AGGRESSIVE BEHAVIOR IN CHILDREN WITH TOURETTE'S SYNDROME AND ASSOCIATED DISORDERS

by

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A thesis submitted in conformity with the requirements
for the degree of Master of Arts
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ABSTRACT

Aggressive behavior has been reported in approximately 33% of Tourette's Syndrome (TS) patients (Spencer, T., Biederman, J., Harding, M., et al., 1995). Attention Deficit Hyperactivity Disorder (ADHD) and /or Obsessive Compulsive Disorder (OCD) have been reported in 50-70% of TS patients (Schuerholz, L. J., et al., 1996). It was unknown whether aggressive behavior was associated with TS directly or found primarily in TS with comorbid ADHD and/or OCD. This study examined aggressive behavior in 25 non-medicated patients with TS (ages 6 to 14) and 5 controls (ages 7 to 12). The analysis revealed that teacher reported aggressive behavior using the Teacher Report Form (TRF; Achenbach, T. M., & Edelbrock, C. ,1983) was significantly higher in participants with TS and comorbidity ($x=1.00$, sd $\pm$ 0.00), $F(12,9)=23.172$, $p<0.05$). These results suggest that the presence of comorbid ADHD and/or OCD increases the risk of aggressive behavior in TS children.

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CHAPTER ONE

INTRODUCTION

Tourette' Syndrome (TS) is a neuropsychiatric disorder usually present in childhood, characterized by unwanted, repetitive, recurring involuntary motor and vocal tics, which persist in a waxing and waning pattern. Onset of TS is often as early as two years of age, with the mean age of onset 7.4 years (Wand, Shady, Broder, Furer, & Staley, 1993).

Tics are generally classified as simple or complex. Simple motor tics include movements, such as eye blinking, facial grimaces, tossing of the head, or neck and shoulder jerks. Motor tics that involve multiple muscle groups often result in more purposeful, complex, and often physically disruptive patterns of movement. These tics may include ocular deviation, facial movement, touching, jumping, smelling or copropraxia (obscene gestures) (Singer & Walkup, 1991). Motor tics usually occur concurrently with a wide spectrum of vocal tics. Examples of vocal tics range from simple sounds (grunting, sniffing, snorting, barking, humming, repetitive clearing of the throat) to more complex vocalizations such as coprolalia (the involuntary utterance of obscenities), palilalia (repetition of one's own utterances and phrases) and echolalia (repetition of someone else's words or phrases) (Shapiro & Shapiro, 1982). Patients can sometimes suppress tics for a limited amount of time, from minutes to hours depending on both the tic and the age of the child, however, subsequently, the patient often experiences a compensatory increase in number of tics and their intensity. Diagnosis is sometimes difficult, and often delayed because

of the confusion of some of the more common tics; sniffing, coughing and throat clearing, with symptoms of colds or allergies. In addition, facial tics, grimaces and blinking are sometimes dismissed as "nervous habits" and are often overlooked until additional tics or behaviors develop (Kurlan, 1994). A diagnosis of TS syndrome must include more than one motor tic, as well as a vocal tic, with symptoms lasting longer than one year (American Psychiatric Association [APA, DSM-III-R], 1987). Previously considered a rare disorder, epidemiological studies now estimate the total prevalence of TS in the general population to be 0.03% (Sandor, P. 1993), similar to the occurrence of schizophrenia. Recent studies have found the prevalence rate of TS to be as high as 3 % of a mainstream general high school population, in contrast to the reported 0.05 % by the local regional mental health authorities (Mason, A., Banerjee, S., Eapen, V., Zeitlin, H, and Robertson, M. M., 1998). These new findings suggest that only approximately 1 in 60 people who fulfill the criteria for TS are actually diagnosed.

Besides the core symptoms of multiple motor and vocal tics, behavioral problems including Attention Deficit Hyperactivity Disorder (ADHD) and Obsessive-Compulsive Disorder (OCD) are commonly comorbid with TS ([APA, DSM-III-R], 1987; Kurlan, R., 1989). Patients with TS, behavioral disorders, and social anxiety from unwanted public attention, often have impaired self-esteem, self-image and relationships (Thibert, A L., Day, H. L., & Sandor, P., 1995).

Although some researchers believe that the accompanying behavioral disorders are a component of the TS itself, others disagree. Shapiro and Shapiro (1982) associated the behavioral problems with the presence of ADHD in children

with TS. Comings et al., (1990) found behavior problems in children with TS pervasive through adolescence and into adulthood. They have reported OCD, Conduct Disorder (CD), inappropriate sexual behaviors, panic attacks and agoraphobia to be at times' comorbid with TS. A large study of 431 participants with TS reported learning disabilities in 31% and severe behavior problems in 25 % of school-aged respondents (Stefl & Rubin, 1985).

ADHD often presents before the onset of tic behaviors, and occurs in about 50-70% of individuals with TS (Schuerholz, L. J., et al., 1996). In addition, OCD, learning difficulties, mood disturbances and aggression (Golden, G ,1984; Singer, H. S., & Rosenberg, L. A., 1989; Dykens, E., et al, 1990; Comings, D., & Comings, B A, 1990; Singer, H. S., Schuerholz, L., & Denckla, M., 1995) are some of the more frequently described conditions associated with TS (Wand, R. R., et al., 1993).

Obsessions are defined as unwanted intrusive thoughts, and compulsions refer to ritualistic activity designed to reduce anxiety or to prevent a negatively interpreted future event. Although adults with OCD usually recognize that their obsessions and compulsions are excessive or unreasonable, children may lack the insight to distinguish these distressing behaviors from necessary routine (American Psychiatric Association, 1994) More than half of those with TS report that obsessions and compulsions are a major component of their disorder (Kurlan, R. 1994). It was estimated that 25% of TS patients seen in a TS clinic meet the diagnostic criteria for OCD (Pauls, D.L., Towbin, K. E., & Leckman, J. F., 1986). Developmental level also appears to have an influence on the type of psychological symptoms expressed by children with TS. Shapiro et al. (1988) reported 33% of

children under 16 years old with TS had comorbid ADHD, while the percentage dropped to 15% in those over 16. In addition, the co-occurrence of OCD increased with the child's age. These findings indicate that similar to age-related changes in other abilities and in the social environment, the behavioral problems associated with TS may also be modified with age. Developmental stage, along with tic severity and the corresponding psychiatric symptoms must all be considered in the diagnosis and prescribed treatment of a child with Tourette's.

Earlier studies examining the association between Tourette's and disruptive behavior problems reported aggression as a factor in up to 67% of adolescents and adults (Moldofsky, H., Tullis et al., 1974). Stefl (1984) in a larger study of 431 individuals with TS, found that 65-75% reported behavior problems, including temper outbursts, hyperactivity, mood swings, and aggression. Although 60% of the sample were taking prescription medications for tic symptoms, the majority of the participants claimed they experienced no relief from the associated behavior problems. According to Comings and Comings (1985), the best descriptor of a behavioral problem in children with Tourette's is "anger." They reported 42% of their TS sample (n=250) as having problems with both anger and violence, with the severity already outside norms by the grade 1 level, to destruction of property and killing animals by grade 2, and legal problems by grade 3. When the addition of hyperactivity with TS was considered, by grade 2, 59% of the children were experiencing severe problems. Interestingly, aggression problems were present in both the male and female participants.

Definitions of Aggression

The distinction between anger and aggressive behavior has been under discussion in the literature, although an accepted universal definition has not yet been developed. Because of this lack of consensus, various definitions have been used in different studies. Baron and Byrne (1984) offer the following traditional interpretation: "Aggression is any form of behavior directed toward the goal of harming or injuring another living being who is motivated to avoid such treatment" (p. 325).

This definition focuses largely on physical aggression or assault. It is typical for aggressive children to be viewed by others as bossy, disruptive, not well liked and often exhibiting socially inappropriate behavior, i.e. a broader concept then simply outbursts of physical aggression. Thus the above definition represents a very limited understanding of aggressive behavior in the TS population.

A more appropriate operational definition of aggressive behavior that has been clinically standardized, and widely accepted appears in the DSM-III-R(APA, 1987). The definition of Oppositional Defiant Disorder (ODD) refers to a recurrent pattern of behavior that is described as defiant, disobedient, negative, and hostile toward authority figures, and that occurs for a minimum of six months. Some of the characteristics of this behavior include losing one's temper, arguing with adults, actively defying or refusing to comply with the requests of adults, deliberately annoying others, not taking the blame for their own mistakes or misbehavior, being touchy, angry , resentful and spiteful. In comparison to other children of the same

age and developmental level, these behaviors must appear more frequently and with greater intensity, as well as causing disruption in the social or academic functioning of the child.

Children with ODD are also often observed to be persistently stubborn, resistant to directions, unwilling to compromise or to develop a sense of "give and take" or successful negotiation with adults or peers. This inability to be flexible in peer relationships, along with consistently blaming others for their own errors, causes great difficulties in maintaining peer friendships. The hostile responses often exhibited by these children are usually verbal, without a physical element. In some children verbal confrontation may lead to physically aggressive ways described in CD. The children with ODD do not usually view themselves as violent or aggressive, but in defense will strive to explain or to justify their aggressive behavior as having been unfairly evoked by someone else. It is noted in the (DSM III-R ; APA, 1987) that a significant proportion of the children with ODD proceed upon a developmental path to later developing CD, however many experts agree that although high proportions of children with CD had ODD in childhood, ODD is not a direct predictor of later CD (Lahey, B. B., & Loeber, R., 1994).

When reviewing the diagnostic criteria for CD, the DSM III-R (APA, 1987) describes the essential behavioral feature as "a repetitive and persistent pattern of behavior in which the basic rights of others or major age-appropriate societal norms or rules are violated" (p. 85). These features of aggression may be categorized as "aggressive conduct that causes or threatens physical harm to other people or animals"; "nonaggressive conduct that causes property loss or damage";

"deceitfulness or theft"; or "serious violations of rules". To meet the criteria for CD the behaviors must have been present within the past 12 months, with an occurrence during the past 6 months. Another criterion for diagnosis is the degree of impairment that these behaviors cause the child. Clinically significant negative impact on the child's social, academic or occupational function must occur as a direct result of these behaviors, across a variety of settings.

The definition of aggression when described in Conduct Disorder or Oppositional Defiant disorder includes verbal aggression, defiance and physical aggression against objects, animals or people which is pervasive across domains, including school and home. For the purpose of this study, the behavior of children described as "aggressive" shall be determined as that which is physically harmful to themselves or others. The aggressive behavior interferes with their social and academic development, and often causes severe disruption in their inter-family relationships and in the relationships of those around them.

It is important to recognize that aggressive behavior is an expression of the underlying emotions, most often anger, but sometimes also fear. Anger, as defined by Kassinove (1995) is a negative internal feeling or state associated with specific cognitive and perceptual distortions and deficiencies (i.e., misappraisals, errors, and attributions of blame, injustice, preventability, and/or intentiality). This emotion is a response to an adverse stimulus which does not always evoke a violent or physical response. The aggressive behaviors that are exhibited by children with TS are often described as "small protests which escalate into uncontrolled rages" (Comings, D. E., 1990, p. 124). The children who experience these rages usually

later regret their verbally and physically explosive outbursts. Between "tantrums" the children may be reasonable and compliant. It is notable that these children usually feel guilty about their recent behavior (Hansen, C. R. Jr., in Haerle,T., 1992) and are unable to predict or describe what may trigger a similar rage in the future. When asked about the importance of the issue that initiated the outburst as compared to the degree of the aggressive response, TS children will usually admit that the response was out of proportion to the provoking events. In this way they differ from non-TS children with conduct disorder who do not recognize or accept their responsibility for a given conflict.

The importance of recognizing aggression in children is supported by Loeber et al., (1988) who stated that most habitual offenders and those who engage in the more violent acts later in life, have usually been highly aggressive as children Additional studies of Canadian children by Loeber et al. (1989) found a relationship between the severity of aggression among the participants (e.g. initiating physical fighting), and the transition to later conduct disorder. Loeber (1995) adds that early identification is critical because the transition from displays of physical aggression to evasive acts of conduct disorder, may occur as early as the elementary years.

Cohen and Leckman (1994) recognized that some children with TS who are approaching adolescence may experience a remission of tics, or an increase in their ability to suppress their tics for longer periods of time, however this suppression may be accompanied by an upsurge of aggression (at self and others). This increase in aggressive tendencies is partially fueled by the frustrating effort at suppressing their tics, emotional problems (anxiety, depression) due to the

increased awareness of the social impact of tics, as well as obsessions and compulsive acts. The adolescent with TS who is attempting to develop autonomy from his parents is frequently met with a selfperpetuating cycle of frustration, anger and aggressive behavior. Contributing factors: frustration of dealing with tics; unwanted attention; immature attitude: impaired impulse control; and a tendency to be perseverative, all underscore the need for parents and clinicians to continually monitor and intervene in the life of the young adult with TS who is struggling with his/her attempts at individuation and separation.

Disruptive behavior directly interferes with a child's academic success, family-peer relationships and psychosocial development. Often the first visit to the clinic occurs because these behavior problems (not tics) intrude on several aspects of the life and development of the child with TS (Cohen and Leckman, 1994; Wand, R. R et al., 1993; Erenberg et al., 1986).

Shapiro et al., (1982) found that male TS participants aged 6-11 with comorbid ADHD, scored higher than controls on all symptom subscales of the Child Behavior Checklist (CBCL; Achenbach & Edelbrock, 1983), including externalizing behavior (aggression, conduct disorder). Randolph et al. (1993) also reported that TS children with ADHD had a higher rating of tic severity than those with TS only. These studies suggest that the presence of ADHD together with a diagnosis of TS was associated with increased aggression and tic severity.

Rationale for this Study

Children who have TS alone may face a different set of behavioral problems than those with TS in combination with ADHD and/or OCD. Identifying the risk of developing aggressive behavior early, as well as predicting severity, based on tic expression and the presence or absence of other comorbid disorders, would be of primary interest to the clinician designing the child's treatment program.

On the basis of the research findings in the current literature, the need was recognized for further clarification regarding comorbidity of TS with aggressive behavior. Aggressive behavior can potentially disrupt the family, school and social life development of children with TS, regardless of how mild their tics may be. Also, TS is a lifelong illness. While the tics tend to wax and wane, and often go into remission during adulthood, the accompanying behavioral problems may affect the child's development, academic potential, and adjustment to their adult role with life-long consequences.

Hypotheses

This pilot study set out to examine the degree of aggressive behavior and tic severity in three groups of participants: TS alone, TS with ADHD, TS with ADHD and OCD, and general population controls.

We wished to test the following hypothesis:

1- Participants with the diagnosis of TS alone will demonstrate a significantly rate in the rate of aggressive behavior compared to controls without TS.

2- The participants who have a comorbid diagnosis of Obsessive Compulsive Disorder, or ADHD, in addition to TS will demonstrate an elevated rate of aggression compared to control and to the participants with TS alone.

3- The participants tic severity will correlate positively with their aggression level in all groups.

CHAPTER TWO

METHOD

Experimental Design

The experimental design was a cross- sectional study of unselected consecutive referrals to the Tourette's Clinic.

Participants

An attempt was made to enroll all eligible consecutive outpatients referred for assessment of TS at the TS Clinic, Toronto Hospital. These children were all recruited during their regular attendance at the Tourette Syndrome clinic. Participants' parents were approached by a research assistant not involved in their care, and informed consent for the participation of their children was obtained (see Appendix A and B). The investigator made it clear to the family that their participation was strictly voluntary, and the decision not to participate would not adversely affect their future care at the clinic. Ethical approval for this study was obtained from the Department of Psychiatry Human Review Committee, University of Toronto Office of Research Services, and also from the OISE/UT Ethics Committee.

Inclusion Criteria:

1. All participants met the DSM-III-R criteria for the diagnosis of TS.
2. Some of the participants met DSM III-R criteria for diagnosis of ADHD and/or Compulsive Disorder in addition to TS.

3. Participants had a good command of the English language and were able to understand and cooperate with the research protocol.
4. Participants were between the ages of 6 years 0 months and 16 years 11 months at the onset of testing.
5. Informed consent was obtained from all participants.

Exclusion Criteria

1. The presence of another significant neurological and/or psychiatric disorder, e.g. brain damage, depression.
2. Treatment with anxiolytic, neuroleptic or antiobsessive medications within two months prior to entering the study.

The control group for this study was recruited from advertisements made to the General Population. Each control subject received twenty (20.00) dollars for travel expenses. The control participants had no history of behavioral disorders.

All participants were medication free at the time of evaluation.

Diagnostic Assessment

Diagnosis of TS was made by an experienced clinician based on DSM III-R criteria (APA, 1987) in the TS Clinic, Toronto Hospital. Diagnosis of ADHD and OCD were made by an experienced clinical interviewer based on the outcome scores of their clinical semi-structured interview using the **Schedule for Affective Disorder and Schizophrenia for School-Age Children Epidemiologic Version** (K-SADS-E, Orvaschel, H., 1995). Each child was then assigned to either the

control, Tourette's, Tourette's and Attention Deficit Hyperactivity, or Tourette's, ADHD and Obsessive Compulsive Disorder Groups.

Parents and teachers of the participants were asked to fill out behavioral questionnaires which included both closed (with 3 and 4 point scales) and open-ended questions.

Dependent measures were scores on the **Achenbach Child and Behavior Checklist** (Achenbach, T. M., & Edelbrock, C. ,1983) (Parent (CBCL) and Teacher (TRF) rating forms) for parent and teacher ratings of aggression. Principal measures also included the scores on the subscales of the **Conner's Rating Scales** that related to aggressive behavior (Conners, C. K., 1973).

Clinical Assessment Considerations

Researchers have recognized a number of difficulties involved in the assessment of patients with TS and Obsessive Compulsive Disorder (Kurlan, 1994; Walkup, Rosenberg, Brown & Singer, 1992). Considerations include: Tics tend to "wax and wane over time" changing in intensity, severity and anatomical location, often influenced by context, patients are sometimes able to suppress tics for limited periods of time, the actual presence of tics is usually underestimated by the patient; compulsive rituals tend to change over time, with one often completely replacing another, it is impossible to directly observe obsessions, it is sometimes difficult to distinguish between compulsions and tic behaviors. This study was designed using standardized measures whenever possible, including observation, questionnaire

and interview formats, as well as a variety of informants (the child, parents, teachers and examiner)

Instruments

i. Clinical Interview

Diagnosis of Obsessive Compulsive Disorder and ADHD were made by a trained Graduate Student interviewer based on the **Schedule for Affective Disorder and Schizophrenia for School-Age Children Epidemiologic Version** (K-SADS-E, Orvaschel, H., 1995), observational and historical data. The semi-structured diagnostic interview was designed to ascertain and record past and current episodes of Axis I psychopathology in children and adolescents (ages 6-17) according to the DSM-III-R criteria. Test-retest reliability for the Conduct Disorder was high (r=.89), reflecting the 17 components items of the scale. The internal consistency was also high (r=.86), indicating that the items of the scale showed covariance to a high degree. This is consistent with understanding that conduct disorder is a true syndrome rather than a collection of disparate behaviors (Chambers et al., 1985).

ii. Assessment for IQ

All participants were administered the **Wechsler intelligence Scale for Children** (WISCIII [Short form] (Wechsler, D., 1991). This short-form combination of the WISC-III contained the Information and Vocabulary (Verbal) as well as the Picture Completion and Block Design (Performance) subtests. The reliability with

the Full Version of the WISC-III is high, (*rtt*=.935), (Atkinson, L., & Yoshida, G., 1989).

iii. Assessment for Obsessive Compulsive Disorder

The Yale-Brown Obsessive Compulsive Scale (Y-BOCS) (Goodman, et al, 1989) was used in conjunction with the K-SADS-E (Orvaschel, 1995) to assess the severity of obsessions and compulsions according to the following criteria: Time spent on obsessions (compulsions); interference from obsessions (compulsions); distress from obsessions (compulsions); and actual control of obsessions (compulsions). Statistically, the Y-BOCS is highly correlated to the Mental Health Global Obsessive Compulsive Scale (NIMH-OC) (r=.67; p< .001, n=20), and the Clinical Global Impression Scale (CGI) (r=.74; p<.001, N=78) independent global measure of OCD, on comparisons of convergent and discriminant validity (Goodman et al., 1989)(see Appendix C).

iv. Assessment for Behavioral Problems

Indices of Aggressive Behavior were determined from parent and teacher reports on the **Achenbach Child and Behavior Checklist** (Achenbach, T. M., & Edelbrock, C. ,1983) (Parent (CBCL) and Teacher forms (TRF)). This questionnaire format instrument assesses a wide range of children's behavioral symptoms (Achenbach, T. M. & Edelbrock, C., 1983). The CBCL and TRF are designed to obtain parental and teacher reports for which there are normative data. Both versions of the questionnaire contain 113 descriptive items. Parents and teachers are asked to indicate whether each item is very true, somewhat true, or not true for

the child. Results are then scored by computer and produce a child behavior profile that consists of nine behavioral problem scales on the parent's form, and eight scales plus adaptive functioning scores on the Teacher rating form. Test-retest reliability of the Teacher Questionnaire ranges from .70 to .90, and test -retest reliability on the parent questionnaire averages .87, with inter-parent correlations averaging .67 (Achenbach & Edelbrock, 1983).

Clusters of symptoms of behavioral disturbances were examined using the **Conners Behavioral Checklist** (Conners, C. K., 1973). The parent version (CPRS), a 93 item questionnaire, is designed to assess symptoms commonly associated with attention disorders and noncompliance in children. The 93 item parent rating instrument includes scales for conduct disorder, anxious-shy, restless-disorganized, learning problem, psychosomatic, obsessive-compulsive, antisocial and hyperactive-immature. A hyperactivity index is scored based on the extent to which the child performed behaviors that were usually considered as indicative of an underlying diagnosis of hyperkinesis.

The long version of the Teacher Rating Scale (CTRS-39) was also used in this study, which clustered the behaviors of the child into the areas of a) classroom behavior, b) group participation and c) attitude toward authority and d) adaptive functioning. It is reported that these factor scores remain relatively stable across ages and a broad range of social classes (Conners, 1973). Scale scores were summed by the weights given for the 39 items that were scored to yield seven factors (hyperactivity, conduct problem, emotional overindulgent, anxious-passive, asocial, daydream-attention problem, 10-item hyperactivity index).

Parents and teacher(s) rated items on a 4 point scale. Item scores were then summed and the percentage of a possible score was determined. The reliability of the Conner's Teacher Rating Scale is fairly high for one-month test-retest (.72 -.91) remaining at a moderate level over a one-year period (.53) and the one-year test-retest on the Parent version of the scale ranged from .40 to .70 (Glow, Glow, & Rump, 1982). In addition, the Hyperactivity Index on the CTRS-39 has been found to be an effective primary screener for ADHD (Boyle & Jones, 1985).

v. Assessment for Motor and Vocal Tics

An estimate of tic severity was made by the investigator using a three (3) point global impression instrument, based on the **Yale Global Tic Severity Scale** (YGTSS), (Leckman, J. F., et al., 1988) which was completed by the parents combined with direct observation (see Appendix D). Also, parents/guardians, teachers and participants were asked to complete a visual analogue scale regarding the severity of tics during the past week. This visual analogue scale consisted of a line 10 cm. long, drawn on a page with descriptors at each end, e g. 'no tics' - 'very severe tics'. A slash was made along the line at the point which reflected the past weeks' symptoms.

vi. Assessment of Social Economic Status

The **Socio-Economic status index for Occupations in Canada** (Blishen, B. 1976) was determined by parental disclosure. Occupations were classified by ranking according to the Canadian labor force and provincial labor force socio-economic index for 1961. Ranking for the purpose of this study was as follows:

1= 4% of the work force, Socio-economic index 70.00+

2= 5% of the work force, Socio-economic index 60.0-69.99

3= 10 % of the work force, Socio-economic index 50.00-59.99

4= 20 % of the work force, Socio-economic index 40.00-49.99

5= 35 % of the work force, Socio-economic index 30.00-39.99

6= 26 % of the work force, Socio-economic index below 30.00.

CHAPTER THREE

RESULTS

Sample Characteristics

Results will be presented both as a comparison of TS to controls, as well as TS with comorbidity to controls. The results will be discussed in relation to general trends, and then as they relate to the hypotheses posed in this study. Twenty-five TS participants, mean age 10.52 (sd ± 2.21) years (range 6-14), and 5 general population controls, mean age 9.4 (sd ± 1.81) years (range 7 to 12) were enrolled in the study.

The participants with Tourette's and comorbidity tended to be somewhat older than those with TS alone or controls. There were no statistically significant differences in age between any of the diagnostic groups. The distribution of participants according to diagnostic group and group mean age is shown in Table 1.

Table 1-Distribution of Participants According to Diagnostic Group and Group Mean Age

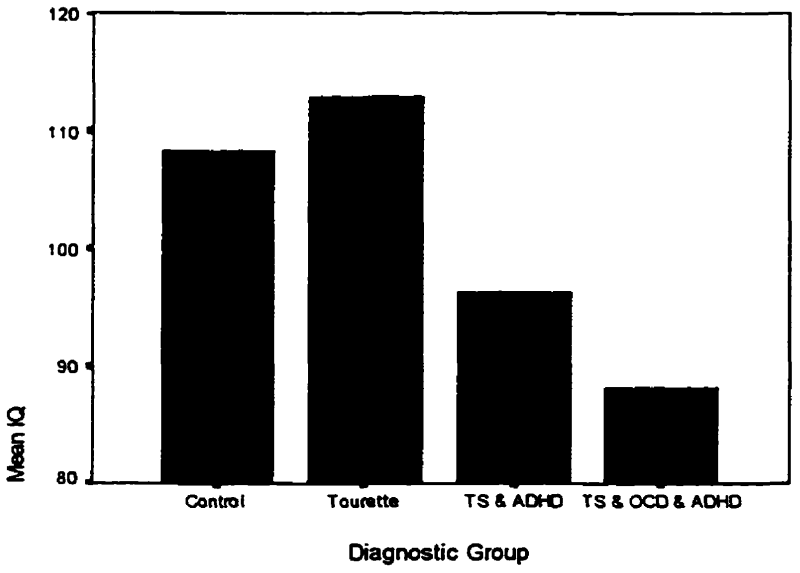
	N	Percent %	Mean Age	SD
Group				
Controls	5	16.7	9.40	1.81
Tourette's	7	23.3	9.71	2.49
TS + ADHD	11	36.7	10.63	2.41
TS/ADHD/OCD	7	23.3	10.83	2.09
Total	**30**	**100.0**		

Four of 25 TS participants (16%) and 1 of 5 control participants (20%) were female. There were no statistically significant differences in SES median ratings by

group, Control = 2.00, TS only = 2.00, TS with ADHD = 2.00, TS with OCD and ADHD = 3.00. As a result of the KSADS-E diagnostic interview, 27.8 % (n=5) of participants with TS and comorbid disorders met the additional criteria for Separation Anxiety, along with 14.3 % (n=1) with TS only and 0.0 % of Controls. The occurrence of Separation Anxiety was significantly higher in TS with comorbidity vs. controls. $F(21,19)=18.54$, $p < .001$.

The IQ scores of all 30 participants ranged from 47 to 147, with a mean of 100.13 (sd. ± 22.46). When examined by group the IQ scores of the controls (x=108.2, sd ± 30.99) was slightly lower than that of the pure TS group (x=112.7, sd ± 17.82), however the difference was not statistically significant. Significant differences in overall IQ scores were observed between the pure TS group and both TS plus ADHD (x=96.18, sd ± 12.84), $F(16,9) = .756$, p ,< .05, and the TS with both OCD and ADHD (x=88.0, sd ± 27.69), $F(16,7) = 6.526$, $p < .05$ (See Figure 1).

Figure 1. Mean Estimated IQ of Participants Categorized by Diagnostic Group *(N=30)*



According to the first hypothesis, participants with TS, as compared to controls, were expected to demonstrate a higher level of aggressive behavior. Sixty per cent of the participants (18/30) returned the teacher forms of the Child Behavior Checklist (TRF). Because of the low cell numbers in the TS and ADHD group (*n*= 11), and TS , OCD and ADHD group (*n*=7), these groups were collapsed to form a TS with any Comorbidity grouping.

Teacher reported aggression scores on the TRF were significantly higher in the TS group with comorbidity (x=1.7, sd ± .948) than the controls (x=1.00, sd ±.00), (F (12,9) = 23.172, p < .05). Similarly there was a significant difference between TS with comorbidity (x=1.7, sd± .948), F(12,9) =23.172, p < .05) and TS alone (x=1.00, sd ±.00). There was no significant difference between TS alone and control groups.

The limited variability in the control and TS only group, including both groups having standard deviations of zero, restricted any further comparisons between groups. Parent aggression ratings (CBCL) did not differ significantly between the control group (x=1.00, sd ±0.0), TS only group (x=1.28, sd ±.48), or TS with comorbidity group (x=1.28, sd ±.755).

The second research question investigated whether the rate of aggressive behavior in participants with one or two comorbid disorders would be different compared to those without comorbidity.

A Pearson Chi-Square test on the CBCL and TRF Aggression Rating Subscales for the TS, and TS with comorbidity groups, was not statistically significant (*Parent ratings* = $p<.46$, *Teacher ratings* = $p<.32$), however an interesting observation did emerge. Examining the groups through Crosstabulation on the CBCL, the TS only and TS with comorbidity participants were represented equally within the normal range of aggressive scores (71% and 78 %). Parents rated 28.6 % (n=2) of the TS only participants Borderline Aggressive, and zero within the Clinical Range, in comparison to the TS with Comorbidity group which parents rated 5.6 % (n=1) of the participants in the Borderline aggression range, and 16.7% (n=3) in the Clinical range.

Similarly, with the Teacher Rating Scale (TRF) , of the 4 TS only participants, 100% fell within the normal range of aggression. In contrast, 60 % (n=6) of the participants in the TS with Comorbidity group were in the normal range, 10 % (n=1) were Borderline, and 30 % of the participants were classified as Clinically Aggressive (n=3). See Table 2.

Table 2. Number of Subjects Scoring in the Normal, Borderline and Clinical Range on the Parent (CBCL) and Teacher (TRF) Subscale of Aggression for TS Only and TS with Comorbidity Participants

Rater	Group	*n*	Normal Range	%	Borderline	%	Clinical Range	%
Parent (CBCL)	TS Only	7	5	71	2	29	0	0
	TS + Comorbid	18	14	78	1	5	3	17
Teacher (TRF)	TS Only	4	4	100	0	0	0	0
	TS + Comorbid	10	6	60	1	10	3	30

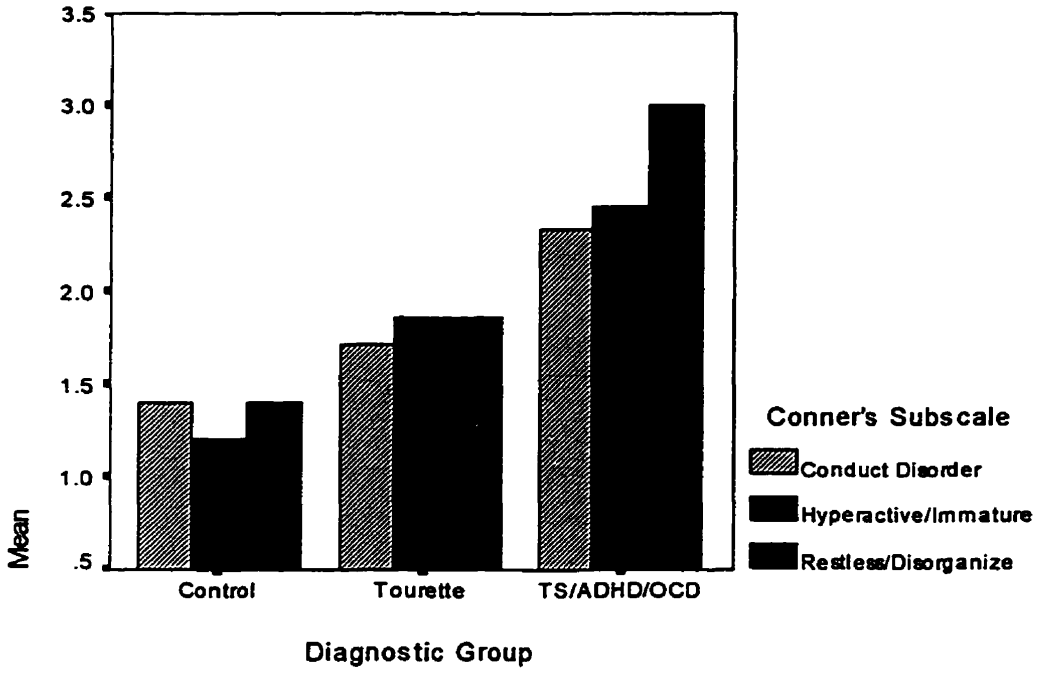
In order to further test the second hypothesis, subtests of the Conner's Rating Scale (CPRS) were analyzed. The limited sample size precluded a multivariate analysis on all the subscales, therefore for the purpose of this analysis, the subtests with similar constructs as CD were selected on face validity, in addition to their strength of correlation to the CD subscale. The Obsessive-Compulsive subscale (r=0.154) and Psychosomatic subscale (r=0.31) were excluded from the analysis based on weak correlations. In addition, learning Problems (r =.647) and Anxious/shy (r=.694) subscales were excluded. Although they moderately correlated to the CD subscale they were not considered related to the target construct.

Pearson Correlations on the remaining Conner's subscales revealed correlations with Conduct Disorder of r=.767 (Hyperactivity/Immaturity), r=.650 (Restlessness/Disorganized), and r=.560 (Antisocial Behavior) ($p < 0.01$). All four subtests were selected for the remaining analysis. TS plus comorbidity group and the TS only group differed significantly on the Aggression score as measured by the four selected subtests of the Conner's Rating Scales ($F(12,75)= 1.912$, $p<.046$).

Examining univariate results on each subscale of the Conner's Rating Scale, the TS with comorbidity group scored significantly higher on the Restless/Disorganized Scale (x= 3.0, sd ± .69) than the control group (x =1.4, sd ±.55) and the TS only group (x=1.86, sd ±.69), (F (1,23) = 7.259, p < .001). The control group did not differ statistically from the TS only group (F(10,9)=.002, p<.963).

On the CPRS Hyperactivity scale, the same relationship was observed with the TS with Comorbidity group having the highest scores (x=1.9, sd ±0.3) compared to the TS only (x=1.7, sd ±0.5) and controls (x=1.2, sd ± 0.4). The relationship between the Controls and the TS with Comorbidity was significant F(5,21) = .904, p<0.001. There was a similar trend on the Conduct Disorder Scale, with Controls (x= 1.4, sd ± .54) and Tourette's (x= 1.7, sd ± .95) not differing significantly (F (3,26) = .1.664, p<.199), however the scores were significantly different between Controls and the TS with Comorbidity group (x=2.33, sd ± 1.08) (F (13,21) = 1.626, p = < .05). See Figure 2.

Figure 2. Four CPRS Subscale Scores (Selected by Aggression Factor) by Diagnostic Group *(N=30)*



Finally, the severity of tics, as measured by the severity score of TS on the **Yale Global Tic Severity Scale** (YGTSS), (Leckman, J. F., et al., 1988), was compared to their rating of aggressive behavior on the four selected subscales of the Conner's Parent Rating Scale. Using a factorial ANOVA with independent factors for group and tic severity and continuous aggression scores as the dependent variable, the main effect of tic severity on aggression was not significant (p <.160).

CHAPTER FOUR

DISCUSSION

The results of this pilot study provide preliminary evidence that the aggressive behavior observed in the population of children with Tourette's Disorder is associated with the presence of comorbid ADHD and/ or OCD. In contrast, children with Tourette's without comorbidity do not differ statistically from controls on any of the measures of aggressive behavior, across both the school and home environments. These findings are consistent with the as yet unpublished study being conducted by Bruun and Budman (as cited in Bruun, R. D., & Budman, C. L., 1997), who have observed that aggression, and explosive symptoms of uncontrollable rage, are highly correlated with comorbid OCD and ADHD, but not with tic severity. It is important to highlight the lack of aggressive behavior in the participants who have TS only. This is consistent with the clinical observation that most TS patients have only minimal symptoms, that do not interfere with their day to day functioning. Only a minority of TS patients with one or more comorbid conditions tend to have difficulties requiring treatment.

Aggression and IQ

Children with TS alone were similar to the controls on the measure of intelligence (IQ). However, in the TS plus ADHD and/or OCD group, the average IQ of the participants was significantly below that of the TS only and control groups. This is consistent with previous studies which have also reported that the IQ scores

of children with TS and comorbid ADHD were significantly lower than that of children with TS alone, (Schuerholz, L. J. et al., 1996; Faraone, S. V., Biederman, J., & Lehman, B. K., 1993). This association suggests that the presence of ADHD in addition to TS may indicate a more pervasive disturbance in brain function.

The observed correlation of both elevated aggressive behavior, and the lower IQ in children with TS and comorbidity, also raises the possibility that children with lower intelligence, combined with impulsivity due to ADHD and/or rigidity due to OCD, may find it more difficult to cope with the demands of the school environment. It is quite likely that the child with comorbid OCD is inhibiting compulsive urges, while also trying to inhibit tic expression. This frustration then may lead to outbursts of aggressive behavior even with trivial provocation.

The increased frequency of aggressive behavior among TS participants with comorbid ADHD and/or OCD may be due to one underlying cause which is activating TS, ADHD and OCD, and also leads to loss of inhibition of the aggressive behavior. Alternately, aggressive behavior may be largely associated with the comorbid conditions (ADHD & OCD). To distinguish between the two hypotheses, it will be necessary to examine participants with OCD only and ADHD only, without history of tic disorder as was envisioned in the thesis proposal. This could not be accomplished in this study due to the limited time and difficulties in accessing those populations in a timely manner.

Separation Anxiety

It is intriguing that in this sample only participants with TS and comorbid disorders presented with Separation Anxiety. Are these children with TS and

comorbid disorders suffering from anxiety because they are attempting to cope with symptoms that may threaten their relationships with the family and peers, and they react out of fear and helplessness? However there may be an organic cause for this association since there are reports that the use of neuroleptics has led to the development of Separation Anxiety in children(Linet, L. S., 1985). It is important to note however that the children in the current study were all medication free. As a result of the initial diagnostic interview, a significantly higher percentage of the participants with TS and comorbidity (27.8 %) met diagnostic criteria for Separation Anxiety as compared to controls (0.0%). These findings are consistent with Coffey and Park (1997) who observed that of 100 consecutive new TS patients at a specialty clinic, 64 % met the criteria for a non-OCD type anxiety disorder, and specifically, 26 % were diagnosed as having comorbid Separation Anxiety. The relationship between Separation Anxiety and aggressive behavior requires further investigation.

Limitations and Future Research Directions

Although the present study supports the hypothesis that aggressive behavior is more likely among the participants with TS and comorbid ADHD and/or OCD, it is not clear whether the participants age or developmental level were relevant factors. The limited number of participants in this study restricted division by age, and therefore both latency period children and adolescents were grouped together. Due to differences in insight and self-control between younger and older children this would have been the preferable way to proceed. Also, the author acknowledges

that the small sample size limited the analysis of participants by specific comorbid disorder. It is also important to note that the participants with TS in this study were all recruited from a specialty TS clinic, and therefore may not be representative of the TS population as a whole. Thus the results can not be generalized to TS subjects in the community.

Future investigations with larger groups would allow the researchers to divide the participants by age grouping and TS comorbidity. In addition, participants with OCD alone and ADHD alone without TS should be included for comparison. Groups of participants without TS would help to clarify whether it is the TS, the comorbid disorder, or the combination of TS with comorbid disorder that is associated with the increased risk of aggressive behavior.

The instruments used to measure aggressive behavior require comment. Both instruments used were pen and pencil reports completed by the parents and teachers. At this time no satisfactory instrument that effectively measures aggressive behavior is available, but perhaps the incorporation of additional methods of measuring the child's aggression would be beneficial. It is recognized that each family has varying interpretations of aggression, in addition to different levels of acceptable aggressive behavior. Incorporation of an instrument or an observational method that would measure the family's aggression threshold would add validity to the construct being measured.

A final note on the design of this the study is necessary. A caveat commonly put forth in many studies with correlational data is the acknowledgment that causality cannot be inferred from the results obtained. This is true for the current

study as well, especially because of the complex nature of this neuropsychiatric disorder. There is insufficient data to identify which factors influence the aggressive behavior in these children.

Implications of this Study

It is vital that those involved in the child's care and management have the knowledge and understanding of what factors may be at work when children behave aggressively. These are considerations that need to be addressed when developing an intervention program for children with TS. Each child must be assessed thoroughly, regarding not only TS, but also the associated disorders.

The implications of each additional diagnosis must be shared with the parents, doctors and teachers involved in the care and management of the child. Because the constellation of symptoms and their severity varies a great deal between individuals, ideally each child with TS would benefit from a multi-disciplinary team of well informed professionals, working together to provide support and intervention in the home and school.

References

Achenbach, T. M., & Edelbrock, C. (1983). Manual for the Child Behavior Checklist. Burlington, VT: Authors.

American Psychiatric Association. (1987). Diagnostic and statistical manual of mental disorders. (DSM-III-R). 3rd ed. Revised. Washington, DC: American Psychiatric Association.

American Psychiatric Association. 1994. Diagnostic and statistical manual of mental disorders. (DSM-IV). 4th ed. Washington, DC: American Psychiatric Association.

Atkinson, L., & Yoshida, G. (1989). A BASIC program for determining reliability and validity of subtest combination short forms. Educational and Psychological Measurement, 49, 141-143.

Baron, R. A., & Byrne, D. (1984). Social psychology: Understanding human interaction (4th ed.) Boston: Allyn & Bacon.

Blishen, B., & McRoberts, H. A. (1976). A revised socio-economic index for occupations in Canada. Rev. Canadian Review of Sociology and Anthropology, 13, 1.

Bornstein, R. A., Stefl, M. E., & Hammond, L. (1989). A survey of Tourette Syndrome patients and their families: The 1987 Ohio Tourette survey. Journal of Neuropsychiatry, 2 (3), 275-281.

Boyle, M. H., & Jones, S. C. (1985). Selecting measures of emotional and behavioral disorders of childhood for use in general populations. Journal of Child Psychology and Psychiatry and Allied Disciplines, 26, 137-159.

Bruun, R. D., & Budman, C. L. (1997). The course and prognosis of Tourette Syndrome. Neurologic Clinics of North America, 15 (2), 291-298.

Budman, C. L., & Bruun, R. D. (1997). [Pilot study]. Unpublished raw data.

Chambers, W. J., Puig-Antich, J., Hirsch, M., Paez, P., Ambrosini, P.J., Tabrizi, M. A., & Davies, M. (1985). The assessment of affective disorders in children and adolescents by semistructured interview: Test-retest reliability of the Schedule for Affective Disorders and Schizophrenia for School-Age Children, Present Episode Version. Archives of General Psychiatry, 42, 696-702.

Coffey, B. J., & Park, K. S. (1997) Behavioral and emotional aspects of Tourette syndrome. Neurologic Clinics of North America, 15 (2), 277-289.

Cohen, D. J., & Leckman, J. F. (1994) Developmental psychopathology and neurobiology of Tourette's Syndrome. Journal of American Academy of Child and Adolescent Psychiatry, 33(1), 2-15.

Comings, D. E., & Comings, B.G. (1985). Tourette Syndrome: Clinical and psychological aspects of 250 cases. American Journal of Human Genetics. 37, 435450.

Comings, D. E., & Comings, B. G. (1990). A controlled family history study of Tourette's syndrome. I. Attention deficit hyperactivity disorder and learning disorders. Journal of Clinical Psychiatry 51, 275-280

Conners, C. K. (1973). Rating scales for use in drug studies with children. Psychopharmachology Bulletin [Special issue Pharmacotherapy of children] 24-84.

Dykens, R., Riddle, M., Hardin, M., Schwaartz, S., & Cohen, D. (1990). Intellectual, academic, and adaptive functioning of Tourette's syndrome children with and without attention deficit disorder. Journal of Abnormal Psychology, 18, 607-615.

Erenberg, G., Cruse, R. P., & Rothner, A. D. (1986). Tourette Syndrome: An analysis of 200 pediatric and adolescent cases. Cleveland Clinic Quarterly, 53, 127-131.

Faraone, S. V., Biederman, J, & Lehman, B. K. (1993). Intellectual performance and school failure in children with attention deficit hyperactivity disorder and in their siblings. Journal of Abnormal Psychology, 102, 616-623.

Glow, R. A., Glow, P. H. ,& Rump, E. E. (1982). The stability of child behavior disorders: A one year test-retest study of Adelaide versions of the Conners Teacher and Parent Rating scales. Journal of Abnormal Child Psychology, 10, 33-60.

Golden, G. (1984). Psychological and neuropsychologic aspects of TS. Neurology Clinical, 2. 91-102.

Goodman, W. G., Price, L. H., Rasussen, S. A., Mazure, C., Delgado, P., Heniger, G. R., & Charney, D. S. (1989). The Yale-Brown obsessive compulsive scale. Archives of General Psychiatry, 46, 1012-1016.

Hansen, C. R., Jr. (1992). What is Tourette syndrome? In T. Haerle (Ed.), Children with Tourette syndrome: A parents' guide (pp. 1-25). Rockville, MD: Woodbine House.

Kurlan R. (1989). Tourette's syndrome. Neurology, 39. 1625-1630.

Kurlan, R. (1994). Hypothesis II: Tourette's syndrome is part of a clinical spectrum that includes normal brain development. Archives of Neurology. 51. 11451150.

Lahey, B. B., & Loeber, R. (1994). Framework for a developmental model of oppositional defiant disorder and conduct disorder. In D. K. Routh (Ed.), Disruptive behavior disorders in childhood (pp. 139-180). New York: Plenum Press.

Leckman, J. F., Riddle, M. A., Hardin, M. T., Ort, S. I, Swartz, K. L., Stevenson, J., & Cohen, D. J. (1988). The Yale global tic severity scale: initial testing of a clinician-rated scale of tic severity. American Academy of Child and Adolescent Psychiatry. 28(4). 566-573.

Linet, L. S. (1985). Tourette syndrome, pimozide, and school phobia: The neuroleptic separation. American Journal of Psychiatry, 142, 613-621.

Loeber, R., & Verhulst, F. C. (1988). Patterns of behavior onset of conduct problems. Western Psychiatric institute and Clinic. University of Pittsburgh, Pittsburgh, PA.

Loeber, R., Tremblay, R. E. Gagnon, C, & Charlebois, P. (1989). Continuity and desistance in early fighting in school. Development and Psychopathology, 1, 39-50.

Loeber, R., Green, S. M., Keenan, K., & Lahey, B. B. (1995). Which boys will fare worse? Early predictors of the onset of conduct disorder in a six year longitudinal study. Journal of American Academy of Child and Adolescent Psychiatry, 34(4). 499-509.

Mason, A., Banerjee, S., Eapen, V., Zeitlin, H, & Robertson, M. M. (1998). The prevalence of Gilles de la Tourette's syndrome in a mainstream school population: A pilot study. Developmental Medicine & Child Neurology, 40, 292-296.

Moldofsky, H., Tullis,C., & Lamon, R. (1974). Multiple tic Syndrome (Gilles de la Tourette's Syndrome). Journal of Nervous Movement Disorders. 159, 282-292.

Orvaschel, H. (1995). Schedule for affective disorders and schizophrenia for school age children-Epidemiologic version, K-SADS-E, Version 5). Nova Southeastern University, Fort Lauderdale, Florida.

Pauls, D. L., Towbin, K E., & Leckman, J. F. (1986). Gilles de la Tourette syndrome and obsessive-compulsive disorder: Evidence supporting a genetic relationship. Archives of General Psychiatry. 43, 1180-1182.

Randolph, C. R., Hyde, T. M., Gold, J. M., Goldberg. T. E., & Weinberger, D R. (1993). Tourette's Syndrome in monozygotic twins: Relationship between tic severity to neuropsychological function. Archives of Neurology, 50,725-728.

Sandor, P. (1993) Gilles de la Tourette syndrome: A neuropsychiatric disorder. Journal of Psychosomatic Research Invited review 37:3, 211-226.

Schuerholz, L J., Baumgardner, T. L., Singer, H. S., Reiss, A. L., & Denckla, M. B. (1996). Neuropsychological status of children with Tourette's Syndrome with and without attention deficit hyperactivity disorder. American Academy of Neurology, 46, 958-965

Shapiro, A. K., Shapiro, E. S., Young, J. G., & Feinberg, T. E. (1988) Gilles de la Tourette Syndrome ,12th Ed. New York: Raven Press.

Singer, H. S., & Rosenberg, L. A. (1989). Development of behavioral and emotional problems in Tourette syndrome. Pediatric Neurology ,5, 41-44.

Singer, H S., Schuerholz, L., & Denckla, M. (1995). Learning difficulties in children with Tourette syndrome. Journal of Child Neurology ,10, S58-S61.

Singer, H. S., & Walkup, J. T. (1991). Tourette syndrome and other tic disorders: Diagnosis, pathophysiology, and treatment. Medicine, 70, 15-32.

Spencer, T. ,Biederman, J., Harding, M. (1995) The relationship between tic disorders and Tourette's syndrome revisited. Journal of American Academy of Child and Adolescent Psychiatry, 34, 1133-1147.

Stefl, M E. (1984) Mental health associated with Tourette's Syndrome. American Journal of Public Health, 74, 1310-1313.

Stefl, M. E, & Rubin, M. Tourette syndrome in the classroom: Special problems, special needs Journal of School health, 55, 72-75.

Thibert, A. L., Day, H. L., & Sandor, P. (1995). Self-concept and self conscientiousness in adults with Tourette syndrome Canadian Journal of Psychiatry, 40, 35-39.

Walkup, J. T., Rosenberg, L. A., Brown, J., & Singer, H. S. (1992). The validity of instruments measuring tic severity in Tourette's syndrome. Journal of the American Academy of Child and Adolescent Psychiatry, 31, 472-477.

Wand, R. R., Matazow, G. S., Shady, G. A., Furer, P., & Staley, D. (1993). Tourette Syndrome: Associated symptoms and most disabling features. Neuroscience and Biobehavioral Reviews, 17, 271-275.

Wechsler, D. (1991). Manual for the Wechsler intelligence Scale for children, Third edition. San Antonio: The Psychological Corporation.

Appendix A

CONSENT FORM
for children and youths aged 6-17
(including Control Participants)

I understand that Robyn Stephens and Dr. P. Sandor are trying to learn more about my condition in children and young adults. They want to find out what makes children with these problems get angry, and sometimes lose their tempers, so that they can help children get along with others better. I agree to fill out the forms with my parents/guardian, along with Robyn Stephens during a one and a half hour interview. My parents and my teacher(s) will also fill out some forms about my behavior at home and in the classroom.

I understand that, I don't have to fill out these forms, and even if I don't want to fill out the forms I will get the best available help with my problems at the Toronto Hospital. I know that I can decide at any time that I don't want to come to the interview, or to stop the interview, even if my parents/guardian say it's okay for me to come. I understand that information about me will be stored in a locked cabinet, in a locked room and will not have my name on it. My privacy will always be protected. My name will not be used if the results are published.

I have spoken to Robyn Stephens or Dr. P. Sandor about the study, and I want to come to the interview. I have been given a copy of this consent form.

_____	_____
Signature	Printed Name
_____	_____
Witness	Date

For further information, contact:

Robyn J. Stephens
Tourette's Syndrome Clinic
Toronto Hospital~Western Div.
(416) 603-5800 x2149

Dr. Paul Sandor
Tourette's Syndrome Clinic
Toronto Hospital~Western
(416) 603-5794

Appendix B

CONSENT FORM
(PARENT/GUARDIAN VERSION)

I, _____, agree to participate in this study along with my child _____ which is being conducted by Robyn Stephens under the supervision of Dr. P. Sandor. The purpose, benefits, risks and method have been explained to me by Robyn Stephens.

The purpose of this study is to obtain a better understanding of children with Tourette's Syndrome, and how additional problems, like Obsessive Compulsive Disorder and/or Attention Deficit Hyperactivity Disorder may affect levels of aggressive behavior. Success in this study may lead to a more adequate therapeutic approach to the above-mentioned disorders.

During a scheduled interview that will take approximately and hour and a half, I will be asked to fill out several questionnaires regarding my child's behavior at home, and will be asked questions about my child's past and present health problems. My child's teacher(s) will also be asked to fill out two questionnaires about my child's behavior in the classroom.

There will be no direct benefits to me or my child from this study. The only possible risk to me or my child, could be upset feeling evoked by talking about sensitive issues but everything possible will be done to make me feel at ease.

I understand that if I refuse to participate in this study, I am assured that my future relationship with the clinic will in no way be compromised, nor will my child's care at the clinic be jeopardized. My child will be asked separately for their assent to participate in this study, and they have the right to refuse any involvement if they so desire.

I,_____, have read and understand the consent form for this study. I have had the purposes, procedures, and technical language of this study explained to me. I have been given sufficient time to consider the above information and to seek advice. I have received a copy of this consent form for my information. If I have further questions or problems regarding this study I will contact Robyn Stephens at (416) 630-5800, ext. 2149, or Dr. P. Sandor at (416) 603-5794.

By signing this consent form, I am indicating that I agree to participate in this study. I have received a copy of this consent form.

_____ _____
Signature of Parent/Guardian Date

_____ _____
Witness Date

If I have concerns regarding my rights as a research subject, I may contact Ms. S. Pilon at (416) 978-5585.

Appendix C

YALE-BROWN SYMPTOM CHECKLIST

Child Name_____ Date_____ Reporter_____

Current Past

Aggressive Obsessions

___ ___ Fear might harm self
___ ___ Fear might harm others
___ ___ Violent or horrific images
___ ___ Fear of blurting out obscenities or insults
___ ___ Fear of doing something else embarrassing
___ ___ Fear will act on unwanted impulses (e.g. to stab friend)
___ ___ Fear will steal things
___ ___ Fear will harm others because not careful enough (e.g. hit/run motor vehicle accident)
___ ___ Fear will be responsible for something else terrible happening (e.g. fire, burglary)
___ ___ Other _____

Contamination Obsessions

___ ___ Concerns or disgust with bodily waste or secretions (eg urine, feces, saliva)
___ ___ Concern with dirt or germs
___ ___ Excessive concern with environmental contaminants (eg asbestos radiation, toxic waste)
___ ___ Excessive concern with household items (eg cleansers, solvents)
___ ___ Excessive concern with animals (eg insects)
___ ___ Bothered by sticky substances or residues
___ ___ Concerned will get ill because of contaminant
___ ___ Concerned will get others ill by spreading contaminant (aggressive)
___ ___ No concern with consequences of contamination other than how it might feel
___ ___ Other _____

Sexual Obsessions

___ ___ Forbidden or perverse sexual thoughts, images, or impulses
___ ___ Content involves children or incest
___ ___ Content involves homosexuality
___ ___ Sexual behaviour toward others (Aggressive)
___ ___ Other _____

Hoarding/Saving Obsessions (distinguish from hobbies and concern with objects of monetary or sentimental value)

___ ___ _____

Religious Obsessions

___ ___ Concern with sacrilege and blasphemy
___ ___ Excess concern with right/wrong, morality
___ ___ Other _____

Obsession with need for symmetry or exactness

___ ___ With magical thinking (eg, worried that another will have accident unless things are in the right place)
___ ___ Not accompanied by magical thinking

Somatic Obsessions

___ ___ Concern with illness or disease
___ ___ Excessive concern with body part or aspect of appearance
___ ___ Other _____

Miscellaneous Obsessions

___ ___ Need to know or remember
___ ___ Fear of saying certain things
___ ___ Fear of not saying just the right thing
___ ___ Fear of losing things
___ ___ Intrusive (nonviolent) images
___ ___ Intrusive nonsense sounds, words, or music
___ ___ Bothered by certain sounds/noises
___ ___ Lucky/unlucky numbers
___ ___ Colours with special significance
___ ___ Superstitious fears
___ ___ Other _____

Cleaning/Washing Compulsions

___ ___ Excessive or ritualized hand washing
___ ___ Excessive or ritualized showering, bathing, tooth brushing, grooming, or toilet routine
___ ___ Involves cleaning of household items or other inanimate objects
___ ___ Other measures to prevent or remove contact with contaminants
___ ___ Other _____

Current Past

Checking Compulsions

____ ____ Checking locks, stove, appliances, etc.
____ ____ Checking that did not/will not harm others
____ ____ checking that did not/will not harm self
____ ____ Checking that nothing terrible did/will happen
____ ____ Checking that did not make mistake
____ ____ Checking tied to somatic obsessions
____ ____ Other _____

Repeating Rituals

____ ____ Rereading or rewriting
____ ____ Need to repeat routine activities (eg in/out door, up/down from chair)
____ ____ Other _____

Counting Compulsions

____ ____ _____

Ordering/Arranging Compulsions

____ ____ _____

Hoarding/Collecting Compulsions (distinguish from hobbies and concern with objects of monetary or sentimental value, (eg carefully reads junk mail, piles up old newspapers, sorts through garbage, collects useless objects)

____ ____ _____

Miscellaneous Compulsions

____ ____ Mental rituals (other than checking/counting
____ ____ Excessive list making
____ ____ Need to tell, ask, or confess
____ ____ Need to touch, tap, or rub
____ ____ Rituals involving blinking or staring
____ ____ Measures (not checking) to prevent:harm to self____; harm to others____; terrible consequences____
____ ____ Ritualized eating behaviours
____ ____ Superstitious behaviours
____ ____ Trichotillomania
____ ____ Other self-damaging or self-mutilating behaviours
____ ____ Other_____

COMPLETE THE YALE-BROWN OBSESSIVE COMPULSIVE SCALE WITH SPECIFIC REFERENCE TO THE CHILD'S PRINCIPAL OBSESSIONS AND COMPULSIONS NOTED ABOVE.

Obsessions Obsession Subtotal: _____

	None	Mild	Moderate	Severe	Extreme
Time spent on Obsessions	0	1	2	3	4
Interference from Obsessions	0	1	2	3	4
Distress of Obsessions	0	1	2	3	4
Resistance	**Definitely resists** 0	1	2	3	**Completely yields** 4
Control over Obsessions	**Complete Control** 0	1	2	3	**No control** 4

Compulsions Compulsion Subtotal: _____

	None	Mild	Moderate	Severe	Extreme
Time spent on Compulsions	0	1	2	3	4
Interference from Compulsions	0	1	2	3	4
Distress of Compulsions	0	1	2	3	4
Resistance	**Definitely resists** 0	1	2	3	**Completely yields** 4
Control over Compulsions	**Complete Control** 0	1	2	3	**No control** 4

Appendix D

ID #: ☐☐☐

Y G T S S

Yale Global Tic Severity Scale

Yale Child Study Center

October 1992 version

NAME:	TODAY'S DATE : / /
RATER:	

MOTOR TIC SYMPTOM CHECKLIST (Check motor tics present during past week.)

•**Simple Motor Tics** (Rapid, Darting, "Meaningless"):

- ❑ Eye blinking
- ❑ Eye movements
- ❑ Nose movements
- ❑ Mouth movements
- ❑ Facial grimace
- ❑ Head jerks/movements
- ❑ Shoulder shrugs
- ❑ Arm movements
- ❑ Hand movements
- ❑ Abdominal tensing
- ❑ Leg, foot, or toe movements
- ❑ Other (describe):

_____

- ❑ Other (describe):

_____

•**Complex Motor Tics** (Slower, "Purposeful"):

- ❑ Eye movements
- ❑ Mouth movements
- ❑ Facial movements or expressions
- ❑ Head gestures or movements
- ❑ Shoulder movements
- ❑ Arm movements
- ❑ Hand movements
- ❑ Writing tics
- ❑ Dystonic postures
- ❑ Bending or gyrating
- ❑ Rotating
- ❑ Leg or foot or toe movements
- ❑ Blocking
- ❑ Tic related compulsive behaviors (touching, tapping, grooming, evening-up)
- ❑ Copropraxia
- ❑ Self-abusive behavior
- ❑ Paroxysms of tics (displays), duration ___ seconds
- ❑ Disinhibited behavior (describe):*

_____

- ❑ Other (describe):

_____

_____

_____

_____

PHONIC TIC SYMPTOM CHECKLIST (Check phonic tics present over the past week.)

•**Simple Phonic Symptoms** (Fast, "Meaningless" Sounds):

- ❑ Sounds, noises (circle: coughing, throat clearing, sniffing, or animal or bird noises)
- ❑ Other (list):

_____

_____

•**Complex Phonic Symptoms** (Language: Words, Phrases, Statements):

- ❑ Syllables (list)

_____

- ❑ Words (list)

_____

- ❑ Coprolalia (list)

_____

- ❑ Echolalia
- ❑ Palalalia
- ❑ Blocking
- ❑ Speech atypicalities (describe)

_____

- ❑ Disinhibited speech (describe)*

_____

* Do not include disinhibitions in ratings of tic behaviors

NUMBER	Motor	Phonic	
None	❑	❑	0
Single tic	❑	❑	1
Multiple discrete tics (2-5)	❑	❑	2
Multiple discrete tics (>5)	❑	❑	3
Multiple discrete tics plus as least one orchestrated pattern of multiple simultaneous or sequential tics where it is difficult to distinguish discrete tics	❑	❑	4
Multiple discrete tics plus several (>2) orchestrated paroxysms of multiple simultaneous or sequential tics that where it is difficult to distinguish discrete tics	❑	❑	5

FREQUENCY	Motor	Phonic	
NONE No evidence of specific tic behaviors	❑	❑	0
RARELY Specific tic behaviors have been present during previous week. These behaviors occur infrequently, often not on a daily basis. If bouts of tics occur, they are brief and uncommon.	❑	❑	1
OCCASIONALLY Specific tic behaviors 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.	❑	❑	2
FREQUENTLY Specific tic behaviors are present on a daily basis. tic free intervals as long as 3 hours are not uncommon. Bouts of tics occur regularly but may be limited to a single setting.	❑	❑	3
ALMOST ALWAYS Specific tic behaviors are present virtually every waking hour of every day, and periods of sustained tic behaviors occur regularly. Bouts of tics are common and are not limited to a single setting.	❑	❑	4
ALWAYS Specific tic behaviors are present virtually all the time. Tic free intervals are difficult to identify and do not last more than 5 to 10 minutes at most.	❑	❑	5

INTENSITY

	Motor	Phonic	
ABSENT	❑	❑	0
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
MILD INTENSITY Tics are not more forceful than comparable voluntary actions or utterances and are typically not noticed because of their intensity.	❑	❑	2
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.	❑	❑	3
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.	❑	❑	4
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.	❑	❑	5

COMPLEXITY

	Motor	Phonic	
NONE If present, all tics are clearly "simple" (sudden, brief, purposeless) in character.	❑	❑	0
BORDERLINE Some tics are not clearly "simple" in character.	❑	❑	1
MILD Some tics are clearly "complex" (purposive in appearance) and mimic brief "automatic" behaviors, such as grooming, syllables, or brief meaningful utterances such as "ah huh," "hi" that could be readily camouflaged.	❑	❑	2
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 behavior or speech (picking, tapping, saying "you bet" or "honey", brief echolalia).	❑	❑	3
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 behavior 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").	❑	❑	4
SEVERE Some tics involve lengthy bouts of orchestrated behavior 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-abusive behavior, or coprolalia).	❑	❑	5

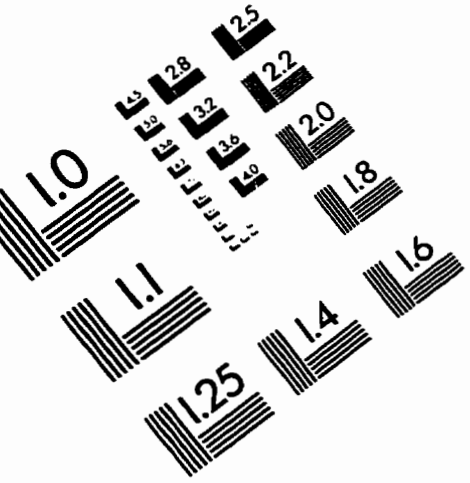
INTERFERENCE

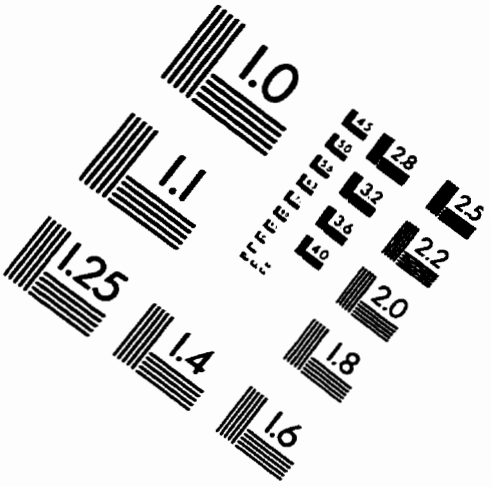
	Motor	Phonic	
NONE	❑	❑	0
MINIMAL When tics are present, they do not interrupt the flow of behavior or speech.	❑	❑	1
MILD When tics are present, they occasionally interrupt the flow of behavior or speech.	❑	❑	2
MODERATE When tics are present, they frequently interrupt the flow of behavior or speech.	❑	❑	3
MARKED When tics are present, they frequently interrupt the flow of behavior or speech, and they occasionally disrupt intended action or communication.	❑	❑	4
SEVERE When tics are present, they frequently disrupt intended action or communication.	❑	❑	5

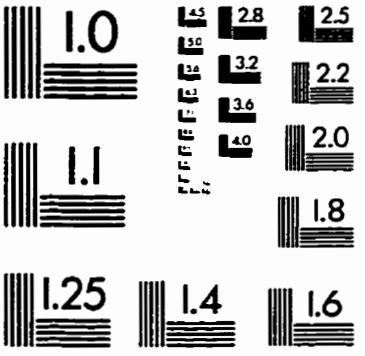
IMPAIRMENT

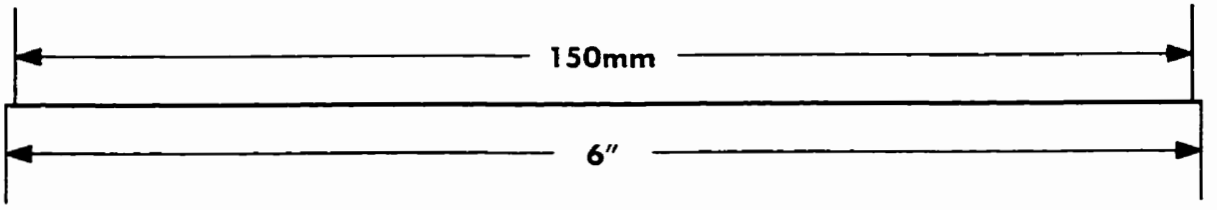
NONE	❑	0
MINIMAL Tics associated with subtle difficulties in self-esteem, family life, social acceptance, or school or job functioning (infrequent upset or concern about tics vis a vis the future, periodic, slight increase in family tensions because of tics, friends or acquaintances may occasionally notice or comment about tics in an upsetting way).	❑	10
MILD Tics associated with minor difficulties in self-esteem, family life, social acceptance, or school or job functioning.	❑	20
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).	❑	30
MARKED Tics associated with major difficulties in self-esteem, family life, social acceptance, or school or job functioning.	❑	40
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 tics - severely restricted life because of social stigma and social avoidance, removal from school or loss of job).	❑	50

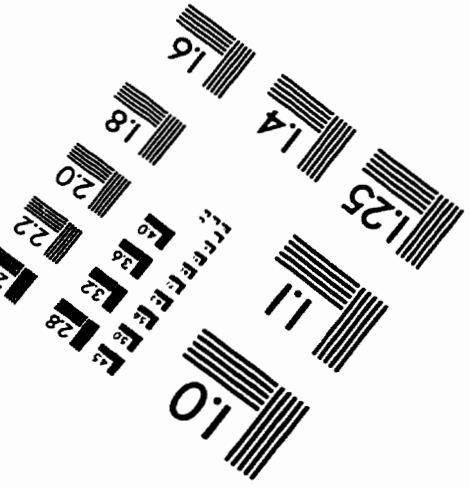
IMAGE EVALUATION
TEST TARGET (QA-3)

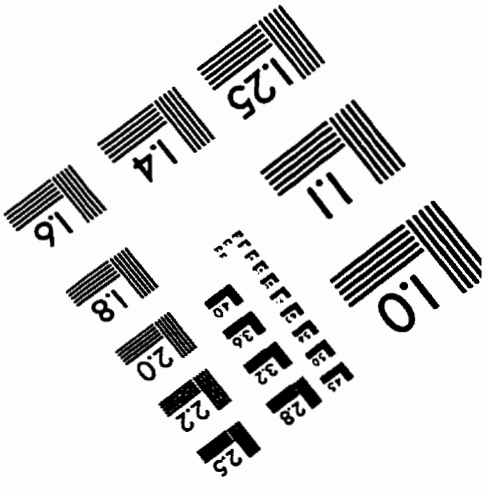












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