development of psychopathology (including personality problems) in adulthood. Furthermore, Shady et. al. (1995) argue that their findings indicate underemployment in adults with TS and others have found academic underachievement in children with TS, particularly those with co-morbid ADHD (e.g. Abwender et. al., 1996). Woodward & Fergusson

(2000) found that problematic peer relationships were related to a variety of school problems, which in turn led to the reduction of educational achievement and so employment opportunities. These findings might partly support the notion that children with TS are at risk from the same problems, progressing into adulthood. Bandura's (1994) argument that reduced self-efficacy may lead to unfulfilled aspirations and so increase the risk of depression and anxiety might also support this hypothesis. Certainly, Elstner et. al. (2001) found that the quality of life of adults with TS was compromised in a number of domains. The process of psychopathology in adults with TS is at present an undeveloped area of research, which could provide additional insight into the connections between childhood and adult psychological distress in those with TS. The following questions may be asked: do poor peer or parental relationships play a part in development or maintenance of psychopathology in adults with TS? Can the nature of these relationships also determine who is likely to experience psychological well-being or later psychological problems? Could these relationships play a part in the development of unhelpful coping styles, which persist into adulthood and which may increase the risk for psychopathology?

**Implications for Treatment of Children and Adults with Tourette Syndrome**

A small number of case studies have been reported which have devised behavioural and cognitive treatment programmes with the main aim of reducing tic symptoms (e.g. Bergin, Waranch, Brown, Carson & Singer, 1998; O'Connor, Gareau & Borgeat, 1997; Kohen, 1995; Evers & van de Wetering, 1994; Peterson & Azrin, 1992; Mitchultka, Blanchard & Rosenblum, 1989). On the whole, the usefulness of these treatment methods have apparently proved inconclusive (Robertson, 2000). Perhaps

this is because the aim of the majority of these studies was to reduce tic frequency and intensity, rather than address underlying psychological difficulties.

As a syndrome with an episodic course that can also be mediated by the stress of external events, it is hypothesised here that treatment approaches for psychopathology in Tourette Syndrome might be best targeted from developmental, psychosocial, interpersonal as well as other psychological perspectives. If it is the case that children and adults with TS experience psychopathology as a result of the interplay between both internal and external influences, then perhaps it is important to identify the precise process mechanisms involved and target treatment on an individual basis depending upon the circumstances of distress.

If the development of psychopathology in children and adults with TS is mediated by their perception of tics and for some ADHD or OCD symptoms, relationships with parents and peers and the extent to which they are able to gain a sense of competence in early life, then perhaps these are the areas where psychological treatment programmes might be most effectively targeted. Further research may also clarify who, amongst the TS population might be most at risk of experiencing additional psychopathology so that a proactive approach to treatment may also be planned. An example of this sort of approach is described by Wigley, Mason, Lambert, Collins, Lask & Christie (2000) who describe group programmes for young children, adolescents and their parents. These groups were facilitated by trained psychological staff and were run on a semi-formal basis. They gave the children an opportunity to share difficult experiences as well build on existing skills like social competence within a supportive environment. Wigley et. al. (2000) report subjective increases in

self-esteem of group members and maintain that success of these groups was also apparent by the high attendance rates. The longer term success of this approach is yet to be reported.

For adults, many of whom may have received a diagnosis of TS later in life, the opportunity to link current schema, unhelpful coping strategies or reciprocal relationships to past experience in order to understand and make sense of current experience might be invaluable. Behavioural management techniques may be useful in the short term, for example with reduction of avoidance behaviour or panic symptoms. However, the complexity of experience accompanying TS and its ever changing nature, probably indicates psychological treatment for this group of adults that may be more exploratory.

**Implications for Further Research**

Application of the model presented in **Figure 1** to further research may clarify a number of issues arising from this review. Furthermore, a series of hypotheses are presented which may be systematically tested in order to elucidate the process mechanisms involved. In particular, the mechanisms by which self-efficacy could mediate both peer and parental relationships as well as internalising and externalising difficulties in the development of psychopathology need investigation. In addition, the relationship between internalising and externalising problems and the role of ADHD and OCD as causal mechanisms for the development of psychopathology beyond neurobiological vulnerability would also warrant further investigation. Further research to identify a link between the possible differing modes of family and

peer relationship dynamics, in the development of specific types of difficulties, might enable treatment to be more proactive or to be targeted with increased precision.

Another observation from the literature reviewed is that many studies have controlled for co-morbidity with ADHD, although few have done so for OCD, which may have a different spectrum of psychological implications, particularly for children. Future research should be mindful of this. The marked occurrence of personality disorder found in one study in people with TS, suggests a pressing need for further research in this area. Another area for further study would be to examine the association between one hand, self-representation and self-efficacy and on the other, the development of psychological difficulties. If one is more dependent upon parental and the other on peer relationship dynamics then this may again allow a proactive approach to treatment.

The evidence currently offered in relation to process mechanisms of psychopathology in adults with TS is diaphanous. A multitude of issues for investigation could be easily identified here. One of the most salient features highlighted from this review is the lack of investigation connecting childhood experience with those in adulthood, in people with TS. A prospective longitudinal study examining both psychosocial and interpersonal processes in children and their contexts, through to adulthood, may elucidate the lifetime process of living with a disorder like TS.

**Conclusions**

Elucidation of evidence associated with the psychological modes of influence on psychopathology in children and adults with TS in this review, appears to answer yes

to the question posed at the outset. There does appear to be a psychological impact arising from the experience of TS. Exactly which process mechanisms of influence may be causally linked is open to debate. Whilst there is evidence to suggest that OCD and ADHD are linked to TS neurobiologically, there is little doubt that both children and adults with TS experience a range of problems such as internalising and externalising problems, anxiety, depression and possibly personality problems that are likely to be beyond these neurobiological vulnerabilities.

A number of factors and issues requiring further clarification have been highlighted that may have a causative effect upon the development of additional psychological difficulties in children and adults with TS. The experience of TS as well as perceptions of peer and parental relationship difficulties, self-concept, self-esteem and the effect of behavioural problems in children with TS have been hypothesised here to be linked to one another as well as with the development of further psychopathology.

The evidence is even less clear from research of adults with TS. More research is important to clarify the issues that contribute to psychopathology in adults with TS. Whether this might be the lasting consequence of the relationship between difficult childhood experience and TS, or whether these findings of increased co-morbidity reflect difficulties with current experience is as yet unknown. However, given reports of reduced quality of life and negative experiences reported by adults with TS in childhood, it is likely that such negative experiences may have longer-term consequences, which do not to date appear to have been adequately studied.

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# Empirical Paper:

## Executive and Attentional Function in Adults with Tourette Syndrome with and without Co-morbid Obsessive Compulsive Disorder

Prepared for the Journal of Clinical and Experimental Neuropsychology

**Abstract**

The objective of this study was to investigate performance on 'everyday' executive and attention function tasks in adults with Tourette Syndrome (TS). Three main hypotheses were tested. The first was that adults with TS would perform more poorly than a matched group of participants without TS on tests of executive and attention function. Secondly, TS participants with co-morbid OCD would perform more poorly on the same measures than TS participants without OCD. Thirdly, that tic severity would be predictive of OCD severity and performance on the same executive and attention tasks. Results revealed significant differences between the TS and non-TS groups on two subtests measuring planning, organisational and self-monitoring ability and auditory-verbal working memory as well as on a self-report measure of behaviours associated with executive function. No further significant differences were observed between groups. There were no differences found on performance of executive function between the TS with OCD and TS without OCD groups. Regression analysis indicated that a subtest measuring mental flexibility and response suppression was a significant predictor of tic severity. Results were discussed with relation to different basal ganglia-thalamo-cortical pathways and with relation to issues of 'ecological validity' and assessment of executive function. Overall, results were interpreted with caution due to the relatively small sample size and the nature of the assessment measures employed.

**Introduction**

Tourette Syndrome (TS) is a neuropsychiatric disorder, with a mean age of onset of 5-7 years old (Leckman, Zhang, Vitale, Lahnin, Lynch, Bondi, Kim & Peterson, 1998; Baron-Cohen, Mortimore, Moriarty, Izaguirre, & Robertson, 1999). It is characterised by multiple motor tics and at least one vocal tic, that have lasted for at least one year, although they need not occur simultaneously (American Psychiatric Association, 2000; World Health Organisation, 1992). A tic is described as a sudden, rapid, recurrent, non-rhythmic, stereotyped motor movement or vocalisation (APA, 2000), which can be simple or complex in nature. TS typically waxes and wanes over the lifespan, although it has been found that between half and two thirds of diagnosed child cases resolve completely or that tic symptoms are significantly reduced by early adulthood (Hedren, 2002; Leckman et al., 1998). Prevalence rates for Tourette Syndrome have been estimated from both clinic and community samples in different countries (Apter, Pauls, Bleich, Zohar, Kron & Ratzoni, 1993; Hornsey, Banjeree, Zeitlin & Robertson, 2001; Kadesjo & Gillberg, 2000), overall agreement seems to have been reached that the accepted rate is around 5 in 10 000 children (Robertson, 2000).

The precise neural mechanisms involved with TS are still unclear. However, it has been proposed that the basal ganglia plays a major role in the manifestation of tic symptoms (Mink, 2001*a,b*; Graybiel & Canales, 2001; Kurlan et. al., 2002; Singer & Wendlandt, 2001, Parent & Hazarati, 1995). In particular, supporting this hypothesis are imaging studies that have consistently found changes in the basal ganglia, thalamic nuclei and anatomically connected, different cortical areas, including frontal regions, in the brains of patients with TS (Stern, Silbersweig, Chee, Holmes,

Robertson, Trimble, Frith, Frackowiak & Dolan, 2000; Peterson, Skudlarski, Anderson, Zhang, Gatenby, Lacadie, Leckman, Gore, 1998; Peterson, Staib, Scahill, Zhang, Anderson, Leckman, Cohen, Gore, Albert & Webster, 2001; Braun, Randolph, Stoetter, Mohr, Cox, Vladar, Sexton, Carson, Herscovitch, Chase, 1996). Excessive levels of dopamine have also been reported specifically in the basal ganglia of TS patients (Mink 2001*a*). This is substantiated by the alleviative effects of dopamine antagonist drugs (Sacks, 1985).

TS has also been found to have high rates of co-morbidity with ADHD and OCD and these have been hypothesised to be neurobiologically linked to TS. The marked co-morbidity of Obsessive Compulsive Disorder (OCD) with TS (Coffey & Park, 1997; Kano, Ohta & Nagai, 1998; Robertson & Gourdie, 1990; Robertson, Channon, Baker, & Flynn, 1993; Carter, Pauls, Leckman & Cohen, 1994; Kadesjo & Gillberg, 2000) has led to hypotheses that these disorders are mediated by the same genetic trait (Miguel, Rosario-Campos, Shavitt, Hounie, & Mercadante, 2001; Pauls, 2001) that is responsible for abnormal functioning of the basal ganglia thalamo cortical pathways, causing deficiency of serotonin (Singer & Wendlandt, 2001). Functional imaging studies of patients with pure OCD have also found consistent patterns indicating abnormal involvement of the basal ganglia thalamo cortical circuitry, particularly involving the orbitofrontal cortex (Rubin, Villanueva-Meyer, Ananth, Trajmar, & Mena, 1992; Swale, Hymans, Lees, & Frackowiak, 1991; Swedo, Pietrini, Leonard, Schapiro, Rettew, Goldgerger, Rappoport, Rappoport, & Grady, 1992). ADHD is another disorder recognised to have a high co-morbidity with TS (Spencer, Biederman, Coffey, Geller, Faraone & Wilens, 2001; Comings & Comings, 1988), which has also been related to dysfunction of the same basal ganglia cortical

pathways (Robertson, 2000). Although beyond the current scope of discussion, Castellanos (2001) provides a detailed discussion of the evidence regarding neurobiological correlates of ADHD.

It has been broadly hypothesised that there are several basal ganglia thalamo cortical pathways that might explain different aspects of cognitive control including attentional and executive functions (Casey, Durston & Fossella, 2001). Executive and attention functions have been described as those that determine the 'how' and 'whether or when' of behaviour as opposed to the 'what' or 'how much' as do other cognitive functions such as memory (Mahone, Koth, Cutting, Singer, Denkla, 2001; Lezak, 1995; Heilman, Voeller, & Nadeau, 1991). It has been further proposed that the dorsolateral prefrontal circuit, might be responsible for a number of higher cognitive 'executive' functions including goal selection, planning, sequencing, set-shifting and cognitive flexibility, concept formation, verbal and spatial working memory, self-monitoring and self-awareness (Royall, Lauterbach, Cummings, Reeve, Rummans, Kaufer, LaFrance, Coffey, 2002; Lezak, 1995). The lateral orbitofrontal circuit has been hypothesised to account for some attentional functions e.g. initiation of behaviour, response suppression and inhibition, as well as emotional regulation, judgement and insight (Royall et. al., 2002). This area has also been specifically implicated in OCD, which has been hypothesised to be a difficulty with suppression of unwanted intrusive thoughts and behaviours (Aycicegi, Dinn & Haris, 2002; Casey et. al., 2001; Saxena, Brody, Schwartz & Baxter, 1998; Cox, 1997). TS and OCD have been hypothesised to reflect an inability to suppress unintentional behaviour and/or cognitions (Ozonoff & Jensen, 1999; Ozonoff et. al., 1998) and so it might also be hypothesised that they share the same neural pathways, namely, the lateral

orbitofrontal circuit. Royall, et. al. (2002) also discusses the anterior cingulate pathway, a third basal ganglia cortical pathway which is also thought to be responsible for attentional functions.

Given the neurobiological correlates and observed behavioural and cognitive difficulties associated with TS, many researchers have attempted to determine whether there is a neuropsychological profile of TS (Dykens, Leckman, Riddle, Hardin, Schwartz & Cohen, 1990; Bornstein, Baker, Bazylewich & Douglass, 1991; Brookshire, Butler, Ewing-Cobbs & Flatcher, 1994; Yeates & Bronstein, 1996; Schuerholz, Singer & Denkla, 1998). On the whole, the results of many of these studies have been inconclusive, owing to varied methodologies, assessment measures, differing operational definitions of the functions being measured, lack of control groups, lack of control for co-morbidity and small sample sizes. Early investigation suggested a discrepancy between measures of Verbal and Performance IQ, poor visuo-spatial construction ability and difficulty with perceptual tasks (e.g. Bornstein, King & Carroll, 1983) although others have not found this to be the case (Brookshire et. al., 1994). It is now recognised that children with TS do not experience intellectual difficulties as a rule and in fact are often recorded as having slightly above average full scale IQ's when compared with normative data (Dykens et. al., 1990; Mahone et. al., 2001). Furthermore, some studies have associated cognitive or academic skills deficits in TS to co-morbidity with ADHD (Dykens et. al., 1990; Swerdlow, Magulac, Filion & Zinner, 1996). One study appears to have done so with OCD (Bornstein, 1991*b*). However, the results regarding co-morbidity remain inconclusive although overall, it has been suggested that ADHD in particular might account for deficits in performance seen in children with TS (Pennington & Ozonoff,

1996). The lack of adequate control for co-morbidity may go some way to explaining the continued inconclusive findings of a specific neuropsychological profile for children with TS (Ozonoff & Jensen, 1999; Cirino, Chapieski & Massman, 2000; Schuerholz et. al., 1996).

With the lack of conclusive evidence to support a global neuropsychological profile of deficit in those with TS, attention has focused more recently to the set of higher cognitive processes termed executive functions (EF). Given the neurobiological correlates of TS and EF, relatively few studies have attempted to assess this set of functions independently. Furthermore, most of these have been conducted with children. Nevertheless, some of the most frequently used tasks to measure different aspects of executive function have been, the Wisconsin Card Sorting Task (WCST; Grant & Berg, 1948), Tower Tests (e.g. Shallice, 1982), Verbal Fluency and a variety of response inhibition paradigms. The WCST was designed to assess cognitive flexibility and concept formation and studies of TS patients have repeatedly found no difference compared with normative data or control groups on this measure (Bornstein, 1991*a*; Ozonoff & Jensen, 1999; Cirino et. al., 2000; Channon, Crawford, Vakili & Robertson, 2003). However, participants with TS with OCD have been found to perform more poorly on the WCST as a function of the severity of OCD symptoms or the presence of TS compared with pure OCD (Bornstein, 1991*b*; Gladstone, Carter, Schultz, Riddle, Scahill & Pauls, 1993). Tower tests of varying difficulty (e.g. London and Hanoi) purport to measure goal selection, planning, sequencing and spatial working memory (Mahone, Cirino, Cutting, Cerrone, Hagelthorn, Hiemenz, Singer & Denkla, 2002; Lezak, 1995). One study to date appears to have used this measure in children with TS with a mean age of 12.6 years

(Ozonoff & Jensen, 1999) and found no significant difference compared with control participants. At least three studies have included measures of verbal fluency and found significantly poorer performance in children with TS (Brookshire et. al., 1994; Schuerholz, Baumgardner, Singer, Reiss & Denkla, 1996; Schuerholz et. al., 1998), another has not (Mahone et. al., 2001).

Neuropsychological research has also assessed response inhibition with the belief that this facet of executive function may be more directly linked to TS and as such, may exert influence on all other cognitive, behavioural and motor action processes (Como, 2001; Casey et. al., 2001). However, results have again been inconclusive with some studies finding differences and others finding no difference compared with control groups (e.g. Ozonoff & Jensen, 1999). It has again been suggested that this inconclusive pattern of results may be likely to be a function of co-morbidity with ADHD or OCD and due to a lack of control for these factors, which was the finding of Ozonoff, Strayer, McMahon & Filloux (1998). Swerdlow et. al. (1996) found that whilst adult TS participants did not differ significantly from controls on an inhibitory and facilitatory visual priming task, children with TS did. These researchers explain this child-adult discrepancy as the probable immaturity of the inhibitory frontal system in children or because of a developmental delay. Swerdlow et. al. (1996) also reported that adults with TS were seen to exhibit a relative deficit on inhibition versus facilitation and propose that these results suggest a deficit in central inhibition in both children and adults with TS.

There is an ongoing debate as to whether executive and attentional functions represent different constructs of higher cognitive function, cognitive and behavioural control or

whether they are facets of the same set of functions that are inseparable by measures of observable process (Royall et. al., 2002). There have been relatively few studies examining EF in TS and as a result, there are a number of facets of EF that have received little or no attention at all. Problem solving is one such EF facet. There has been one study to date assessing problem solving in adults with TS. Channon et. al. (2003) assessed "Real-life type" problem solving and found that TS participants produced significantly fewer problem solutions and selected poorer final solutions than controls. Channon et. al. also reported that even though TS participants selected poorer final solutions their satisfaction with their responses was not significantly different from control participants. This was interpreted as a possible subtle impairment of awareness or acknowledgement of their own performance, and possible problems with insight were also supported by discrepancies between self and other ratings on the DEX questionnaire of different facets of EF (Burgess, Alderman, Wilson, Evans & Emslie, 1996).

There also remains uncertainty as to whether the neuroanatomical, chemical and imaging studies evidence basal ganglia cortical pathways which remain separate or whether they share an ultimate common basal ganglia process. If either were the case, cortical pathways may differ, giving rise to different clinical presentations which could be seen as tics, compulsions, executive or attentional problems although the latter might result in a more mixed clinical presentation (Mink, 2001). In addition, there is a lack of clarity regarding the dysfunction of the basal ganglia cortical pathways, and how this might affect a variety of executive control functions in TS (Como, 2001).

The current study set out to determine a variety of facets of executive and attentional function in adults with TS using measures designed to be more 'ecologically' valid. This is because traditional neuropsychological tests have been criticised for being unable to bridge the gap between experimental performance and actual everyday performance measures. Everyday performance might use measures of that allow some determination of functional outcome (Sbordone, 2000). Although the Wisconsin Card Sorting Test has been proposed as a 'gold standard' for determining executive function performance (Royall et. al., 2002), it is an abstract task carried out in controlled conditions, unlikely to be encountered in everyday life. This and other traditional measures have also been criticised for underestimating functional outcomes given the complexity of stimuli encountered in everyday life (Sbordone, 2000). The current study attempts to bridge this gap by assessing in particular, everyday planning, organisational ability, novel problem solving, and ability to monitor one's own performance. In addition, given the debate that both symptoms of OCD and ADHD may represent a sub-threshold manifestation of TS itself (Robertson, 2000), the current study also sets out to examine attentional function in those with a clinical diagnosis of TS, who have not received an additional diagnosis of ADHD. Furthermore, the relationship between co-morbidity with OCD and neuropsychological performance has not been studied in adults with TS, although two studies have done so with children (Gladstone et. al., 1993; Bornstein, 1991*b*).

Three main hypotheses will be tested, firstly that adults with TS will perform more poorly than matched controls on measures of everyday executive function and attention. Secondly, adults with TS and co-morbid OCD will perform more poorly than adults with TS alone on measures of executive function. Thirdly that

performance on executive and attention measures and severity of OCD can be predicted by tic severity.

## Method

*Participants*

Fifty-three adults over 16 years old were identified as meeting the inclusion criteria for the study from the clinic list of a regional centre for Tourette Syndrome in the UK. After receiving information[1] about the study by post, people were asked to opt in to the research by returning a slip giving the researcher permission to contact them by telephone. Twenty-five adults with Tourette Syndrome responded but 6 of those were ultimately unable to take part.

Seventeen healthy participants without TS were recruited for comparison with the TS sample (16 men and 1 woman) (age range 16-59 years), with permission, from the non-medical or psychological staff pool of the psychiatric hospital from which the TS participants were recruited. The TS and control participants did not differ significantly in age ($t = -0.30$, $p = 0.76$; TS mean 29.59, SD 12.92; control group mean 30.88, SD 12.10), age of leaving education ($t = -0.59$, $p = 0.56$; TS mean 18.94, SD 3.49; control group mean 19.82, SD 5.21) or in IQ ($t = -1.33$, $p = 0.19$; TS mean, 102.33, SD 14.97; control group mean, 109.18, SD 15.46). The two groups did however differ significantly in their levels of reported anxiety ($t = 2.81$, $p = 0.008$; TS mean 8.88, SD 3.64; control group mean 5.53, SD 3.41) and depression ($t = 2.84$, $p < 0.01$; TS mean 4.33, SD 3.64 control group mean 2.18, SD 2.13). All participants gave written informed consent to participate in the study.

[1] Information leaflet see Appendix 2

*Procedure and Measures*

In all, 36 TS and non-TS control participants took part in the research. TS participants were interviewed in order to ascertain lifetime and current tic severity status using the Diagnostic Confidence Index (DCI) (Robertson et. al., 1999) and Yale Global Tic Severity Scale respectively (YGTSS) (Leckman et. al., 1989). Participants were also administered the Yale-Brown Obsessive Compulsive Scale (Y-BOCS) (Goodman et. al., 1989), in order to ascertain diagnosis and severity of OCD. Scores obtained from these measures were verified by case note review by the Consultant Neuropsychiatrist responsible for their care. One participant was excluded from the study on the basis that their DCI score was below 50. Eighteen participants (16 men, 2 women) (age range 16-56 years) who met the ICD-10 criteria for Tourette Syndrome and who did not have any psychiatric diagnoses other than Obsessive Compulsive Disorder, took part in the study. Out of 18 participants, 8 were identified as having co-morbid OCD. Participants were also excluded if they had a history of, or were currently suffering from any psychiatric or medical condition that might affect their ability to think or remember, or if they were colour blind. Other information collected at initial interview included age of onset of tics and age at TS diagnosis, for the TS group and for both groups; family history of tics, age of finishing education, highest qualification attained and current occupation.

All participants were administered the following assessment measures in the order indicated, Wechsler Abbreviated Scale of Intelligence, Test of Everyday Attention, Behavioural Assessment of the Dysexecutive Syndrome, and the Hospital Anxiety and Depression Scale. All assessment measures were administered and scored according to the instructions of each respective manual.

*Assessment Measures* [2]

1. The Yale Global Tic Severity Scale (Leckman et. al., 1989) was designed for use by clinicians to assess the current number, frequency, intensity, complexity and interference of motor and phonic tic symptoms. It yields total motor and phonic tic severity scores (maximum 25 points for each sub-scale) and an impairment score with a maximum of 50 points. Therefore an overall severity score out of 100 is obtained. The YGTSS is reported to have good construct, convergent and discriminant validity as well as high rates of internal consistency (0.8-0.91) (Leckman et. al., 1989)

2. The Diagnostic Confidence Index (Robertson et. al., 1999) was designed to measure lifetime tic symptoms and severity, independent of current severity of symptoms. A total confidence of diagnosis score out of 100 is obtained. The DCI is a relatively new measure which has high correlations with established measures of tic severity (e.g. YGTSS) (Robertson et. al., 1999)

3. The Yale-Brown Obsessive Compulsive Scale (Goodman et. al., 1989*a*) was designed to be a measure of the severity of obsessions and compulsions, yielding an overall severity score out of 40. Studies have shown the YBOCS to good validity (Goodman et. al., 1989*b*) and rates of reliability (Goodman et. al., 1989*a*) and internal consistency (0.78-.096) (Nakagawa et. al., 1996)

4. Wechsler Abbreviated Scale of Intelligence (4-subtest version) (Wechsler, 1999) was developed as a shorter, screening form of the full version of the

[2] All record forms for assessments can be found in appendix 3

Wechsler Adult Intelligence Scales and is particularly recommended for matching of cognitive ability in research samples. It has been nationally standardised, has good reliability and validity and correlates highly with the WAIS-III (Wechsler, 1999). Subtests include vocabulary, block design, similarities and matrix reasoning.

5. Test of Everyday Attention (TEA) (Robertson et. al, 1994) was designed as a battery of attentional measures using everyday materials. The following subtests were administered; Elevator Counting, measuring sustained attention; Elevator Counting with Distraction, measuring auditory selective attention; Elevator Counting with Reversal, measuring auditory-verbal working memory; Telephone Search, measuring visual selective attention; Telephone Search while Counting, measuring divided and sustained attention. Each subtest has been age standardised with a normal population and cross-validated with a neurologically impaired sample. It has good discriminant validity and correlates well with other measures of attention (Robertson et. al., 1996; Chan, 2000)

6. The Behavioural Assessment of the Dysexecutive Syndrome (BADS) (Wilson et. al., 1996) is a battery of subtests designed to assess different facets of executive function. In particular subtests measure ability to shift set, novel problem solving, planning, organisation and self-monitoring ability and temporal judgement. All subtests were administered. Also used was the DEX self-rating questionnaire, measuring participant's self-reported behavioural difficulties associated with a range of executive functions. This measure was

not used to calculate the overall Age Adjusted scores because it is an additional measure to the BADS battery, not included with the standardisation sample. Therefore, the sum of items checked per person was used in the analysis. The BADS has been standardised with a normal population and cross-validated with a brain injured population. An overall age-standardised score is derived from performance on all subtests. Internal consistency for all subtests has been reported at between 0.88-1.0 (Wilson et. al., 1996). Construct and discriminant validity has been reported as high and comparable to other standard tests of executive function (Norris & Tate, 2000).

7. Hospital Anxiety and Depression Scale (HADS) (Snaith & Zigmund, 1983) was designed to be a short measure of current anxiety and depression. It has been extensively used for research purposes, is well validated and correlates well with other measures of anxiety and depression (Bjelland et. al., 2002).

8. An additional Interview Schedule was designed in order to obtain demographic variables and to ensure adherence to the exclusion criteria.

## Results

*Tests of Normal Distribution*

Using the Kolmogorov-Smirnov one sample test, the distribution of all profile scores from subtests on the BADS were significantly different for that expected from a normal distribution curve, except the zoo map subtest. Overall Age Adjusted scores from the BADS, DEX scores, IQ scaled scores and HADS scores were normally distributed. All standard scores from subtests on the TEA were normally distributed except Elevator Counting and Elevator Counting with Distraction. Non-parametric, Mann-Whitney U tests (for one-tailed hypothesis) were employed for skewed data and parametric independent t-tests were employed for normally distributed data.

*BADS Scores*

**Table 1** gives the descriptive characteristics of BADS data. The differences between profile scores on the BADS for all of the TS and control group participants were evaluated. All Mann-Whitney U analyses were for a one-tailed hypothesis. Only the 6 Elements subtest indicated a significant difference between the groups ($U = 82$, $z = -2.79$, $p = 0.01$), with the control group obtaining higher scores than the TS group. A trend for significance was seen for the Action Programme ($U = 127$, $z = -1.74$, $p = 0.08$) and Temporal Judgement subtests ($U = 101$, $z = 1.72$, $p = .06$), with the TS group performing better than the control group on the Temporal Judgement subtest. The difference between profile scores for the Rule Shift subtest ($U = 153$, $z = 0.0$, $p = 1.0$), Key Search subtest ($U = 147.50$, $z = -0.18$, $p = 0.86$) or Zoo Map subtest ($t = -1.2$, $df = 33$, $p = 0.22$), were all non-significant.

Further analysis compared overall Age Adjusted scores on the BADS between the TS and control groups. The Age Adjusted scores correlated with FSIQ ($r = 0.47$, $p = 0.004$), and so ANCOVA with FSIQ as a covariate was calculated. There was no significant difference between the TS group and control group on overall Age Adjusted scores for overall performance on all subtests of the BADS ($F(1, 32) = 0.44$, $p = .50$).

**Table 1: Descriptive Statistics for BADS and TEA Subtests***

| | *TS Group* | | | | | | *Control Group* | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | **N** | **Mean** | **Median** | **SD** | **Interquartile Ranges 25%ile** | **75%ile** | **N** | **Mean** | **Median** | **SD** | **Interquartile Ranges 25%ile** | **75%ile** |
| **Elevator Count** | 18 | | 7 | | **6** | **7** | 17 | | 7 | | **7** | **7** |
| **Elevator Count Distract** | 18 | | 10 | | **8.75** | **13** | 17 | | 13 | | **10** | **13** |
| **Elevator Count Reversal** | 18 | 6.77 | | 3.66 | | | 17 | 9.53 | | 2.81 | | |
| **Tele Count** | 18 | 7.33 | | 3.58 | | | 17 | 9.41 | | 3.41 | | |
| **Tele Search Count** | 18 | 9.61 | | 4.06 | | | 17 | 11.65 | | 4.62 | | |
| **Rule Shift** | 18 | | 4 | | **3.75** | **4** | 17 | | 4 | | **3.50** | **4** |
| **Action Programme** | 18 | | 4 | | **4** | **4** | 17 | | 4 | | **4** | **4** |
| **Key Search** | 18 | | 4 | | **2** | **4** | 17 | | 4 | | **2.50** | **4** |
| **Temporal Judgement** | 18 | | 3 | | **2** | **3** | 17 | | 2 | | **2.00** | **3** |
| **Zoo Map** | 18 | 2.11 | | 1.53 | | | 17 | 2.65 | | .931 | | |
| **6 Elements** | 18 | | 3 | | **2.75** | **4** | 17 | | 4 | | **4** | **4** |
| **DEX** | 18 | 30.33 | | 13.87 | | | 17 | 20.35 | | 11.27 | | |
| **BADS Overall Age Adjusted Score** | 18 | 99.77 | 102 | 15.69 | **88.50** | **114.25** | 17 | 105.29 | 108 | 10.23 | **97.5** | **113** |

* Means and Standard Deviations provided for normally distributed data; Median and Interquartile Ranges provided for non-normally distributed data. DEX not included in BADS Overall Age Adjusted Score.

Within the TS group, analysis was performed to ascertain whether there was a difference between those with TS with co-morbid OCD, and those with TS alone on individual profile scores from subtests of the BADS. No significant differences were seen on any subtests; Rule Shift ($U = 39.50$, $z = -.057$, $p = .97$), Action Programme ($U = 34$, $z = -.83$, $p = .63$), Key Search ($U = 29.50$, $z = -1.03$, $p = .36$), Temporal Judgement ($U = 25.5$, $z = -1.39$, $p = .20$), Zoo Map ($t = .02$, $df = 15$, $p = .98$), 6 Elements ($U = 35$, $z = -.48$, $p = .70$). Furthermore, no significant difference was found for TS with OCD and TS with out OCD on overall age adjusted scores from the BADS ($U = 33.5$, $z = -.58$, $p = .57$).

There was found to be a significant difference between groups on the DEX self-report measure of behavioural difficulties associated with executive function, with the TS group scores being higher than the control group ($t = 2.33$, $df = 33$, $p < 0.05$).

*Test of Everyday Attention Scores*

The difference between age-adjusted scores for subtests from the TEA for the TS and control group participants were evaluated using Mann-Whitney U and Independent t-tests.

There were no significant differences between the TS and control group participants on Elevator Counting ($U = 119$, $z = -1.12$, $p = 0.26$), Elevator Counting with Distraction ($U = 112$, $z = -1.35$, $p = 0.18$) and Telephone Search whilst Counting subtests ($t = -1.39$, $df = 33$, $p = 0.17$).

The Elevator Counting with Reversal (r = .62, p >0.01) and Telephone Search (r = .43, p = >0.01) subtests correlated with FSIQ and so were analysed separately using ANCOVA. With IQ as a covariate there was found to be a significant difference between the TS and control group on the Elevator Counting with Reversal subtest from the TEA (F (1, 32) = 4.08, p = .005). However, no significant difference was seen between groups on the Telephone Search subtest (F (1, 32) = 1.65, p = 0.20).

*The Relationship Between Tic Severity, OCD Severity And Measures Of Cognitive Functioning*

Regression analysis was performed in order to assess the relationship between tic severity, OCD severity and a number of measures of cognitive functioning. The Ordinary Least Squares Regression Model (OLS) has the advantage of estimating parameters for predictor variables with associated standard errors and significance tests. However, due to the small sample size (N=18) and relatively large number of possible predictor variables (i.e. sixteen) the OLS method is at risk of overfitting (i.e. a regression model which is well fitted to the characteristics of the particular sample but which does not generalise to the population from which the sample is obtained). Overfitting can result in artificially inflated $R^2$ values and biased parameter estimates.

Therefore, the Partial Least Squares Regression (PLS) model was chosen because it does not impose the same restrictions employed by other generalisations of the linear model (e.g. discriminant analysis, principle components regression or canonical correlation). PLS is a linear regression method that forms components (i.e. factors or latent variables determined by both the dependent variable and the predictor variables) as new independent variables in a regression model. A regression model

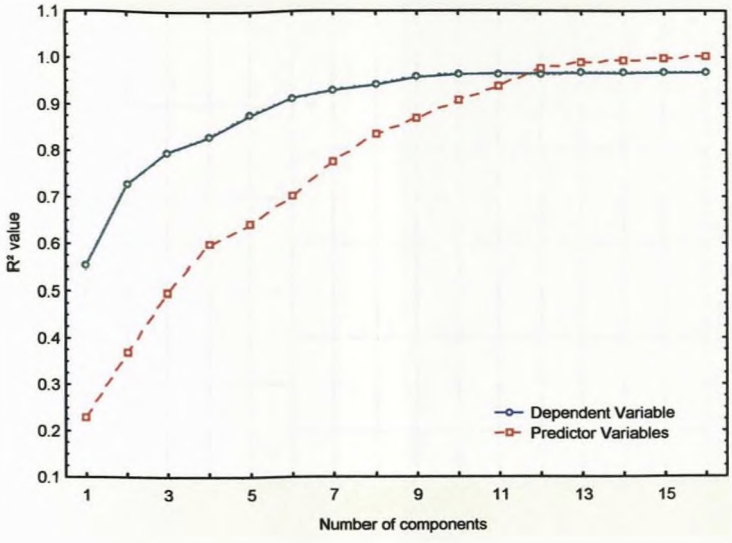
from PLS can be expected to have a smaller number of components without appreciable $R^2$ value. The flexibility of the PLS method means that it can be used in situations where traditional multivariate methods are extremely limited, such as when there are fewer observations than predictor variables (StatSoft, 2001). An extreme example is given by Wold (1989) who analysed 27 variables using two latent constructs with a data set consisting of ten cases.

The fitted components of a PLS regression model can then be used to reconstruct parameter estimates for the original predictor variables. Monte Carlo simulation studies have shown that, in small sample situations, parameter estimates from Partial Least Squares regression models have better correspondence to large sample estimates than do estimates derived from the OLS regression method (Chin, Marcolin, and Newsted, 1996). Because it is not possible to estimate the standard error of these parameter estimates, associated tests of significance are not possible.

A two-phase analysis strategy was therefore employed. In the first phase the PLS Regression model was fitted and parameter estimates were derived. In the second phase, the OLS regression model and parameter estimates were calculated. The parameter estimates from these two regression methods were compared. Parameter estimates showing substantial differences (i.e. greater than 0.1) were considered to have been overfitted and therefore unreliable estimates of population parameters.

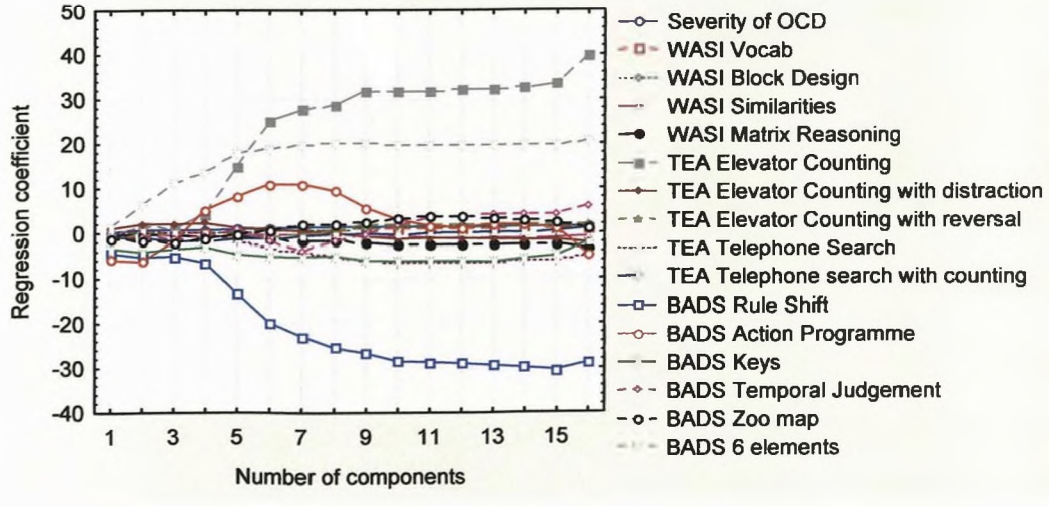
The PLS regression model resulted in a fit of sixteen potential components. As can be see from **Figure 1**, a ten-component solution resulted in retaining 92% of the variation of the predictor variables and 96% of the variation of the dependent variable.

**Figure 1: $R^2$ values by number of components**




Also, **Figure 2** illustrates that a ten-component solution showed stability with the estimated regression coefficients. Thus, a ten-component solution was selected for the estimation of regression parameters. These parameter estimates are presented in **Table 2** along with the OLS regression estimates.

**Figure 2: Change of Regression Coefficients By Number Of Components**



The OLS regression Model resulted in an $R^2 = 0.97$ ($F(16,1) = 1.82$; $p = 0.53$) and an adjusted $R^2 = 0.44$. The discrepancy between the $R^2$ and adjusted $R^2$ values indicated considerable redundancy in the measures of cognitive functioning and OCD severity.

**Table 2: Ordinary Least Squares and Partial Least Squares Regression Parameters***

| | PLS Beta | OLS Beta | OLS SE | OLS-PLS Discrepancy | PLS B | OLS B | OLS SE. | t(1) | p |
|---|---|---|---|---|---|---|---|---|---|
| **Intercept** | | | | | | -21.58 | 112.47 | -0.19 | 0.88 |
| **YBOCS Severity** | 0.65 | 0.69 | 0.41 | 0.03 | 1.36 | 1.42 | 0.85 | 1.68 | 0.34 |
| **Vocabulary** | 0.52 | 0.52 | 0.92 | 0.00 | 1.03 | 1.02 | 1.82 | 0.56 | 0.67 |
| **Block Design** | 0.51 | 0.52 | 0.53 | 0.01 | 1.39 | 1.42 | 1.45 | 0.98 | 0.51 |
| **Similarities** | -0.49 | -0.49 | 0.68 | 0.01 | -0.97 | -0.96 | 1.34 | -0.71 | 0.61 |
| **Matrix Reasoning** | -0.73 | -1.08 | 2.09 | -0.35 | -2.43 | -3.61 | 7.00 | -0.52 | 0.70 |
| **E Counting** | 0.77 | 0.97 | 1.30 | 0.20 | 31.82 | 39.99 | 53.74 | 0.74 | 0.59 |
| **E Counting D** | -0.13 | -0.22 | 0.64 | -0.09 | -1.29 | -2.14 | 6.25 | -0.34 | 0.79 |
| **E Counting R** | 0.29 | 0.28 | 0.40 | -0.01 | 1.96 | 1.87 | 2.70 | 0.69 | 0.61 |
| **Tele Search** | -0.93 | -0.71 | 1.45 | 0.22 | -6.42 | -4.88 | 9.99 | -0.49 | 0.71 |
| **Tele Search Counting** | -0.03 | 0.13 | 0.68 | 0.16 | -0.19 | 0.79 | 4.13 | 0.19 | 0.88 |
| **Rule Shift** | -0.89 | -0.90 | 0.63 | -0.01 | -28.48 | -28.81 | 20.09 | -1.43 | 0.39 |
| **Action Programme** | 0.05 | -0.08 | 0.77 | -0.12 | 3.03 | -4.81 | 49.40 | -0.10 | 0.94 |
| **Key Search** | -0.31 | -0.10 | 1.37 | 0.22 | -6.16 | -1.92 | 26.74 | -0.07 | 0.95 |
| **Temporal Judgement** | 0.06 | 0.17 | 0.55 | 0.11 | 2.03 | 6.11 | 19.61 | 0.31 | 0.81 |
| **Zoo Map** | 0.21 | 0.05 | 0.98 | -0.15 | 3.33 | 0.84 | 15.70 | 0.05 | 0.97 |
| **6 Elements** | 0.82 | 0.86 | 0.41 | 0.04 | 19.69 | 20.65 | 9.77 | 2.11 | 0.28 |

*Highlighted rows show substantial OLS- PLS discrepancy and should be considered unreliable population estimates.

Nevertheless, it might be conservatively estimated that the measures of cognitive functioning and OCD accounted for approximately 44% of tic severity. The OLS and PLS regression parameters are presented in **Table 2.** None of the OLS Beta coefficients reached the criteria for statistical significance. However, given the small sample size and relatively large number of predictor variables there is a substantial possibility of a Type 2 error. If the PLS beta coefficients are rank ordered then two measures of executive functioning (6 Elements and Rule Shift) and OCD

symptomatology were most related to tic severity. These three predictor variables were then entered into another reduced OLS regression model. This model resulted in an $R^2 = 0.63$ ($F(13,4) = 3.13$; $p = 0.06$) and an adjusted $R^2 = 0.40$. The discrepancy between the reduced model adjusted $R^2$ and complete model adjusted $R^2$ values (i.e., 0.44-0.40 = 0.04) indicates that there is only a small loss of accuracy in the reduced regression model. **Table 3** presents the regression coefficients for the reduced model. It can be seen from **Table 3**, that the beta coefficient for the Rule Shift task showed a significant negative association with tic severity (beta= -0.49; $t(14) = -2.15$; $p=0.05$).

**Table 3: Ordinary Least Squares Regression Parameters**

| | Beta | SE | B | SE | t(14) | p |
|---|---|---|---|---|---|---|
| **Intercept** | | | 83.05 | 27.58 | 3.01 | 0.009 |
| **YBOCS Severity** | 0.31 | 0.21 | 0.63 | 0.44 | 1.44 | 0.16 |
| **Rule Shift** | -0.49 | 0.22 | -15.65 | 7.28 | -2.14 | 0.04* |
| **6 Elements** | 0.3523 | 0.22 | 8.48 | 5.33 | 1.59 | 0.13 |

* Significant at $p<0.05$

**Discussion**

The first hypothesis sought to answer the question of whether the TS group as a whole, would perform worse than the control group on measures of executive function and attention. Overall, this hypothesis was not accepted, although on separate subtests of the BADS and TEA there was one significant result for each. Analysis of individual profile scores on the BADS revealed a significant group difference on the 6 Elements subtest, with the control group obtaining higher scores. This suggests that adults with TS may have more difficulty than adults without TS on a task involving self-monitoring as well as planning and organisational ability. Given this significant difference on the 6 Elements subtest, it is surprising that none of the other subtests, nor overall age adjusted scores showed significant between group differences, particularly those supposedly measuring similar functions e.g. Key Search and Zoo Map subtests. This may be indicative of caution when interpreting these results. However, overall totalled individual scores on the self-report DEX questionnaire measure revealed significant group differences, with the TS group endorsing overall higher scores on items relating to a variety of behavioural difficulties associated with executive function than the control group.

On measures of attention, only the Elevator Counting with Reversal subtest was found to be significantly different across groups. The Elevator Counting with Reversal subtest is a complex task, which measures auditory-verbal working memory and these results suggest that people with TS who do not have an additional diagnosis of ADHD may have reduced capacity here compared with a matched control group. Other measures of attention were not found to be significantly different between the TS and control groups.

The second hypothesis sought to determine whether the TS with OCD group would perform more poorly on measures of executive function than the TS without OCD group. There were no significant differences observed on any subtests of the BADS or on the overall age adjusted score between these groups. The previous findings of Bornstein (1991*b*) and Gladstone et. al. (1993) that performance of the WCST was associated with OCD is partly unsupported by these findings, if the functions measured by the WCST are accepted to be cognitive flexibility and concept formation.

The third hypothesis was to determine if there was any predictive power of tic severity on the measures of cognitive function and on OCD severity. Unfortunately, restrictions of sample size introduced the threat of overfitting the data and consequently drawing erroneous conclusions regarding the relative importance of the potential predictor variables. As a result, a two-stage analysis strategy was undertaken in an attempt to identify parameter estimates that may have been confounded by overfitting. The three variables that most related to tic severity from the Partial Least Squares Regression method were entered again into an Ordinary Least Squares model. The Rule Shift task was found to be significantly negatively associated with tic severity. This suggests that ability to shift from one rule to another and to suppress unnecessary information whilst doing so might decrease as tic severity increases.

Regarding hypotheses concerning particular basal ganglia cortical pathways, results from the current study offers little in the way of additional insight. However, if it is accepted that functions measured by the 6 Elements and Elevator Counting with Reversal subtests (i.e. planning, organisation, self-monitoring and auditory-verbal

working memory), might be neurobiologically mediated by the dorsolateral-prefrontal-basal ganglia pathway (Royall et. al., 2002; Casey et. al., 2001), then the group differences observed here might lend some support to the hypothesis that adults with TS may have an impairment of these 'higher cognitive' executive functions than adults without TS. Moreover, the presence of additional OCD in participants with TS made no difference to this finding, indicating that all participants with TS performed similarly when compared with the non-TS group. Another finding of note is that the Rule Shift task was predictive of tic severity. If the purported function being measured here as defined in the BADS manual is mental flexibility (measured by the ability to shift thinking from one rule to another), then the implication is that as tic severity increases then so does a possible impairment in this ability which is mediated by the dorsolateral prefrontal cortex. However, clinical and research experience assigns a response inhibition function to this subtest too. If this is true, then it may also be the case that as tic severity increases, so does a possible impairment with response inhibition thought to be mediated by the orbitofrontal cortex (Royall et. al., 2002). Although Swerdlow et. al., (1996) argue for the presence of a deficit of central inhibition in TS and this pathway has been implicated in OCD (Aycicegi, Dinn & Harris, 2002), there is not enough evidence from the current data to comment upon a possible association between TS, OCD and the basal ganglia orbitofrontal pathway. However, the possible multiple pathways that have been implicated by the functions mentioned above, might lend support to the hypothesis that the different basal ganglia thalamo-cortical loops may share common neurobiological connections.

Another finding of note is the non-significant differences between the control and TS participants on measures of attention. It has been hypothesised elsewhere that TS, by

its nature, is a disorder that is internally distracting, and therefore that deficits in attention may be observed on a functional level, as an inevitable consequence of the syndrome (Robertson, 2000). If working memory is functionally assigned to the neurobiologically determined dorsolateral basal ganglia pathway (Royall et. al., 2002), then the hypothesis that attentional difficulties may be an inevitable consequence of TS in those without ADHD is unsupported here, as no differences were observed between the TS and control groups on other measures of attention.

A number of methodological issues arise from this study. One is the use of the BADS. Although the BADS reportedly measures EF in a more 'ecologically valid' way, the subtests evidence a ceiling effect and consequently a skewed distribution of most of the subtests. This reduced sensitivity limits its use in a normal population and for those who may have subtle EF deficits. In addition, the individual subtest scores are not age adjusted and are converted to profile scores, resulting in a small range of possible scores. The apparent discrepancy between non-significant group differences on the BADS and significant group differences on the DEX measure, might indicate that the DEX questionnaire is a better measure of everyday EF than the subtests themselves. However, it is also possible that findings of significant difference on the DEX between groups may have been confounded by higher scores, reflecting actual TS symptoms, rather than behavioural signs of executive function. Together with the fact that there is no normative data for this part of the BADS, this finding needs to be interpreted with caution. However, completion of the 'independent rater' version of the DEX (which was not feasible in this study) might have added additional insight regarding others' experience of executive function difficulties in adults with TS. (The BADS manual discusses the utility of comparing 'self' with 'independent rater'

versions of the DEX.) A way of cross-validating the BADS battery would have been to include a number of 'traditional' tests of EF, for example, WCST, Stroop, Tower Test or the Trail Making Test. This may also have allowed comparison between present and previous findings as well as with the BADS battery.

Another possible confounding problem with results may be that significant differences between groups on self-reported levels of anxiety and depression may have directly influenced performance on the tests. This would be a very difficult factor to control for given the very high reported rates of co-morbidity occurring in people with TS (Robertson, 2000).

A major drawback of this study is the sample size. A small sample means that statistical analysis is less sensitive to significant differences in the population and that medium to small experimental effect sizes may be obscured. Furthermore, the small sample size placed restrictions on the number of variables that could be simultaneously analysed.

Another step that could improve the methodology in this study would be to include a community sample as ascertainment bias is a problem in the majority of TS research. The assumption is that accessing a sample through clinical populations may be likely to result in the participants having particular characteristics relating to those who need or seek specialised treatment, rather than those who do not.

Although the majority of findings here have been non-significant, there was a significant difference between the TS and control groups on a measure of planning,

organisational and self-monitoring ability as well as on a measure of working memory. The difficulty here is that most measures of executive function are not readily generalisable to everyday function or may not detect subtle impairments that may have substantial impact upon everyday functioning. These are perhaps functions that are not readily elicited by clinical interview and clinicians should be mindful of this. In terms of treatment for subtle executive function impairments there are a number of methods involving simple behavioural and cognitive and psychoeducational training that could be employed.

Results often report significant results and seemingly discount non-significant ones. The results of the present study indicate that overall, adults with TS perform comparably to adults without TS on tests of everyday executive and attention function. However, the clinical and research experience of undertaking these assessment measures have enabled observations of qualitative differences in performance between the two groups on some of these measures that were not highlighted by the manualised scoring criteria.

The intended purpose of assessing executive function in a way that makes it applicable to everyday ability has been a compounding yet insightful experience. This was compounded by the fact that only one assessment battery was available purporting to measure executive function in an 'ecologically valid' way. This makes comparison with other studies difficult due to the reductionist nature of experimentally designed measures of executive function. Even so, there is great debate about the function or set of functions measured by different EF tasks. An insight gained here is that executive function is likely to be a multifaceted set of

abilities that are probably not functionally discriminable because often one part of an EF set requires another in order to carry out the whole of a given task. If this hypothesis is true, then subtle executive functions might be best measured by functional ability rather than by individual task analysis.

Most neuropsychological research in TS has focused on children. If executive functions in particular are not likely to be fully developed before early adolescence (Giedd, Blumenthal, Jeffries, Castellanos, Liu, Zijdenbos, Paus, Evans & Rapoport, 1999), then it is possible that the inconclusive evidence of executive function deficit in TS may be a reflection of this. Further research is needed to clarify the presence and nature of executive function impairments in adults. Specifically targeted research of everyday executive function, particularly planning, organisational abilities and self-monitoring may also elucidate the presence of subtle executive functions, in adults with TS which may have significant impact on everyday functioning. In addition, such research would enable further hypothesis testing of a neuropsychological model of TS and how this may relate to neuroanatomic and neurochemical correlates. A relative wealth of research has studied the effect of co-morbid ADHD and neuropsychological variables, however few have investigated co-morbidity with OCD in this way. Future research might clarify this issue, particularly as pure OCD is purported to involve similar frontal regions and to have a particular neuropsychological profile of executive impairment (Aycicegi, Dinn & Harris, 2002).

The findings of the present study do not indicate any overall differences in performance between those with and without TS. However, significant group differences found here on a measure of planning, organisational ability, self-

monitoring and on working memory, need to be interpreted with caution because of the small sample size and the nature of the tests used. This does not mean that executive function difficulties do not exist in a TS population. Instead, the results of this study indicate that no overall discernible group differences were found on the BADS battery between TS and non-TS participants, and this may have been for a number of reasons.

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**Appendix: 1 Public Domain Briefing Paper**

Literature Review:

Is There Any Evidence For Psychologically Mediated Psychopathology In Tourette Syndrome: A review and hypothesis of process mechanisms involved

Empirical Paper:

Executive and Attentional Function in Adults with Tourette Syndrome with and without Co-morbid Obsessive Compulsive Disorder

**Public Domain Briefing Paper**

Thesis Titles

Literature Review:

**Is There Any Evidence For Psychologically Mediated Psychopathology In Tourette Syndrome: A review and hypothesis of process mechanisms involved**

Empirical Paper:

**Executive and Attentional Function in Adults with Tourette Syndrome with and without Co-morbid Obsessive Compulsive Disorder**

## Outline

This study was undertaken by Caroline Formby in partial fulfilment of requirements for the degree of Doctor of Clinical Psychology (Clin.Psy.D) at the University of Birmingham. The research was supervised by Dr Chris Jones and Andy Papadopolous with collaboration from Dr. Rickards at the Queen Elizabeth Psychiatric Hospital (QEPH). Two papers were submitted, a literature review and an empirical paper. The literature review sought to determine whether there was evidence for psychologically mediated psychopathology in people with Tourette Syndrome (TS). It was highlighted that a number of factors indicated that this was so. A hypothetical psychological model was provided as a framework in which the reciprocal relationships between TS and co-morbid OCD and ADHD as neurological vulnerabilities, peer and family relationships, internalising and externalising behavioural difficulties might be mediated by self-representation and self-efficacy to result in the development of psychopathology. Clinical implications and directions for future research were discussed. The empirical paper was a neuropsychological study, which sought to determine executive and attention function ability in adults with TS with and without co-morbid Obsessive Compulsive Disorder (OCD), compared with a matched group of control participants. In addition, the predictive power of tic severity upon all cognitive variables and OCD severity was investigated. Overall, there were found to be few differences between the groups on measures of executive and attention function. However, performance on two individual subtests measuring planning, organisational skills and self-monitoring and auditory-verbal working memory was significantly poorer in the TS group. No differences were seen between the TS with and TS without OCD groups. Furthermore, tic severity was found to be significantly predictive of a task measuring cognitive flexibility and response suppression, suggestion that as tic severity increases, cognitive flexibility and response suppression decreases. These results were interpreted with caution because of the relatively small sample size and the nature of the assessment measures employed.

## Background

Tourette Syndrome (TS) is a neuropsychiatric disorder, with a mean age of onset of 5-7 years old. It is characterised by multiple motor tics and at least one vocal tic, that have lasted for at least one year. TS is typically episodic over the lifespan, although it

has been found that between half and two thirds of diagnosed child cases resolve completely or that tic symptoms are significantly reduced by early adulthood (Leckman et al., 1998). Prevalence rates for Tourette Syndrome have been estimated to be around 5 in 10 000 children (Robertson, 2000).

The precise neural mechanisms involved with TS are still unclear. However, it has been proposed that the basal ganglia plays a major role in the manifestation of tic symptoms. The marked co-morbidity of Obsessive Compulsive Disorder (OCD) with TS, has led to hypotheses that these disorders might be mediated by the same genetic trait that is responsible for abnormal functioning of the basal ganglia thalmo cortical pathways, causing deficiency of serotonin. It has been broadly hypothesised that there are several basal ganglia thalmo-cortical pathways that might explain different aspects of cognitive control including attentional and executive functions, three of which are the prefrontal, orbitofrontal and anterior cingulate pathways (Casey, Durston & Fossella, 2001).

Executive and attention functions have been described as those that determine the 'how' and 'whether or when' of behaviour as opposed to the 'what' or 'how much' as do other cognitive functions such as memory. There is a lack of conclusive evidence to support a global neuropsychological profile of deficit in those with TS, and attention has focused more recently to the set of higher cognitive processes termed executive functions (EF). Given the neurobiological correlates of TS and EF, relatively few studies have attempted to assess this set of functions independently results have again been inconclusive with some studies finding differences and others finding no difference compared with control groups (e.g. Ozonoff & Jensen, 1999). The current study set out to determine a variety of facets of executive and attentional function in adults with TS using measures designed to be more 'ecologically' valid. This is because traditional neuropsychological tests have been criticised for being unable to bridge the gap between experimental performance and actual everyday performance measures (Sbordone, 2000).

## Empirical Paper

*Aims*

Three main hypotheses were tested, firstly that adults with TS would perform more poorly than matched controls on measures of everyday executive function and attention. Secondly, adults with TS and co-morbid OCD would perform more poorly than adults with TS alone on measures of executive function. Thirdly that performance on executive and attention measures and severity of OCD would be predicted by tic severity.

*Participants*

Twenty-five adults with Tourette Syndrome were recruited via a specialist clinic for people with TS at the QEPH in Birmingham. Nineteen participated and one was excluded, leaving a total of eighteen participants with TS. Seventeen healthy participants without TS were recruited for comparison.

*Measures*
All participants were interviewed to ensure they met the inclusion criteria. The following assessment measures were undertaken:

1. Yale Brown Obsessive Compulsive Scale (Goodman, 1989). Measured presence and severity of obsessive or compulsive symptoms.
2. Wechsler Abbreviated Scale of Intelligence (4-subtest version) (Wechsler, 1999) was employed to measure full scale, verbal and performance IQ's.
3. Test of Everyday Attention (TEA) (Robertson et. al, 1994) was used to assess attention. The following subtests were administered; Elevator Counting; Elevator Counting with Distraction; Elevator Counting with Reversal; Telephone Search; Telephone Search while Counting.
4. The Behavioural Assessment of the Dysexecutive Syndrome (BADS) (Wilson et. al., 1996) was used to assess different facets of executive function. All subtests were administered. Also used was the DEX self-rating questionnaire.
5. Hospital Anxiety and Depression Scale (HADS) (Snaith & Zigmund, 1983) measured current levels of anxiety and depression.

Participants with Tourette Syndrome were also administered the following:

1. The Yale Global Tic Severity Scale (Leckman et. al., 1989). This was designed for use by clinicians to assess the current number, frequency, intensity, complexity and interference of motor and phonic tic symptoms.
2. The Diagnostic Confidence Index (Robertson et. al., 1999). Measured lifetime tic symptoms and severity, independent to current severity of symptoms.

**Summary of Findings**
- Two subtest items were significantly different with the TS group performing more poorly than the control group. These measured planning, organisational ability, and self-monitoring and working memory. There were no other differences observed.
- No difference was found on executive function measures between the TS with and TS without OCD groups.
- Tic severity was most predicted by a subtest of the executive function battery measuring cognitive flexibility and response suppression.

**Limitations of the Study**
- Relatively small sample size
- Low ceiling effect for the BADS battery

**Implications and Future Directions**
The results were interpreted with caution due to the small sample size. Future research might use 'ecologically valid' measures in conjunction with traditional measures of executive function to enable cross-validation of results. Further research needs to be conducted with adults with TS as most research studies have focused on children. This is especially true if full development of executive function doesn't happen before the onset of adolescence. More neuropsychological research needs to

focus on assessing TS co-morbid with OCD as there is little empirical evidence in this area.

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Sbordone, R. J. (2000). Ecological validity issues in neuropsychological Testing. *Brain Injury Journal, 4,* 10-12.

**Further Information**

The literature review and empirical paper findings are reported in:

Literature Review Paper

Formby, C. J. (2003). Is There Any Evidence For Psychologically Mediated Psychopathology In Tourette Syndrome: A review and hypothesis of process mechanisms involved. *Clin.Psy.D., Volume I,* School of Psychology, University of Birmingham.

Empirical Paper

Formby, C. J. (2003). Executive and Attentional Function in Adults with Tourette Syndrome with and without Co-morbid Obsessive Compulsive Disorder. *Clin.Psy.D., Volume I,* School of Psychology, University of Birmingham.

## Appendix 2: Patient Information Leaflet

# Patient Information

**A Study Investigating the Neuropsychology of Tourette Syndrome**

## What is the study about?

Some research that has already been done has suggested that sometimes, people with Tourette Syndrome might experience particular kinds of difficulties. These could be things like problems with planning ahead, doing lots of things at once, controlling impulses (e.g. feeling you have to do something and not being able to stop yourself) or solving difficult problems. This study aims to find out who may be more likely to experience such problems and whether the severity of tics affects these things.

## So if I do take part, what will the benefits be?

Even though lots of people with Tourette Syndrome do not experience these difficulties, this research will allow clinicians to find out who is most likely to. This would then allow staff to work with clients to build on their strengths and to learn strategies to help them with the things that they are not so good at. This will help people to have more self-confidence and to have an overall better quality of life.

## What will I have to do, if I take part?

You will be asked to answer a series of questionnaires with the researcher. Then you will be asked to complete a number of tasks designed to assess the sorts of abilities outlined above, like doing more than one thing at a time, planning etc. The whole assessment should take about 2 hours. The researcher will be able to see you on a day when you are already attending the [redacted] for a follow-up or appointment, or if you prefer, in your own home.

## So what exactly are these tasks?

The tasks you will be asked to do will be things like; answering general knowledge type questions, solving new problems and reasoning, searching a map, finding your way round a map with instructions etc. Some people find some these tasks a bit boring but most people do find them fun to do!

## Are there any risks to taking part in this research?

This study does not involve taking any drugs or any other invasive procedures. Taking part will not affect the treatment you are already receiving at the [redacted] from [redacted].

Everyone will be offered feedback on their performance over the telephone if they wish. If the assessment has discovered any difficulties that may be affecting you, further information and advice will be offered by the researcher, under the supervision of a qualified Clinical Psychologist, about how best to cope with these and what strategies you could use to maximise your strengths. Or you might be referred back to [redacted] if any other problems are found.

## What will happen to my information afterwards?

All information given by you will be treated as strictly confidential. Your name will not appear on anything. Once information has been gathered from everyone taking part, it will be entered into a computer and analysed by the researcher.

The findings of the study will be written up by the researcher, handed in and assessed by an examiner at the University of Birmingham. It will then be sent to a professional journal to be published.

For everyone who said they wanted to receive them, a summary of these findings will also be sent to people who took part in the study.

## What if I do not want to take part?

There is no pressure for you to take part in this research and the treatment you are already receiving will in no way be affected by your decision.

## Who else will be taking part?

Other clients who attend [redacted] Tourette Syndrome Clinic will also be taking part in this study.

## What if I have more questions?

If you have further questions about this research, you can contact the researcher's supervisor, Mr Andy Papadopolous or the researcher at the addresses given below.

## What if I say yes, then change my mind?

If you do not want to take part, or want to pull out of the research at any time, that is okay. All you have to do is let the researcher know. Again, this will in no way affect the treatment you already receive.

## Who is the researcher?

The researcher is Caroline Formby, a Trainee Clinical Psychologist based at the University of Birmingham. You can contact her or her supervisor Andy Papadopolous, at the addresses below:

Caroline Formby
[redacted]

Andy Papadopolous
[redacted]

**Appendix 3: Interview Schedules and Procedures for Both TS and Control Groups**

(i) **Interview Schedule**

(ii) **Yale Global Tic Severity Scale**

(iii) **Diagnostic Confidence Index**

(iv) **Yale-Brown Obsessive Compulsive Scale**

(v) **WASI Record Form**

(vi) **TEA Record Form**

(vii) **BADS Record Form**

(viii) **DEX Questionnaire**

(ix) **HADS Scale**

(x) **Consent Forms**

# Participant Interview Schedule

Participant number

Date of assessment:.....

Gender M F

Age in years ..... Months.....

Age at diagnosis of TS..... Age Onset.....

Diagnosis of OCD? Family History.....

Diagnosis of ADHD?

Suffer from other medical condition that might affect ability to think or remember, e.g. head injury, addiction?

Currently suffer from any other psychological / psychiatric condition e.g. depression, anxiety, psychosis etc?

Current medication?

Handedness R L both

Reading Glasses Y N

Hearing Aid Y N

Colour Blindness Y N

Age finish education .....

O'level/GCSE A'level Diploma Degree higher

Current occupation (if unemployed, last occupation)

Consent form signed? Y N

Feedback phone summary of findings by post

# Yale Global Tic Severity Scale[1]

Instructions: This clinical rating scale is designed to rate the overall severity of tic symptoms across a range of dimensions (number, frequency, intensity, complexity and interference). The final rating is based on all available information and reflects the clinician's overall impression for each of the items rated.
The clinician should first compile a list of motor and phonic tics present during the past week as reported by the patient and observed during the evaluation. Then, the clinician proceeds with the questions concerning each rating dimension, using the content of the anchor point as a guide.
As reproduced here, the scale is intended for clinical rather than research use.

**NUMBER OF TICS** **Motor score**_____ **Phonic score** _____

**0** = None.
**1** = Single tic.
**2** = Multiple discrete tics (2-5).
**3** = Multiple discrete tics (>5).
**4** = Multiple discrete tics plus at least one orchestrated pattern of multiple simultaneous or sequential tics where it is difficult to distinguish discrete tics.
**5** = Multiple discrete tics plus several (>2) orchestrated paroxysms of multiple simultaneous or sequential tics where it is difficult to distinguish discrete tics.

**FREQUENCY** **Motor score**_____ **Phonic score** _____

**0** = None, no evidence of specific behaviours.
**1** = Rarely, specific tic behaviours have been present during previous week. These behaviours occur infrequently, often not on a daily basis. If bouts of tics occur, they are brief and uncommon.
**2** = Occasionally, specific tic behaviours are usually present on a daily basis, but there are long tic-free intervals during the day. Bouts of tics may occur on occasion and are not sustained for more than a few minutes at a time.
**3** = Frequently, specific tic behaviours are present on a daily basis. Tic-free intervals as long as 3 hours are not uncommon. Bouts of tics occur regularly, but maybe limited to a single setting.
**4** = Almost Always, specific tic behaviours are present virtually every waking hour of every day, and periods of sustained tic behaviours occur regularly. Bouts of tics are common and are no limited to a single setting.
**5** = Always, specific tic behaviours are present virtually all of the time. Tic-free intervals are difficult to identify and do not last more than 5 to 10 minutes, at most.

**INTENSITY** **Motor score**_____ **Phonic score** _____

**0** = Absent.
**1** = Minimal intensity, tics not visible or audible (based solely on patient's private experience) or tics are less forceful than comparable voluntary actions and are typically not noticed because of their intensity.

---

[1] Leckman et. al., (1989)

**2** = Mild intensity, tics are not more forceful than comparable voluntary actions or utterances and are typically not noticed because of their intensity.
**3** = Moderate intensity, tics are more forceful than comparable voluntary actions, but are not outside the range of normal expression for comparable voluntary actions or utterances. They may call attention to the individual because of their forceful character.
**4** = Marked intensity, tics are more forceful than comparable voluntary actions or utterances and typically have an "exaggerated" character. Such tics frequently call attention to the individual because of their forceful and exaggerated character.
**5** = Severe intensity, tics are extremely forceful and exaggerated in expression. These tics call attention to the individual and may result in risk of physical injury (accidental, provoked or self-inflicted) because of their forceful expression.

**COMPLEXITY** **Motor score**_____ **Phonic score** _____

**0** = None, if present, all tics are clearly "simple" (sudden, brief, purposeless) in character.
**1** = Borderline, some tics are not clearly "simple" in character.
**2** = Mild, some tics are clearly "complex" (purposive in appearance) and mimic brief "automatic" behaviours, such as grooming syllables or brief meaningful utterances such as, "ah huh," "hi," that could be readily camouflaged.
**3** = Moderate, some tics are more "complex" (more purposive and sustained in appearance) and may occur in orchestrated bouts that would be difficult to camouflage, but could be rationalized or "explained" as normal behaviour or speech (picking, tapping, saying "you bet" or "honey," brief echolalia).
**4** = Marked, some tics are very "complex" in character and tend to occur in sustained orchestrated bouts that would be difficult to camouflage and could not be easily rationalized as normal behaviour or speech because of their duration and/or their unusual, inappropriate, bizarre, or obscene character (a lengthy facial contortion, touching genitals, echolalia, speech atypicalities, longer bouts of saying "what do you mean" repeatedly, or saying "fu" or "sh").
**5** = Severe, some tics involve lengthy bouts or orchestrated behaviour or speech that would be impossible to camouflage or successfully rationalize as normal because of their duration and/or extremely unusual, inappropriate, bizarre or obscene character (lengthy displays or utterances often involving copropraxia, self-abuse behaviour or coprolalia).

**INTERFERENCE** **Motor score**_____ **Phonic score** _____

**0** = None.
**1** = Minimal, when tics are present, they do not interrupt the flow of behaviour or speech.
**2** = Mild, when tics are present, they occasionally interrupt the flow of behaviour or speech.
**3** = Moderate, when tics are present, they frequently interrupt the flow of behaviour or speech.
**4** = Marked, when tics are present, they frequently interrupt the flow of behaviour or speech, and they occasionally disrupt intended action or communication.
**5** = Severe, when tics are present, they frequently disrupt intended action or communication.

**IMPAIRMENT** (Rate overall impairment for motor and phonic tics)

**0** = None.
**10** = Minimal, tics associated with subtle difficulties in self-esteem, family life, social acceptance, or school or job functioning (infrequent upset or concern about tics vis-à-vis the future, periodic, slight increase in family tensions because of tics, friends or acquaintances may occasionally notice or comment about tics in an upsetting way).
**20** = Mild, tics associated with minor difficulties in self-esteem, family life, social acceptance, or school or job functioning.
**30** = Moderate, tics associated with some clear problems in self-esteem, family life, social acceptance, or school or job functioning (episodes of dysphoria, periodic distress and upheaval in the family, frequent teasing by peers or episodic social avoidance, periodic interference in school or job performance because of tics).
**40** = Marked, tics associated with major difficulties in self-esteem, family life, social acceptance, or school or job functioning.
**50** = Severe, tics associated with extreme difficulties in self-esteem, family life, social acceptance or school or job functioning (severe depression with suicidal ideation, disruption of the family, separation, divorce, residential placement, disruption of social ties---severely restricted life because of social sigma and social avoidance, removal from school or loss of job).

# TOURETTE'S SYNDROME DIAGNOSTIC "CONFIDENCE INDEX"[1]

III. RATE DIAGNOSTIC CONFERENCE FACTORS

Enter the weighting score (shown in parentheses) for each item present. If absent, enter zero. If unknown, enter "N".

A. Coprolalia (15) IIIA. ☐☐

B. Echophenomena

1. Echopraxia (5) IIIB1. ☐
2. Echolalia (5) IIIB2. ☐
3. Palilalia (5) IIIB3. ☐

C. Complex Tics (Simple Tics Must be Present)

1. Complex Motor Tics (7) IIIC1. ☐
2. Complex Vocal Tics (12) IIIC2. ☐

D. Temporal Features

1. Age at Onset < 12 years (4) IIID1. ☐
2. Waxing and Waning Course (7) IIID2. ☐
3. Tics Began IIID3. ☐
4. 5 or more Motor Tic Types (2) IIID4. ☐
5. 3 or more Vocal Tic Types (2) IIID5. ☐
6. Multiple Motor Tic Body Locations (2) IIID6. ☐
7. Tics have varied with some tics disappearing and new tics appearing over time (4) IIID7. ☐
8. Environment Dependent (not only during stress) (1) IIID8. ☐

[1] Robertson et. al. (1999)

E. Subjective and Cognitive Experiences

| | | |
|---|---|---|
| 1. | Tics Voluntarily Suppressible (1) | IIIE1. ☐ |
| 2. | Attempts to Suppress Tics (1) | IIIE2. ☐ |
| 3. | Rebound Effect After Suppression (2) | IIIE3. ☐ |
| 4. | Premonitory Sensations (4) | IIIE4. ☐ |
| 5. | Sense of Relief After Tics (1) | IIIE5. ☐ |
| 6. | Tics are Suggestible (2) | IIIE6. ☐ |

F. Tics Severity

| | | |
|---|---|---|
| 1. | Frequent Tics (> 1 pre minute at times) (2) | IIIF1. ☐ |
| 2. | Orchestrated Sequences of Tics (4) | IIIF2. ☐ |
| 3. | Causes Distress (2) | IIIF3. ☐ |
| 4. | Sought Treatment or Diagnosis (2) | IIIF4. ☐ |

G. Duration of Tics 2 or more years (2) IIIG. ☐

H. Tics confirmed by 1 or more reliable observers (4) IIIH. ☐

I. Absence of other medical problems that might cause tics (e.g. prior stimulant therapy, history of encephalitis) (1) III I. ☐

TOTAL "CONFIDENCE" SCORE ☐☐☐
(ADD IIIA I)

# Yale-Brown Obsessive Compulsive Scale

Patient Name: Date:

| | None | Mild | Moderate | Severe | Extreme |
|---|---|---|---|---|---|
| 1. Time spent on obsessions | 0 | 1 | 2 | 3 | 4 |
| 2. Interference from obsessions | 0 | 1 | 2 | 3 | 4 |
| 3. Distress of obsessions | 0 | 1 | 2 | 3 | 4 |
| | Definitely resists | | | | Completely yields |
| 4. Resistance | 0 | 1 | 2 | 3 | 4 |
| | Complete Control | Much Control | Moderate Control | Little Control | No Control |
| 5. Control over obsessions | 0 | 1 | 2 | 3 | 4 |

Obsession Subtotal (Add items 1-5) =

| | None | Mild | Moderate | Severe | Extreme |
|---|---|---|---|---|---|
| 6. Time spent on compulsions | 0 | 1 | 2 | 3 | 4 |
| 7. Interference from compulsions | 0 | 1 | 2 | 3 | 4 |
| 8. Distress of compulsions | 0 | 1 | 2 | 3 | 4 |
| | Definitely resists | | | | Completely yields |
| 9. Resistance | 0 | 1 | 2 | 3 | 4 |
| | Complete Control | Much Control | Moderate Control | Little Control | No Control |
| 10. Control over compulsions | 0 | 1 | 2 | 3 | 4 |

Compulsion Subtotal (Add items 6-10) =

**TOTAL Y-BOCS Score =**

| | Excellent | | | | Absent |
|---|---|---|---|---|---|
| 11. Insight into O-C | 0 | 1 | 2 | 3 | 4 |

# WASI™ Record Form

WECHSLER *ABBREVIATED* SCALE OF INTELLIGENCE™

Name _____ ID _____

Address/School _____ Grade/ Highest Education _____

Examiner _____

| | Year | Month | Day |
|---|---|---|---|
| Date of Testing | | | |
| Date of Birth | | | |
| Age | | | |

## Subtest Scores

| Subtest | Raw Score | T Score |
|---|---|---|
| Vocabulary | | |
| Block Design | | |
| Similarities | | |
| Matrix Reasoning | | |

Sums of *T* Scores

| Verbal | Performance | |
|---|---|---|
| 4-Subtest | | 2-Subtest |
| Full Scale | | |

## WASI IQ Scores

| | Sum of *T* Scores | IQ | Percentile | ___% Confidence Interval |
|---|---|---|---|---|
| Verb. | | | | – |
| Perf. | | | | – |
| Full–4 | | | | – |
| Full–2 | | | | – |

## Prediction Intervals

| | WISC–III | | WAIS–III | |
|---|---|---|---|---|
| | 90% | 68% | 90% | 68% |
| Verb. | – | – | – | – |
| Perf. | – | – | – | – |
| Full–4 | – | – | – | – |

## Profile of Subtest Scores

| | Verbal | | Performance | |
|---|---|---|---|---|
| | V | S | BD | MR |
| 80 | | | | |
| 75 | | | | |
| 70 | | | | |
| 65 | | | | |
| 60 | | | | |
| 55 | | | | |
| 50 | | | | |
| 45 | | | | |
| 40 | | | | |
| 35 | | | | |
| 30 | | | | |
| 25 | | | | |
| 20 | | | | |

## Profile of IQ Scores

| | VIQ | PIQ | FSIQ |
|---|---|---|---|
| 160 | | | |
| 155 | | | |
| 150 | | | |
| 145 | | | |
| 140 | | | |
| 135 | | | |
| 130 | | | |
| 125 | | | |
| 120 | | | |
| 115 | | | |
| 110 | | | |
| 105 | | | |
| 100 | | | |
| 95 | | | |
| 90 | | | |
| 85 | | | |
| 80 | | | |
| 75 | | | |
| 70 | | | |
| 65 | | | |
| 60 | | | |
| 55 | | | |
| 50 | | | |

THE PSYCHOLOGICAL CORPORATION®
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2 3 4 5 6 7 8 9 10 11 12 A B C D E

# 1. Vocabulary


**Start Point**

Ages 6–8: Item 5
Ages 9–89: Item 9

**Reverse Rule**

**All Ages:** Administer Items 1–4 in forward sequence if score of 0 or 1 on Item 5 or 6.

**Ages 9–89:** Administer Items 5–8 in reverse sequence if score of 0 or 1 on Item 9 or 10.

**Discontinue Rule**

After 5 consecutive scores of 0


**Stop Point**

Ages 6–8: After Item 30
Ages 9–11: After Item 34
Ages 12–16: After Item 38
Ages 17–89: No stop point

**Scoring Rule**

Items 1–4: 0 or 1
Items 5–42: 0, 1, or 2

| Item | Response | Score |
|---|---|---|
| 1. Fish | | (0 or 1) |
| 2. Shovel | | |
| 3. Map | | |
| 4. Shell | | |
| 5. Shirt | | (0, 1, 2) |
| 6. Shoe | | |
| 7. Flashlight | | |
| 8. Car | | |
| 9. Bird | | |
| 10. Calendar | | |
| 11. Number | | |
| 12. Bell | | |
| 13. Lunch | | |
| 14. Police | | |
| 15. Vacation | | |
| 16. Pet | | |
| 17. Balloon | | |
| 18. Transform | | |
| 19. Alligator | | |

Continue

# 1. Vocabulary *(Continued)*

| Item | Response | Score (0, 1, 2) |
|---|---|---|
| 20. Cart | | |
| 21. Blame | | |
| 22. Dance | | |
| 23. Purpose | | |
| 24. Entertain | | |
| 25. Famous | | |
| 26. Reveal | | |
| 27. Decade | | |
| 28. Tradition | | |
| 29. Rejoice | | |
| 30. Enthusiastic | | |
| 31. Improvise | | |
| 32. Impulse | | |
| 33. Haste | | |
| 34. Trend | | |
| 35. Intermittent | | |
| 36. Devout | | |
| 37. Impertinent | | |
| 38. Niche | | |
| 39. Presumptuous | | |
| 40. Formidable | | |
| 41. Ruminate | | |
| 42. Panacea | | |

STOP

STOP

STOP

**Maximum Raw Score**
Ages 6–8: 56
Ages 9–11: 64
Ages 12–16: 72
Ages 17–89: 80

**Total Raw Score**

# 2. Block Design

**Start Point**

Ages 6–8: Design 1
Ages 9–89: Design 3

**Reverse Rule**

Ages 9–89: Administer Items 1–2 in reverse sequence if score of 0 or 1 on Item 3 or 4.

**Discontinue Rule**

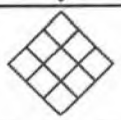
After 3 consecutive scores of 0

**Scoring Rule**

Items 1–4: 2 for a correct design on Trial 1
1 for a correct design on Trial 2
0 for incorrect designs on Trials 1 & 2

Items 5–13: 0–7

Examinee

| Design | Time Limit | Incorrect Design | Completion Time in Seconds | Correct Design | Score (Circle the appropriate score for each design.) |
|---|---|---|---|---|---|
| 6–8 1.  | 30" | Trial 1  Trial 2  | | Y N | 0 1 2 |
| 2.  | 60" | Trial 1  Trial 2  | | Y N | 0 1 2 |
| 9–89 3.  | 60" | Trial 1  Trial 2  | | Y N | 0 1 2 |
| 4.  | 60" | Trial 1  Trial 2  | | Y N | 0 1 2 |
| 5.  | 60" |  | | Y N | 0 21"–60" 4 16"–20" 5 11"–15" 6 1"–10" 7 |
| 6.  | 60" |  | | Y N | 0 21"–60" 4 16"–20" 5 11"–15" 6 1"–10" 7 |
| 7.  | 60" |  | | Y N | 0 21"–60" 4 16"–20" 5 11"–15" 6 1"–10" 7 |
| 8.  | 60" |  | | Y N | 0 21"–60" 4 16"–20" 5 11"–15" 6 1"–10" 7 |
| 9.  | 60" |  | | Y N | 0 21"–60" 4 16"–20" 5 11"–15" 6 1"–10" 7 |
| 10.  | 120" |  | | Y N | 0 66"–120" 4 46"–65" 5 31"–45" 6 1"–30" 7 |
| 11.  | 120" |  | | Y N | 0 76"–120" 4 56"–75" 5 41"–55" 6 1"–40" 7 |
| 12.  | 120" |  | | Y N | 0 76"–120" 4 56"–75" 5 41"–55" 6 1"–40" 7 |
| 13.  | 120" |  | | Y N | 0 76"–120" 4 56"–75" 5 41"–55" 6 1"–40" 7 |

Examiner

**Maximum Raw Score**
All Ages: 71

**Total Raw Score** ☐

# 3. Similarities


**Start Point**

Ages 6–8: Item 1
Ages 9–11: Item 5
Ages 12–89: Item 7

**Reverse Rule**

**Ages 9–89:** Administer Items 1–4 in forward sequence if score of 0 or 1 on Item 5 or 6.

**Ages 12–89:** Administer Items 5 & 6 in reverse sequence if score of 0 or 1 on Item 7 or 8.

**Discontinue Rule**

After 4 consecutive scores of 0


**Stop Point**

Ages 6–8: After Item 20
Ages 9–11: After Item 24
Ages 12–89: No stop point

**Scoring Rule**

Items 1–4: 0 or 1
Items 5–26: 0, 1, or 2

| | Item | Response | | | | | Score |
|---|---|---|---|---|---|---|---|
| | | | | | | | (0 or 1) |
| 6–8 | 1. Four-Wheeled | Ship | **BUS** | Bike | Train | | |
| | 2. Dining Items | **SPOON** | Pan | Bowl | Can Opener | | |
| | 3. Clothing | Jump Rope | Ball | **SHOES** | Crayons | | |
| | 4. Fruits | **BANANA** | Bean | Pumpkin | Potato | | |
| | | | | | | | (0, 1, 2) |
| 9–11 | 5. Red–Blue | | | | | | |
| | 6. Circle–Square | | | | | | |
| 12–89 | 7. Grapes–Strawberries | | | | | | |
| | 8. Cow–Bear | | | | | | |
| | 9. Plane–Bus | | | | | | |
| | 10. Shirt–Jacket | | | | | | |
| | 11. Pen–Pencil | | | | | | |
| | 12. Bowl–Plate | | | | | | |
| | 13. Love–Hate | | | | | | |
| | 14. TV–Newspaper | | | | | | |
| | 15. Smooth–Rough | | | | | | |
| | 16. Shoulder–Ankle | | | | | | |
| | 17. Sit–Run | | | | | | |
| | 18. Child–Adult | | | | | | |
| | 19. Steam–Cloud | | | | | | |
| | 20. Bird–Flower | | | | | | |
| 6–8 STOP | 21. More–Less | | | | | | |
| | 22. Photograph–Song | | | | | | |

Continue

# 3. Similarities *(Continued)*

| Item | Response | Score (0, 1, 2) |
|---|---|---|
| 23. Peace–War | | |
| 24. Capitalism–Socialism | | |
| 25. Tradition–Habit | | |
| 26. Freedom–Law | | |

9–11 STOP

**Maximum Raw Score**
Ages 6–8: 36
Ages 9–11: 44
Ages 12–89: 48

**Total Raw Score**

# 4. Matrix Reasoning


**Start Point**

Administer Sample Items A and B first.

Ages 6–8: Item 1
Ages 9–11: Item 5
Ages 12–44: Item 7
Ages 45–79: Item 5
Ages 80–89: Item 1

**Reverse Rule**

Ages 9–11 and Ages 45–79: Administer Items 1–4 in reverse sequence if score of 0 on Item 5 or 6.

Ages 12–44: Administer Items 1–6 in reverse sequence if score of 0 on Item 7 or 8.

**Discontinue Rule**

After 4 consecutive scores of 0 or after 4 scores of 0 on 5 consecutive items


**Stop Point**

Ages 6–8: After Item 28
Ages 9–11: After Item 32
Ages 12–44: No stop point
Ages 45–79: After Item 32
Ages 80–89: After Item 28

**Scoring Rule**

Items 1–35: 0 or 1

| Item | Response Options (Circle One) | Score (0 or 1) |
|---|---|---|
| A. | 1 2 3 4 5 DK | |
| B. | 1 2 3 4 5 DK | |
| 1. | 1 2 3 4 5 DK | |
| 2. | 1 2 3 4 5 DK | |
| 3. | 1 2 3 4 5 DK | |
| 4. | 1 2 3 4 5 DK | |
| 5. | 1 2 3 4 5 DK | |
| 6. | 1 2 3 4 5 DK | |
| 7. | 1 2 3 4 5 DK | |
| 8. | 1 2 3 4 5 DK | |
| 9. | 1 2 3 4 5 DK | |
| 10. | 1 2 3 4 5 DK | |
| 11. | 1 2 3 4 5 DK | |
| 12. | 1 2 3 4 5 DK | |
| 13. | 1 2 3 4 5 DK | |
| 14. | 1 2 3 4 5 DK | |
| 15. | 1 2 3 4 5 DK | |
| 16. | 1 2 3 4 5 DK | |
| 17. | 1 2 3 4 5 DK | |

| Item | Response Options (Circle One) | Score (0 or 1) |
|---|---|---|
| 18. | 1 2 3 4 5 DK | |
| 19. | 1 2 3 4 5 DK | |
| 20. | 1 2 3 4 5 DK | |
| 21. | 1 2 3 4 5 DK | |
| 22. | 1 2 3 4 5 DK | |
| 23. | 1 2 3 4 5 DK | |
| 24. | 1 2 3 4 5 DK | |
| 25. | 1 2 3 4 5 DK | |
| 26. | 1 2 3 4 5 DK | |
| 27. | 1 2 3 4 5 DK | |
| 28. | 1 2 3 4 5 DK | |
| 29. | 1 2 3 4 5 DK | |
| 30. | 1 2 3 4 5 DK | |
| 31. | 1 2 3 4 5 DK | |
| 32. | 1 2 3 4 5 DK | |
| 33. | 1 2 3 4 5 DK | |
| 34. | 1 2 3 4 5 DK | |
| 35. | 1 2 3 4 5 DK | |

6–8 80–89 STOP

9–11 45–79 STOP

**Maximum Raw Score**
Ages 6–8: 28
Ages 9–11: 32
Ages 12–44: 35
Ages 45–79: 32
Ages 80–89: 28

**Total Raw Score**

# The Test of Everyday Attention — Scoring sheet

Thames Valley Test Company

## Subject and test details

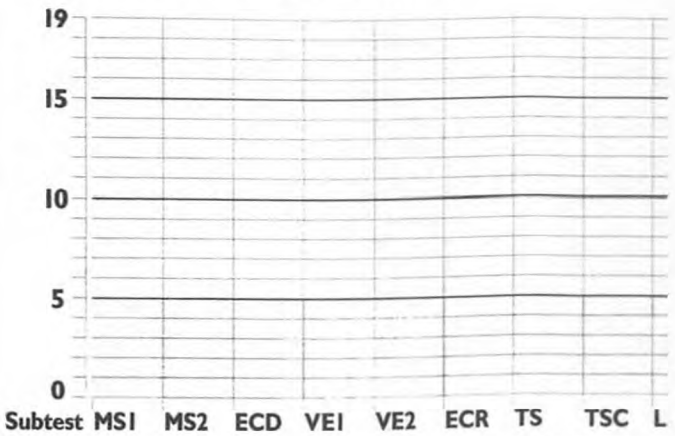
Name _____

Age _____

Date of test

Version A B C

Scaled-scores summary plot




## Score summary and scaled-scores

- **Subtest 1: Map Search** — Scaled-scores **MS1** — Percentile

  Symbols circled in one minute — see Appendix 1 Manual page 23 — **MS2**

  Symbols circled in two minutes — see Appendix 2 Manual page 24

- **Subtest 2: Elevator Counting**

  Correctly-counted strings — 7 = normal; 6 = possibly abnormal; ≤5 = abnormal

- **Subtest 3: Elevator Counting with Distraction** — Scaled-score **ECD** — Percentile

  Correctly-counted strings (Rule out hearing impairments) — see Appendix 3 Manual page 25

- **Subtest 4: Visual Elevator** — Scaled-scores **VE1** — Percentile

  Raw accuracy score — see Appendix 4a Manual page 25 — **VE2**

  Timing score — see Appendix 4c Manual page 26

- **Subtest 5: Elevator Counting with Reversal** — Scaled-score **ECR** — Percentile

  Correctly-counted strings — see Appendix 5 Manual page 27

- **Subtest 6: Telephone Search** — Scaled-score **TS** — Percentile

  Time per target score — see Appendix 6 Manual page 27

- **Subtest 7: Telephone Search While Counting** — Scaled-score **TSC** — Percentile

  Dual task decrement — see Appendix 7 Manual page 28

- **Subtest 8: Lottery** — Scaled-score **L** — Percentile

  Number of responses with *at least* one letter correct and in the correct position — see Appendix 8 Manual page 29

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**Thames Valley Test Company**
7–9 The Green, Flempton
Bury St Edmunds
Suffolk IP28 6EL England

*'We are interested in your concentration on a range of everyday tasks. I want you to imagine that you are on a long trip to Philadelphia (United States). I will ask you to do various tasks such as looking at maps and looking up telephone directories while you are on this imaginary trip. Let me explain the first task.'*

## Subtest 1: Map Search

For full text and procedure see Manual page 13.

Show subject the target symbol cue (version A, B or C in the test materials book).

- *'The symbol here shows where restaurants / garages / gas (petrol) stations can be found in the Philadelphia area. There are many symbols like this on the map.'*
- Show map and then turn over.
- *'Let's say you are with a family member or a friend...'*
- Give subject the map and red pen.
- After one minute swap red pen for blue pen.
- Stop after one further minute.

**Raw score**
Total symbols circled in one minute (red)

**Raw score**
Total symbols circled in two minutes (red + blue)

## Subtest 2: Elevator Counting

For full text and procedure see Manual page 14.

- *'Imagine you are in an elevator (lift) in your hotel...'*
- Play first example (ensure you are using tape A, B or C).
- *'That's right, you would be on the third floor.'*
  (Or play tape again.)
- Play second example.
  (Rewind and repeat, counting with the subject at first, until they get the correct answer on their own.)
- *'Now I would like you to do the same thing with another series of elevator tones.'*

| | 1 | 2 | 3 | 4 | 5 | 6 | 7 |
|---|---|---|---|---|---|---|---|
| A | 3 | 5 | 6 | 8 | 11 | 9 | 14 |
| B | 4 | 6 | 7 | 9 | 12 | 14 | 10 |
| C | 5 | 7 | 6 | 8 | 10 | 14 | 12 |

**Raw score**
Score 1 for each correctly-counted string (maximum = 7)

## Subtest 3: Elevator Counting with Distraction

For full text and procedure see Manual page 15.

- *'This time you will hear the same elevator tone but now there are also higher pitched tones...'*
- Play first example.
- *'That's right, you would be on the third floor.'*
  (Or play tape again.)
- Play second example.
  (Rewind and repeat, counting with the subject at first, until they get the correct answer on their own.)
- *'Now I would like you to do the same thing with another series of elevator tones.'*

| | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 |
|---|---|---|---|---|---|---|---|---|---|---|
| A | 2 | 4 | 6 | 8 | 7 | 10 | 9 | 12 | 11 | 14 |
| B | 3 | 5 | 6 | 7 | 8 | 11 | 10 | 14 | 12 | 9 |
| C | 4 | 3 | 7 | 6 | 8 | 12 | 11 | 9 | 10 | 14 |

**Raw score** Score 1 for each correctly-counted string

## Subtest 4: Visual Elevator

For full text and procedure see Manual page 16.

- *'Try to imagine that during your trip, you decide to stay in a large hotel...'*
- Show subject the first Visual Elevator practice item (labelled 'Practice 1' in version A, B and C in the test materials book).
- *'Look at this series of pictures...'*
- Go through Practice 1.
- Repeat Practice 1 as often as necessary until the subject gets the correct answer on their own.
- *'That's right, you would be on the second floor.'*
- *'Now try this next example.'*
- Show 'Practice 2' (next page of test materials book).
- *'That's right, you would be on the fourth floor.'*
- *'Now try and do the same with the next set of pictures...'*
- Prepare stopwatch to time each item.
- Subjects are allowed to make *one* correction on *each* item.

| | | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 |
|---|---|---|---|---|---|---|---|---|---|---|---|
| **Response** | | | | | | | | | | | |
| ✓/✗ | | | | | | | | | | | |
| **Time** *(sec)* | | | | | | | | | | | |
| Answers | **A** | 8 | 5 | 6 | 6 | 5 | 8 | 6 | 6 | 4 | 10 |
| | **B** | 8 | 3 | 2 | 5 | 8 | 4 | 9 | 2 | 10 | 6 |
| | **C** | 4 | 3 | 8 | 6 | 8 | 4 | 6 | 8 | 10 | 4 |
| Switches | **A** | 3 | 2 | 3 | 4 | 5 | 3 | 6 | 6 | 4 | 4 |
| | **B** | 4 | 2 | 3 | 3 | 4 | 3 | 5 | 3 | 6 | 4 |
| | **C** | 3 | 2 | 3 | 4 | 4 | 3 | 4 | 6 | 4 | 4 |

**Raw accuracy score**
Score 1 for each correctly-counted item (maximum = 10)

**Total time taken for correct items** (seconds) ÷ **Total number of switches for the correct items** = **Timing score** (seconds per switch)

## Subtest 5: Elevator Counting with Reversal

For full text and procedure see Manual page 18.

- Do not test subjects with severe brain damage.
- *'Now we are going to try something similar but a bit more complicated. Look again at what you did here.'*
  Point to Visual Elevator Practice 1 item.
- *'Remember how the big arrows tell you whether the elevator is going up or down? Now we are going to try an auditory (sound) version of this...'*
- Play first practice item, and count out loud: *'one – two – up – three – four – down – three – two...: so the answer is two.'*
- Rewind and play again for subject, pausing after each beep.
- Play second example. *'The answer is three.'*
- Play third example. *'The answer is three.'*
- Repeat the examples until the subject gets the correct answers on their own.

| | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 |
|---|---|---|---|---|---|---|---|---|---|---|
| A | 3 | 1 | 3 | 5 | 8 | 2 | 6 | 2 | 6 | 8 |
| B | 3 | 5 | 3 | 3 | 8 | 2 | 8 | 4 | 6 | 8 |
| C | 1 | 5 | 1 | 5 | 8 | 2 | 8 | 2 | 6 | 8 |

**Raw score** Score 1 for each correctly-counted string

## Subtest 6: Telephone Search

For full text and procedure see Manual page 19.

- *'In this exercise, you should imagine that you are using a telephone directory to look up various services while you are on your trip.'*
- *'Here we have the yellow pages you would see in a telephone directory, in this case it lists plumbers/restaurants/hotels.'*
- Show subject the target symbol cues (in the test materials book), the relevant yellow pages sheet, and a pen.
- *'Imagine that during your vacation (holiday)...'*
- Prepare stopwatch.
- *'Begin'*
- Start watch as subject makes first mark.
  Stop watch when subject puts cross in box.

**Time taken** (seconds)

÷ = **A** **Raw score** (time per target score)

**Total number of correctly-circled symbols** (ignore any false positives)

## Subtest 7: Telephone Search While Counting

For full text and procedure see Manual page 20.

- *'Now you will search through a different set of yellow pages for the same double symbols as in the last subtest. But this time, I will ask you to do a second and equally important task at the same time – counting a number of series of tones on the tape recorder...'*
- Show subject the restaurants/hotels/plumbers yellow pages.
- Play practice item, and count with the subject.
- *'So you will be looking for the same double symbols...'*
- *'Get ready...'*
- Prepare stopwatch.
- Subject starts when tape-voice says *'Ready...'*
- Start watch as subject makes first mark.
  Stop watch when subject puts cross in box.
- Note the number of strings of tones which the subject attempts in the box marked **C** below.

| | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 |
|---|---|---|---|---|---|---|---|---|---|
| **Response** | | | | | | | | | |
| ✓/✗ | | | | | | | | | |
| Answers **A** | 4 | 8 | 3 | 6 | 1 | 12 | 2 | 5 | 7 |
| **B** | 6 | 1 | 12 | 2 | 5 | 7 | 5 | 9 | 2 |
| **C** | 2 | 5 | 7 | 5 | 9 | 2 | 6 | 11 | 3 |

| | 10 | 11 | 12 | 13 | 14 | 15 | 16 | 17 | 18 |
|---|---|---|---|---|---|---|---|---|---|
| **Response** | | | | | | | | | |
| ✓/✗ | | | | | | | | | |
| Answers **A** | 5 | 9 | 2 | 6 | 11 | 3 | 6 | 3 | 5 |
| **B** | 6 | 11 | 3 | 6 | 3 | 5 | 5 | 4 | 8 |
| **C** | 6 | 3 | 5 | 5 | 4 | 8 | 3 | 6 | 12 |

**Time taken** (seconds)

÷ = **B** Time per target score

**Total number of correctly-circled symbols** (ignore any false positives)

Number of strings of tones correctly counted

÷ **C** = **D** Proportion correctly counted

Number of strings of tones attempted

Re-enter **B** here

÷ = **E** Time per target weighted for accuracy of tone-counting

Re-enter **D** here

Re-enter **E** here

– = **Dual task decrement**

**Subtract** Re-enter **A** here

## Subtest 8: Lottery

For full text and procedure see Manual page 21.

- *'While you are on your trip, you become interested in the state lottery...'*
- Show subject the target cues (version A, B or C in the test materials book) and give the subject a piece of paper and a pen.
- *'The radio programme goes on for quite a long time...'*
- Play the tape.
- Stop after first number ending in 55 / 88 / 33.
  Rewind and repeat until the subject responds correctly.

| | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 |
|---|---|---|---|---|---|---|---|---|---|---|
| **A** | HH | EA | LV | DR | CF | QO | TS | FN | FA | XT |
| **B** | WG | WA | LW | CT | YK | UF | CM | UA | RN | HY |
| **C** | FN | AT | XW | YG | EA | WN | RC | FO | HU | IT |

**Raw score** Score 1 for each response with *at least* one letter correct *and* in the correct position (maximum = 10)

BADS

# Scoring sheet

Thames Valley Test Company

## Subject and test details

Name _____

Age _____

Date of test

## Before you start the test battery

- Ensure that you have all the test materials, a stopwatch, a tape recorder, set of coloured pens, a pencil, eraser, spare paper, and water for the action program.

## Profile score summary

**Test 1: Rule shift cards** ____

**Test 2: Action program** ____

**Test 3: Key search** ____

**Test 4: Temporal judgement** ____

**Test 5: Zoo map** ____

**Test 6: Modified six elements** ____

**Total profile score** (max = 24)

**Standardised score**
(Manual Table 5, p.16)

**Age corrected standardised score**
(Manual Table 5, p.16)

**Overall classification**

- ☐ Impaired
- ☐ Borderline
- ☐ Low average
- ☐ Average
- ☐ High Average
- ☐ Superior
- ☐ Very superior

## Test 1: Rule shift cards

For full text and procedure see Manual p. 8

### Trial 1

- Put the playing card booklet, unopened, between you and the subject and have the rule sheet ready.
- *'This is a booklet of playing cards. I am going to turn over...'*
- Place Rule 1 in front of the subject ('Say 'yes' to red, 'no' to black').
- Remember to omit page 0 for this trial – start with the 2 of ♦.
- Time the trial.

| | Correct response | Subject's response |
|---|---|---|
| 1 | Y | |
| 2 | N | |
| 3 | N | |
| 4 | N | |
| 5 | Y | |
| 6 | Y | |
| 7 | Y | |
| 8 | Y | |
| 9 | N | |
| 10 | Y | |
| 11 | Y | |
| 12 | N | |
| 13 | Y | |
| 14 | N | |
| 15 | N | |
| 16 | N | |
| 17 | Y | |
| 18 | N | |
| 19 | Y | |
| 20 | N | |

**Total errors** ____

**Time taken** ____

**Note that Trial 1 is not used to calculate the profile score**

### Trial 2

- *'I am going to turn over the set of cards again now...'*
- Place Rule 2 in front of the subject ('Say 'yes' if the card is the same colour as the last one, otherwise say 'no' ').
- Remember to start on page 0 – the 4 of ♥.
- Time the trial.

| | Correct response | Subject's response |
|---|---|---|
| 1 | Y | |
| 2 | N | |
| 3 | Y | |
| 4 | Y | |
| 5 | N | |
| 6 | Y | |
| 7 | Y | |
| 8 | Y | |
| 9 | N | |
| 10 | N | |
| 11 | Y | |
| 12 | N | |
| 13 | N | |
| 14 | N | |
| 15 | Y | |
| 16 | Y | |
| 17 | N | |
| 18 | N | |
| 19 | N | |
| 20 | N | |

**Total errors** ____

| Total errors | Profile score |
|---|---|
| 0 | 4 |
| 1–3 | 3 |
| 4–6 | 2 |
| 7–9 | 1 |
| ≥10 | 0 |

**Time taken** ____

If time taken is greater than 67 seconds subtract 1 from profile score

**Total profile score** ☐

## Test 2: Action program

For full text and procedure see Manual p. 8

- Fill the beaker to two-thirds full of water (out of sight of the subject) and place the equipment in front of the subject.
- *'If you look at the bottom of this tube you will see a small cork...'*
- Start stopwatch
- If necessary, prompt after 2 minutes *'I'll give you some help'*, and remove the lid with the wire hook. *'Try to complete the task now'.*
- If necessary, prompt after a further 2 minutes by attaching the screw top to the container.

**Tick each stage completed independently**

- ☐ Removes lid from beaker using wire hook
- ☐ Attaches screw top to container
- ☐ Fills container with water
- ☐ Pours one containerful of water into tube containing cork
- ☐ Pours second containerful of water into tube containing cork

**Raw score**

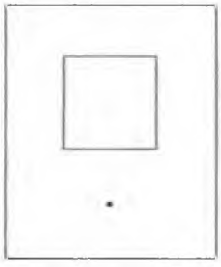
| Raw score | Profile score |
|---|---|
| 5 | 4 |
| 4 | 3 |
| 3 | 2 |
| 2 | 1 |
| ≤1 | 0 |

**Total profile score** ☐

## Test 3: Key search

For full text and procedure see Manual p. 9

- Place a photocopy of the response sheet in front of the subject.
- *'I want you to imagine that this square is a large field...'*
- *'Starting from this dot I want you to draw a line with the pen to show me where you would walk to search the field...'*
- If the subject does not grasp the idea, demonstrate on another piece of paper.
- *'Although I will be timing you there is no time limit...'*
- Start the stopwatch.
- Make notes here. These could indicate, for example, the order in which the subject makes marks. This will help you to calculate the score later.
- For scoring criteria see Appendices 9.1 and 9.2 in Manual pp. 20–22.



**Entering the field** **Raw score**

- within 10mm of a corner (base of square) = 3
- base of square (other than within 10mm of corners) = 2
- somewhere else = 1

**Finishing the search**

- within 10mm of any corner = 3
- base of square (other than within 10mm of corners) = 2
- somewhere else = 1

**Making a continuous line** = 1

**Making all parallel lines** = 1

**Making all vertical/horizontal lines** = 1

**Search patterns**

- followed one of our pre-defined search patterns (see Appendix 9.1, Manual pp. 20–21) or super-imposed one pre-determined pattern over another = 5 or 3
- duplicated or combined one or more of our pre-defined search patterns = 2
- followed some other obviously systematic, but inefficient and/or unsuccessful search pattern = 1
- ad hoc – not systematic or pre-planned = 0

**Has made an obvious effort to cover all the ground** = 1

**Using their chosen pattern, they would find the keys (95% certainty)** = 1

**Time taken** **Total raw score** **Profile score**

| Raw score | Profile score |
|---|---|
| 14–16 | 4 |
| 11–13 | 3 |
| 8–10 | 2 |
| 5–7 | 1 |
| ≤4 | 0 |

**If time taken is greater than 95 seconds subtract 1 from profile score** –

**Total profile score** ☐

## Test 4: Temporal judgement

For full text and procedure see Manual p. 9

- *'I'm going to ask you to estimate how long it takes to do four things...'*

**Question 1**
How long does it take to do a routine dental check up?

**Raw score**
If between 5 & 15 mins score 1, otherwise 0

**Question 2**
How long does it take a window cleaner to clean the windows of an average size house?

If between 15 & 25 mins score 1, otherwise 0

**Question 3**
How long do most dogs live for?

If between 9 & 15 years score 1, otherwise 0

**Question 4**
How long does it take to blow up a party balloon?

If between 50 & 70 secs score 1, otherwise 0

**Total raw score** **= total profile score** ☐

## Test 5: Zoo map

For full text and procedure see Manual p. 9

### Version 1

- Place a photocopy of Zoo map version 1 in front of the subject.
- *'Here is a map of a zoo. Your task is to plan a route around the zoo to visit all the places indicated in the instructions...'*
- Allow the subject to read the instructions (aloud).
- Clarify the rules by reading them again.
- *'While I will use this stopwatch to see how long it takes you to do the task, the time really is not important...'*
- Start the stopwatch.
- For scoring criteria see Appendix 9.3 in Manual p. 23.

| Note subject's sequence | Each correct scores 1 | Correct responses | Occasions each path used more than once |
|---|---|---|---|
| | | Entrance | A |
| | | Llamas/Cafe/Elephants | B |
| | | Elephants/Cafe | C |
| | | Cafe/Elephants/Llamas | D |
| | | Bears | E |
| | | Lions | F |
| | | Bird sanctuary | G |
| | | Picnic area | H |
| | | | I |
| | | | J |
| | | | K |
| | | | L |
| | | | M |

**Sequence score** **Total**

**Planning time** **Total time**

**Errors**

Total number of occasions paths used more than once (from above)

Number of deviations from the path (i.e. cutting across the grass)

Number of failures to make a continuous line

Number of inappropriate places visited

**Total errors**

**Version 1 raw score = sequence score minus total errors**

### Version 2

- Place a photocopy of Zoo map version 2 in front of the subject.
- *'The next day you go back to the zoo for another visit...'*
- Clarify the rules and record timings as in version 1.

| Note subject's sequence | Each correct scores 1 | Correct response | Occasions each path used more than once |
|---|---|---|---|
| | | Entrance | A |
| | | Llamas | B |
| | | Elephants | C |
| | | Cafe | D |
| | | Bears | E |
| | | Lions | F |
| | | Bird sanctuary | G |
| | | Picnic area | H |
| | | | I |
| | | | J |
| | | | K |
| | | | L |
| | | | M |

**Sequence score** **Total**

**Planning time** **Total time**

**Errors**

Total number of occasions paths used more than once (from above)

Number of deviations from the path (i.e. cutting across the grass)

Number of failures to make a continuous line

Number of inappropriate places visited

**Total errors**

**Version 2 raw score = sequence score minus total errors**

**Add version 1 and version 2 raw scores**

| Raw score | Profile score |
|---|---|
| 16 | 4 |
| 11–15 | 3 |
| 6–10 | 2 |
| 1–5 | 1 |
| ≤0 | 0 |

**Profile score**

If planning time on version 2 is greater than 15 seconds subtract 1 from profile score

If total time on version 2 is greater than 123 seconds subtract 1 from profile score

**Total profile score** ☐

## Test 6: Modified six elements

For full text and procedure see Manual p. 10

- Arrange the test materials.
- *'You get ten minutes for this next test, and in this test you will be doing three different kinds of task...'*
- Go through each task with the subject.
- *'During the next ten minutes I would like you to try to complete at least some of each of the six individual parts...'*
- *'However, there is one rule you must obey...'*
- *'Now, tell me what you must do.'*
- Set the timer for 10 minutes.
- Start the stopwatch and timer.

**Record the order of sub tasks attempted and the subject's start and stop times**

| Sub task | Time started | Time stopped | Time on subtask |
|---|---|---|---|
| | | | |
| | | | |
| | | | |
| | | | |
| | | | |
| | | | |
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| | | | |
| | | | |
| | | | |
| | | | |
| | | | |

**Summary of time spent on each sub task and number of correct responses**

| Sub task | Total time on sub task |
|---|---|
| **Dictation A** | |
| **Dictation B** | |
| **Pictures A** | |
| **Pictures B** | |
| **Arithmetic A** | |
| **Arithmetic B** | |

**Number of sub tasks attempted** (max = 6)

**Minus number of sub tasks where rules were broken** (max = 3)

**Raw score**

| Raw score | Profile score |
|---|---|
| 6 | 4 |
| 4 or 5 | 3 |
| 2 or 3 | 2 |
| ≤ 1 | 1 |

**Profile score**

If total time on any one sub task is greater than 271 seconds, subtract 1 from profile score –

**Total profile score** ☐



**Thames Valley Test Company**
7–9 The Green, Flempton
Bury St Edmunds
Suffolk IP28 6EL England

BADS

# Dex Questionnaire
## Self-rating

TVTC
Thames Valley Test Company

Subject's name _____

Date _____

**This questionnaire looks at some of the difficulties that people sometimes experience. We would like you to read the following statements, and rate them on a five-point scale according to your own experience:**

1 I have problems understanding what other people mean unless they keep things simple and straightforward

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

2 I act without thinking, doing the first thing that comes to mind

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

3 I sometimes talk about events or details that never actually happened, but I believe did happen

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

4 I have difficulty thinking ahead or planning for the future

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

5 I sometimes get over-excited about things and can be a bit 'over the top' at these times

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

6 I get events mixed up with each other, and get confused about the correct order of events

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

7 I have difficulty realizing the extent of my problems and am unrealistic about the future

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

8 I am lethargic, or unenthusiastic about things

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

9 I do or say embarrassing things when in the company of others

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

10 I really want to do something one minute, but couldn't care less about it the next

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

11 I have difficulty showing emotion

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

12 I lose my temper at the slightest thing

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

13 I am unconcerned about how I should behave in certain situations

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

14 I find it hard to stop repeating saying or doing things once I've started

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

15 I tend to be very restless, and 'can't sit still' for any length of time

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

16 I find it difficult to stop myself from doing something even if I know I shouldn't

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

17 I will say one thing, but will do something different

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

18 I find it difficult to keep my mind on something, and am easily distracted

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

19 I have trouble making decisions, or deciding what I want to do

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

20 I am unaware of, or unconcerned about, how others feel about my behaviour

☐0 ☐1 ☐2 ☐3 ☐4

Never Occasionally Sometimes Fairly often Very often

 BADS, ISBN 1 874261 95 4
**Thames Valley Test Company**, 7–9 The Green, Flempton, Bury St Edmunds, Suffolk, IP28 6EL, England.

# Hospital Anxiety and Depression Scale (HADS)


Name: _____ Date: _____

FOLD HERE

Clinicians are aware that emotions play an important part in most illnesses. If your clinician knows about these feelings he or she will be able to help you more.

This questionnaire is designed to help your clinician to know how you feel. Read each item below and **underline the reply** which comes closest to how you have been feeling in the past week. Ignore the numbers printed at the edge of the questionnaire.

Don't take too long over your replies, your immediate reaction to each item will probably be more accurate than a long, thought-out response.

FOLD HERE

A D

**I feel tense or 'wound up'**
- Most of the time — 3
- A lot of the time — 2
- From time to time, occasionally — 1
- Not at all — 0

**I still enjoy the things I used to enjoy**
- Definitely as much — 0
- Not quite so much — 1
- Only a little — 2
- Hardly at all — 3

**I get a sort of frightened feeling as if something awful is about to happen**
- Very definitely and quite badly — 3
- Yes, but not too badly — 2
- A little, but it doesn't worry me — 1
- Not at all — 0

**I can laugh and see the funny side of things**
- As much as I always could — 0
- Not quite so much now — 1
- Definitely not so much now — 2
- Not at all — 3

**Worrying thoughts go through my mind**
- A great deal of the time — 3
- A lot of the time — 2
- Not too often — 1
- Very little — 0

**I feel cheerful**
- Never — 3
- Not often — 2
- Sometimes — 1
- Most of the time — 0

**I can sit at ease and feel relaxed**
- Definitely — 0
- Usually — 1
- Not often — 2
- Not at all — 3

A D

**I feel as if I am slowed down**
- Nearly all the time — 3
- Very often — 2
- Sometimes — 1
- Not at all — 0

**I get a sort of frightened feeling like 'butterflies' in the stomach**
- Not at all — 0
- Occasionally — 1
- Quite often — 2
- Very often — 3

**I have lost interest in my appearance**
- Definitely — 3
- I don't take as much care as I should — 2
- I may not take quite as much care — 1
- I take just as much care as ever — 0

**I feel restless as if I have to be on the move**
- Very much indeed — 3
- Quite a lot — 2
- Not very much — 1
- Not at all — 0

**I look forward with enjoyment to things**
- As much as I ever did — 0
- Rather less than I used to — 1
- Definitely less than I used to — 2
- Hardly at all — 3

**I get sudden feelings of panic**
- Very often indeed — 3
- Quite often — 2
- Not very often — 1
- Not at all — 0

**I can enjoy a good book or radio or television programme**
- Often — 0
- Sometimes — 1
- Not often — 2
- Very seldom — 3

**Now check that you have answered all the questions**

A D

TOTAL [ ][ ]

# CONSENT FORM

## A study investigating the neuropsychology of Tourette Syndrome.

Tick (✓yes) as appropriate

- **I have read the information leaflet about the study** ☐
- **I have had enough opportunity to ask questions about, and discuss the study** ☐
- **I have had satisfactory answers to my questions** ☐
- **I have received enough information about the study** ☐

**I understand that I do not have to take part in this study and that if I do decide to take part, I am free to withdraw:**

- ► **at any time**
- ►**without having to give any reason**
- ► **without my usual medical treatment being affected** ☐

**• I AGREE TO TAKE PART IN THIS STUDY YES / NO**

**Name (print):**.....

**Signed:**.....

**Date:**.....

**Name of Researcher:**.....

**Signed:**.....

**Date:**.....

Participant number | C |

# CONSENT FORM: Control Group

## A study investigating the neuropsychology of Tourette Syndrome.

Tick (✓yes) as appropriate

- **I understand that all of my answers will be confidential** ☐
- **I have had enough opportunity to ask questions about, and discuss what I will be asked to do** ☐
- **I have had satisfactory answers to my questions** ☐
- **I have received enough information about the study** ☐

**I understand that I do not have to take part in this study and that if I do decide to take part, I am free to withdraw:**

- ► **at any time**
- ►**without having to give any reason** ☐

- **I AGREE TO TAKE PART IN THIS STUDY** **YES / NO**

**Name (print):**.....

**Signed:**.....

**Date:**.....

**Name of Researcher:**.....

**Signed:**.....

**Date:**.....

**Appendix 4: Statistical Analyses**

**4 (a) Independent t-tests for Age, FSIQ, Age Finish Education, anxiety and depression and DEX Scores**

**4 (b) Tests of Normal Distribution using Kolmogrov-Smirnov One sample Test**

**4 (c) Histograms Showing Distribution for Individual Subtests on the BADS**

**4 (d) t-tests for BADS, TEA and IQ Scores**

**4 (e) Mann-Whitney U tests for All Scores**

**4 (f) ANCOVA with FSIQ as covariate, Illustrating Between Group, Overall Age Adjusted Score on the BADS;**

**4 (g) Mann-Whitney U and t-tests for differences between OCD and no OCD in TS Group**

**4 (h) Correlation Matrix for Subtests on the TEA with Age and FSIQ**

**4 (i) ANCOVA with FSIQ as covariate, Illustrating Between Group Differences on Elevator Counting with Reversal**

**Appendix 4 (a): Independent t-tests**

(i) Age and FSIQ

| | Mean | Std.Dev. | Mean | Std.Dev. | t-value | df | p |
|---|---|---|---|---|---|---|---|
| **AGE** | 29.59 | 12.920 | 30.88 | 12.105 | -0.302 | 33 | 0.7642 |
| **FSIQ** | 102.33 | 14.974 | 109.18 | 15.465 | -1.330 | 33 | 0.1927 |

(ii) Age Finish Education

| | Mean | Mean | t-value | df | p |
|---|---|---|---|---|---|
| AGEEDFIN | 18.94444 | 19.82353 | -0.589583 | 33 | 0.559486 |
| Valid N | Valid N | Std.Dev. | Std.Dev. | F-ratio | p |
| 18 | 17 | 3.489012 | 5.210990 | 2.230671 | 0.110710 |

(iii) Anxiety and Depression

| | Group 1 | | Group 2 | | | | | | |
|---|---|---|---|---|---|---|---|---|---|
| | Mean | Std.Dev. | Mean | Std.Dev. | t-value | df | p | Valid N | Valid N |
| **HADSANX** | 8.888889 | 3.644317 | 5.529412 | 3.411701 | 2.811252 | 33 | 0.008240 | 18 | 17 |
| **HADSDEP** | 4.333333 | 2.351470 | 2.176471 | 2.128241 | 2.839471 | 33 | 0.007678 | 18 | 17 |

(iv) DEX

| | Mean | Std.Dev. | Mean | Std.Dev. | t-value | df | p | Valid N | Valid N |
|---|---|---|---|---|---|---|---|---|---|
| **DEX** | 30.33 | 13.87 | 20.35 | 11.27 | 2.327300 | 33 | 0.026231 | 18 | 17 |

## Appendix 4 (b): Tests of normal Distribution

### (i) BADS subtest scores

| | N | max D | K-S |
|---|---|---|---|
| **RULESHIF** | 35 | 0.448507 | p < .01 |
| **ACTIONP** | 35 | 0.532876 | p < .01 |
| **KEYS** | 35 | 0.335832 | p < .01 |
| **TEMPJ** | 35 | 0.280603 | p < .01 |
| **ZOOMAP** | 35 | 0.213699 | p < .10 |
| **ELEMENTS** | 35 | 0.383843 | p < .01 |

### (ii) DEX and Age Classification Scores

| | N | Normal Parameters | | Most Extreme Differences | | | Kolmogoro v-Smirnov Z | Asymp. Sig. (2-tailed) |
|---|---|---|---|---|---|---|---|---|
| | | Mean | Std. Deviation | Absolute | Positive | Negative | | |
| DEX | 35 | 25.4857 | 13.47846 | .142 | .142 | -.082 | .840 | .481 |
| AGECLASS | 35 | 102.4571 | 13.42154 | .146 | .123 | -.146 | .863 | .446 |

a Test distribution is Normal.
b Calculated from data.

### (iii) IQ Scaled Scores

| | *N* | *max D* | *K-S* |
|---|---|---|---|
| FSIQ | 35 | 0.155748 | p > .20 |
| VERIQ | 35 | 0.115484 | p > .20 |
| PERIQ | 35 | 0.122673 | p > .20 |
| VOCAB | 35 | 0.102895 | p > .20 |
| BD | 35 | 0.171755 | p > .20 |
| SIMS | 35 | 0.108399 | p > .20 |
| MR | 35 | 0.097390 | p > .20 |

### (iv) TEA Scores

| | N | max D | K-S |
|---|---|---|---|
| EC | 35 | 0.492432 | p < .01 |
| ECD | 35 | 0.285277 | p < .01 |
| EDR | 35 | 0.110172 | p > .20 |
| PS | 35 | 0.130528 | p > .20 |
| PSCOUNT | 35 | 0.154227 | p > .20 |

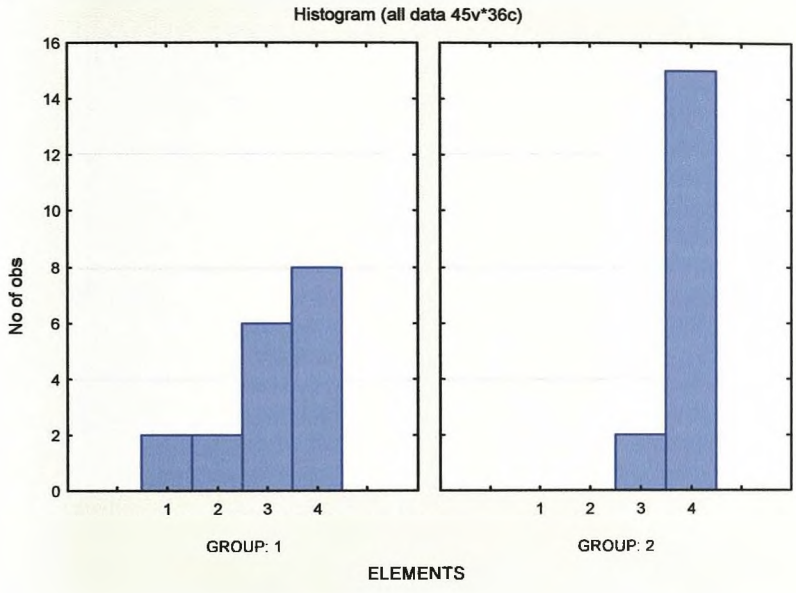
### (v) HADS Scores

| | N | Normal Parameters | | Most Extreme Differences | | | Kolmogoro v-Smirnov Z | Asymp. Sig. (2-tailed) |
|---|---|---|---|---|---|---|---|---|
| | | Mean | Std. Deviation | Absolute | Positive | Negative | | |
| HADSANX | 35 | 7.2571 | 3.87559 | .091 | .091 | -.074 | .540 | .932 |
| HADSDEP | 35 | 3.2857 | 2.46829 | .137 | .137 | -.092 | .811 | .526 |
| HADSTOT | 35 | 10.5429 | 5.91778 | .098 | .098 | -.060 | .578 | .891 |

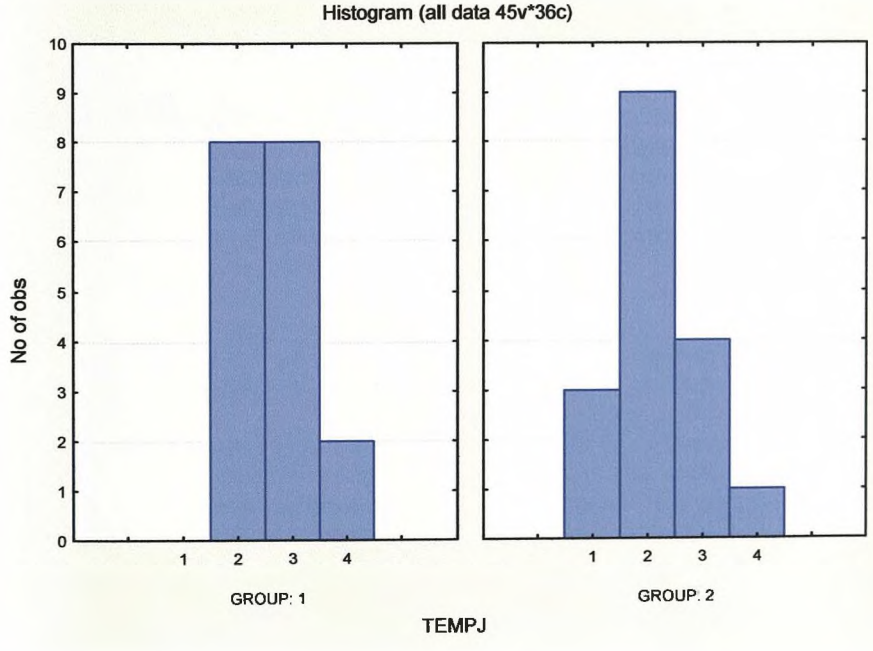
a Test distribution is Normal.
b Calculated from data.

## Appendix 4 (c): Histograms of Individual subtests of the BADS

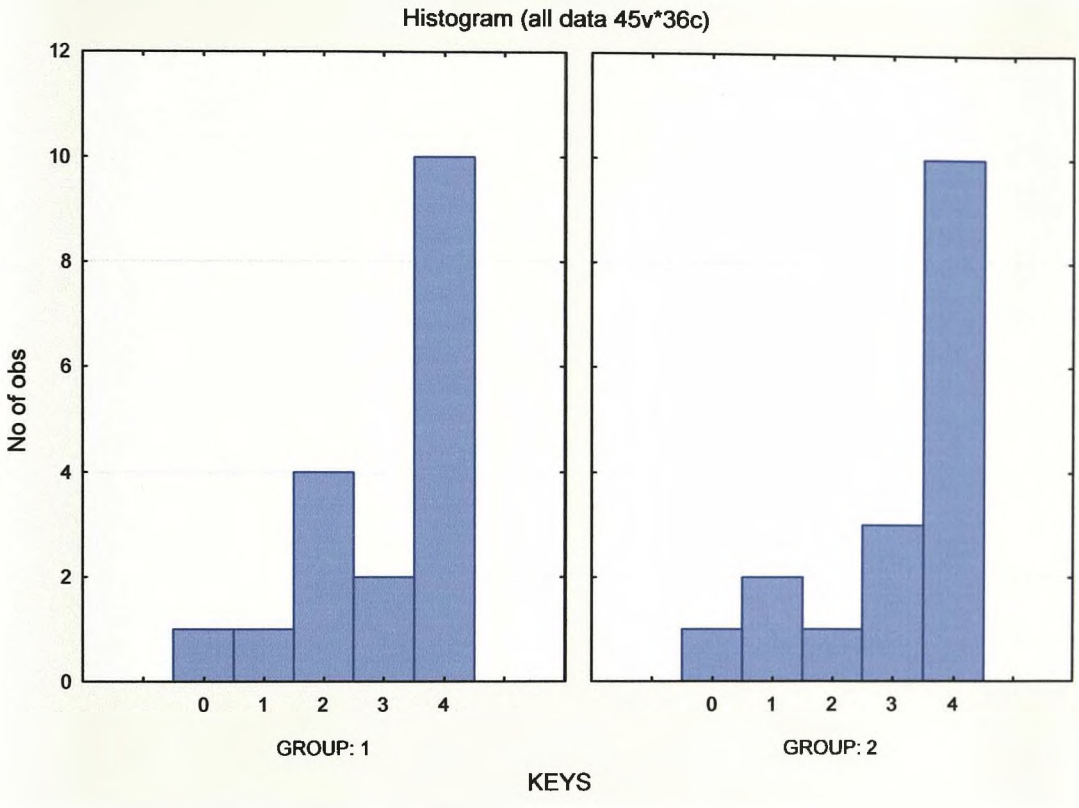
(i) 6 Elements



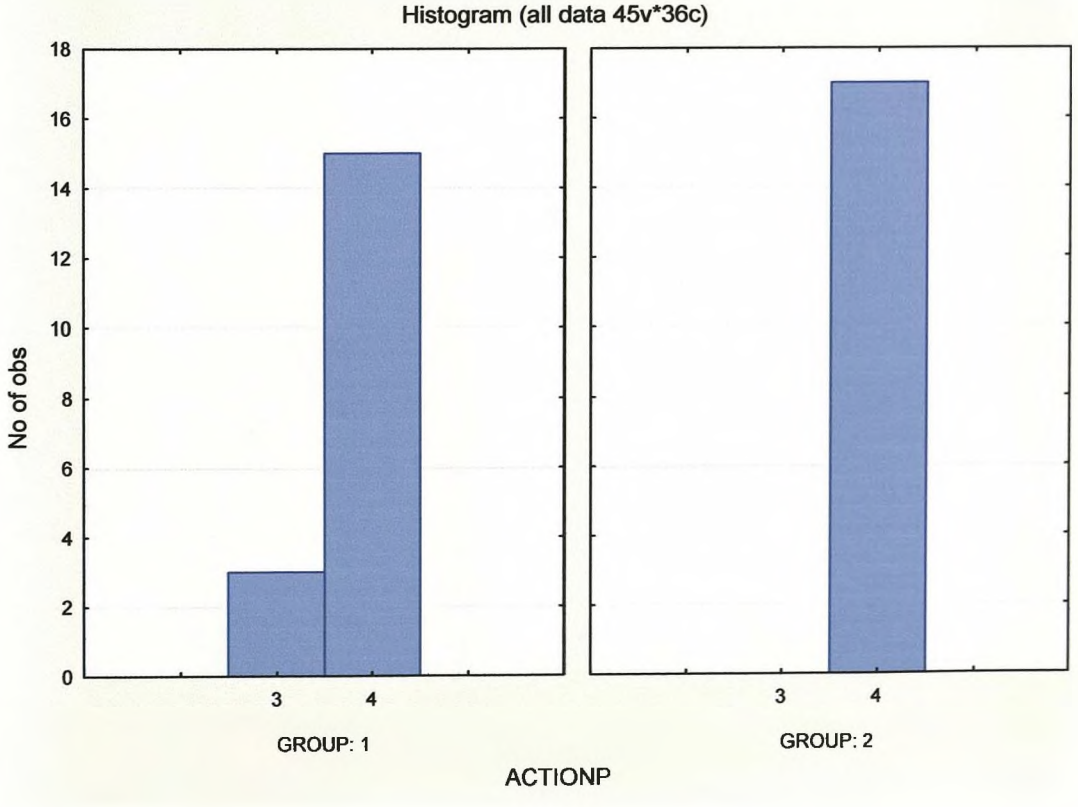

(ii) Temporal Judgement



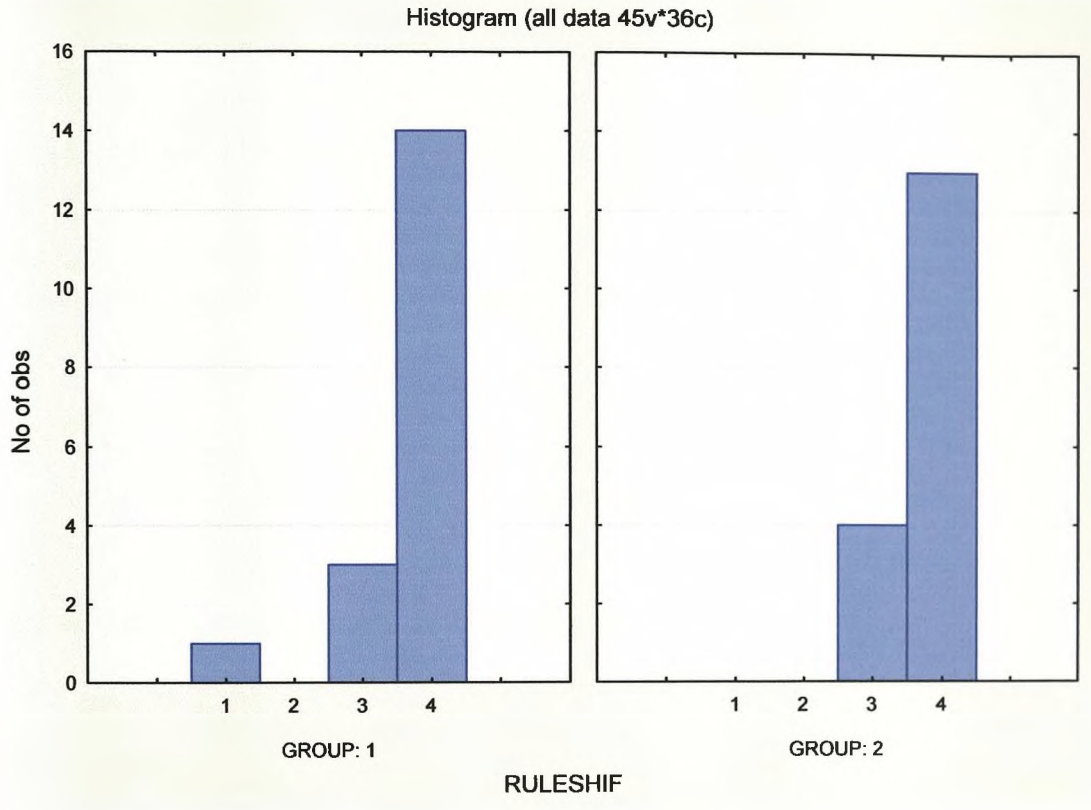
(iii) Key Search



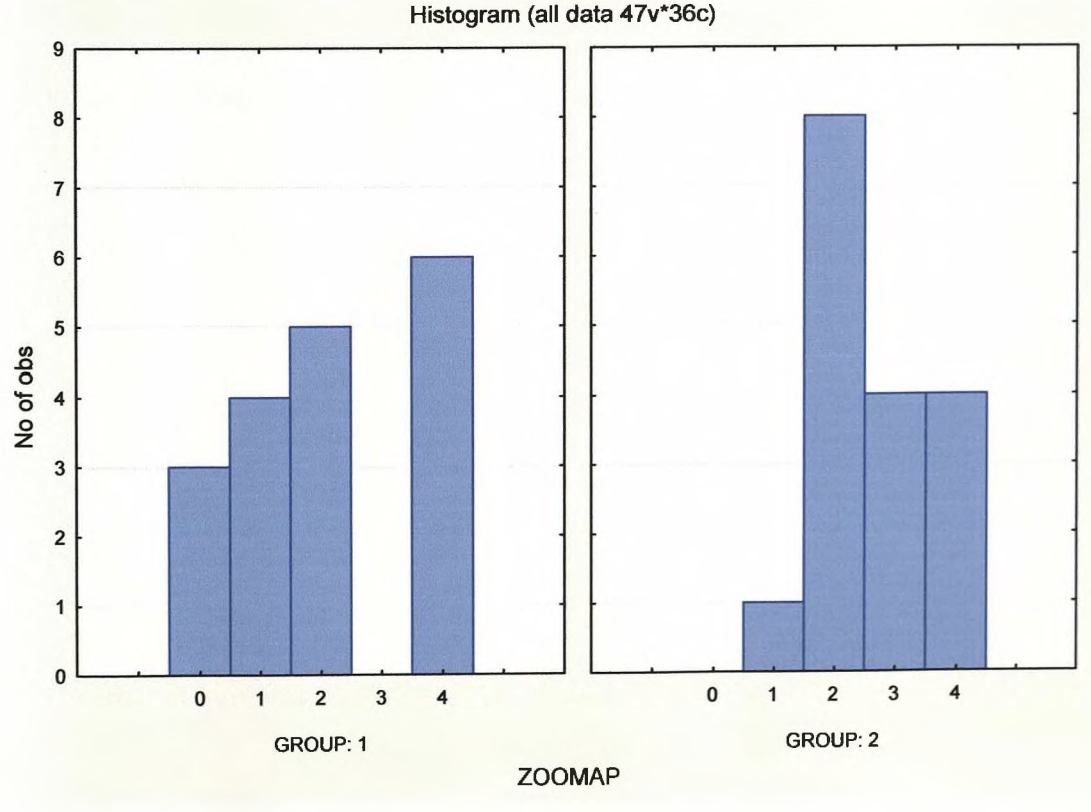

(iv) Action Programme



(v) Rule Shift




(vi) Zoo Map



## Appendix 4 (d): t-tests for BADS, TEA and IQ Scores

(i) BADS

| | Group 1 N = 18 | | Group 2 N = 17 | | | | |
|---|---|---|---|---|---|---|---|
| | Mean | Std.Dev. | Mean | Std.Dev. | t-value | df | p |
| RULESHIF | 3.67 | 0.77 | 3.76 | 0.44 | -0.5 | 33 | 0.65 |
| ACTIONP | 3.83 | 0.38 | 4.00 | 0.00 | -1.8 | 33 | 0.08 |
| TEMPJ | 2.67 | 0.69 | 2.18 | 0.81 | 1.9 | 33 | 0.06 |
| ZOOMAP | 2.11 | 1.53 | 2.65 | 0.93 | -1.2 | 33 | 0.22 |
| ELEMENTS | 3.11 | 1.02 | 3.88 | 0.33 | -3.0 | 33 | 0.01 |
| KEYS | 3.06 | 1.26 | 3.12 | 1.32 | -0.14 | 33 | 0.89 |

(ii) TEA

| | Group 1 N = 18 | | Group 2 N = 17 | | | | |
|---|---|---|---|---|---|---|---|
| | Mean | Std.Dev. | Mean | Std.Dev. | t-value | df | p |
| EC | 6.67 | 0.594 | 6.94 | 0.243 | -1.77 | 33 | 0.086 |
| ECD | 10.33 | 2.521 | 11.59 | 2.265 | -1.55 | 33 | 0.132 |
| EDR | 6.78 | 3.655 | 9.53 | 2.809 | -2.49 | 33 | 0.018 |
| PS | 7.33 | 3.581 | 9.41 | 3.411 | -1.76 | 33 | 0.088 |
| PSCOUNT | 9.61 | 4.060 | 11.65 | 4.623 | -1.39 | 33 | 0.175 |

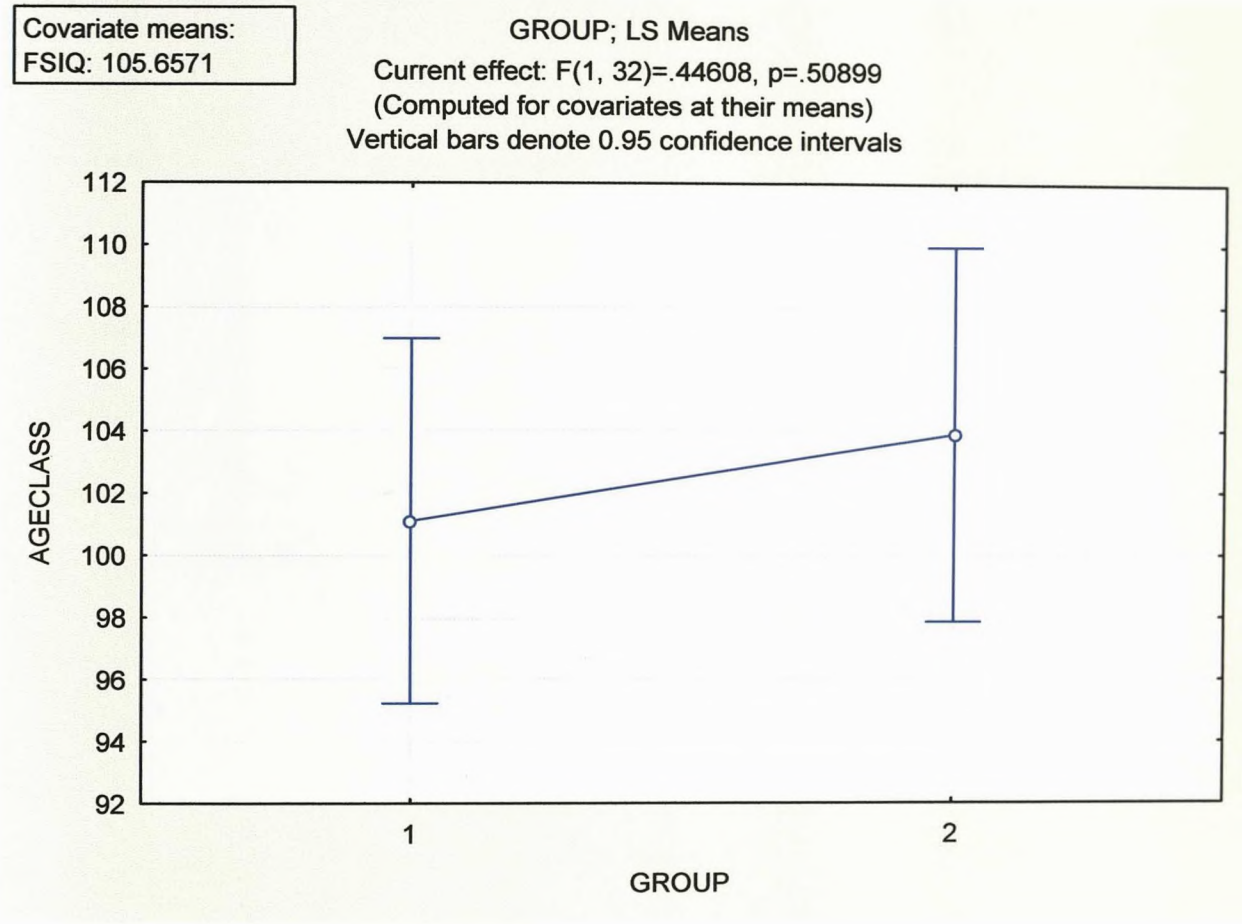
(iii) IQ Scores

| | Mean | Std.Dev. | Mean | Std.Dev. | t-value | df | p | Valid N | Valid N |
|---|---|---|---|---|---|---|---|---|---|
| FSIQ | 102.3333 | 14.97449 | 109.1765 | 15.46462 | -1.32995 | 33 | 0.192654 | 18 | 17 |
| VERIQ | 100.7222 | 18.34892 | 106.8235 | 17.84389 | -0.99639 | 33 | 0.326309 | 18 | 17 |
| PERIQ | 106.4444 | 12.32512 | 110.2941 | 12.59844 | -0.91367 | 33 | 0.367518 | 18 | 17 |
| PV Discrep | 5.7222 | 16.59485 | 3.4706 | 15.32203 | 0.41636 | 33 | 0.679845 | 18 | 17 |
| VOCAB | 48.6667 | 12.44754 | 52.8235 | 13.75334 | -0.93848 | 33 | 0.354819 | 18 | 17 |
| BD | 54.5556 | 9.05033 | 57.5882 | 7.11564 | -1.09761 | 33 | 0.280322 | 18 | 17 |
| SIMS | 49.1667 | 12.46289 | 54.5882 | 9.06269 | -1.46439 | 33 | 0.152550 | 18 | 17 |
| MR | 54.3333 | 7.35647 | 54.2353 | 9.18238 | 0.03496 | 33 | 0.972323 | 18 | 17 |

## Appendix 4 (e): Mann-Whitney U Tests for all Scores

| | Rank Sum | Rank Sum | U | Z | p-level | Z | p-level | Valid N | Valid N | 2*1sided |
|---|---|---|---|---|---|---|---|---|---|---|
| YGTSEV | 477.00 | 153.00 | 0.00 | 5.05 | 0.000 | 5.37 | 0.000 | 18 | 17 | 0.00 |
| FSIQ | 282.00 | 348.00 | 111.00 | -1.39 | 0.166 | -1.39 | 0.165 | 18 | 17 | 0.17 |
| VERIQ | 298.00 | 332.00 | 127.00 | -0.86 | 0.391 | -0.86 | 0.391 | 18 | 17 | 0.40 |
| PERIQ | 307.50 | 322.50 | 136.50 | -0.54 | 0.586 | -0.55 | 0.586 | 18 | 17 | 0.59 |
| PV Discrep | 325.50 | 304.50 | 151.50 | 0.05 | 0.961 | 0.05 | 0.960 | 18 | 17 | 0.96 |
| VOCAB | 304.50 | 325.50 | 133.50 | -0.64 | 0.520 | -0.64 | 0.519 | 18 | 17 | 0.52 |
| BD | 298.00 | 332.00 | 127.00 | -0.86 | 0.391 | -0.86 | 0.390 | 18 | 17 | 0.40 |
| SIMS | 285.50 | 344.50 | 114.50 | -1.27 | 0.204 | -1.27 | 0.203 | 18 | 17 | 0.21 |
| MR | 325.50 | 304.50 | 151.50 | 0.05 | 0.961 | 0.05 | 0.960 | 18 | 17 | 0.96 |
| EC | 290.00 | 340.00 | 119.00 | -1.12 | 0.262 | -1.71 | 0.087 | 18 | 17 | 0.27 |
| ECD | 283.00 | 347.00 | 112.00 | -1.35 | 0.176 | -1.45 | 0.148 | 18 | 17 | 0.18 |
| EDR | 255.50 | 374.50 | 84.50 | -2.26 | 0.024 | -2.27 | 0.023 | 18 | 17 | 0.02 |
| PS | 273.50 | 356.50 | 102.50 | -1.67 | 0.096 | -1.67 | 0.094 | 18 | 17 | 0.10 |
| PSCOUNT | 281.50 | 348.50 | 110.50 | -1.40 | 0.161 | -1.41 | 0.158 | 18 | 17 | 0.16 |
| RULESHIF | 324.00 | 306.00 | 153.00 | 0.00 | 1.000 | 0.00 | 1.000 | 18 | 17 | 1.01 |
| ACTIONP | 298.50 | 331.50 | 127.50 | -0.84 | 0.400 | -1.74 | 0.083 | 18 | 17 | 0.40 |
| KEYS | 318.50 | 311.50 | 147.50 | -0.18 | 0.856 | -0.20 | 0.840 | 18 | 17 | 0.86 |
| TEMPJ | 376.00 | 254.00 | 101.00 | 1.72 | 0.086 | 1.87 | 0.062 | 18 | 17 | 0.09 |
| ZOOMAP | 288.00 | 342.00 | 117.00 | -1.19 | 0.235 | -1.24 | 0.216 | 18 | 17 | 0.24 |
| ELEMENTS | 253.00 | 377.00 | 82.00 | -2.34 | 0.019 | -2.79 | 0.005 | 18 | 17 | 0.02 |
| AGECLASS | 296.00 | 334.00 | 125.00 | -0.92 | 0.355 | -0.93 | 0.352 | 18 | 17 | 0.37 |
| CLASSIF | 298.00 | 332.00 | 127.00 | -0.86 | 0.391 | -0.93 | 0.353 | 18 | 17 | 0.40 |
| DEX | 394.00 | 236.00 | 83.00 | 2.31 | 0.021 | 2.31 | 0.021 | 18 | 17 | 0.02 |

**Appendix 4 (f): ANCOVA with FSIQ as covariate, Illustrating Between Group, Overall Age Adjusted Scores from the BADS**




| | AGECLASS |
|---|---|
| **FSIQ** | .4796 |
| | p=.004 |

| | SS | Degr. of | MS | F | p |
|---|---|---|---|---|---|
| **Intercept** | 2450.54 | 1 | 2450.54 | 16.8611 | 0.00026 |
| **FSIQ** | 1207.85 | 1 | 1207.85 | 8.3107 | 0.00699 |
| **GROUP** | 64.83 | 1 | 64.83 | 0.4461 | 0.50899 |
| **Error** | 4650.79 | 32 | 145.34 | | |

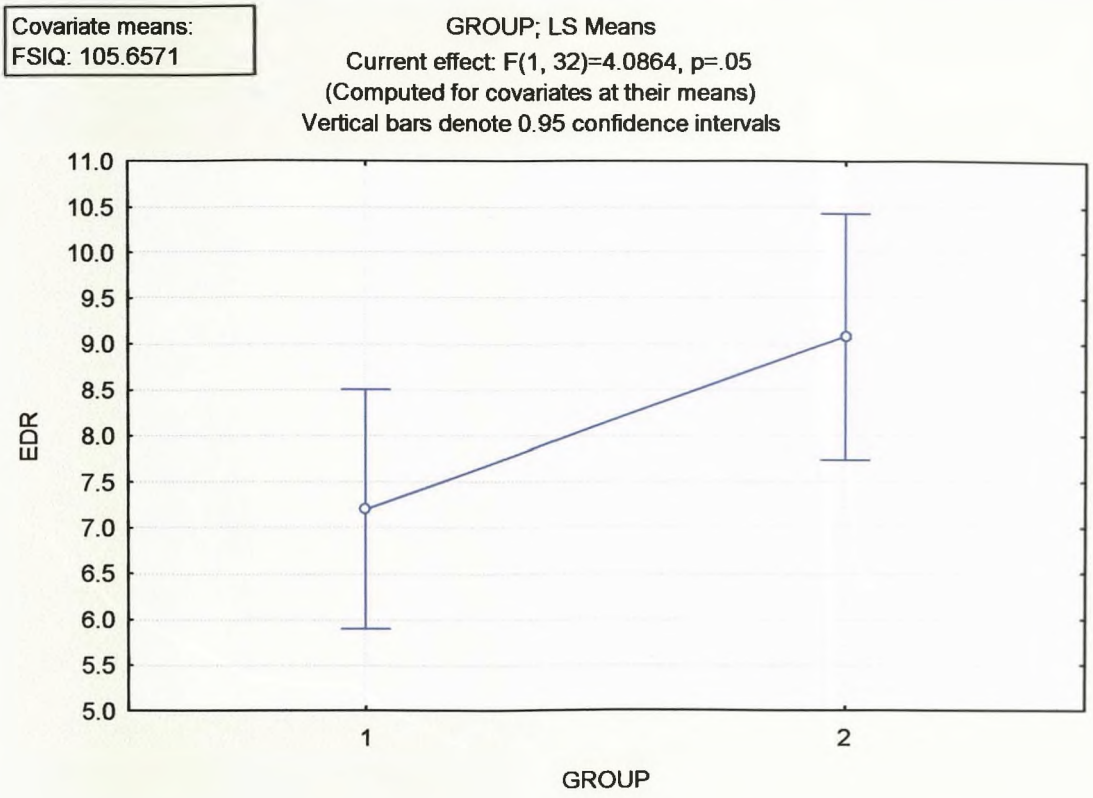
**Appendix 4 (g): Mann-Whitney U tests for differences between BADS scores for OCD and non-OCD within the TS Group.**

| | YBOCS | N | Mean Rank | Sum of Ranks |
|---|---|---|---|---|
| RULESHIF | no | 10 | 9.45 | 94.50 |
| | yes | 8 | 9.56 | 76.50 |
| | Total | 18 | | |
| ACTIONP | no | 10 | 10.10 | 101.00 |
| | yes | 8 | 8.75 | 70.00 |
| | Total | 18 | | |
| KEYS | no | 10 | 10.55 | 105.50 |
| | yes | 8 | 8.19 | 65.50 |
| | Total | 18 | | |
| TEMPJ | no | 10 | 8.05 | 80.50 |
| | yes | 8 | 11.31 | 90.50 |
| | Total | 18 | | |
| ZOOMAP | no | 10 | 9.60 | 96.00 |
| | yes | 8 | 9.38 | 75.00 |
| | Total | 18 | | |
| ELEMENTS | no | 10 | 10.00 | 100.00 |
| | yes | 8 | 8.88 | 71.00 |
| | Total | 18 | | |
| AGECLASS | no | 10 | 10.15 | 101.50 |
| | yes | 8 | 8.69 | 69.50 |
| | Total | 18 | | |

**Appendix 4 (h): Correlation Matrix for Subtests on the TEA with AGE and FSIQ**

| | EC | ECD | EDR | PS | PSCOUNT |
|---|---|---|---|---|---|
| AGE | .2094 | .2861 | .1067 | .2596 | -.1075 |
| | p=.227 | p=.096 | p=.542 | p=.132 | p=.539 |
| FSIQ | .2733 | .1127 | .6152 | .4338 | .1421 |
| | p=.112 | p=.519 | p=.000 | p=.009 | p=.415 |

## Appendix 4 (i): ANCOVA with FSIQ as covariate, Illustrating Between Group Differences on Elevator Counting with Reversal




| | Elevator Counting with Reversal | | | | | Telephone Search | | | | |
|---|---|---|---|---|---|---|---|---|---|---|
| | SS | df | MS | F | p | SS | df | MS | F | p |
| **Intercept** | 18.23 | 1 | 18.23 | 2.52 | 0.12 | 1.02 | 1 | 1.01 | 0.09 | 0.75 |
| **FSIQ** | 122.10 | 1 | 122.10 | 16.89 | 0.0003 | 63.01 | 1 | 63.01 | 5.91 | 0.02 |
| **GROUP** | 29.53 | 1 | 29.53 | 4.08 | 0.05** | 17.61 | 1 | 17.61 | 1.65 | 0.20 |
| **Error** | 231.25 | 32 | 7.23 | | | 341.11 | 32 | 10.66 | | |

## Appendix 5: South Birmingham Research Ethics Committee Approval Letters

Birmingham and The Black Country NHS
Health Authority

**South Birmingham Research Ethics Committee**
[redacted]

*Tel:* [redacted]
*Fax:* [redacted]

Administrator: Mrs A P McCullough

Our ref: APM/mbt/DD/04/07
**Please Quote: 2002/066**

Ms Caroline Formby
[redacted]

Dear Ms Formby

**REC reference number 2002/066**
**Neuropsychological profile of executive function in adults with Gilles de la Tourette Syndrome: the relationship with co-morbid obsessive-compulsive disorder and the predictive value of tic severity on neuropsychological performance**
**Research Protocol - Undated**
**Letter to Participant**
**Volunteer Information Sheet**
**Consent Form**
**Y-BOCS Symptom Checklist**
**Yale Global Tic Severity Scale**
**BADS Scoring Sheet**
**Wechsler Abbreviated Scale of Intelligence**
**The Test of Everyday Attention Scoring Sheet**
**Tourette's Syndrome "Diagnostic Confidence Index"**
**Hospital Anxiety and Depression Scale**

South Birmingham Research Ethics Committee are happy to Approve your Study subject to the following:

- Satisfactory Indemnity arrangements being in place.
- Clearance from your Trust or relevant employer.
- That you produce an annual review in line with the Good Clinical Practice Guidelines.
- Active Approval is required until the Study has been completed.
- The Committee would wish to be kept informed of Serious Adverse Events, Amendments and any modifications to Patient Information Leaflets and Consent Forms.

Approval is valid for three years, however, if it is intended to continue the Study after THREE YEARS from the date of this letter South Birmingham Research Ethics Committee would wish to re-examine it.

Would you please communicate this approval immediately to all members of the investigating team and where appropriate the sponsoring commercial company. Please also advise your Research and Development Office of this approval.

Yours sincerely [redacted]

**Chairman**
**Research Ethics Committee**
cc: File
Appropriate Trust

Research Ethics Committee
2 [illegible] 2002
APPROVED

Birmingham and The Black Country NHS
Strategic Health Authority

**South Birmingham Research Ethics Committee**
[redacted]

*Tel:* [redacted] *Fax:* [redacted]

**Chairmen:** Mr R K Vohra

**Our ref:** DD/mbt/jb/04/07
**Date:** 5 February 2003
**Please Quote:** 2002/066

Ms C Formby
[redacted]

Dear Ms Formby

**REC reference number 2002/066**
**Neuropsychological profile of executive function in adults with Gilles de la Tourette Syndrome: the relationship with co-morbid obsessive-compulsive disorder and the predictive value of tic severity on neuropsychological performance**
**Research Protocol - Undated**
**Letter to Participant**
**Volunteer Information Sheet**
**Consent Form**
**Y-BOCS Symptom Checklist**
**Yale Global Tic Severity Scale**
**BADS Scoring Sheet**
**Wechsler Abbreviated Scale of Intelligence**
**The Test of Everyday Attention Scoring Sheet**
**Tourette's Syndrome "Diagnostic Confidence Index"**
**Hospital Anxiety and Depression Scale**

Thank you for your letter dated 21 January 2003 requesting an amendment to the research Protocol for the above Study.

It will be acceptable for a control group to be included in the Study. The information Sheet and Consent Form for this group are approved.

Yours sincerely

[redacted]

**Vice Chairman**
**Research Ethics Committee**

## Appendix 6: Information for Authors

(i) **Neuroscience and Biobehavioural Reviews**
(ii) **Journal of Clinical and Experimental Neuropsychology**

authorGATEWAY for Elsevier Journals

**Neuroscience & Biobehavioral Reviews**

print page close wind

## Guide for Authors

**Submission of Papers**
Authors are requested to submit their original manuscript and figures with two copies to one of the Editors-in-Chief, **Verity J. Brown**, School of Psychology, University of St. Andrews, St. Andrews KY16 9JU, UK; **Ann E. Kelley**, Department of Psychiatry, University of Wisconsin-Madison Medical School, 6001 Research Park Blvd, Madison, WI 53719, USA.

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**Types of Contributions**
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**General**: Manuscripts must be typewritten, double-spaced with wide margins on one side of white paper. Good quality printouts with a font size of 12 or 10 pt are required. The corresponding author should be identified (include a Fax number and E-mail address). Full postal addresses must be given for all co-authors. Authors should consult a recent issue of the journal for style if possible. An electronic copy of the paper should accompany the final version. The Editors reserve the right to adjust style to certain standards of uniformity. Authors should retain a copy of their manuscript since we cannot accept responsibility for damage or loss of papers. Original manuscripts are discarded one month after publication unless the Publisher is asked to return original material after use.

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**Drugs**: Proprietary (trademarked) names should be capitalized. The chemical name should precede the trade, popular name, or abbreviation of a drug the first time it occurs.

**Anesthesia**: In describing surgical procedures on animals, the type and dosage of the anesthetic agent should be specified. Curarizing agents are not anesthetics; if these were used; evidence must be provided that anesthesia of suitable grade and duration was employed.

**Units and abbreviations**: All dimensions and measurement must be specified in the metric system. Standard nomenclature, abbreviations and symbols, as specified by Royal Society Conference of Editors. Metrification in scientific journals, Am. Scient. 56:159-164; 1968, should be used throughout.

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In the text refer to references by a number in square brackets on the line (e.g. [1]) and the full reference should be given in a numerical list at the end of the paper.
References should be given in the following form:

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[1]Van der Geer J, Hanraads JAJ, Lupton RA. The art of writing a scientific article. J. Sci COmmun 2000;163:51-9.

Reference to a book:
[2]Strunk JR W, White EB. The elements of style. 3rd ed. New York: Macmillan; 1979.

Reference to a chapter in an edited book:
[3]Mettam GR, Adams LB. How to prepare an electronic version of your article. In: Jones BS, Smith RZ, editors. Introduction to the electronic age. New York: E-publishing Inc; 1999, p.281-304.

**Illustrations**: All illustrations should be provided in camera-ready form, suitable for reproduction (which may include reduction) without retouching. Photographs, charts and diagrams are all to be referred to as "Figure (s)" and should be numbered consecutively in the order to which they are referred. They should accompany the manuscript, but should not be included within the text. All illustrations should be clearly marked on the back with the figure number and the author's name. All figures are to have a caption. Captions should be supplied on a separate sheet.
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