RELATIONSHIP OF SEVERITY OF SYMPTOMS IN

GILLES DE LA TOURETTE SYNDROME

TO INDIVIDUAL AND FAMILY FUNCTIONING

BY

LAURA M. CHAMPION, M.A.

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Acknowledgements

DEDICATION

To Katherine Furlong Adkock

with deepest love and appreciation

and to

Patrick Edward Champion

William Andrew Champion

and

Jessica Champion Carr

who are the next generation.

Abstract

Nineteen male and three female Tourette Syndrome patients, aged 8 -
16, and their parents were assessed to evaluate the relationship among
severity of TS symptoms, individual functioning, and family functioning. TS
subjects were administered the Rorschach Inkblot Test, the Piers-Harris
Children's Self Concept Scale (PH), and the Family Adaptability and
Cohesion Scales III (FACES III). Their parents completed an History and
Information Questionnaire, the Tourette Syndrome Global Scale (TSGS), FACES
III, the Family Satisfaction Scale, the Family Crisis Oriented Personal
Evaluation Scale (F-COPES), the Family Strengths Scale, and the Child
Behavior Checklist (CBCL). Clinicians at the St. Boniface Tourette Syndrome
Clinic rated the severity of TS symptoms in each subject using the TSGS in
order to assign subjects to High and Low TS severity groups. A multiple
regression analysis indicated that there was no significant predictive
relationship among TS symptom severity and selected predictor variables.
An Hotelling's T-Squared Test indicated that there were no significant
differences between High TS and Low TS groups on any of the variables
measured in this study. Significant differences between TS subjects and the
normative sample were found on a number of the measures. The data suggested
that TS subjects have more behavior problems than the normative sample and
that families wth a TS member are less cohesive and less flexible than
those in the normative sample. The data also indicated that TS subjects and
their parents have developed a number of adaptive strategies for coping
with the distress of this disorder. Implications for treatment are
discussed and future research directions are suggested.

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I. INTRODUCTION AND HISTORY OF THE STUDY OF TOURETTE SYNDROME

Tourette Syndrome (TS) is a relatively rare, chronic movement disorder. The distinguishing symptoms of TS are phonic and motor tics that vary in degree of intensity, frequency and complexity. This is a lifelong disorder, although people with TS may experience remissions that last months or even years. Current research indicates a neurological basis for TS (Shapiro & Shapiro, 1982), and there has been an emphasis on physiological aspects of TS for the past twenty-five years. Recently, however, a more integrative approach to medical practise has emerged, and some recent studies on TS reflect this perspective (Cohen, Detlor, Shaywitz & Leckman, 1982; Grossman, Mostofsky & Harrison, 1986; Wilson, Garron, Tanner & Klawans, 1982). The current study was designed to evaluate the effect, if any, of certain aspects of familial functioning on TS symptom severity, and to determine whether or not there are some individual and familial characteristics that are common among people with TS.

The history of the study of TS as a distinct disorder spans about 100 years. Shapiro, Shapiro, Bruun and Sweet (1978) divide the study of TS into six phases, punctuated by shifts between emphasis on physiological and psychological factors of the disorder. A seventh phase, characterized by the recently emerging trend towards integration of psychological and physiological factors, could be added to the original six.

The first phase became identifiable as such when the syndrome was clinically described by Gilles de la Tourette in 1885 (Shapiro, et. al.,

1978). Tourette described the syndrome as a nervous affliction, and noted that the symptoms were generalized motor tics and noises, accompanied by echolalia and coprolalia. He further noted that the illness began early in life (usually before puberty), afflicted more males than females, and that the symptoms were progressive in that new tics would be added to replace old ones. Although Tourette was initially impressed with the emotional stability of the nine patients he described in his original paper, he later revised his thinking about the social and emotional functioning of "tiquers". By 1899 he observed that there was always some history of nervous disorders in the families of the tiquers and had found that the tiquers themselves were afflicted by numerous anxieties, fantasies and fears. Consequently he came to believe that the tiquers were emotionally unstable and classified them as "higher degenerates". The designation of "higher degenerates" rather than "degenerates" reflected his observation that despite their emotional or mental "instability", these patients were frequently of average or above average intelligence and maintained high social and/or professional positions. Tourette believed that the prognosis for patients with this syndrome was related to the severity of the symptoms and to the recurrence of the symptoms after periods of remission. These variables determined the nature and extent of the interference of the tics in the physical and social life of the tiquer. He was firm, however, in his belief that despite any variability in types and severity of symptoms, TS was a lifelong disorder.

The etiology of the illness was not clear to Tourette, although he did assert that the illness was hereditary. Exactly what was inherited was not specified; possibly he was referring to "neuropathic antecedents" (i.e.,

family history of "mental instability") that would render the patient susceptible to developing a disorder such as TS. Because the disorder was thought to be caused by hereditary antecedents, Tourette and other investigators of his time placed little importance on psychological factors as causal agents in TS.

In sum, the hallmarks of the first stage of scientific investigation into TS, as described by Shapiro et al. (1978), were an emphasis on a description of the disorder and a conceptualization of the disorder that emphasized heredity as the etiological factor. Treatment was rest, isolation, and avoidance of fatigue.

The second stage, which began around the turn of the century, was identified by a marked shift towards emphasis on psychological factors of TS. A developing interest in these factors culminated in the treatise by Meige and Feindel (1902, translated by Wilson, 1907). In this highly influential paper, the authors defined a tic as a coordinated purposive act, initially provoked by some external cause or idea. The repetition of the tic led to its becoming habitual, such that it would be involuntarily reproduced in a nonpurposive fashion. Thus the tic would eventually assume the characterstics of a convulsive movement. These authors further noted that the tic was often preceded by an irresistible impulse, the suppression of which was associated with "malaise". Finally, they observed that when patients were distracted or made an effort to suppress a tic voluntarily, the effect was to "diminish its activity". This observation is consistent with current findings that absorption in some task attenuates tics (Bruun, 1984) and that tics may be voluntarily suppressed for minutes to hours (DSM-III, 1980). Meige and Feindel further described the person with TS as

having pronounced hereditary or neuropathic tendencies as well as numerous psychic or physical anomolies. They also observed a tendency towards multiple phobias and obsessions. Like Tourette, they postulated hereditary etiological factors, but they were vague about both the mechanism of inheritance and the precise nature of what it was that was inherited.

In both the first and second stages of conceptualizing and studying TS the evidence for inheritance of tics was based on familial history of "mental instability". Such evidence is clearly insufficient to explain the etiology of the behavior or inheritance of a tendency toward ticquing. To their credit, writers in both of these early stages made many astute observations of their patients. For example, the obsessive (or phobic) characteristics of many people with TS that were described by these writers has recently been studied in some depth (Pauls & Leckman, 1986).

Thus, the second phase in the study of TS reflected an increased emphasis on the psychological features of the disorder. Heredity, in some nebulous fashion, was considered to be the crucial etiological factor. Treatment was basically "rest and relaxation".

The shift to the next phase in the study of Tourette Syndrome was marked by Ferenczi's (1921) psychoanalytically oriented discussion of tics and Tourette Syndrome. He postulated that tics had an erotic origin. In this model, tics were understood as a displacement of genital sensation onto other body parts, resulting in increased pleasure in muscular movements. Abraham (1921) and Klein (1925) saw tics as a conversion symptom at the anal-sadistic level. Fenichel (1945) discussed the psychogenic tic, also positing that the underlying mechanism was conversion. He differentiated the conversion of the tic from the

conversion of hysteria. Fenichel maintained that the person with TS had both a compulsive character and a narcissistic orientation. He believed that many tics had a connection to compulsions, but also noted that some tics occurred without volition. He explained this by suggesting that the origin of such tics lay in compulsive motor patterns of such long standing history that they had become involuntary, automatic acts. The tics might serve a defensive function against certain emotions (rage, for example) or impulses (towards hostile actions, for example). The treatment literature of this era is primarily based on single cases. Treatment failure was essentially blamed on the patients who were thought to be unresponsive to treatment because of their narcissistic orientations.

The third phase evolved out of the second phase, with a stronger emphasis on psychological factors. In keeping with a psychoanalytic model, heredity was supplanted by early childhood experiences as the key to development of the disorder. Psychoanalysis was the prescribed, albeit generally unsuccessful, treatment.

The fourth phase, which is called the epidemiological phase, was marked by reviews of the literature on TS to that point, augmented by the collection and study of retrospective data on large numbers of TS patients (Shapiro et al., 1978). Although interest in the role that psychological factors play in the etiology and maintenance of tics remained dominant, an important result of this phase was a renewal of interest in possible organic factors in TS.

The foundation for the fifth phase was laid in the 1950's, when psychopharmacological treatment for mental disorders was introduced. In the 1960's, a number of researchers found that haloperidol, an

antipsychotic, was also effective in alleviating TS symptoms (Challas & Brauer, 1963; Seignot, 1961). The discovery of haloperidol, a potent antagonist of the dopamine receptor site, as an effective supressor of TS symptoms, fueled a growing trend towards defining TS as an organically-based disorder. Evidence from further success with psychopharmacological treatments of TS suggests that this disorder is indeed neurologically based. The effects of this phase are still apparent in current research and treatment trends; it might be argued that this phase has not been marked by a relatively distinct ending point, but has instead merged with the later phases.

The search for an effective treatment for TS digressed briefly in the early 1970's, when behavior therapists attempted to modify TS symptoms using behavioral techniques. The rationale for using such techniques was based on the observation that TS patients can temporarily control or completely suppress their tics. From this observation, some therapists and researchers questioned the growing acceptance of TS as a neurological disorder, and refocused their attention on psychological factors in the etiology of TS. This avenue of treatment and research has proven to be a blind alley since behavioral techniques have generally shown themselves to be ineffective in the long-term attenuation of TS symptoms (Shapiro et al., 1978). Behavioral techniques are sometimes still used in conjunction with medication, since such techniques can be effective in teaching people with TS to relax, and/or to temporarily suppress symptoms. Symptom suppression, however, is not without its price; there is almost always a rebound effect. After suppressing symptoms for a period of time, the person with TS will usually experience a temporary increase in the frequency and intensity of symptoms (Brunn, 1984).

The sixth, and last, phase that Shapiro et al. (1978) describe is a data-oriented one. There was a shift from theoretical or conceptual research to the development of an empirical data base. Shapiro et al. (1978) note that this shift in TS research occurred concurrently with a shift in general psychiatric research to a focus on "data orientation". They attribute this, in large part, to advances in the methodology of clinical studies, including double blind procedures, the use of more sophisticated statistical procedures, advances in the use of computer facilities, more reliable and valid measures of subjective states, and the development of placebo studies. According to Shapiro & Shapiro (1982), this became the "dominant approach to the study of TS" (p. 18).

The developments in the study of TS have been marked by an "either/or" approach to understanding the etiology and the nature of this disorder. The pendulum has swung back and forth between the dominance of physiological and psychological perspectives. In the psychoanalytic tradition, tics are thought to be related only to psychological factors. At the other end of the contiuum, Shapiro et al. (1978, 1981) contend that psychological factors play no role in the development, maintenance, or severity of TS. They contend that there is no evidence of patterns of psychological dysfunction or impairment in TS patients and maintain that, as a group, TS patients function surprisingly well (Shapiro et al., 1978).

Recently, a more interactive perspective has begun to emerge. Cohen, Detlor, Shaywitz and Leckman (1982) discuss the interaction between biological and psychological factors in TS. Golden (1984), Wilson, Garron, Tanner and Klawans (1982), and Grossman, Mostofsky and Harrison (1986) have challenged Shapiro et al.'s assertion that TS patients show little or no

impairment in psychological and social functioning. Although none of these researchers suggest in any way that TS is not a neurological disorder, all emphasize the role that psychological factors can play in the course of the disorder. The possibility that level of psychological functioning can affect to some degree the nature of specific tics, the severity of the symptoms, and the ability to cope with the stress of the disorder has been raised. Further, the likelihood that the disorder itself can affect the level of psychological and social functioning of the person with TS has been postulated (Grossman et al., 1986).

In summary, the study of TS as a specific disorder spans over a hundred years. The systematic study of large numbers of subjects, using more sophisticated designs and statistical analysis has emerged only in the last thirty to forty years. The course of the study of TS has been marked by abrupt, mutually exclusive shifts in the conceptualization of the disorder; these shifts have typically followed the current zeitgeist in conceptualizing mental disorders in general. Recently, a trend towards an integration of the physiological and psychological factors of TS has emerged, and the current study is a part of this most recent phase in the study of TS.

II. DEFINITION AND DESCRIPTION OF TOURETTE SYNDROME

A. Diagnostic Criteria

The diagnostic criteria for Tourette Syndrome, according to the Diagnostic and Statistical Manual of Mental Disorders Third Edition: Revised (DSM-III-R, 1987) are:

1. Both multiple motor and one or more vocal tics have been present at some time during the illness, although not necessarily concurrently.

2. The tics occur many times a day (usually in bouts) nearly every day or intermittently throughout a period of more than one year.

3. The anatomic location, number, frequency, complexity and severity of the tics change over time.

4. Age of onset before age 21.

5. Occurrence not exclusively during psychoactive substance intoxication, or known central nervous system disease, such as Huntington's chorea and postviral encephalitis.

According to the DSM-III-R (1987), the median age of onset is seven years; the majority of patients have an onset before 14 years. TS symptoms may, however, appear as early as one year. The current estimated lifetime prevalence of TS is a minimum of 0.5 per thousand. TS is at least three times more common among males than females. This is a lifetime disorder, although there may be periods of remission lasting weeks to years.

B. Symptoms

The initial symptom is frequently a single tic (such as an eyeblink).
Other tics include tongue protrusion, sniffing, squatting, hopping, throat
clearing or stuttering. Multiple tics often develop and can include any
combination of the above. Copropraxia, echolalia, echokinesis, and
palilalia are also symptoms of TS. Symptoms usually begin with the head or
face, then move to the torso and limbs. One of the most dramatic and
well-known symptoms, coprolalia (involuntary speaking or shouting of
obscene or socially unacceptable words or phrases) is seen in 30 to 50
percent of the cases (Bruun, 1984; Shapiro & Shapiro, 1982) and rarely
emerges before age 10 (Fernando, 1967). Bruun (1984) notes that the words
uttered are not always strictly coprolalic. Frequently, the words and
phrases are articulations of thoughts that the person with TS would like to
keep hidden. Thus this symptom may be a reflection of a breakdown in
inhibition and control as suggested by Cohen, Detlor, Shaywitz and Leckman
(1982). People with TS often develop complex motor sequences that may
include touching oneself or others, hitting, kicking, jumping, et cetera.
In addition, compulsive, ritualized behaviors are not unusual and
self-mutilating behaviors have been observed (Van Woert, Rosenbaum & Enna,
1982). Shapiro and Shapiro (1981) describe a fluctuation in symptoms, as
old symptoms may disappear and reappear, and with new symptoms being added
to old ones. Symptoms are thought to be exacerbated by stress (Bruun, 1984;
Grossman et al., 1986) and may be alleviated by absorbing activities
(Bruun, 1984). Although it was previously thought that tics disappeared
during sleep, some researchers have presented data suggesting that tics do
occur during sleep (Champion, Fulton & Shady, 1988; Glaze, Frost, &

Jankovic, 1983; Stefl, 1984). This data is equivocal, however, and the question of whether or not tics are present during sleep remains open. Barabas, Matthews, and Ferrari (1984a) have found that children with TS are significantly more prone to disorders of arousal (sleep walking and night terrors) than are children with seizure disorders or learning disabilities. Both Stefl (1984) and Champion, Fulton and Shady (1988) found, that both children and adults with TS are likely to experience somnabulism, insomnia, night terrors and nocturnal enuresis. Glaze et al. (1983) also found an inordinate amount of sleep disturbances among people with TS and related this finding to the maturational lag in the development of the central nervous system (CNS) in TS patients suggested by Shapiro et al. (1978).

C. Treatment

Several researchers, using separate reviews of the literature on pharmacological approaches to the treatment of TS, have concluded that haloperidol and pimozide, which are dopamine antagonists, are the most effective medications for the attenuation of TS symptoms (Shapiro & Shapiro, 1982; Van Woert, Rosenbaum & Enna, 1982). There is little support in the literature for psychologically-oriented treatment of TS except in conjunction with psychopharmacological agents (Shapiro et al., 1978). The work of Grossman, et al. (1986), Matthews et al. (1985), Cohen, et al. (1982), Wilson et al. (1982) and Golden (1984) suggest that attention needs to be paid to the psychological stress and/or dysfunction that may accompany TS. None of the above researchers suggest, however, that non-pharmacological treatment will alleviate the primary symptoms of TS.

III. ASSOCIATED FEATURES

Associated features of TS include mental coprolalia, obsessions and compulsions (DSM-III-R, 1987). According to DSM-III-R (1987), Attention-deficit Hyperactivity Disorder (ADHD) and Obsessive and Compulsive Disorder (OCD) are frequently associated with TS in clinical samples. Cohen et al. (1982) and Hagin, Beecher and Pagano (1982), among others, have found a high incidence of learning disabilities among people with TS.

A. Mental Coprolalia

Mental coprolalia is defined in the DSM-III-R (1987) as "sudden, intrusive, senseless thoughts of socially unacceptable or obscene words, phrases or sentences that differ from true obsessions in that no attempt is made to ignore, suppress or neutralize the thoughts" (p. 79). Patients at the TS Clinic at St. Boniface Hospital have also described intrusive, perseverative fantasies which may be related to mental coprolalia (G.A. Shady, personal communication, June, 1987). These patients describe their thoughts as being very distressing and perceive them to be out of their control.

B. Obsessions and Compulsions

In the DSM-III-R (1987), obsessions are defined as "persistent ideas, thoughts, impulses or images that are experienced, at least initially, as intrusive and senseless" (p. 245). Compulsions are defined as "repetitive, purposeful and intentional behaviors that are performed in response to an obsession, according to certain rules, or in a stereotyped fashion" (p. 245). The patient "generally recognizes the senselessness of the behavior, and does not derive pleasure from carrying out the activity, although it does provide a release of tension" (p. 245). Some of the more complex tics certainly resemble compulsions. Bruun (1984) notes that many TS patients "speak of an 'inner tension' which is almost continually present... The release of a tic will bring at least a momentary release of this tension" (p. 127).

C. Obsessive Compulsive Disorder

A number of researchers have found a high incidence of obsessive compulsive disorder (OCD) among people with TS and their first degree biological relatives (Fernando, 1967; Kidd & Pauls, 1982; Montgomery, Clayton & Friedhoff, 1982; Nee, Caine & Polinsky, 1980; Nee, Polinsky & Ebert, 1982; Pauls & Leckman, 1986; Pauls, Towbin, Leckman, Zahner, & Cohen, 1986). These researchers hypothesize a genetic relationship among OCD, chronic tics, and TS. The specific mode of inheritance has not been established, although Kidd and Pauls (1982) suggest that a single major locus model provides the best statistical fit to the available data.

D. Attention-deficit Hyperactivity Disorder

The essential features of Attention-deficit Hyperactivity Disorder (ADHD), which replaces Attention Deficit Disorder (ADD) in the DSM-III-R (1987), are developmentally inappropriate degrees of inattention, impulsiveness and hyperactivity. Comings and Comings (1984) have found that in families with one or more diagnosed TS patient, there was also a greater than normal prevalence of ADD. The relationship between these two disorders is of clinical importance because Golden (1984) and Lowe, Cohen, Detlor, Kremenitzer and Shaywitz (1982) suggest that stimulant medications can precipitate TS symptoms. Both Golden (1974) and Lowe et al. (1982) note that early TS symptoms may be difficult to distinguish from ADD. Comings and Comings (1984) suggest that TS and ADD co-exist in some patients and caution against calling ADD a "misdiagnosis" in these cases. Their data and that of Shapiro and Shapiro (1981) indicate that TS symptoms would develop with or without the use of stimulant medications, and these researchers contend that stimulant medications do not cause, provoke or precipitate TS symptoms. In the absence of a clear resolution of this controversy, it seems reasonable that caution should be exercised in prescribing stimulant drugs for the treatment of ADHD if there is a family history of TS or chronic tics, or if mild tics are present.

E. Learning Disabilities

Several researchers have found that children with TS show patterns of learning problems that could impede academic progress (Hagin et al., 1982; Ferrari, Matthews & Barabas, 1984; Incagnoli & Kane, 1981, 1983; Sutherland, Kolb, Schoel, Whishaw & Davies, 1982). These studies and those

of Shapiro et al. (1978) and Shapiro, Shapiro and Clarkin (1977) indicate that TS children tend to show deficits primarily in visuographic skills. For example, Hagin et al. (1982) found that as a group children with TS obtained scores on the Arithmetic subtest of the Weschler Intelligence Test for Children--Revised (WISC-R) that were consistent with other verbal subtest scores, but performed poorly on the Arithmetic subtest of the Wide Range Achievement Test (WRAT). Thus, although the subjects were able to mentally perform orally presented computations they had difficulty doing similar computations on paper. Incagnoli and Kane (1981, 1983) and Shapiro et al. (1974, 1978) found that TS children performed poorly on the Bender-Gestalt Test. A significant discrepancy between Verbal and Performance IQ scores on the WISC-R, with the Performance IQ generally (but not always) lower is common among TS patients (Hagin et al., 1982; Incagnoli & Kane, 1981, 1983; Shapiro et al., 1974, 1978).

IV. CURRENT RESEARCH

A. Physiological Factors

TS research has been plagued by some noteworthy methodological problems. The relative rarity of TS has made obtaining large samples difficult, and many studies have been based on small samples (N=1 in some cases). Other methodological problems are related to the nature of TS symptoms themselves. First, it has been shown (Shapiro & Shapiro, 1978) that TS symptoms wax and wane of their own accord. This means that results of drug studies may be difficult to interpret because it is unclear whether the alleviation of symptoms is due to a natural "downswing" in the cycle or to the effect of the drug (Caine et al., 1986). It is only recently that the diagnosis of TS has been standardized (DSM-III, 1980; DSM-III-R, 1987). Second, TS can be difficult to diagnose. Early symptoms, frequently resemble attention deficit disorders. TS symptoms also may closely resemble obsessive-compulsive symptoms, and distinguishing between the two disorders may pose some problems. Third, TS patients are able to suppress symptoms for minutes to hours (Bruun, 1984), and may do so in the doctor's office, thus making a medical diagnosis more difficult.

Furthermore, the precise etiology of TS has not yet been established. Following the success of haloperidol in the treatment of TS by Seignol in 1961, and by others since then (Shapiro et al., 1978), an overwhelming proportion of TS research has uncovered circumstantial evidence that strongly suggests an organic basis for this disorder (Shapiro et al., 1978;

Van Woert, Rosenbaum & Enna, 1982). The nature of TS, including the waxing and waning of both severity and type of symptoms, the subtle, non-specific neurological abnormalities (Caine, 1986), as well as the differential response of patients to medications has presented an etiological conundrum to TS researchers. Some of the research on the role of neurotransmitters and on the role of genetics in TS has shed light on the etiology of this syndrome.

The role of Neurotransmitters

The findings of neurochemical and psychopharmacological studies indicate that neurotransmitters play a role in TS symptoms. According to Devinsky (1983) and Feinberg and Carroll (1979), it has been consistently found that drugs that block postsynaptic dopamine receptors tend to alleviate TS symptoms, whereas drugs that increase dopaminergic activity tend to exacerbate TS symptoms. Haloperidol and pimozide, powerful dopamine antagonists, are the most frequently prescribed medications for TS, and are rated by patients as the most effective in alleviation of TS symptoms (Fulton, Shady and Champion, 1988). Levodopa (L-dopa), on the other hand, is a precursor to dopamine that has been found to exacerbate TS symptoms (Messiha & Carlson, 1983). Van Woert et al. (1982) suggest that haloperidol binds preferentially to the dopamine Z (D-2) receptors, thus blocking the release of dopamine.

Glaze, Frost and Jankovic (1983) and Barabas, Matthews and Ferrari (1984a, 1984b) have linked disorders in arousal among children with TS to disturbances in the metabolism of serotonin. Brody and Share (1957) found that inhibition of serotonin synthesis has resulted in general excitation

and increased locomotor activity in animals. Cohen et al. (1978) and Cohen et al (1979) also have also suggested that abnormalities in serotonin metabolism may be linked to TS symptoms. They found decreased levels of 5-hydroxyindoleacetic acid (5-HIAA) in the cerebrospinal fluid (CFS) of TS patients. Such a finding may indicate either a loss of serotonergic neurons and/or a hypersensitivity of serotonin receptors, thus leading to compensatory inhibition of serotonin synthesis.

Norepinephrine has also been circumstantially linked to TS symptoms. Cohen et al. (1978) have found that some TS patients respond to clonodine, an alpha-adrenergic agonist that reduces the turnover of norepinephrine (NE) in the CSF, possibly through feedback inhibition (Svensson, Bunney, Aghajanian, 1978).

Caine, Ludlow and Polinsky (1986) found that L-amphetamine tended to exacerbate vocal symptoms, whereas d-amphetamine tended to exacerbate motor symptoms. Golden (1982), Lowe, Cohen, Detlor, Kremintzer and Shaywitz (1982) also found that the stimulant drugs prescribed for hyperactivity frequently precipitated TS symptoms. Comings and Comings (1984) and Shapiro and Shapiro (1981) refute these findings, asserting that the TS symptoms would be manifested anyway. Caine et al. (1986) attempt to resolve this controversy by hypothesizing that some children with TS are sensitive to amphetamines, while others are not. Such mixed results are typical of TS psychopharmacological studies. No single medication has been found to have complete efficacy in alleviating TS symptoms, and some TS patients do not respond well to any medications (Barabas, Matthews, & Ferrari, 1984a). Efficacy ratings for haloperidol, for example, range from 50-90% (Shapiro & Shapiro, 1982). These findings have made establishing a single etiology for TS quite elusive.

Taken together, these data suggest that hyperarousal may play an important role in TS (A. Yazowitz, personal communication,November,1988).The tics themselves, which may be described as increased locomotor activity, as well as the associated features such as hyperactivity, emational lability, anxiety, behavioral problems, distractability, sleep disorders, and obsessive-compulsive behaviors may all be associated with cerebral hyperaruosal. Stimulant medications may exacerbate or precitate TS symptoms by increasing the level of arousal to some critical point. Dopamine is associated with level of arousal (Sacks,1987) and Haldol appears to work by decreasing the level of arousal in the patient. Indeed, some patients find that the side effects of mental and physical torpor experienced while on Haldol are more difficult to bear than are the tics (Fulton, 1988; Sacks, 1987).

The Role of Genetics

Another source of support for the belief that TS is an organic disorder comes from the study of the role of genetics in this disorder. In the late 1970's, a number of investigators began to suspect that TS was hereditary because a large number of patients had a family history of TS (Eldridge, Sweet, Lake, Zeigler, & Shapiro, 1977; Nee, Caine & Polinsky, 1980; Nee, Polinsky & Ebert, 1982; Shapiro et al., 1978). More recently, investigators have attempted to study the mode of genetic transmission (Baron, Shapiro, Shapiro & Rainer, 1981; Comings, Comings, Detlor, & Cloninger, 1984; Devor, 1984; Kidd & Pauls, 1984; Pauls & Leckman, 1986). As yet, a specific mode of transmission has not been established. The most recent data available (Pauls & Leckman, 1986) suggests that TS is

"inherited as a highly penetrant, sex-influenced, autosomal dominant trait" (p. 993).

Genetic studies have provided a wealth of information about TS patients and their families. One consistent finding (Pauls & Leckman, 1986; Pauls et al., 1981) is that relatives of female probands are statistically at much higher risk for developing TS than are relatives of male probands. Pauls and Leckman (1986) estimate that about 10% of all TS patients are phenocopies (i.e., do not show any family history of TS or associated behaviors). Genetic studies have uncovered a correlation among TS, chronic tics (CT) and OCD (Kidd & Pauls, 1982; Nee et al., 1980; Pauls et al., 1981; Pauls, Kruger, Leckman, Cohen and Kidd, 1984 and Pauls and Leckman, 1986). Pauls and Leckman hypothesize that these disorders are etiologically related. They also concluded that transmission of these disorders is sex-linked, and that there may be sex differences with regard to specific symptoms. For example, more fathers of probands were affected with CT whereas more mothers were affected with OCD. Pauls and Leckman do not suggest that all people who exhibit obsessive-compulsive behaviors have a disorder that is etiologically related to TS. They cite recent research that suggests that patients with OCD may be divided into a minimum of two groups: those with and those without family histories of tics.

To date, the search for an etiology of TS has been based primarily on circumstantial evidence, and there are a number of apparent inconsistencies and paradoxes in the data. Cohen et al. (1979) have attempted to resolve the inconsistencies by suggesting that TS has a multifactorial etiology in all patients and/or that TS symptoms are caused by different factors in different patients. An explanation such as this would be consistent with

the differential responses of people with TS to a variety of medications.
These investigators point out that any "adequate biological model for TS
must account for various types of metabolic, clinical, and pharmacological
data". They suggest that a combination of serotonergic, noradrenergic and
dopaminergic neurotransmitter systems may be responsible for TS symptoms.
The authors further hypothesize that the specific loading may vary in
importance from patient to patient, resulting in a differential response to
medication. They also suggest that the differential loading of factors may
reflect distinct genetic subgroups. This is an intriguing possibility that
could be investigated by gathering careful family data using a large sample
in a longterm, prospective study.

B. Psychological Factors

Although the data strongly suggest that physiological factors
predominate in the etiology of TS, recent research and accounts of clinical
experiences suggest that psychological factors may play a role in certain
aspects of the manifestation of this disorder. Both Bruun (1984) and
Grossman et al. (1986) have noted that stress can precipitate or exacerbate
TS symptoms. Wilson, Garron, Tanner and Klawans (1982) found that children
with TS exhibit a greater degree of behavioral disturbance than do children
in general and that their level of behavioral disturbance is comparable to
that of children in special education classes. They suggest that this may
be due to subtle neurological dysfunctions. Hagin, Beecher and Pagano
(1982) have documented a relatively high incidence of learning disabilities
(LD) among children with TS. A number of investigators have found a high
incidence of low self-esteem among LD children (Blood, 1985; Bryan & Pearl,

1979; Leviton & Kiraly, 1975). Given the above findings, it is likely that at least those TS children who also have LD would suffer from a lowered self-esteem. Comings and Comings (1984) found that 62% of their sample of 250 TS patients also had Attention Deficit Disorder (ADD). Cohen, Shaywitz and Leckman (1982) note that people with TS describe feeling "trapped" within their bodies and by their obsessions, and unable to move forward or backward when caught in the midst of a TS ritual. Matthews, Eustace, Grad, Pelcovitz and Olson (1985) found that families of TS children tend to have below average communication and that TS children view their families as less flexible and adaptable than do nonpatient children.

Other indicators that psychological factors may be relevant to the understanding and treatment of TS come from the literature on another group of people with a highly visible neurologically based disorder. Research on people with epilepsy and their families suggest that both children and adults with epilepsy, their siblings, and parents (especially mothers), are at risk to show for psychological distress or behavior problems (Hoare, 1984a, 1984b, 1984c; Herman & Whitman, 1984). Dorenbaum, Cappelli, Keene and McGrath (1985) found that children with epilepsy showed significant problems in social adjustment, as measured by the Child Behavior Checklist. Herman (1982) found that there was an association between level of behavioral and social functioning and level of neuropsychological functioning in children with epilepsy. Ferrari, et al. (1983) found that children with epilepsy have significantly lower self-concepts than either healthy children, or children with a non-neurological chronic illness such as diabetes.

Given the above findings, it seems probable that psychological factors are relevant to TS, even if an organic etiology for the disorder is established. Recently researchers have begun to evaluate which psychological factors are most relevant to TS, to assess their interaction with organic factors, and to explore what effect this interaction might have on some aspects of TS symptoms. Current research findings suggest the presence of some neuropsychological, individual, social and familial factors that affect how a person with TS copes with TS and how s/he functions on a day to day basis. These factors may also influence, to some degree, the manifestation of this disorder in terms of type, frequency and intensity of symptoms.

C. Neuropsychological Findings

Newman, Borth and Zillman (1986) conclude that no specific or definitive patterns of neuropsychological deficits have emerged from test data on children and adolescents with TS. This is an accurate assessment of the test data in that there is no one pattern of deficits that is common to all or even an appreciable majority of people with TS. Test data does, however, indicate that a large percentage of people with TS do show some neuropsychological deficits and some statements about general tendencies can be made. Consistent findings associated with neurological deficits include a male:female ratio of at least 3:1 (DSM-III-R, 1987), a significant number of left-handed TS patients (Shapiro & Shapiro, 1982; Sutherland, Kolb, Schoel, Wishaw & Davies, 1982), and a high frequency of verbal-performance discrepancies, particularly as measured by the Wechsler Intelligence Test for Children (WISC) and the WISC-R (Bornstein, King, &

Carroll, 1982; Incagnoli & Kane, 1981, 1982; Joschko & Rourke, 1982; Shapiro et al., 1978). As noted above, and of relevance to the current study, a significant proportion of children and adolescents with TS have been found to have LD and ADHD.

Learning Disabilities

Shapiro et al.(1982) note that the levels of overall intellectual functioning of people with TS span a broad range. The accumulation of data indicates, however, that children and adolescents with TS tend to show deficits in visual processing and especially in visuographic skills (Bornstein, et al. 1982; Ferrari, Matthews & Barabas, 1984; Golden, 1984; Incaguoli & Kane, 1982; Shady, Fulton & Champion, 1988; Shapiro & Shapiro, 1982). Shapiro and Shapiro (1982), in a review of available data, found that a large percentage of people with TS had significant Verbal-Performance IQ differences, with the Performance IQ's generally lower than Verbal IQ. The researchers cited above have found a consistent pattern of relatively depressed Coding subtest scores on the WISC and WISC-R. They have also found that the Arithmetic subtest score on the WRAT and the Wide Range Achievement Test-Revised (WRAT-R) tends to be lower than the Reading and Spelling subtest scores on the WRAT and WRAT-R, as well as lower than the Arithmetic subtest on the WISC-R. Below age level scores on the Bender-Gestalt Test have also been documented by the above researchers. Ferrari et al. (1984) interpret these findings to suggest that children with TS demonstrate impaired visual-perceptual skills.

These findings suggest that a high proportion, though not all, of children with TS show deficits in areas that could interfere with academic

functioning. A number of researchers have found that children with learning problems tend to have lower self esteem and lower peer status in the classroom. Bryan (1974) found that children with learning problems tend to have low social status in the classroom while Seraficz and Harway (1979) and Hutten and Polo (1976) also found that LD children tend to have low peer status. Primavera, Simon and Primavera (1974), Coopersmith (1967), and Leviton and Kiraly (1975) all have found a correlation between self-concept and academic achievement. Empirical data supports the notion that LD children have lower self concepts than do non-LD children (Blood, 1984; and Champion et al., 1984). Hagin et al. (1982) found that TS children tend to feel isolated in the classroom because they feel "different". The impact of the nature of TS symptoms, coupled with a learning deficit in a high proportion of TS children might set such children apart from their peers in a negative fashion. When this is added to the proportion of TS children who also show attentional deficits, the implications for acquisition of both academic and social skills become quite serious.

Attention Deficit Disorder

Cohen et al. (1982) found that ADD, characterized by "difficulty in focusing attention, impulsivity and poor regulation of motor activity" (p. 32) was the first sign of a developmental disorder in 50% of the TS children whom they have studied. Bruun (1984) also found a high percentage of TS children with ADD, and suggested that an attentional deficit impaired a child's ability to learn, socialize and work effectively. Both Shapiro and Shapiro (1978) and Incagnoli and Kane (1981, 1982, 1983) found that a

high percentage of children with TS did poorly on the Coding subtest of the WISC-R. Among other factors, this subtest provides a measure of attention and concentration (Sattler, 1974). Comings and Comings (1984) found that 62% of TS patients in a large sample were also diagnosed as ADD. They found that the TS patients in general had a low frustration tolerance and a short attention span. Shady et al, (1988) found that TS patients frequently complained about problems with attention and concentration. According to the DSM III-R (1987), ADHD is frequently associated with TS.

In summary, a review of the literature suggests that there is a close association amon ADD, or ADHD, and TS. Patients with TS are consistently described as impulsive, even in the absence of a concurrent diagnosis of ADD. While the cause of the attentional deficit in these cases has not been established, it is possible that it is linked to the subtle neurological dysfunction that frequently accompanies TS (Wilson et al., 1982).

The data consistently suggest that, as a group, children with TS show some neuropsychological impairment. Specific deficits which would interfere with academic progress have been found and a close association between TS and ADHD has been established. It has been noted that children with learning problems tend to have both low peer and social status in the classroom as well as low self-esteem. The impulsivity that characterizes ADD is likely to lead to problems with both academic and social learning. These findings suggest that children with TS would be at risk for developing some sort of behavioral problems.

D. Behavioral Problems

Shapiro et al. (1978) and Shapiro and Shapiro (1982) contend that people with TS as a group do not show signs of psychological distress or dysfunction. More findings have suggested that children and adolescents with TS do show evidence of behavioral problems and that adults with TS also show signs of psychological dysfunction (Ferrari et al, 1984; Grossman et al., 1986; Matthews et al., 1985; Newman, Barth & Zillner, 1986; Wilson et al., 1982). In addition, both Stefl (1984) and Champion et al. (1988) found that people with TS self-reported both behavior problems, and a perception of being in psychological distress. Harcherick, Leckman, Detlor, and Cohen (1984), who developed the Tourette Syndrome Global Scale (TSGS), note that:

> Although the relation of...behavioral symptoms (including attentional problems, impulsiveness, poor frustration tolerance, motor restlessness and compulsive behaviors) to TS is controversial, we consider them to be an integral part of the clinical presentation which should be rated along with the more classical features of the disorder (p. 153).

Leckman, Detlor and Cohen (1983) note that onset of impulsivity and diminished ability to concentrate frequently precede onset of motor and phonic tics. They have found that behavioral symptoms are frequently seen in TS and that such symptoms may be subject to the same waxing and waning found in the phonic and motor tics. These investigators note that behavioral symptoms may also persist when the more classic symptoms are in periods of waning, or even remission. Frequently seen behavioral symptoms include increased motor restlessness, impulsivity, diminished frustration tolerance, argumentativeness, diminished ability to concentrate, and subsequent poor academic performance, and disabling compulsive actions. Leckman et al. (1983) have found that obsessive-compulsive behaviors

generally appear relatively late in the developmental course of the syndrome (mean age 10.8 years), and can become extreme and disabling. Elaborate rituals may disrupt effective functioning and obsessive doubting may render even simple decision making difficult. Comings and Comings (1984) describe TS patients as being easily angered, as having low frustration tolerance and short attention span, as being difficult to discipline, as having exhibitionistic tendencies, as being confrontative, and as having low self-esteem as well as learning disorders.

Cohen et al. (1979), Cohen, Detlor, Shaywitz and Leckman (1982) and Wilson et al. (1982) discuss the failure of inhibition that is characteristic of TS and relate it to behavioral problems. Cohen et al. (1979) suggest that during periods of developmental transition, such as adolescence, psychological structures of self-control may weaken. At this time, behavioral symptoms, including distractability, extremes of mood, heightened anger and anxious preoccupations may emerge. Children with TS, who may have pre-existing problems in regulating attention and impulsivity, may have more than the normal difficulty in inhibiting motoric discharges or conscious awareness of thoughts and feelings. Thus the excitability, coprolalia, corpopraxia and other dramatic symptoms of TS might be understood in terms of a failure of inhibition that is more muted and better modulated in children who do not have TS, or similar disorders. Wilson et al. (1982) suggest that subtle brain dysfunction may be at work in the behavioral problems exhibited by Tourette sufferers. They note that other children with subtle brain dysfunction appear to be at increased risk for the development of behavioral problems. Further, according to Wilson et al. (1982), patients with extrapyramidal movement disorders are more

likely to exhibit psychopathology than are neurologic patients in general, and they relate this to a loss in striatal inhibition.

Wilson et al. (1982) tested their hypothesis by comparing TS children with children in special classes and with unselected public school children. These investigators found that children and adolescents with TS showed significantly elevated behavioral problems, as measured by the Behavioral Problem Checklist (BPC). Further, the TS children were more similar to children in special education than to the unselected public school children. They found that the severity of the behavioral problems was related to the severity of TS, as measured by a Tourette Symptom Checklist that they constructed, based on a literature review of common TS symptoms. In their sample (N=16), Wilson et al found that Verbal IQ predicted severity of behavioral problem, but not severity of TS symptoms. They interpreted this finding as supporting their hypothesis that the movement disorder and the putative associated subtle brain dysfunction increase the risk of behavioral problems, with verbal skills acting as a potential mediating factor. They caution that, at this time, this is a speculative model.

Stefl (1984) and Champion et al. (1988) used surveys to assess self-reports of behavioral problems. In both surveys, over 50% of the respondents reported numerous behavior problems. including obsessive compulsive behaviors, hyperactivity, extreme anxiety, temper tantrums, and extreme mood swings. Data from both surveys also revealed that well over a third of the respondents believe that they have some problems in coping. Similarly, over one-third in both surveys reported their overall well-being to be only fair or poor. Stefl (1984) compared the TS respondents to the

general population on this question, and found that TS respondents rated their own well-being significantly lower than the general population rated their own well-being. Champion et al. (1988) also found that 41% of people with TS reported problems in making and keeping friends as a result of having TS symptoms. Almost one half of the older respondents reported problems with dating and developing opposite sex relationships, again as a result of having TS. Thus, a large percentage of people with TS report problems in social interactions and relate these problems to having TS. Both Stefl (1984) and Hubka, Fulton, Shady and Champion (1988) found that more than half of their survey samples had sought some sort of counseling for problems arising from TS symptoms. From these data, it seems clear that a large proportion of people with TS perceive themselves, and/or are perceived by their families as being in psychological distress.

Ferrari et al. (1984) used the Child Behavior Checklist (CBCL) (Achenbach & Edelbrock, 1983) to assess level of behavioral functioning in children with TS. They found that parents rated their children as having low social competence (mean score at approximately 30th percentile). These children were also found to have a higher level of internalizing than externalizing problems. Parents as a group rated their children at the 93rd percentile for depression, at the 92nd percentile for obsessive compulsive behavior, and at the 94th percentile for uncommunicative behavior. These data suggest that the psychological or behavioral problems found in this study may be, in part, a reaction to having TS. If the behavioral problems were primarily related to the neurological correlates of TS such as impulsivity, hyperactivity and attention-deficit, it would be expected that the rate of externalizing problems would exceed the rate of

internalizing problems. Therefore, these data suggest that the psychological or behavioral problems indicated by this study are not solely a function of the disorder itself.

Matthews et al. (1985) administered the Personality Inventory for Children-Revised (PIC-R) (Wirt, Lachar, Klinedinst & Seat, 1984) to 20 mothers of children with TS and 20 mothers of nonclinical control children. They found that the mothers of children with TS rated their children as having poorer communication with other family members than did the controls. It is not likely that the problems in communication found in this study and in that of Wilson et al. (1982) are due to language deficits, as there is no evidence of consistent patterns of such deficits among people with TS.

Grossman et al. (1986) administered the Minnesota Multiphasic Personality Inventory (MMPI) to 29 adults with TS and 29 adult normal controls, matched for age and sex. They found that Tourette subjects scored significantly higher on the Schizophrenia, Depression, Psychopathic Deviate, Psychasthenia and Hypochondriasis scales. Again, the results suggest an elevation of psychological distress among people with TS.

In conclusion, data from standardized psychometric measures suggest the presence of an interaction between the physiology and psychology of people with TS. The result is a combination of both neurologically-oriented behavioral problems that may be associated with hyperarousal (such as impulsiveness, excitability, extreme mood swings, quickness to anger and obsessive-compulsive behaviors) and emotional distress in reaction to those problems, and possibly to the nature of the other TS symptoms. Although not all people with TS demonstrate or report

behavioral problems and/or psychological distress, there appears to be strong group tendency towards such problems and distress. Some individuals with TS may indeed, as suggested by Shapiro and Shapiro (1982) function well, and even exceptionally well. Current data does not, however, support the contention that psychological factors play little or no part in TS.

E. Family Functioning

To date, there has been only one study in which the level of functioning of families with a TS member was explored (Matthews et al., 1985). Given the nature of the disorder, however, and the related individual behavioral problems and psychological distress discussed above, it is likely that having a TS member would have an effect on the families. The finding of a higher than expected frequency of OCD (Pauls et al., 1986) and ADD (Comings & Comings, 1984) among TS family members also would suggest that families with TS members might have a higher than average array of problems with which to cope.

Clinical findings.

Bruun (1984) suggests, based on her considerable clinical experience with TS, that a number of issues need to be explored in the family context. She has found that family support and acceptance are crucial to the well-being of the TS child and are especially important to the development of self-esteem. She suggests that parents may react to the diagnosis of TS with guilt and shame, and may tend to deny the diagnosis and/or the symptoms. She has found that families may need help in sorting out reasonable from unreasonable expectations and indicated that it is important that parents neither overprotect nor set unreasonably high expectations for their child.

Cohen et al. (1982) found that no parent reacts neutrally to the diagnosis of TS. The reaction of both parents and child to onset and diagnosis of TS reflects an interaction among individual personalities, abilities to cope with uncertainty and stress, and availability of social and medical support. Some parents may overprotect their child with TS in order to create an accepting and protective structure around the child. Other parents may react with anxiety, shame, and/or anger. Cohen et al. emphasized that the nature of the parent's response will affect how the child sees himself/herself, as well as his/her ability to develop effective coping skills. These investigators suggest that TS children and adolescents cope best when they are expected to function socially with normal children, when the disorder is seen as medical and not fully under the child's or family's control, and when parents are able to maintain both a deep commitment to the child, as well as an appropriate distance, such that they are able to perceive the tics and other symptoms as belonging to the child, and not to themselves. Cohen et al. (1982) further posit that the degree to which parents perceive the child's TS symptoms as insults to their own egos will determine how well they are able to help the child feel worthwhile despite TS symptoms. Parker (1985) notes that family adjustment to TS do not come easily. Parents typically react with varying amounts of shame, anxiety, anger, and guilt to any long-term illness. She suggests that open communication, as well as open acceptance, are the keys to helping children cope with TS. She recommends that parents find individual ways of helping their child with TS cope with his/her specific symptoms. Cohen, Ort, Leckman, Riddle and Hardin (1988) note that "all authentic therapy for a patient with TS is, in some sense, therapy of the patient's family" (p. 190).

The above findings, which are primarily based on clinical experience, illustrate the burden that families face when a family member has TS. At a minimum, families must be able to recognize that a problem (the TS symptoms) exists, to obtain an accurate diagnosis, to begin pharmacological treatment if prescribed, and to maintain the medical contact involved in monitoring medication. In addition, parents with TS children who are also diagnosed with LD or ADHD must obtain appropriate assessment and diagnosis, ascertain that appropriate measures are taken in the school setting and remain in regular contact with the school to monitor their child's progress. Families who choose to do so may also undertake the education of school personnel about TS. These above tasks, time and energy-consuming as they may be, are only the instrumental ones. The family must also cope with the emotional aspects of all of these tasks. Accepting and adapting to a life-long, debilitating disorder such as TS is not likely to be an easy task. Prior to diagnosis, families are faced with a child who presents with an array of unusual motoric and phonic behaviors, which change over time and which sometimes appear to be under the child's control. Research data have also suggested that the child with TS may be impulsive, inattentive, difficult to discipline, have trouble sleeping at night, have trouble in school and with getting along with peers and family members.

Thus it is likely that, at best, families with a TS member will have a number of stressors with which to cope. Clinical findings suggest that the manner in which these families do cope will depend on the family, and on the particular level of communication among family members (Cohen et al., 1982; Brunn, 1984). Again, an interactional model is suggested. Not only

does the nature of TS place demands and constraints upon the family, but the way the family is able to cope with these demands will affect the well-being of the person with TS. Bruun (1984) has found that both psychological and physiological stressors will exacerbate TS symptoms, just as Temkin and Davis (1984) have found that negative stress increases the frequency of epileptic seizures. It is possible that poor familial coping could lead to greater stress for the individual with TS. A feedback cycle (Hoffman, 1981) might be activated, with the individual's or the family's stress leading to an exacerbation of the TS symptoms, leading to more demands on the family whose resources are inadequate to cope with more demands; thus the level of coping deteriorates, which leads back to more individual stress and so on.

Empirical data.

The impact of TS on a family's structure and level of functioning, and the impact of the family on the individual with TS's level of social and intrapersonal functioning has received little empirical attention. The literature on family therapy and studies of families of children with epilepsy, however, provide direction in this area.

Liebman, Minuchin, Baker, and Rosman (1976) define psychosomatogenic families as those in which "the development of severe psychosomatic symptoms in a child is related to the presence of certain patterns of family organization and functioning" (p. 313). The characteristics of such families include enmeshment, overprotectiveness, rigidity, lack of conflict resolution, and involvement of the child in parental conflict. It is important to note that the existence of disorders such as asthma or diabetes is not caused by familial dysfunction. The disorders themselves

are understood to be organic in origin. It is the severity of the disorder and the exacerbation of the symptomatology that is related to these familial characteristics. Thus, in milder form, it is possible that family characteristics such as enmeshment, overprotection, and rigidity, could serve to exacerbate TS symptomatology by creating more stressors for the child with TS. Similarly, the nature of TS symptoms could pull for parental over-involvement (enmeshment), overprotectiveness (to create a haven for the child with TS), and rigidity (to compensate for the unpredictability of TS symptoms). Such a model is consistent with the clinical findings discussed above.

Two questions may be posed here. First, do TS families present with some sort of distress and/or dysfunction? Second, what is the nature of the distress/dysfunction? Hubka et al. (1988) found that TS families do indeed perceive themselves to be in distress. Over 60% of TS families in their Canadian survey (N=230) reported that having TS in the family interfered with daily activities, and about one half reported that they had sought counselling as a result of having a family member with TS.

The question about the nature of the distress/dysfunction may be more difficult to answer in the absence of empirical data. Three studies, however, shed some light on this issue.

Ferrari, Matthews and Barabas (1980) compared families of children with epilepsy to the families of children with diabetes and to families of children with no known chronic illness. The three groups (N=15 each) were matched as closely as possible on the variables of the target child's current age, age at onset of illness (for the epileptic and diabetic groups), sex, socioeconomic status and academic standing. Subjects with

confounding variables of mental retardation, secondary medical problems, or gross physical impairments were excluded from the study. Significant differences were found between the families with epileptic children and the other two groups. First, families with epileptic children reported themselves to be significantly less close (i.e. low family cohesion) than families with either diabetic or nonchronically ill children. Second, communication between parents and children in the epileptic families was reported to focus on specific issues or potential problems whereas communication in the other two groups of families was reported to be more general in nature. Third, parents of children with epilepsy reported that these children more frequently complained about feeling rejected, were more likely to act babyish or immature and had more frequent periods of emotional distress, than was reported by the parents in the other two groups. Thus it can be seen that having a child with epilepsy affects both the family and the child in unique ways. The families are perceived by the parents as being less cohesive and as having poorer communication, and the child is perceived as experiencing more behavioral problems and emotional distress.

Hartlage and Green (1972) found that when parents of children with with epilepsy encouraged passivity and delay to self-initiated behavior, the child was quite likely to underachieve at school. Hoare (1984c) found that children with chronic epilespy were much more likely to be described by their parents as dependent than were children in the general population or children with chronic diabetes. Hoare (1984b) also found that siblings of children with chronic epilespy were more likely to show evidence of psychiatric disturbance than were the siblings of children with chronic

diabetes, children with newly diagnosed epilepsy or diabetes, or children in the general population. Thus a picture of vulnerability to familial as well as individual dysfunction has begun to emerge in the literature on children with epilepsy and their families.

These findings are relevant to families with a TS member to the extent that, with both of these neurologically-based disorders, the symptoms are unpredictable, appear to be in some measure out of the patient's control, may be imperfectly controlled by medication, and may assert themselves in public settings. Attentional deficits and academic disorders are also associated with both groups of children (Harcherik, Carbonari, Shaywitz, Shaywitz, & Cohen, 1982).

Matthews et al. (1985) compared 20 TS children and their mothers to 20 nonclinical children and their mothers. They found that family communication between TS child and other family members was perceived by their mothers to be poor, whereas the control mothers perceived no problems in communication between child and family. The TS children perceived their families to be less flexible and adaptable than did the control children. This is consistent with the work of Farber (1960) who suggests that chronic illness can interfere with the developmental process in the family life cycle. Matthews et al. (1985) suggest that the needs of the TS child drain the emotional resources needed to meet the changing developmental needs of the family. They suggest that, since TS creates control-oriented conflicts, both for parents and within the child, it is likely that more rigid interactional patterns, though less effective and adaptive, serve to provide some measure of predictability and regularity in family functioning and interaction. Thus far, the data suggest both dysfunctions in both

communication and level of structural rigidity in TS families. Available empirical data do not support Liebman et al.'s (1976) model for a psychosomatogenic family for this population, although the paucity of empirical findings does not as yet allow for rejection of that model. Together the clinical and empirical findings support the suggestion by Cohen et al. (1982) and Cohen et. al. (1988) that the family needs to be considered as part of the complex picture of TS.

F. An Integrative Approach

Cohen et al. (1982) suggest that the interaction between biological and psychological factors in TS presents a model for understanding how the interplay among genetic, neurophysiological, behavioral and environmental factors affect the development and course of neuropsychiatric disorders in childhood. The data presented in the preceding sections suggest that all of these factors do indeed affect TS. A thorough comprehension of all these factors and how they interact among themselves is necessary in order to understand and appropriately treat this disorder. Any model that emphasizes only biological factors or only psychological factors runs the risk of neglecting important information needed to understand the etiology, the nature of the symptomatology, and the factors that influence successful treatment. Cohen et al. (1982) point out that TS is probably a biologically heterogeneous disorder, "reflecting genetic and other biological determinants, with different subgroups manifesting various neurochemical abnormalities and responding differently to medication" (p. 39). This hypothesis is supported by the literature. It was noted above that individuals with TS respond differentially to the various available

medications, and a group of neurotransmitters, rather than one single neurotransmitter, has been implicated by research findings. Family genetic studies have suggested that there may be genetically determined TS subgroups including some 10% who show no previous family history of TS or related disorders of OCD, CT, or ADHD.

Cohen et al. (1982) note that an individual's development reflects interactions between biological and experiential factors and dysfunction. In TS the biological factors include degree of severity of the disorder and the presence or absence of concurrent learning deficits, attentional deficits and such neurologically-related features as impulsivity, excitability and disinhibition. Experiential factors in TS might include family style and level of functioning, socio-economic factors, academic and peer experiences, internal reactions to the disorder (for example, depression, anxiety, anger), as well as level of individual coping skills.

A review of the TS literature to this point indicates that there is an emerging trend towards a more integrative approach in understanding this complex disorder. An integrative model for the study of TS prompts a number of questions, and thus encourages the exploration of how the various factors interact with one another in this disorder.

Several specific questions were of particular interest for the current study. First, is the severity of TS symptomatology related to the level of intrapersonal and social functioning of the person with TS? Second, is the severity of TS symptomatology related to the level of functioning in the TS family? Third, is the level of functioning of the child with TS related to familial functioning? Fourth, are there specific structural styles common to TS families? Fifth, are there specific patterns of intrapersonal and

social functioning common to children with TS? The literature does suggest some answers to the above questions. Matthews et al. (1985) found that TS families are perceived by the TS child to be less adaptable and flexible than "normal" families. Mothers of TS patients in this study also perceived intrafamilial communication to be poor. Wilson et al. (1982) found that severity of TS symptoms does predict level of behavioral disturbance, with level of intelligence acting as a mediating factor. The purpose of the current study was to extend the work of Wilson et al. (1982) and Matthews et al. (1985) by investigating the relationship among severity of TS symptomatology, level of individual intrapersonal and social functioning, and level of familial functioning.

V. RATIONALE FOR THE CURRENT STUDY

A number of researchers (Bruun, 1984; Cohen et al., 1982; Grossman et al., 1986; Matthews et al., 1985; Wilson et al., 1982) have emphasized the importance of assessing and treating the TS patient within the context of taking a holistic or systemic approach to the understanding of this disorder. This is consistent with the emergence of a contextual approach to the study of disorders such as epilepsy and diabetes. The effect of environmental factors such as stress and familial interaction on the intensity and/or frequency of symptoms in epilepsy has been documented (Hermann & Whitman, 1983). The negative effect of epilepsy on family functioning has been documented by Hoare (1984a, 1984b, 1984c). The mediating characteristics of social competence in metabolic control of diabetes has been explored by Hansen, Henggeler and Burghen (1987) as has the relationship between level of neuropsychological functioning and severity of behavior problems in children with epilepsy (Hermann, 1982). These studies are relevant to the study of TS in that they provide both a model for an holistic approach to exploring an organically-based syndrome, and clues as to which psychosocial factors may be important to provide the fullest understanding and treatment of this disorder. So far, the emphasis has been on the biological, medical, genetic and/or neuropsychological aspects of TS. As discussed above, some attention has been paid to the academic context; one study has assessed the family context; a few studies have addressed the psychological or behavioral aspects of TS. To date,

however, only one study (Wilson et al., 1982) has assessed the relationship of the severity of TS symptoms to the level of behavioral problems in children with TS. One weakness of the Wilson et al. (1982) study was that the measure of severity was based on a questionnaire on which no reliability or validity data were reported.

In the current study, the Tourette Syndrome Global Scale (Harcherik et al., 1984) was used. This is a standardized scale, for which validity and reliability has been established (Harcherik et al., 1984). In the family study (Matthews et al., 1985), no measure of severity was obtained; therefore no information about the relationship between severity of TS symptoms and family functioning is currently available. Similarly, Wilson et al. (1982) reported no psychometric information about level of family functioning. None of the above TS studies used direct psychometric information about the child's self-image, tolerance for stress or interpersonal orientation.

This current study represents an attempt to provide a more cohesive picture of the interaction among severity of TS symptoms, interpersonal functioning of the child with TS and level of functioning in the TS family. Four aspects of the TS child's world were assessed. First, the severity of TS symptoms was assessed. Second, the level of the TS child's intrapersonal functioning was evaluated in terms of degree of impulsivity, tolerance for stress, self-image and intrapersonal orientation. Third, parental perceptions of the child with TS were measured, by having parents fill out a standardized questionnaire. Fourth, level of family functioning was assessed. Parents and child were asked to fill out four family measures, which provided information about perceptions of family

adaptability and cohesion, coping skills, strengths and level of satisfaction with family functioning.

A circular model of relatedness of these factors was hypothesized. Nichols (1984) defines circularity as "the idea that events are related through a series of interacting loops or repeating cycles" (p. 583). Hoffman (1981) distinguishes between a linear concept of causality and circularity. In a linear model, it is assumed that A causes B causes C. In a circular model, it is hypothesized that A, B, and C are interactive, such that more of A might engender more of B and/or C which in turn might engender more of A and so forth. Simplistically, in a linear model it might be assumed that severe TS symptoms cause severe intrapersonal dysfunction which causes severe familial dysfunction. In a circular model, it is assumed that all of these factors interact concurrently such that a dysfunctional family, a dysfunctional child and severe TS symptoms are seen together. Similarly, a more functional family, a more functional child and milder TS symptoms might be expected to coincide. Clearly, one or another of these factors may have more power in the interaction such that it would be possible to find severe TS symptoms in a well functioning family with a well functioning Tourette individual.

VI. HYPOTHESES

Because the available data on the role of intrapersonal factors, level of social functioning and level of family functioning in TS is limited, the current study was primarily exploratory in nature. One major hypothesis was tested and several research questions were evaluated.

The hypothesis tested was that level of individual and familial functioning would predict severity of TS symptomatology. Specifically, it was predicted that a combination of the level of self-esteem, the level of tolerance for stress, the level of behavioral problems reported by parents and the level of familial coping skills (as reported by the parents) would predict the severity of the child/adolescent's TS symptomatology, as rated by clinicians in the Tourette Clinic. Thus, it was expected that higher levels of individual and family functioning would be associated with milder TS symptomatology, and that lower levels of individual and family functioning would be associated with more severe TS symptomatology.

The research questions were as follows:

1. Would severity of TS symptomatology be associated with a) level of impulsivity and, b) interpersonal orientation in terms of expectations of negative or hostile interpersonal interactions?

2. Would severity of TS symptomatology be associated with level of parents' perceptions of the child/adolescent's level of social competency?

3. Would severity of TS symptomatology be associated with a) level of
    family functioning; b) parental degree of satisfaction with family
    functioning?

The purpose of the above research questions, as well as the primary
hypothesis, was explore the relationship, if any, between individual and
family functioning and severity of TS symptomatology. Post hoc analyses
were conducted to further clarify the interaction among these factors.

VII. METHOD

A. Subjects

Subjects were 22 children and adolescents with Tourette Syndrome, recruited through the Tourette Syndrome Clinic (TS Clinic) at St. Boniface General Hospital, Department of Psychiatry, Winnipeg, Manitoba. Participation was voluntary, and the children and their parents were informed regarding the nature of the research. Recruitment and dissemination of information was through direct contact with the researcher. No deception of the subjects was employed. Participants were free to drop out of the study at any point.

Subjects met the following inclusion criteria:

1. Ages between 7 and 16.

2. Diagnosis of TS by staff psychiatrist or other experienced medical personnel, using DSM III-R criteria.

3. Absence of other primary mental or physical handicaps.

4. All subjects and their parents signed an informed consent agreement to participate in this research (Appendix A).

Nineteen males and three females participated in the study. Thirteen intact, six single parent (five mother only, and one father only), and three remarried (all with the natural mother in the home) families participated. In the case of remarried families, the natural mother and step-father were asked to complete the relevant measures.

Subjects ranged in age from 8-16 years. The mean age was 12 years; modal age was 14 years, and the median age was 12 years. Twenty-seven subjects and their parents agreed to participate in the project. Two families dropped out of the study, and three families did not complete the family questionnaires. Twelve subjects were from Winnipeg, Manitoba, seven were from small towns in southern Manitoba, and three were from rural areas in southern Manitoba.

Ninety-five percent of the subjects reported current motor tics; 72.7% reported current phonic tics. The majority of subjects' parents reported the presence of some behavior and sleep problems. About one-third of the subjects reported allergies, and about one-third reported a current diagnosis of Hyperactivity of the time of the study. No subjects were currently on medication for Hyperactivity. About one-third of the subjects reported that they had repeated at least one grade and almost 60% were currently in some sort of special education classes (including resource classes). A variety of in-school problems were reported. A majority of the subjects reported involvement in organized sports as well as reported age-appropriate peer relationships.

B. Instruments and Measures

The instruments used in the current study were chosen to measure severity of TS symptoms, level of intrapsychic functioning and level of family functioning. These instruments were chosen on the basis of prior clinical and empirical use in measuring these variables. A history and information questionnaire was also completed by the parents of the TS children. Data were coded to ensure confidentiality, and research data on

individuals was not available to clinicians working with subjects who were also patients, unless the child and his/her parents specifically requested that it be made available. Such information was made available to clinicians only after all data were gathered on all subjects. Individual and mail-out feedback was made available to individual children and their parents after all data had been gathered on all subjects, except when data had been gathered as part of the initial clinical assessment of the TS patient. In that case, feedback was given by a clinician other than the primary researcher at the time of assessment.

History and Information Questionnaire.

An history and information questionnaire (Appendix B) was completed by one of the parents of the TS subject. This questionnaire included optional questions about racial origin and religious affiliation. Other questions included: birth date; academic history of the TS subject; concurrent diagnosis of ADD or ADHD; past and current medications for TS, ADD and ADHD and/or allergies taken by the TS subjects. Although this information was not used in testing the specific hypotheses of this study, it provided additional data which were used to amplify or clarify the findings.

Tourette Syndrome Global Scale

The Tourette Syndrome Global Scale (TSGS) (Appendix C) (Harcherik, Leckman, Detlor & Cohen, 1984) is a multidimensional scale "specifically designed to permit reliable and valid cross-subject comparisons on several dimensions of TS symptomatology including behavioral symptoms, motor restlessness and school or occupational performance, as well as simple and

complex motor and phonic tics" (p. 153). It was included in this study to provide a measure of severity of TS symptomatology and level of social functioning. Harcherik et al. (1984) describe the scale as being

> comprised of 8 individually rated dimensions summed into an overall global score. The scale ranges from 0, which represents no symptoms, to 100, representing the worst possible TS symptoms, consisting of constant and debilitating motor and phonic symptoms, unacceptable social behavior, nonstop motor restlessness and an inability to function in school or work situations (p.154).

Scores for the TSGS range from 0 to 100, distributed normally with a mean of 40.2 and a standard deviation of 15.2. Harcherik et al. (1984) divide severity of TS symptomatology into four subgroups based on the overall score: Mild (0-24); Moderate (25-39); Severe (40-59); and Extreme (60-100).

Scores from the two major domains contribute equally to the total TSGS score. The first domain, TS symptomatology, consists of motor and phonic tics. Tics are rated according to their frequency and complexity and to degree of disruptiveness. Inter-rater reliability for this domain is 0.65-0.85 (p < .001) (Harcherik et al., 1984). The second major domain, social functioning, is comprised of three areas: behavioral problems, motor restlessness and level of school or occupational functioning. Each of these areas is scored on a scale of 0 to 25. The sum of these three scores, multiplied by 2/3, to give the overall social functioning score. Inter-rater reliabilities for the behavioral problems, school performance, and motor restlessness are: 0.87 (p < .01); 0.93 (p < .01); and 0.32 (p = N.S.), respectively (Harcherik et al., 1984). Inter-rater reliability for the TSGS total score was 0.89 (p < .01) (Harcherik et al., 1984).

Validity for the TSGS was measured in two ways (Harcherik et al., 1984). First, six TS patients were rank ordered in terms of overall severity of TS symptomatology by four different raters. Agreement with the consensus global score of the TSGS was 0.46 to 0.99 (p < .05), suggesting that the TSGS accurately differentiates degree of TS symptomatology. Second, scores on the TSGS were compared to scores on the Children's Global Assessment Scale (C-GAS). A high level of agreement between these scores was obtained (0.76 - 0.89; p < .01).

The TSGS was completed by both parents and by the clinicians in the Tourette Clinic so that inter-rater reliability could be established for this study. The clinicians' TSGS rating was used for the main analysis, and to divide the subjects into "Low" and "High" TS subgroups. The parents' ratings were compared to each other, and to the clinicians' ratings.

Rorschach Inkblot Test--Exner Comprehensive System

The Rorschach Inkblot Test (Rorschach, 1921) consists of 10 cards on which inkblots are printed. The test was administered and scored using the Exner Comprehensive System (Exner, 1974, 1986). This system is based on a standardized procedure for the administration and coding of the test. Each subject was shown the inkblots one at a time and was asked to tell the examiner what each inkblot looked like to him/her. After going through the cards once in this fashion, the examiner showed the cards again, asking the subject to tell the examiner what it was about the blot that made it look like that. The test took about forty-five minutes to an hour to administer.

The Rorschach Test was chosen because it provides a wealth of information about the psychological characteristics of the subject (Exner, 1986). Although the use of the Rorschach in empirical studies historically has been controversial, Exner (1974, 1986) has devised a standardized administration and coding system which has minimized the "personal approach" to this test. Normative data are available for children and adolescents aged 5-16 for most of the coding categories and for the ratios and derivations that are obtained from the coding categories. Coding categories include: Location, Developmental Quality, Determinants, Form Quality, Pair Responses, Content, Popular Response, Organizational Level and Special Scores. Codes from the above categories are summarized into ratios and derivations. The ratios or derivations which were of primary interest in this study were the Lambda and the D Score. One Special Score, Aggressive Movement, was also evaluated.

Lambda is a ratio that reflects the use of Pure Form (F) responses in responding to the inkblots (Exner, 1986). Rorschach (1921) suggested that the use of F was related to the attention-concentration features of the individual's thinking. According to Exner (1986) a low proportion of pure F responses is related to an inability "to promote the necessary delays required for the formulation of the pure F answer (as in the organic or characterological style prone toward impulse display)" (p. 313). Inter-rater reliability for scoring pure F responses is high (90-91% agreement). The test-retest stability for Lambda in children is also respectable (0.82 for 8 year olds; 0.84 for 9 year olds).

The D Score, according to Exner (1986) provides a measure of the individual's ability to tolerate stress. It is derived from movement,

color and shading determinants. These determinants have interscorer reliabilities ranging from 0.87 to 0.99 (Exner, 1986). Test-retest reliability of the D score is 0.93 for 8 year olds and 0.91 for 9 year olds (Exner, 1986). An average D score indicates that the individual's available resources are generally sufficient to meet the emotional and environmental demands being made on him/her. A low D score suggests that the individual's available resources are not sufficient to meet these. These individuals are consequently at risk for becoming overloaded by perceived stimulus demands (Exner, 1986).

The Aggressive Movement Response is a Special Score that reflects an increased likelihood of aggressive behaviors (either verbal or nonverbal) as well as relatively negative and/or hostile attitudes towards others. Inter-rater reliability for this score is 0.96 (Exner, 1986).

Other data from the Rorschach were analyzed to amplify or clarify these findings. The number of Pure Human (H) responses, for example, provides information about the individual's reality-based, or experientially-based orientation towards others (Exner, 1986). The Affective Ratio (Afr) provides information about the individual's responsiveness to emotional stimulation. A high Afr would suggest that the subject may be easily excitable, whereas an average Afr suggests a more modulated or subdued response to emotional stimulation (Exner, 1986). The Rorschach was administered to all TS subjects by the researhcer, who had been trained in the Exner Comprehensive System. All responses were coded by both the researcher and another clinician, also trained in the Exner System, to minimize coding errors and so that inter-rater agreement scores could be established. When coding the Rorschach responses, the coders were

blind to the TSGS severity rating of the subject, to all other test data, and to the coding of those Rorschach responses by the other researcher/clinician. A third clinician, who was also trained in the Exner Comprehensive System, was available to arbitrate coding disagreements.

Piers-Harris Children's Self-Concept Scale

The Piers-Harris Children's Self-Concept Scale (Piers, 1984) (see Appendix E) is an 80 item scale designed to measure self-concept in children and adolescents. The items are read aloud to younger children (up to age 12), while older children (12-18) may complete the scale independently. Each item was answered "yes" or "no". Raw scores may be converted to percentiles, stanines and/or T-scores. The scale was normed on children aged 8-18.

The test took about 10-20 minutes to administer. Higher scores are associated with higher self-concept (Piers, 1984). Scores are summarized into a Total Scale Score. Six Cluster Scale Scores may also be obtained. These are: Behavior; Intellectual and School Status; Physical Appearance and Attributes; Anxiety; Popularity; and Happiness and Satisfaction. Raw scores may be converted to percentiles, stanines and/or T-scores. The scale was normed on children aged 8-18.

Test-retest reliability on the PH ranges from .42 (testing interval of 8 months) to .96 (testing interval of 3 to 4 weeks), with a median test-retest reliability of .73 (Piers, 1984). Correlations with other measures of children's self concept range from .32 to .85; correlations between children's self report on the PH and peer and teacher ratins of behavior or self-concept range from -.03 to .55.

<u>Child Behavior Checklist</u>

The Child Behavior Checklist (CBCL) (Achenbach and Edelbrock, 1983) (see Appendix F) was "designed to record in a standardized format the behavioral problems and competencies of children aged 4 through 16, as reported by their parents" (Achenbach & Edelbrock, 1983, p. 7). The CBCL consists of 113 behavior problem items, scored on a 3-step response scale, plus 20 social competence items. Both Total Social Competence and Total Behavior Problem scores can be obtained. A Child Behavior Profile, consisting of 9 behavior problem scales can also be obtained. Total raw scores on the Social Competence Scale, the Total Behavior Problem Scale and the 9 subscales are converted to T-scores. The scales are arranged under three main headings: Internalizing Syndrome, Mixed Syndrome and Externalizing Syndrome. These clinical scales are based on extensive factor analysis of the behavior problem items (Achenbach & Edelbrock, 1983). The position of each of the 9 scales in relation to the main headings depends on the age and the sex of the child. For Example, the behavior problem scales are arranged as follows for boys aged 12-16:

<u>Internalizing Syndromes</u>

Somatic Complaints

Schizoid

Uncommunicative

Immature

Obsessive-Compulsive

<u>Mixed Syndromes</u>

Hostile Withdrawal

<u>Externalizing Syndromes</u>

Delinquent

Aggressive

Hyperactive

The CBCL is reported to be a highly reliable instrument (Achenbach and Edelbrock, 1983). The authors used intraclass correlation coefficients (ICC's) to establish test-retest and interscorer reliability of the behavior problem and social competency items of the scale. Short-term (less than 3 months) test-retest reliability was 0.95 for the behavior problem items and 0.99 for the social competence items. Longer term (more than 3 months) test-retest reliability was 0.84 and 0.97 for the behavior problem and social competence items, respectively. Inter-parent reliability was 0.98 for the behavior problem items and 0.98 for the social competence items.

The authors assessed the content, construct and criterion validity of the CBCL. Content validity was assessed by determining whether or not the items were related to expressed clinical concern of the parents. Behavior problem and social competence items were found to be signifcantly related to clinical status established independently of CBCL (p < .01 for behavior problem and social competence items, respectively). Construct validity was established by comparing the Total Behavior Problem Score with scores on other established parent rating instruments. Achenbach and Edelbrock (1983) found correlations between the CBCL and and other similar instruments to be as high as correlations usually found among the groups of tests that measure specific abilities (such as general intelligence scales). Criterion-related validity was evaluated by using referal for mental health services as the criterion. Using demographically similar groups, the CBCL differentiated between referred and non-referred groups (p < .001), on both behavior problem and social competence items.

Thus the CBCL provides a measure of the parent perceptions of their child's level of functioning, and this perception has been shown to be consistent with external evaluations of the child's behavior. In this study, the CBCL was completed by both parents of all TS subjects living in a two parent home so that an inter-parent reliability score could be obtained. The average of the parents' scores was used to compute the Total Behavior Problems Score, the Social Competence Score, and the Child Behavior Profile. If there was only one parent living in the home with the child, then that parent's scores were used alone.

Family Measures

Four family scales were used to assess level of family functioning. These measures were designed to measure a broad range of the factors involved in family functioning (Olson,, 1985). The use of standardized instruments in family assessment is relatively new, and the interactive qualities of the family unit make standardized assessment problematic (Riskin & Faunce, 1970). The instruments chosen represent the best available measures at this time (Skinner, 1986).

Family Adapatibility and Cohesion Evaluation Scales III. The Family Adaptability and Cohesion Scales III (FACES III) (Olson, Portner & Lavee, 1985) (Appendix G) is a 20 item Likert-type scale. Family cohesion is defined as "the emotional bonding that family members have toward one another" (Olson, McCubbin, Barnes, Larsen, Muxen & Wilson, 1985, p. 4). Family adaptability is defined as "the ability of a marital or family system to change its power structure, role relationships and relationship rules in response to situational and developmental stress" (Olson et al.,

1985, p. 4). Thus FACES III is designed to measure family functioning along two dimensions: the degree to which families are separated from or connected to each other, and the extent to which the family system is flexible and able to change. The authors suggest that balanced families will function more effectively than extreme families, and that it would be expected that families who fall in the extreme range on both dimensions would have more difficulty in coping with developmental and situational stress. Thus, too little or too much cohesion and/or adaptability would be seen as dysfunctional in the family system, whereas a balance between the two extremes would enhance family functioning.

FACES III is the third version of the FACES scale. The primary goal in developing this version was to improve the reliability, validity and clinical utility of the scale. Test-retest reliability is reported to be 0.83 for cohesion and 0.80 for adaptability. Internal consistency is reported to be .77, .62 and .68 for cohesion, adaptability and the total scores, respectively (Olson et al., 1985).

Olson et al.'s (1985) Circumplex Model of family functioning assumes independence (orthogonality) between the two dimensions of cohesion and adaptability. The correlation between these two scales is reported to be 0.03 (Olson et al., 1985), suggesting that these two dimensions, as measured by the FACES III, are indeed orthogonal. A factor analysis of the scale revealed that items designed to measure cohesion loaded on the Cohesion factor, whereas items designed to measure adaptability loaded on the Adaptability Factor. Thus data suggesting that there is construct validity for this scale.

Each TS subject completed FACES III. The test was read aloud to children 12 and under; older children completed the scale independently. Both parents also completed FACES III. Scores from parents and TS subjects were averaged for a Total Family Score and normative data was available for comparison purposes (Olson et al., 1985).

Family Satisfaction. The Family Satisfaction Scale (Olson and Wilson, 1985) (Appendix H) is a 14-item true-false scale designed to measure degree of satisfaction with level of family cohesion and adaptability. Normative data was available for parents and for adolescents. In terms of reliability, Olson et al. (1985) report a Cronbach alpha coefficient of .91 for the total score. Test-retest reliability was 0.75 (Olson et al. 1985). They suggest using only the total score for research purposes, because their factor analysis suggests that family satisfaction is unidimensional and because reliability and validity are strongest for the total score. In the current study, only the parents completed the scale, since normative data were not available for a combined family score nor for younger children.

Family Crisis Oriented Personal Evaluation Scales. The Family Crisis Oriented Personal Evaluation Scales (F-COPES) (McCubbin, Larsen & Olson, 1981) (Appendix I) is a 30-item Likert-type scale created "to identify effective problem solving and behavioral strategies utilized by families in difficult or problematic situations" (Olson et al., 1985). In addition to a total score, five subscale scores can be obtained: The subscales are: Acquiring Social Support; Reframing; Seeking Spiritual Support; Mobilizing Family to Acquire and Accept Help; and Passive Appraisal. In terms of reliability, a Cronbach's Alpha for the total scale was .86, and ranged

from .62 through .83 for the subscales. Test-retest reliability was 0.81 for the total scale and ranged from 0.61 through 0.95 for the subscales (Olson et al., 1985).

Both parents completed the F-COPES and their scores were averaged to obtain the Total Family Score (Olson et al., 1985).

Family Strengths. Family Strengths (Olson, Larsen & McCubbin, 1982) (Appendix J) is a 12-item true-false scale designed to measure family pride and family accord. Family pride is defined as "Family attributes relating to respect, trust, and loyalty within the family" (Olson et al., 1985, p. 141). The authors define family accord is defined as "Attributes relating to a family's sense of mastery or competency" (Olson et al., 1985, p. 141). Norms are available for combined parents' score and for a total family score. Test-retest reliability for the total score is 0.58, and Cronbach's alpha for the total score is .83 (Olson et al., 1985).

Both parents completed the Family Strengths Scale and their scores were averaged to obtain a Total Family Score (Olson et al., 1985).

C. Procedures

All TS subjects were administered the Rorschach Inkblot Test, the Piers-Harris Children's Self-Concept Scale and the FACES III. All parents were asked to complete the History and Information Questionnaire, the TSGS, the CBCL, the FACES III, F-COPES, the Family Satisfaction Scale and the Family Strength Scale. Clinical staff at the Tourette Clinic also completed a TSGS on each child, and the clinican's ratings were as the severity measure in the main analysis.

All subjects were given both group and individual feedback on the results of the study. Feedback was only given after all data had been collected and coded.

Most of the TS subjects were tested at the TS Clinic at St. Boniface Hospital. Parents were given their test forms at the time of assessment, and were asked to complete these at home, independently of each other. A stamped, addressed envelope was provided so that the form could be retrieved by mail. A coding system was used to insure anonymity. If the test forms were not returned within three weeks, a follow-up phone call was made by the researcher.

The researcher was not involved in any therapeutic interventions with the TS subjects or their parents. Participation in the study was completely separate from therapeutic interventions; i.e., other clinicians in the TS Clinic were not made aware of their patient's agreement to participate in the study, nor of any decisions to withdraw from the study.

Every effort was made to bring TS subjects into the TS Clinic for the assessment procedure. In the case of out of town subjects for whom travel to Winnipeg was impossible, the researcher went to their homes to administer the tests. Out of town subjects were included to obtain the broadest range of TS subjects and their families.

D. Design

The primary purpose of the current study was to evaluate the interactional relationship among severity of TS symptomatology, level of individual functioning and level of family functioning. To explore this type of question, a one group posttest only design was deemed appropriate

(Cook & Campbell, 1979). Posttest generally refers to some sort of treatment, so that the questions asked concern the impact of the treatment on a group of subjects; in a broader sense, posttest may refer to some variable common among the group members, such as having committed a crime (Cook & Campbell, 1979), or, as in the case of the proposed study, having TS.

A one group posttest only design does not allow for direct causal inferences to be made (Cook & Campbell, 1979). The authors note, however, that this design is frequently used in social and clinical settings, in which a wealth of contextual information is available on the group being studied. This contextual information allows for some reasonable causal inferences to be made (Cook & Campbell, 1979). In the case of the current study, the review of the literature indicates that a wealth of contextual information is indeed available on individuals with TS. More specifically, the work of Cohen et al. (1982), Wilson et al. (1982), Bruun (1984), Matthews et al. (1985), and Grossman et al. (1986) strongly suggest that having TS has a significant effect on both individual and family functioning. The relationship of severity of TS symptomatology to these factors has not yet been explored. Thus the salient contribution of the current study is the examination of the relationship among these variables. The questions being explored, and the design of the study are consistent with a circular causality model of functioning, as discussed above. Since the literature presented here indicates that TS does have an effect on levels of individual and family functioning, the critical question in the current study is whether or not the severity of TS symptomatology interacts with level of individual and family functioning, such that level of functioning can predict severity of TS symptomatology.

Because the focus of this study was the comparison between milder and more severe TS symptomatology an external comparison group was not utilized. The availability of normative data of most measures allowed for the analysis of some patterns between both TS groups, and between TS subjects and normative samples.

Cook and Campbell (1979) also note that a one group posttest only design is useful in generating new hypotheses about the phenomenon being studied.

E. Analysis

In the first analysis, a multiple regression analysis was used to evaluate the relationship among the three primary variables under investigation. This type of an analysis yields information about the predictive value of a set of variables (Tabachnick & Fidell, 1983). The hypothesis tested statistically was that level of individual and family functioning predict severity of TS symptomatology. Thus the dependent variable (DV) was severity of TS symptomatology and the predictor variables were levels of individual and family functioning.

Tabachnik and Fidell (1983) suggest that a subject to predictor variable ratio of 20:1 is optimal, and that a ratio of 5:1 is the minimum requirement. Given the rarity of TS, a 20:1 ratio is beyond the scope of the present study. Four predictor variables, yielding a ratio of 6:1, were chosen based on their hypothesized importance to the predictive equation. These variables included two individual and two family variables. A combination of family and individual variables was chosen to reflect the expectation that both types of functioning will interact in affecting the

TS severity variable. These were the D score on the Rorschach, the Total Score on the Piers-Harris Children's Self Concept Scale, the Total Score on the CBCL and the parents' Score on F-COPES. The D Score provides a measure of the individual tolerance for stress, and the Total Score on the Piers-Harris provides a measure of individual self concept.

The CBCL provides a measure of parents' perception of their child's overall level of functioning, and the F-COPES provides a measure of the family's ability to cope with stress and change.

In a second analysis, other comparisons were made, using an Hotelling's T-squared analysis. This statistic was chosen because it is designed to minimize chance significant findings with a large number of comparisons. Comparisons were made between subjects with mild to moderate (Low) and severe to extreme (High) TS symptoms (based on TSGS scores), and between the TS subjects and the normative sample on a number of measures.

Finally, frequency and mean data from the Rorschach, cluster scores from the Piers-Harris, and Family Type and Range from FACES III were also visually inspected to determine whether or not differences between existed Low and High TS subjects, or between TS subjects' scores and normative data.

The above analyses were used to clarify and amplify the primary findings, as well as to generate hypotheses for future research.

VIII.   RESULTS

A. Regression Analysis

The hypothesis that level of individual and familial functioning would predict severity of TS symptomatology was tested using a hierarchical multiple regression. The predictor variables were Total Score on the Piers-Harris, the D-score on the Rorschach, Total Score on the CBCL and parents' average Total Score on the F-COPES. The predicted, or dependent variable was the clinician's rating on the TSGS.

The resulting Multiple R of .434 (R - Squared = .188; Adjusted R - Squared = -.003; F = .984; p < .44) indicates that the independent variables accounted for less than 19% of the variability in the TSGS scores. None of the independent variables alone contributed significantly to the prediction of the TSGS score (Table 1). Therefore, the null hypothesis cannot be rejected on the basis of these findings.

B. History and Information Questionnaire

Data from the History and Information Questionnaire were descriptive in nature (Tables 2 - 6). Information about medication was not used to assign TS subjects to comparison groups. Several parents reported that they had recently taken their child off medication, had changed the dosage, or had recently begun medication. Thus, it was believed that valid comparisons between medicated and non-medicated groups could not be made.

Table 1

Predictability of TS Symptom Severity

Independent Variable	Regression Coefficient	Standard Error	T (df=17)	Partial R↑2	p
D	− 2.83	3.95	− .715	.03	.48
PH	.53	.54	.996	.06	.33
TCBCL	.65	.41	1.613	.13	.13
FCOPES	− .06	.16	− .410	.01	.69
CONSTANT	−47.97				

Dependent Variable : TSGS score

Standard Error of Estimation = 18.44

Adjusted R squared = − .003

R squared =   .188

Multiple R =   .434

Table 2

Type and Frequency of Tics Reported by TS Subjects

Type	Number	% Reported
MOTOR	21	95.5
Face	5	22.7
Neck	12	54.5
Arm	9	40.9
Leg	6	27.3
Torso	8	36.4
PHONIC	16	72.7
Sniffing	4	18.2
Coughing	9	40.9
Swearing	15	71.4
Barking	3	13.6
"Calling out"	9	40.9

Table 3

Reported Behavior and Sleep Problems

	Never	Rarely	Some times	Often	Almost Always	TOTAL
	%	%	%	%	%	%
Behavior Problems						
Temper outbursts	4.5	22.7	40.9	31.8	0.0	95.5
Obsessive-compulsive	33.3	9.5	23.8	23.8	9.5	66.7
Hyperactive	23.8	28.6	14.3	23.8	9.5	76.2
Extreme anxiety	9.5	19.0	42.9	23.8	4.8	90.5
Extreme mood swings	9.5	23.8	38.1	19.0	9.5	90.5
Aggressive behavior	19.0	23.8	19.0	33.3	4.8	81.0
Coprolalia	71.4	4.8	14.3	9.5	0.0	28.6
Running away	85.0	10.0	5.0	0.0	0.0	15.0
Lying/stealing	47.6	23.8	9.5	14.3	4.8	52.4
Sleep Problems						
Getting to sleep	22.7	18.2	18.2	27.3	13.6	77.3
Staying asleep	52.4	14.3	14.3	14.3	4.8	47.6
Bad dreams	63.6	13.6	18.2	0.0	4.5	36.4
Bed wetting	95.2	0.0	4.8	0.0	0.0	4.8

Note. Total percent excludes the "never" responses.

Table 4

History of Allergies or Hyperactivity of TS Subjects

	% Reported
Allergies	
Currently troubled by allergies	36.3
Previously on antihistamines	29.4
Currently on antihistamines	10.0
Hyperactivity	
Past	44.4
Previously on medication for Hyperactivity	31.8
Currently Hyperactive	31.8
Currently on medication for Hyperactivity	0.0
Hyperactive at home	76.9
Hyperactive at school	76.9

Table 5

Frequency of Academic Problems

Problem	% Reported
Repeated a grade	31.9
Special education classes	59.1
Poor work habits	45.5
Inattentiveness	40.9
Poor use of time/materials	54.5
Doesn't listen	36.4
'Careless' written work	50.0
Poor self discipline	50.0
Other	22.7

Table 6.

Social and Peer Activities Reported by TS Subjects

Activity	% Reported
Social	
Organized sports,school	59.1
Organized sports,community	68.2
Church groups	31.8
YMCA,YWCA,YMHA	9.1
Other	36.4
Peer	
No friends	0.0
Some friends, none close	27.3
1-2 close friends	68.2
More than 3 close friends	4.5
Older friends	4.5
Younger friends	4.5
Same age friends	90.9

C. Tourette Syndrome Global Scale

The TSGS was used to assess overall level of TS symptomatology, and to separate the TS subjects into two groups, Low TS and High TS. The clinicians' ratings were used to assign TS subjects into these groups (Table 7). Eighteen of the 22 subjects (82%) fell into the mild-to-moderate range, and four subjects who fell into the severe-to-extreme range. The small size of this latter group precluded statistically valid comparisons. Parents' averages could not be used, as there were only 16 intact families, and therefore only 16 true average scores. Furthermore, parents' ratings revealed a similarly skewed distribution (Table 8). Therefore the median score of 19 (based on clinicians' ratings) was used to distinguish between Low TS and High TS groups. This yielded two groups of 11 subjects each.

The combined mean rating in this sample was 22.13, with a standard deviation of 16.17. This was more than one standard deviation below the mean score of 40.2 (S.D. 15.2) repoted by Harcherik et al. (1984). The population on which the test was normed (N=45) had a much broader age range (6-56 years) than the current sample (8-16 years), but the mean age of the normative sample (14.0 years) was fairly close to the mean age of the current sample (12 years). Harcherik et al reported a normal distribution of scores, whereas the scores in the current sample were slightly skewed (sk = 1.08) in a positive direction. Some possible reasons for these differences between samples will be discussed below.

Inter-rater agreement on the TSGS score ranged from 76.18% to 80.64% (Table 9). This was comparable to the 89% agreement reported by Harcherik et al. (1984).

Table 7

Tourette Syndrome Global Scale Scores

	Mean	SD	Range
Clinician's Ratings (n = 22)			
Total Score	25.0	18.4	1.0 - 74.0
Tics	9.7	8.2	1.0 - 40.0
Behavior Problems	15.3	12.3	0.0 - 46.2
Mother's Ratings (n = 21)			
Total Score	20.9	15.0	1.0 - 52.0
Tics	9.4	7.9	0.5 - 24.0
Behavior Problems	11.5	8.9	0.0 - 29.0
Father's Ratings (n = 17)			
Total Score	19.4	14.4	0.5 - 50.7
Tics	8.3	6.5	0.5 - 21.0
Behavior Problems	11.1	8.9	0.0 - 29.7

Table 8

Tourette Syndrome Global Scale Severity Ratings by Percent

	Severity (%)			
Rater	Mild	Moderate	Severe	Extreme
Clinicians (n = 22)	59.1	22.7	13.6	4.5
Mothers (n = 21)	60.0	30.0	10.0	0.0
Fathers (n = 17)	71.4	14.3	14.3	0.0
Average	62.5	23.2	12.5	1.8

Table 9

Tourette Syndrome Global Scale Inter-rater Percent Reliability

	Mean	SD	Range
Clinicians - Mothers	76	23.12	10 - 100
Clinicians - Fathers	77	26.12	10 - 100
Mothers - Fathers	90	26.25	54 - 100

D. Rorschach Inkblot Test

The Rorschach data was analyzed using an Hotelling's T-squared statistic. In the first analysis Low TS and High TS groups were compared to each other; in the second analysis all the TS subjects' scores were compared to the normative data. Variables that were found to differentiate significantly between the TS group and the normative sample are summarized in Table 10.

Because the normative data varies among age groups, the raw scores were transformed so that meaningful comparisons could be made. The normative mean was assigned a value of 5. Scores that were within one standard deviation of the subject's normative age group mean were also assigned a value of 5. Scores that were between one and two standard deviations above the normative mean were assigned a value of 6, scores between two and three standard deviations were assigned a value of 7, and scores at or above three standard deviations above the normative mean were assigned a value of 8. Scores between one and two standard deviations below the normative mean were assigned a value of 4. Scores between two and three standard deviations below the normative mean were assigned a value of 3, and scores three or more standard deviations below the normative mean were assigned the value of 2. This transformation also removed any outliers from the data set, as extremely deviant scores were included with those in the $\pm$ 3 standard deviations from the mean. This also served to increase the validity of the comparison, as it is unlikely that a score of e.g., $\pm$ 6 standard deviations from the normative mean would be more clinically relevant than one that was $\pm$ 3 standard deviations from the normative mean.

The results of the analysis of all Rorschach variables are summarized in Table 11. Inter-rater agreement ranged from 33% to 100% (Table 12). In most cases, disagreements were due to coder error. Disagreements based on factors other than error were settled by referring to the coding criteria. A third coder was available for arbitration, but this proved to be unnecessary. It should be noted that the low inter-rater agreements were all on codes that occurred infrequently in this sample, so that disagreement on one or two responses significantly lowered the percent agreement.

Validity of the Protocols

The mean number of responses to the Rorschach inkblots given the TS subjects fell within the normal range. No subjects gave fewer than ten responses, which is the cut-off point for interpretive validity suggested by Exner (1986). Intellectual assessment devices were not administered to the subject in the current sample at the time that the data for this study was gathered. Information from previous assessments, at either the TS Clinic or from other sources, as well as interactions with the subjects, indicated that all subjects were at or above the Low Average Range of intellectual functioning. Thus, the Rorschach protocols in this sample can be considered valid for interpretation.

Rorschach Coding Variables

Contrary to expectation, the average D score was within the normal range for all age groups. Only five subjects (22.7%) had D scores that were below the normative average for their age group, whereas thirteen

subjects (59.1%) had average D scores and four subjects (18.2%) had above average D scores. When the D score was adjusted (ADJ D) to minimize those components that are related to situational stress, only two subjects (9.1%) had below average scores, whereas five subjects (22.7%) had above average scores. The small difference between the D score and ADJ D reflects another finding: both inanimate movement and shading codes were not elevated in this sample, contrary to expectation.

In the current sample, none of the variables that provide an index of the individual's interpersonal orientation fell outside the cutoff point of one standard deviation above or below the mean across all age groups. One of the variables, Pure Human content (H) did, however, fall at the low end of the normal range for most age groups and significantly differentiated between TS subjects and the normative samples. Although the trend towards low H content was not as strong as expected, this trend was bolstered by the finding that 6 (27.3%) of the TS subjects gave no H contents and that another 5 (22.7%) gave only one H content response. The mean expected H content across ages ranges from 2.18 to 3.26 (Exner, 1986). The Isolate: Response ratio (Isolate:R) and Aggressive Special Score (Ag), which are also related to interpersonal oriention, fell within normal limit.

The indices of perceptual accuracy (X + %, F + % and X-%) were all significantly lower than would be expected. The Affective Ratio (Afr) was also significantly lower than would be expected. The Lambda ratio did not reliably differentiate between TS subjects and the normative sample. This was not due to a tendency for the subjects to score within the normal range on this variable. Instead, a bimodal distribution, with subjects tending to score either above or below the normative mean scores, was found on this variable.

The egocentricity ratio significantly differentiated between TS subjects and the normative sample. The lowest normative mean score for this ratio is .44 (SD=.10); only six subjects (27.3%) had an egocentricity ratio of .44 or above.

The TS subjects as a group scored within normal limits on all of the Special Scores. The Depression Index (Depri) was also within normal limits. The Schizophrenia Index (Sczi), on the other hand, was elevated and differentiated between TS subjects and the normative sample ($p < .01$). It should be noted that the maximum Sczi is 5, and that a Sczi of at least 4 is required to reliably distinguish between schizophrenics and non-schizophrenics (Exner, 1986). In the current sample, the mean Sczi = 2.36 (S.D. = 1.22). The Sczi for 14 subjects (63.7%) in the current sample was 1 or 2 and only five subjects (22.7%) had a Sczi of 4 or 5.

Table 10.

Results of Hotelling's T-test for Selected Rorschach Variables and T-test
for All TS Subjects vs Normative Data

	F-value	df	p
TS Low vs High	0.98	8,13	.53
TS vs Normative	49.38	8,35	.01 *

TS vs Normative Sample

Item	T-statistic
X +%	-17.79 *
F +%	- 8.90 *
X -%	+ 7.66 *
Pure H	- 3.74 *
Schizophrenia Index	4.54 *
Affective Ratio	- 3.19 *
Egocentricity ratio	- 4.81 *
Lambda	- 1.47

* p < .01

Note: Degrees of freedom = 21.

Table 11.

Descriptive Statistics for All Rorschach Variables

Variable	Mean	S.D	Range	
Response	19.55	11.64	11 - 48	
Zf	10.96	5.72	4 - 26	
Zsum	34.73	20.69	11 - 86	(d)
Popular	3.96	1.56	1 - 7	(b)
Pairs	6.09	4.61	0 - 17	(e)
Reflections	0.18	0.39	0 - 1	(f)
Location				
Whole	8.36	4.52	3 - 19	
Detail	9.96	9.07	0 - 31	
Unusual Detail	2.36	2.67	0 - 9	
Space	2.00	1.38	0 - 6	
Developmental Quality				
Synthesized	4.45	3.58	0 - 12	
Synthesized/vague	0.46	0.80	0 - 3	(f)
Ordinary	14.86	9.38	2 - 39	(e)
Vague	0.86	1.25	0 - 4	(f)
Form Quality				
Superior/overelaborated	0.23	0.61	0 - 2	
Ordinary	8.82	3.87	3 - 17	(b)
Unusual	6.09	4.46	0 - 20	(a)
Minus	5.45	4.43	0 - 17	(a)
No Form	0.05	0.21	0 - 1	(d)
M-Quality				
Superior/overelaborated	0.91	0.29	0 - 1	(d)
Ordinary	1.23	1.07	0 - 3	(d)
Unusual	0.68	0.84	0 - 3	(d)
Minus	0.77	1.34	0 - 5	(e)
Movement				
M	2.82	2.06	0 - 8	
FM	1.91	1.57	0 - 5	(f)
M	0.96	1.09	0 - 3	(e)
Color				
C	0.18	0.39	0 - 1	
CN	0.00	0.00	0 - 0	
CF	1.41	1.14	0 - 4	
FC	1.41	1.56	0 - 6	

(table continues)

Variable	Mean	S.D	Range
Shading			
C'	0.00	0.00	0 - 0
C'F	0.14	0.35	0 - 1
FC'	0.55	0.96	0 - 4
T	0.00	0.00	0 - 0
TF	0.00	0.00	0 - 0
FT	0.73	1.12	0 - 4
V	0.00	0.00	0 - 0
VF	0.14	0.35	0 - 1
FV	0.18	0.39	0 - 1
Y	0.00	0.00	0 - 0
YF	0.27	0.55	0 - 2
FY	0.54	0.74	0 - 2
FD	1.77	1.48	0 - 5
F	10.86	7.75	1 - 32
Content (primary only) _____ (d)			
Whole Human	1.82	1.65	0 - 5
Fictional Human	1.32	1.17	0 - 4
Human detail	1.27	1.45	0 - 4
Fictional Human Detail	0.32	0.65	0 - 2
Animal	8.22	4.84	2 - 18
Fictional Animal	0.59	0.91	0 - 3
Animal detail	2.55	2.56	0 - 10
Fictional Animal Detail	0.18	0.39	0 - 1
Abstraction	0.04	0.21	0 - 1
Alphabet	0.00	0.00	0 - 0
Anatomy	0.59	1.26	0 - 5
Art	0.09	0.29	0 - 1
Archaeology	0.05	0.21	0 - 1
Blood	0.00	0.00	0 - 0
Botany	0.77	1.19	0 - 4
Clothing	0.23	0.43	0 - 1
Cloud	0.00	0.00	0 - 0
Explosion	0.05	0.21	0 - 1
Fire	0.09	0.43	0 - 2
Food	0.14	0.35	0 - 1
Geography	0.14	0.35	0 - 1
Household	0.18	0.59	0 - 2
Landscape	0.96	0.95	0 - 3
Nature	0.18	0.50	0 - 2
Science	0.68	0.99	0 - 3
Sex	0.00	0.00	0 - 0
X-ray	0.00	0.00	0 - 0
Ideographic content	1.46	1.68	0 - 7

(table continues)

Variable	Mean	S.D	Range	
Special Scores				
DV	0.36	0.66	0 - 2	
Incom	1.14	2.44	0 - 11	(e)
DR	0.04	0.21	0 - 1	(f)
Fabcom	0.50	1.10	0 - 5	
ALOG	0.05	0.21	0 - 1	(e)
CONTAM	0.00	0.00	0 - 0	
WSUM6	4.86	9.03	0 - 42	
AG	0.41	0.96	0 - 4	(f)
CONFAB	0.05	0.21	0 - 1	(e)
CB	0.00	0.00	0 - 0	
MOR	0.68	1.04	0 - 4	(f)
PER	0.18	0.66	0 - 3	(f)
PSV	0.32	0.78	0 - 3	
Ratios, Percentages				
and Derivations				
Zd	0.25	4.18	-7 - +6	
EB (total M)	2.82	2.06	0 - 8	
(weighted color)	2.30	1.74	0 - 6	
EA	5.11	3.30	1 - 13.5	(f)
eb (FM + M)	2.86	2.18	0 - 7	
(total shading)	2.27	1.86	0 - 7	
es	5.50	3.25	1 - 14	
D	0.05	1.05	-2 - +3	
ADJD	0.27	0.88	-1 - +3	
a	3.50	2.70	0 - 10	
p	1.86	2.08	0 - 9	
Ma	2.00	2.12	0 - 7	
Mp	0.91	0.92	0 - 4	
Depi	1.27	1.12	0 - 4	
Sczi	2.36	1.22	1 - 5	
FC	1.18	1.59	0 - 6	
F+C	1.64	1.26	0 - 4	
PureC	0.18	0.39	0 - 1	
Afr	0.64	0.30	0.33- 1.27	(b)
3r + (2)/R	0.31	0.15	0 - 0.60	(b)
L	1.42	1.35	0.11- 5.25	(c)
Blends	2.59	2.18	0 - 8	(f)
X +%	0.46	0.12	0.27- 0.67	(b)
X -%	0.23	0.13	0 - 0.45	(a)
F +%	0.46	0.22	0 - 0.80	(b)
Isolate	2.09	1.88	0 - 9	(d)
AB+Art	0.14	0.35	0 - 1	(d)
AN+XY	0.59	1.26	0 - 5	(d)
A +(H)	3.00	2.09	0 - 8	
Hd +(Hd)	1.41	1.37	0 - 4	
Pure H	1.82	1.65	0 - 5	(f)

(table continues)

Note:

 (a) = greater than one standard deviation above the normative mean, across all ages.

 (b) = greater than one standard deviation below the normative mean, across all ages.

 (c) = bimodal distribution.

 (d) = normative data not available.

 (e) = above the normative mean across all ages, but within one standard deviation for at least one age.

 (f) = below the normative mean across all ages, but within one standard deviation for at least one age.

Table 12.

Rorschach Inter-Rater Agreement

VARIABLE	% AGREEMENT
Location	99
(Z)	99
P	93
FQ	97
DQ	99
H	99
(H)	99
A	99
Special Scores	
INCOM	96
DR	87
FABCOM	73
AG	33
PSV	57
PER	75

Note. Inter-rater agreement was 100% for those variables not listed above.

Note. Disagreements in most cases were found to be due to coder error.

E. Family Measures

The Family Measures data were analyzed using Hotelling's T-squared analysis, first to compare Low TS with High TS families, and then to compare TS family scores with the normative samples.

The Hotelling's T-squared for Low vs High TS families did not differentiate between these two groups. Significant differences between TS families and the normative samples were demonstrated and are presented in Table 13. Data on mothers and fathers separately and on parent averages were compared on the F-COPES, Family Satisfaction and Family Strength measures. Data from FACES III allowed a family average to be used in addition to the above categories.

Inter-rater reliability on the family measures ranged from 63.8 to 85% (Table 14). On the FACES III the agreement among raters was higher than that reported by Olson et al. (1984). These researchers found correlations of .44 between mothers and fathers, 0.44 between fathers and adolescents, and 0.38 between mothers and adolescents. Agreement between mothers and fathers was not reported for the other family measures.

F. Piers-Harris Children's Self-Concept Scale

The Data from the Piers-Harris was analyzed using an Hotelling's T-squared statistic in a comparison between Low and High TS subjects. No significant results were obtained.

The cluster scores on the Piers-Harris were tabulated and visually inspected to determine whether or not TS subjects differed from the normative groups (Table 15). As a group, TS subjects fell within the normal range in all scoring categories. Almost all individual t-scores fell at or

above T=40, which is the lower limit for the normal range. One subject fell below this level on the Behavior subscale; another fell below this level on the Physical Attributes and Appearance subscale, and a third subject fell below this level on the Popularity subscale. All individual Total Scores fell between T=42 and T=79.

G. Child Behavior Checklist

An Hotelling's T-squared statistic was used to analyze data from the CBCL. Because different behavior problem categories are assigned for males and females, and for different age groups, only those categories that were the same across age and sex were included in the analysis. These were: Total Score, Total Social Competence Score, Internalizing, Externalizing, Somatic Complaints, Schizoid, Delinquent, and Aggressive. The Hotelling's T-test was used to compare Low with High TS subjects, and to compare all TS subjects' scores to normative data. A separate analysis, using only male subjects was also performed, in order to evaluate more of the subscales (see Table 16).

Inter-rater reliability was established using the percent agreement scores between parents for the CBCL Total Score, the Total Social Competency Score, and the Behavior Problem Total only (see Table 17).

Table 13

Results of Hotelling's T-test for Family Questionnaires.

	F-value	df	p
TS Low vs TS High	0.19	20,1	.97
TS vs Normative Samples	57.67	21,22	.01 **

TS vs Normative Sample

	Mean	SD	T	
FACES III				
Parents' Cohesion	35.8	5.2	9.70	*
Parents' Adaptability	22.8	5.3	9.24	*
Family's Cohesion	35.4	5.3	26.06	**
Family's Adaptability	23.6	3.5	25.02	**
F-COPES				
Parents Average				
Total	87.9	12.6	- 2.33	*
Acquiring social support	28.2	5.0	0.96	
Reframing	28.3	3.9	- 2.37	*
Seeking spiritual support	10.9	5.0	- 4.89	**
Mobilizing family	13.1	2.3	2.19	*
Passive appraisal	7.5	1.9	- 2.71	**
FAMILY SATISFACTION				
Mother Total Score	42.7	8.4	- 2.41	*
Father Total Score	44.2	9.1	- 2.94	**
Average Total Score	42.6	7.9	- 2.62	*
FAMILY STRENGTHS				
Mother Total Score	39.2	7.7	- 4.01	**
Father Total Score	21.8	7.9	- 3.60	**
Average Total Score	39.7	7.4	- 4.13	**

* p < .05.

** p < .01.

Table 14

Family Inter-Rater Percent Reliability

	N	Mean	SD
FACES III			
Mother-Child	21	63.8	14.5
Father-Child	17	71.3	11.4
Mother-Father	16	80.7	11.2
F-COPES			
Mother-Father	16	74.9	13.2
FAMILY SATISFACTION			
Mother-Father	16	85.0	15.5
FAMILY STRENGTHS			
Mother-Father	16	79.7	21.8

Table 15

Piers-Harris T-Scores

Score	Mean T-Score	S.D.	Range
Behavior	54.5	9.3	36-66
Intellectual & School status	55.2	8.4	41-70
Physical Appearance & Attributes	56.0	9.5	37-69
Anxiety	58.0	8.6	41-69
Popularity	55.0	11.0	29-70
Happiness & Satisfaction	57.1	6.7	42-63
Total	57.5	9.3	42-79

Note. Mean T-score = 50; S.D. = 10.

Table 16

Results of Hotelling's T-test for Child Behavior Checklist

	F-value	df	p
TS Subjects High vs Low (N = 22)	1.35	8,13	.30
TS Subjects vs Normative (N = 22)	2.09	8,35	.06
Male TS Subjects vs Normative (n = 19)	1.84	10,27	.10

Subscale Scores	T-value, all TS vs Norm	p	TS Males vs Norm	p	
Total Social competence	- 2.03	.06	- 2.05	.06	
Internalizing	3.05	.01	2.63	.02	
Externalizing	2.02	.06	2.04	.06	
Somatic complaints	3.52	.01	3.01	.01	*
Schizoid	2.89	.01	2.67	.02	
Uncommunicative	--	--	2.38	.03	
Obsessive-Compulsive	--	--	2.69	.02	
Delinquent	2.00	.06	2.02	.06	
Aggressive	2.13	.04	2.00	.06	
Hyperactive	--	--	2.79	.01	*
Total Behavior Problems	2.61	.02	2.33	.03	

* $p < .01$.

Table 17

Child Behavior Check List Inter-Rater Percent Reliability

	Mean	SD
Total Score	69.1	11.5
Social Competency	67.3	10.7
Behavior Problem only	69.6	13.2

Additional Analyses

Because there has been relatively little evaluation of the possible relationships among severity of TS symptoms and level of family, individual and social functioning, several post hoc analyses were performed on the data gathered in this study. The purpose of these analyses was to determine whether or not there were any trends, not revealed in the primary analyses, that would be productive to pursue in future research. The results of these analyses must be interpreted with extreme caution, as the combination of the relatively small number of subjects in the study and the large number of post hoc analyses makes the risk of spurious findings quite high.

In order to determine whether or not there were correlations between severity of TS symptoms and scores on any of the measures, a series of regression analyses was performed. In each of these analyses the three independent (i.e., predictor) variables were the three scores from the TSGS: Total, Tic, and Social Functioning scores. The Tic and Social

Functioning domains were included as separate predictor variables in order to assess the role, if any, that each of these domains would play in predicting the various dependent variables. The dependent (i.e., predicted) variable in each regression was a specific score or subscore from the measures used in this study. The independent variables were entered in a stepwise fashion, with $p < .100$ as the criterion for entry into the equation. All of the scores and subscores on all of the measures, except for the History and Information Questionnaire, were analyzed in this way.

The subscores that showed the highest correlation to the TSGS were the T-scores from the Child Behavior Checklist. Among all the subjects, the Externalizing, Delinquency, Aggression subscores and the total score were strongly associated with the TSGS Social Functioning domain. The Total Social Internalizing and Schizoid subscales had weaker but still positive correlations with the Social Functioning domain. The Delinquency subscale was strongly associated with the Tic domain: and Internalizing, Externalizing and Aggression were positively correlated with the Total Score on the TSGS. When only male subjects were included in the analysis, associations between the obsessive-compulsive and hyperactive subscale and both the tic and Social Functioning domains were found. In this subsample, a strong association between Total CBCL and Total TSGS scores was also found.

On the Piers-Harris Children's Self-Concept Scale, negative associations between Total CBCL and Total TSGS scores was also found.

On the Piers-Harris Children's Self-Concept Scale, negative associations between the Happiness and Satisfaction subscale and the Social Functioning domain, and between Total Scores on the Piers-Harris and TSGS were found.

On the Rorshach, associations between number of FY (Shading), X-%, and F+% and the Tic domain, and between Deviant Responses (DR), number of Reflection Responses (FRRF), and the Social Functioning Domain were found.

Post hoc analyses of the family questionnaires were not indicative of any meaningful trends.

IX. DISCUSSION

It is apparent from the analyses of the data that, contrary to expectation a combination of self-esteem, tolerance for stress, parental self-reported coping strategies, and parents' perceptions of the TS child's level of functioning did not predict severity of TS symptomatology. The analysis of these data indicates that these factors have little, if any, bearing on symptom severity. The data are not supportive of the circular model hypothesis that severity of TS symptoms would be associated with level of family and individual funnctioning. Further research would be needed to clarify what, if any, systemic factors might contribute to severity of TS symptomotology.

Further analysis of the data revealed that none of the measures used in this study distinguished between Low TS and High TS subjects. This is consistent with a recent study by Sverd, Curley, Jandorf and Volkersz (1988), who found that diagnosis of behavioral and attentional disorders did not distinguish between mild and severe TS boys. Other findings from the current study indicated, however, that TS children as a group demonstrate differences from the normative samples on a number of variables. In general, the data from this study suggest that the crucial factor in predicting deviance from the norm is having a diagnosis of TS, rather than the severity of the TS symptomotology.

In this section, a discussion of the results of the current study will be followed by some comments about the clinical implications. Suggestions

for future research will also be made.   It should be noted that, due to the relatively small sample size, these findings should be interpreted with some caution.

A. History and Information Questionnaire

The overwhelming majority (95.5%) of TS subjects were reported by their parents to be currently experiencing motor tics. These included facial tics, such as blinking, mouth grimaces and tongue protrusion; neck twisting and jerking; arm and leg jerking and lifting; and torso movements. A majority of the TS subjects (72.7%) were also reported to be currently experiencing phonic tics, such as sniffing; coughing; swearing; barking; and "calling out" in the classroom. The percentage of TS subjects experiencing coprolalia was 71.4, which was much higher than the 30% to 50% reported by Bruun (1984) and Shapiro and Shapiro (1982). The reason for this difference is not clear, although the relatively high mean (12 years) and modal (14 years) ages of this sample may be a contributing factor. This symptom does tend to emerge at a later age than do other symptoms (Fernando, 1967). It is possible that parents may have interpreted an adolescent prediction for swearing as coprolalia. This explanation does not appear to be a good one, as clinical observation of this symptom indicates that it is readily distinguishable from "ordinary" swearing. Coprolalic swearing tends to be sharp, explosive and not usually contextually appropriate.

The patterns of behavioral and sleep problems described by the parents are consistent with previous survey results (Stefl, 1984; Champion et al., 1988). These findings indicate that parents perceive their TS children and

adolescents to have behavioral problems, including temper outbursts, obsessive-compulsive rituals, a high level of activity, extreme anxiety and mood swings, and aggressive behaviors, that are similar to those that have been reported by other researchers (Champion et al., 1988; Stefl, 1984; Wilson et al., 1982). A high rate of sleep problems is also consistent with data from other researchers (Glaze et al., 1982, 1983). Nearly one-third (31.9%) of the TS subjects were reported to have failed at least one grade and well over one-half (59.1%) of the subjects were reported to be in some kind of special education classes (primarily resource classes). Large percentages (22.7% - 54.5%) were described as having problems functioning effectively in the classroom (for example, poor organization of time and material, inattentiveness, poor self discipline). Four subjects (8.2%) were reported to do exceptionally well in school. Intellectual assessment data were available on 21 of the 22 subjects. One subject was reported to be in the Low Average Range of intellectual functioning; twelve were in the Average Range; five were in the Above Average Range; two were in the Superior Range; and one was in the Very Superior Range.

Approximately one-third (31.8%) of the subjects were reported to have a current diagnosis of Hyperactivity, and 44.4% were reported to have been diagnosed as Hyperactive in the past. Although 31.8% had previously been on medication for Hyperactivity, none were currently using such medication. This suggests that, at least among the TS population that has been accurately diagnosed, use of stimulant mediations for hyperactive behavior has declined.

Taken together, the above data are consistent with previous findings that children and adolescents with TS as a group experience significant

behavioral, academic and sleep problems. On a positive note, the majority of TS subjects in this sample were repored to be involved in community activities, and to have at least some age-appropriate peer relationships. Thus, despite the above-mentioned problem areas, some level of adaptive functioning is apparent in most of these individuals. As a group, these subjects may be more successful in social than in academic activities.

B. Tourette Syndrome Global Scale

As noted previously, the combined mean rating of the current sample was more than one standard deviation below the mean score of the normative sample. A number of explanations for this will be discussed.

The normative population consisted of 45 drug-free outpatients whereas a percentage of the subjects in the current sample were on medication for TS symptoms. Thus, the lower mean in the current sample could reflect some control of symptoms by medication. The variability in time on or off medication, in types of medications used, and in the dosages did not, however, allow for valid analysis of the effect of medication on the level of symptom severity in the current sample.

Another factor that may be related to the differences between the normative and the current samples was that the former appears to have included only recently diagnosed TS patients, whereas the latter included both recently diagnosed TS patients and patients who had been associated with the TS Clinic for as long as six years. It is likely that most recently diagnosed TS patients would tend to have been referred at a time of intense symptomatology, whereas the current sample may have reflected a broader range of severity, including some subjects in a relatively quiescent phase of symptomatology.

One final difference between these two groups lies with the raters. For the normative study, only experienced clinicans were used as raters (Harcherik et al., 1984). In the current sample, raters included both experienced clinicians and parents (although only clinicians' ratings were used to separate subjects into High TS and Low TS subgroups). The effect of this difference was probably not the salient factor as the clinicans' mean rating in the current study was 25.02, which is still a full standard deviation below the normative mean.

C. Rorschach Inkblots

Contrary to expectations, the Rorschach scoring variables did not distinguish between Low TS and High TS subgroups. Further analysis of the data, however, revealed some significant differences between TS subjects and the normative sample. The ways in which TS subjects did and did not differ from age norms will be discussed next.

Tolerance for stress.

It was expected that children and adolescents would show diminished tolerance for stress, as measured by the D score on the Rorschach. These data suggest that this group of TS subjects does not have diminished tolerance for stress; indeed their stress tolerance as a group is well within the average range. The small difference between the D score and ADJ D reflects the finding that inanimate movement and shading codes were not elevated in this sample. Both of these codes are associated with situational stress and/or anxiety (Exner, 1986). This finding suggests that the TS subjects may not perceive themselves to be in a state of acute stress and/or anxiety. Another possible explanation is that they have

developed the coping skills necessary to deal effectively with what appears to be a very stressful disorder.

Coping style.

The ratios that reflect coping style, "EA" and "es", are subject to the development changes that occur as the child matures (Exner & Weiner, 1982). "EA" reflects coping strategies that are readily accessible, whereas "es" is a reflection of need states that are impinging on the individual (Exner, 1986). The number of subjects in each age group was too small to analyze these ratios by age. "EA" and "es" will therefore not be discussed except to note that these ratios and their component variables fell within one standard deviation across all age groups. Thus, the current data does not suggest that the TS subjects show any unusual patterns in terms of preferred coping styles.

Interpersonal orientation.

Several variables provide an index of the individual's interpersonal orientation. These are Pure Human content responses (H), the Isolate:Response ratio (Isolate:R), and the Aggressive (Ag) Special Score (Exner, 1986). In the current sample, none of these variables fell outside the cutoff point of one standard deviation above or below the mean across all age groups. H was at the low end of the normal range for most age groups, and signifantly differentiated between TS subjects and normative data (p < .01). This suggests a trend towards interpersonal avoidance. Although this trend was not as strong as expected, it is bolstered by the finding that six (27.3%) of the TS subjects gave no H responses, and another five subjects (22.7%) gave only one H response. The mean expected H response across ages ranges from 2.18 to 3.26 (Exner, 1985).

The Isolate:R ratio was within normal limits, indicating that, contrary to expectation, the TS subjects in this sample do not experience a strong sense of social isolation. The Ag Special Score also fell within the normal range, which was again contrary to expectation. Thus, the subjects in the current sample do not show evidence of a hostile interpersonal orientation (Exner, 1986).

These data indicate that, while the TS subjects in this sample show a slight tendency towards interpersonal avoidance, there is no evidence of significantly impaired interpersonal orientation. Thus, the expectation that TS subjects would show an avoidant or hostile interpersonal orientation was not met.

Perceptual accuracy.

The X+%, F+%, and X-% ratios reflect an individual's perceptual accuracy, and have implications for the individual's ability to make appropriate judgements (Exner, 1986). As expected, all of these variables differentiated between TS subjects and the normative sample ($\underline{p}$ < .01). According to Exner (1986), the X+% for children and adult nonpatients tends to hover around .80 with a standard deviation of about .10. The mean F+% for children ages 8-16 ranges from .80 to .85, with a standard deviation range of .9 to .12 (Exner, 1985). The X-% for these age groups ranges from .5 to .9, with a standard deviation ranging from .4 to .6 (Exner, 1985). F+% and X+% less than one standard deviation from the mean are interpreted as reflecting a significant impairment in perceptual accuracy (Exner, 1986). The F+% reflects the perceptual accuracy of those responses that are based only on use of the contour features of the blot. The X+%, on the other hand, reflects the perceptual accuracy of the total record. A low X+%

may reflect perceptual distortion, an idosyncratic perceptual style, or a failure to modulate affective responses (Exner, 1986). To assess which of these factors (separately or in combination) has contributed to a low X+%, other Rorschach variables must be examined.

The first of these variables is the X-%. This ratio reflects the proportion of responses that are based on a distorted perception of the blot, and are scored minus for Form Quality. Such responses are difficult, if not impossible, for the examiner to see, even when the subject points them out and describes them in detail. Most children and adults give one such response per protocol, but several minus responses in one protocol are rare in nonpatient populations (Exner, 1986). The significantly high mean X-% of this sample suggests that the responses generated by the subjects in this sample are based on perceptual distortions rather than on idiosyncratic perceptual styles. The TS subjects did not give more than the expected No Form responses, which would have reflected failure to modulate or control emotion-based responses. Thus, failure to modulate cannot be considered to have contributed to the low X-% ratio.

Finally, Popular responses (P), which are responses that are frequently given to particular blots or blot areas, provide a measure of the subject's ability to see the world as others do (Exner, 1986). The subjects in this sample gave fewer P responses across all age groups. This suggests that the TS subjects do not see the world as others do, and is consistent with the difficulties in perceptual accuracy, as discussed above.

In sum, the combination of significantly below expected X+% and F+% ($\underline{p}$ < .01) and elevated X-% ($\underline{p}$ < .01) indicated that the subjects in this

sample do not see the world, or understand events, in the same way that their peers do. This may have negative implications for their ability to make good judgements based on the available information. A tendency to make relatively poor judgements may be related to the behavioral problems, including impulsivity, that have been found in children and adolescents with TS.

Responsiveness to the environment.

It was expected that the TS subjects would show high levels of excitability, or be highly responsive to emotionally provocative stimuli, and to have difficulty modulating their responses. The data, however, did not support this expectation. In fact, the TS subjects as a group appeared to inhibit their responses to emotionally provoking stimuli.

As a group, the Ts subjects' Affective Ratio (Afr) was significantly lower than would be expected in a non-clinical population which may reflect some emotional constriction in these subjects (Blood, 1985). Exner (1986) suggests that a low rate of response to emotionally-loaded stimuli may be the "product of concerns about control" (p. 381). A low Afr may also be associated with difficulty in responding in affectively appropriate way to the environment. Thus, the subjects in this sample may tend to constrict their emotional responses in order to exert some control, in order to avoid the negative consequences of over-responding. The Lambda ratio may shed some further light on this interpretation. A high Lambda would be associated with a tendency to avoid stimulus complexity, and to respond in a rather simplistic, or perhaps avoidant fashion. A low Lambda, on the other hand, may reflect an inability to "back off", or to avoid complexity (Exner, 1986). In either case, some concerns about control of

responsiveness may be present. When Lambda is high, the subject may tend to be overcontrolled in his/her responsiveness to his/her world, whereas when Lambda is low, the tendency may be towards undercontrol (or impulsivity).

In the current sample, the Lambda score did not differentiate between TS subjects' scores and the normative group.This was not due to a tendency on the part of the subjects to score within the normal range on this variable. Instead, the Lambda ratio was distributed bimodally, with the responses of the subjects in the current sample tending to fall either above or below the normative mean age scores (usually at least one standard deviation above or below the mean). This suggests a tendency to either over- or under-inhibit responsiveness to stimuli. This finding is interesting in light of the suggestion by Wilson et al. (1982) that children and adolescents with TS have difficulty modulating and inhibiting impulses, actions, and thought.

The bimodal distribution of Lambda may reflect two separate types of TS subjects: those who do show impulsivity and failure of inhibition, and those who do not. Another explanation is that most of the subjects in the current sample do have problems with impulsivity and inhibition, and that some of these subjects have learned to cope with this by imposing rather rigid controls on their impulsivity. The low mean Afr, as discussed above, tends to support this explanation. Other support for this explanation is found in the trend toward a low (but within normal limits for at least some age groups) number of Blend responses. A Blend refers to the use of more than one determinant in generating the Rorschach response. A low number of Blends reflects a tendency to back away from complexity (Exner 1986). The combination of the low Afr, the tendency towards few Blend responses, and

the bimodally distributed Lambda suggests that some of the TS subjects in the current sample may tend to back away from complexity and to inhibit emotional responsiveness. It is possible that this is a way of coping with a biological hyperarousal or tendency to overrespond. Further exploration of this hypothesis, using a larger sample size, would be warranted.

Self-image.

The egocentricity ratio, which was less than one standard deviation from the normative mean for all age groups, differentiated between TS subjects and normative data. This ratio is frequently considered to be an index of self-esteem. A more precise description of this ratio, however, is that it reflects the degree of an individual's self-centeredness, or self-focus (Exner, 1986). As a group, the TS subjects showed a low degree of self-focus.

The low egocentricty ratio, does not, however, appear to be associated with a negative self-image in most of the TS subjects. These subjects did not show any elevations in morbid content, which is associated with a negative or "damaged" self image and possibly a pessimistic self-orientation (Exner, 1986). Anatomy (An) and X-ray (Xy) content responses are usually associated with bodily concern and often distress (Exner, 1986). Despite the physical nature of their TS symptoms, the subjects in the current sample did not show any elevations in these content categories.

Thus, although the expected low Egocentricity ratio was found, it does not appear to be associated with a negative, damaged, or pessimistic self-image. Blood (1985) found that children with learning disabilities (LD) had low Egocentricity ratios, accompanied by a negative or "damaged" self-image. LD and TS subjects therefore appear to share a low degree of self-focus, but apparently to have divergent self images.

Special Scores and Special Indices.

The TS subjects as a group scored within normal limits on all of the Special Scores. The Depression Index (Depri) was also within normal

limits, which is consistent with the finding that TS subjects as a group do not have negative self-images. The Schizophrenia Index (Sczi), on the other hand, was elevated and differentiated between TS subjects and the normative sample This appears to be related primarily to the low X+% and the elevated X-%. It should be noted that the maximum Sczi is 5, and that a Sczi of at least 4 is required to reliably distinguish between schizophrenics and non-schizophrenics (Exner, 1986). In the current sample, the mean Sczi = 2.36 (S.D. = 1.22). The Sczi for 14 subjects (63.7%) in the current sample was 1 or 2 and only five subjects (22.7%) had a Sczi of 4 or 5. Thus, as a group, the TS subjects cannot be considered to resemble a schizophrenic population.

Summary and Conclusions.

Analysis of the Rorschach data indicated that the TS subjects had a number of strengths. Although it was expected that the subjects in this sample would share a lowered tolerance for stress, this proved not to be the case. It is possible that, as a group, children with TS develop the skills necessary to cope with what appears to be a very stressful disorder. Although the Egocentricity ratio was significantly depressed for all subjects, this appears to reflect a lack of self-focusing, as there was no evidence of the expected negative self-image. Similarly, above average bodily concern was not found in this sample. Hostile or aggressive interpersonal orientations were not found in this sample, although there was a tendency towards interpersonal avoidance.

The TS subjects as a group showed a significant tendency to avoid both complexity in their world and emotionally provoking stimuli. A significant tendency towards perceptual inaccuracy was also found. This can lead an

individual towards making poor judgements. It is possible, however, that the tendencies towards simplifying one's world, avoiding emotional arousal and seeing the world differently from the way that others do can work in the TS subject's favor. Simplifying his or her world and avoiding emotional arousal may help the person with TS to control a possible biological tendency towards reactivity and impulsivity. A tendency towards overresponsiveness, impulsivity, or failure of inhibition is not likely to have been positively reinforced in the subject's daily life and thus learning to steer clear of stimuli that are overly provoking could be an asset for the TS child. Perceptual distortion may allow the person with TS to deny or minimize the ways in which he or she is different from others. In this way, a child or adolescent with TS may be able to minimize, or reframe, the effect of poor grades, placement in resource classes, and continual tiquing. A tendency to avoid self-focusing may also allow the TS subject to avoid dwelling on his/her differences from his/her peer group. This would leave the subject free to participate in social activities, as many of the subjects in this study were reported to do.

Although some significant deviations from normative samples were found, it cannot be said that the TS subjects as a group resemble a psychologically dysfunctional population. Some of their areas of deviation may, in fact, serve to insulate them from some of the possibly distressing effects of having TS.

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D. Family Measures

Contrary to expectation, the family measures did not differentiate between Low and High TS subjects, but these measures did, on the whole, differentiate between families in the TS sample and families in the normative sample. Because of the high level of agreement that were found between parents, and among family members, only the combined scores will be discussed.

FACES III.

The parents in the TS groups were found to be less cohesive, and to have less flexibility (adaptability) than the parents in the normative sample. Family cohesion and flexibility were also lower in the TS sample than in the normative sample. This was consistent with the findings of Mathews et al. (1985) that TS children perceive their families to be less flexible than nonpatient children perceive their families to be. These results are also consistent with the findings of Ferrari et al. (1980) that families of a child with epilepsy tend to be less cohesive than the normative families. It is important to note that the data suggest that a psychosomatic family model, which assumes elevated family cohesion (enmeshment), is not appropriate for understanding families with a TS member (Liebman et al., 1976). These data must be interpreted with some caution due to the relatively high mean (12 years) and modal (14 years) ages in this sample population. Ackerman (1980) suggests that the emergence of adolescence is likely to test the flexibility of a family's organization. Family cohesion is also likely to be lower in families with adolescents, as adolescents become more involved in peer activities and less involved with family activities. Thus, the possible effect of the high

proportion of adolescents in this sample on family cohesion and flexibility must be considered. The normative sample did include families across the life cycle, including those with adolescents, so it is not likely that having a large subsample of TS adolescents would account for all of the differences between TS families and normative families.

F-COPES.

In terms of overall coping strategies, as measured by the F-COPES, parents in the TS sample also scored below the normative parents. They were, by their report, less adept at using such strategies as reframing problems to make them less noxious and/or more manageable; and seeking support and/or guidance from religion and religious institutions. On the other hand, the TS parents were less likely than the normative parents to passively acknowledge and appraise problems. The TS parents were as likely to acquire supports outside the family to help cope with problems, and more likely to mobilize the entire family to seek and accept such support. The TS parents differed most sharply from the normative parents in their tendency <u>not</u> to seek religious guidance. This may be due to a possible bias in the normative sample, which was gathered from a predominantly Lutheran population in Minnesota. It is possible seeking religious guidance and/or support is more common in such a population.

The coping skills that were employed by the TS sample may also reflect a sampling bias. These parents have all had at least some contact with the TS Clinic, and many of them have had experience in advocating for their children in school. Thus, their tendency to seek out and to mobilize their families to accept support from outside their family may be a reflection of skills acquired in the diagnosis and treatment of TS. Nonpatient families

may not have had to develop such skills. This interpretation of the data is not meant to suggest that these skills do not represent a real strength in the TS families. Rather, this finding suggests that, at least in those families in which TS has been diagnosed, the families are able to acquire and use appropriate coping strategies.

Family Satisfaction.

The data indicate that the TS parents were less satisfied with their level of cohesion and flexibility than were the normative parents. Again, some caution must be used in interpreting these data, due to the high proportion of adolescents in this sample. Ackerman (1980) points out that the emergence of adolescence can be a confusing and difficult time for families and that "truly, it is impossible to raise teenagers" (p. 148). Family rules must be adapted to meet the changing needs of the individuals and the family unit, and relationships between parents and adolescents undergo fundamental changes. Thus, in addition to the problems presented by the child or adolescent with TS, the "normal" adolescent problems must also be addressed. It is likely that the relatively low satisfaction reported by the parents in this sample is a result of a combination of issues relating to coping with TS, plus issues relating to living with adolescents.

Family Strengths.

The TS parents also reported less overall pride in and accord among their families than did the normative parents. Again, the effect of adolescence on these factors may be a confounding element in this finding. This finding is, however, consistent with a relatively low level of family satisfaction.

<u>Summary and Conclusions</u>.

Overall, analysis of these data indicate that the parents of the TS subjects find their families to be less cohesive, less adaptive, as having fewer overall coping strategies and are less satisfied with their families than the families in the normative sample. Matthews et al. (1985) also found TS families to be less flexible. These researchers suggest that such families may find their resources to be overtaxed, and respond to control-related conflicts by becoming more rigid. The current findings are also consistent with those of Ferrari et al. (1983), in which families of children with epilepsy were found to be less cohesive than the norm. Findings such as these indicate that a psychosomatic family model is not appropriate for understanding families with a TS member. The interpretation of the current results is supported by the finding that level of family functioning is not predictive of severity of TS symptomatology. Finally, it has been suggested that the relatively large proportion of adolescents in this sample may account for some but not all of these findings in that the presence of adolescents in the family may lower both family cohesiveness and family satisfaction.

E. Piers-Harris Children's Self-Concept Scale

The scores in the Piers-Harris did not differentiate between Low and High TS subjects. The mean total T-Score and all mean T-scores for the subscales fell within the average range. In fact, all but three of the individual subjects' T-scores fell within or above the average range. This finding was quite unexpected, as children with academic problems such as those reported on these subjects have frequently been found to have poor

self-concepts (Blood, 1985; Bryan, 1984; Champion et al., 1984). These results are consistent, however, with the findings of Ferrari et al., (1984) who reported a Piers-Harris mean percentile score of 63.0 for their sample of 10 children with TS.

One possible explanation for this finding was that the subjects were distorting their responses to this measure in a socially desirable fashion. Piers (1984) suggests that protocols with a total raw score of 70 or above be interpreted with caution. This was the case in eight (36.4%) of the TS subjects in this sample. Thus the contribution of social desirability to the mean score cannot be dismissed.

Another contributing factor to the unexpectedly high mean T-scores may be that a proportion of subjects showed "a lack of critical self-evaluation" (Piers, 1984; p. 33). This would be consistent with the Rorschach data discussed above. The apparent contradiction between the normal range of Piers-Harris self-concept scores and the low Egocentricity ratio may actually reflect a low level of self-focus in TS subjects and therefore a lack of critical self-evaluation. Certainly this would be consistent with the Rorschach results that indicated that the TS subjects in general do not have negative or pessimistic self-images.

F. Child Behavior Checklist

Analysis of the data from the CBCL indicated that this measure did not reliably differentiate between Low TS and High TS subjects, nor did it reliably differentiate between TS subjects and the normative population. Some significant differences between TS subjects and the normative sample in specific subscales and on the Total Score were found, however.

The finding that the CBCL did not differentiate between Low TS and High TS subgroups is consistent with the recent findings of Sverd et al. (1988). These researchers found that clinical diagnosis of behavioral disorders and attentional deficits did not differentiate between boys with a low number of TS symptoms and boys with a high number of TS symptoms.

The differences between the TS subjects and the normative sample will be interpreted with some caution, since the overall analysis of this data did not differentiate significantly between the two groups. It should be noted that the differences did approach significance (p < .06) on those variables that did not achieve statistical significance.

As expected, the Total Behavior Problem scores differentiated between all TS subjects and the normative sample, and between male TS subjects and the normal sample. Thus, the TS parents as a group rated their children with TS as showing more behavior problems than most children would be expected to show. This data is consistent with previous findings for children with TS (Champion et al., 1988; Comings & Comings, 1984, 1985; Stefl, 1984; Sverd et al., 1988; Wilson et al. 1982).

The subscales that significantly differentiated between TS subjects and the normative sample (p < .05) were: Internalizing, Somatic Complaints, Schizoid, and Aggressive. The subscales that also differentiated between TS male subjects and the normative sample (p < .05) were: Uncommunicative, Obsessive Compulsive, and Hyperactive. The finding that the Obsessive Compulsive and Hyperactive scales were elevated is consistent with the findings reported by other researchers (Champion et al., 1988; Stefl, 1984; Wilson et al., 1982). The finding that the Aggressive and Somatic Complaint subscales were elevated was expected, but

somewhat inconsistent with the findings from the Rorschach data. This apparent contradiction in results may best be understood in terms of the level at which the Rorschach and the CBCL measure these constructs. The Ag Special Score on the Rorschach reflects an aggressive or hostile interpersonal orientation, whereas the Aggressive subscale on the CBCL is a reflection of parents' reports of the subjects' aggressive behaviors. Aggression against objects and animals is included in the Aggression subscale. This subscale includes such items as showing off, screaming, swearing, demanding attention, as well as items that reflect interpersonal aggressiveness (Achenbach & Edelbrock, 1983). It is possible that the aggressive behavior reported by the TS subjects' parents does not reflect a hostile interpersonal orientation so much as it reflects aggressive behavior against objects. Similarly, the Somatic Complaints subscale on the CBCL reflects parents' reports of somatic behaviors and includes items such as nausea, pains, headaches, accident proneness, worrying and slow moving (Achenbach & Edelbrock, 1983), whereas the Anatomy and X-ray content codes on the Rorschach, on the other hand, reflects the subjects' inner concern about their body and body image (Exner, 1986).

The Schizoid subscale includes such items as "feels guilty", "too neat", "needs to be perfect", "worries", "fears school", and "clings to adults" as well as "hears things" (Achenbach & Edelbrock, 1983). Thus this scale reflects social avoidance, as well as age-inappropriate behavior. These data are consistent with a depression on the Total Social Competence scale which reflects the subjects' degree of involvement in sports, hobbies, organizations and peers as well as academic achievement and performance of household chores. The finding that the TS subjects'

performance in these areas tends to be somewhat lower than the norm is consistent with the elevated Schizoid subscale, and with the Rorschach findings that have the TS subjects as a group showing a tendency towards interpersonal avoidance. These data suggest that, although many of the TS subjects were reported to be involved in social activities, they may not be as adept at such activities as their peers.

The above finding is also consistent with parents' reports that the TS subjects in this sample tend to have poor academic achievement and with other researchers' findings of low academic achievement in children with TS (Hagin et al., 1982; Incagnoli & Kane, 1983).

As expected, the TS subjects were reported to have more behavioral problems than would be expected in a normal sample. The trends were consistent with the findings of other researches, as discussed above and elsewhere in this paper. These findings also indicate that children with TS and children with epilepsy share some behavioral problems, including depressed social competency, as measured by the CBCL (Ferrari et al., 1983). These results, however, must be interpreted with some caution, as the results of the overall analysis did not achieve statistical significance.

Additional Analyses

Results of the post hoc analysis suggested that TSGS Social Functioning domain was associated with the CBCL Total Score and with the Externalizing, Delinquency, and Aggression subscores of the CBCL. This is not surprising, given that the TSGS Social Functioning domain and the CBCL are both used to measure level of social functioning. The finding that CBCL scores were not

associated with the TSGS TS symptomatology domain strengthens the above discussed finding that severity of TS symptoms are not directly associated with behavioral problems.

G. Clinical Implications

The results of this study indicate that children with TS and their families show more problems in living than would be expected in a normative sample. The degree of these problems were not associated with severity of TS symptoms. Therefore, the critical factor in predicting and treating these problems in living would be diagnosis of TS, not severity of TS symptomatology.

The results of this study have several implications for working with TS children and their families. First, data suggest the families with a TS member tend to perceive themselves to be less cohesive and less flexible and to feel less satisfied with their families than do nonclinical families. On the other hand, these families have developed some appropriate strategies for coping with having a TS member. Therefore, family therapy strategies might best be addressed to helping families who want to do so to develop more flexibility. Since these families do not appear to fit the model of the psychosomatic families in terms of enmeshment or overly elevated cohesion, it does not appear that the TS is serving a function of keeping the family unit together. Therefore strategies that increase cohesion would not be inappropriate in most TS families. These families, as a group, also are adept at seeking out and accepting help from outside the family. Thus, they may be open to family therapy, if needed, and might benefit from learning coping strategies for dealing with problems within the family. This might serve to increase family satisfaction levels.

One of the problems that families with a TS member are likely to face is having a child with more than the usual number of behavioral problems. The results of this study suggest that children with TS are more likely to experience academic problems, to be hyperactive, to behave aggressively, to be obsessive-compulsive, to have a number of somatic complaints and to be less socially skilled than their peers. Wilson et al. (1982) suggest that such behavior problems are associated with the subtle neurological impairments that are often found in movement disorders such as TS. It is not likely, therefore, that these problems will entirely disappear with even the best family and/or individual therapy. Instead, families with a TS member may need help in acknowledging that these problems may continue to be a source of some stress. One strategy that might be useful would be to define their child's behavioral problems as part of the overall TS picture. In this way, these problems can be reframed and understood to be much less under the control of either the child with TS or his or her parents that would be expected in a non-TS child. Thus, the behavioral problems could be removed, at least to some extent, from the control-oriented conflicts that Ferrari et al. (1983) suggest are characteristic of TS families. Learning to cope effectively with the problems and the stress would be more appropriate than trying to eliminate the behavioral problems. Research on assessment of families with epileptic children suggest that expecting the child to behave as normally as possible increases the child's academic social success (Hoare, 1984). Thus, a combination of accepting that some behavior problems are part of TS, coping effectively with the problems, and maintaining normal activities (such as sports) might be most effective in minimizing the problems and maximizing a good adjustment for the child with TS and his/her family.

The data from this study suggest that the child or adolescent with TS is likely to have some problems with good decision-making, because their decisions will probably be based on inaccurate perceptions of their world. In addition, their attempts to steer clear of emotionally-arousing stimuli may leave them somewhat uncommunicative and constricted in their expression of their feelings. This may be associated with poorly developed skills for self-expression and with a concommitant tendency to have temper tantrums or to otherwise express themselves inappropriately when they do react to their world. Frequently, helping children to be more expressive of their feelings is a goal of individual therapy. Such a goal might need to be approached more cautiously in children with TS, as their emotional constriction may serve an important purpose. For the neurologically less inhibited, more reactive child with TS constricting one's response to the world may allow him or her to function more effectively in school, with peers, and in the family. Just as the suppression of tics tends to lead to intensified tiquing at a later date, however, emotional constriction often leads to later explosiveness. A strategy in individual therapy may be to help the child or adolescent with TS learn to "let off steam" in appropriate ways. Sports activities, the therapy session itself, letting off steam to an understanding significant adult, and fostering communication about more minor daily stresses, frustrations and joys may be arenas for discharging pent-up emotional energy.

When a child's perceptual accuracy is impaired, it may appear that therapy should focus on improving that accuracy. Again, caution may be in order. The TS child's perceptual inaccuracies may serve the functioning of cushioning his or her self-esteem. Perceptual inaccuracies that hamper

decision-making, or that lead to negative social interactions (for example feeling unrealistically rejected by peers) may of course need to be dealt with. Pressing a child (or more likely adolescent) with TS to become more critically self-evaluative may, on the other hand, be counter-productive.

Another facet of the clinical implications relates to building on the strengths exhibited by this group of children and adolescents and their families. Parents who have learned to accept outside help can be encouraged to use their skills in appropriately advocating for their children in school and in the community. The lack of negative or pessimistic self-images demonstrated by the TS subjects in the sample can provide a foundation on which to build a positive self-image. In a similar vein, although the TS subjects as a group were not reported to be as socially adept as their peers, they also did not evidence as strong a sense of social isolation, nor as intensely avoidant or hostile interpersonal orientations as might have been expected. It is therefore likely that some improvements in social skills could be fostered, perhaps in group therapy or structured group activities.

The results of this study provide some therapeutic clues in approaching the problems presented by the child or adolescent with TS and their families. Obviously, these approaches will not in any way "cure" the condition, nor are they likely to have s significant effect on the severity of the TS symptoms. Rather, the suggestions above are intended to address the associated behavioral, social, familial and intrapersonal features of TS.

H. Future Research

The results of this study have suggested some answers to some of the questions about the nature of the associated features of TS. Some other questions have not been answered, and new ones have been raised. Future research can be designed to address these questions.

A major question not fully answered in this study is whether or not children and adolescents with TS are different from children with LD, from children with other neorologically-based disorders, such as epilepsy, or from children with medical disorders such as diabetes. Some similarities between children with epilepsy and children with TS, and between their families have been found by other researchers, as discussed above. A study that directly compares TS subjects and subjects from one or more of the above groups would be needed to assess more specific similarities and differences.

A question raised by the current study is how having an adolescent with TS affects family cohesiveness, flexibility, and satisfaction. To address this question, it would be necessary to study families with adolescents with TS and families with younger children with TS in large enough numbers that valid comparisons could be made between these two groups.

The measures in this study do not provide any information about the perceived level of stress and/or number and degree of stressful events for the family or the child with TS. It would be useful to assess level of individual and familiar stress and perhaps to compare that to severity of TS symptomotology, and to degree of of associated behavioral and school problems.

One possible source of bias in this study is that all of the subjects and their families have contact with the TS clinic, and many have been involved with the Manitoba Society for Tourette Syndrome (MSTS). The degree of involvement has varied from initial assessment at the TS clinic to individual group and/or family therapy, school consultations, and peer support provided by the MSTS. The small sample size and the variability in involvement made it impossible to parcel out the impact that the various types of therapy, consultation and/or peer support may have had on the subject in this study. A longitudinal study with a larger sample would be useful in determining the impact of various interventions, including those recommended in the previous section, on TS patients and their families. A longitudinal study, with the same measures taken at intervals, would also be useful in determining the stability of the findings of the current study.

The data from the post hoc analyses also point to possible directions for future research. The relationship between the Social Function domain of the TSGS and scales on the CBCL could be further explored using a larger sample size. Other measures related to the CBCL, such as the Teacher's Report Form Scale, the Direct Observation Form, and the Youth Self-Report (Achenbach & Edelbrock, 1983) might also be used to clarify the relationship between behavioral problems and the TSGS Social Functioning domain.

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Appendix A

INFORMED CONSENT AGREEMENT

Relationship of Severity of Symptoms in Gilles de la Tourette's
Syndrome to Individual and Family Functioning[1]
<u>PARAPHRASE AND CONSENT FORM</u>

1. I understand that this is a study to find out how I am getting
    along, how well I get along with other people, how my parents think
    I am getting along, and how my family gets along. This study will
    also look at how severe my Tourette symptoms are and try to find out
    whether or not the severity of my symptoms makes a difference to how
    well I get along, or how well my family gets along. All testing
    will be done by a qualified, experienced examiner. I understand
    that I will take the Rorschach Inkblot Test, which is a personality
    test, and it will take about one hour to complete. I will also fill
    out a paper and pencil questionnnaire about my family, and a
    questionnaire about how I feel about myself. My parents will fill
    out one questionnaire about me, four about our family and one about
    my Tourette symptoms. They will fill out one of these forms twice
    -- once as they see our family now, and once as they think other
    families would fill it out. It will take them about three to four
    hours to complete the forms.

2. If information about me is taken only as part of this study, I
    understand that the information will not be seen by my therapist (if
    I have one) unless my parent(s) and I ask that my therapist see it.

3. I have the right to receive feedback about the information on me,
    even if the information is taken only as part of this study.
    Feedback will be given by a qualified, experienced professional or
    professional in training.

4. If the information is taken as part of a clinical assessment of my
    Tourette symptoms, I still have the right to receive feedback, as
    above.

5. I have the right to withdraw from the study at any time. A decision
    to do so will not affect in any way my standing at the Tourette
    Clinic, nor will it affect the type and quality of services I
    receive.

6. Information about me that is taken for this study will be kept in a
    locked filing cabinet away from clinical information. My name will
    not be on the test forms, nor will it be entered onto a computer,
    nor will it be published in any way. My test forms will be
    identified by a code number only, and the code number/name sheets
    will be stored in a locked filing cabinet away from forms used in
    the study and away from the clinical files. The code number/name
    sheets will be kept so that the researchers can keep track of who
    has completed the study, and so that, should I later request
    information about me, it will be available.

7. I have read all of the above, or it has been read to me, and I have
    had a chance to ask any questions I might have and I understand I
    can talk to the researcher if I think of any more questions. I
    agree to participate in this study.

_____	_____
Witness	Name

_____
Date

     I have read all of the above, or it has been read to me and I have had a chance to ask any questions I might have. I understand I can contact the researcher if I have any further questions or concerns. I agree to participate in this study and to allow my child to participate in this study.

_____	_____
Witness	Name

_____
Date

Appendix B

HISTORY AND INFORMATION FORM

HISTORY AND INFORMATION

NAME: _____
     Last name   Given name(s)

DATE OF BIRTH: _____ AGE: _____ SEX: _____
                Year   Month   Day

LANGUAGE SPOKEN IN OUR HOUSE IS:
(Please indicate)

_____

ORIGIN (Ethnic Background) (Optional):
(Please indicate)

_____

RELIGION (Optional):
(Please indicate)

_____

DOES THE PERSON WITH TOURETTE SYNDROME EXPERIENCE:

1. Motor tics   ____Yes   ____No

    Part(s) of the body involved:

    _____

    _____

    Please describe tics:

    _____

    _____

    _____

2. Phonic tics   ____Yes   ____No

    Please describe:

    _____

2. Phonic tics (continued)

_____

_____

PLEASE DESCRIBE ANY OTHER SYMPTOMS WHICH YOU FEEL RELATE TO TOURETTE SYNDROME:

_____

_____

_____

DO ANY OTHER FAMILY MEMBERS (IMMEDIATE AND/OR EXTENDED) HAVE TICS?

____Yes   ____No   Relationship to patient: _____

_____

_____

_____

HAVE ANY OTHER FAMILY MEMBERS (IMMEDIATE AND/OR EXTENDED) EVER BEEN DIAGNOSED AS:

1. Tourette Syndrome   ____Yes   ____No

Relationship to patient: _____

_____

_____

_____

Comment: _____

_____

_____

2. Chronic Tic   ____Yes   ____No

Relationship to patient: _____

_____

_____

_____

2. Chronic Tic (continued)

   Comment: _____

   _____

   _____

3. Transient Tic of Childhood   ____Yes   ____No

   Relationship to patient: _____

   _____

   _____

   _____

   Comment: _____

   _____

   _____

4. Obsessive-Compulsive Disorder   ____Yes   ____No

   Relationship to patient: _____

   _____

   _____

   _____

   Comment: _____

   _____

   _____

HOW OFTEN HAS EACH OF THE FOLLOWING BEEN A PROBLEM FOR THE PERSON WITH TOURETTE?

	ALMOST NEVER	RARELY	SOMETIMES	OFTEN	ALMOST ALWAYS
a) Extreme temper or temper tantrums	1	2	3	4	5
b) Obsessive-compulsive behaviour	1	2	3	4	5
c) Hyperactive behaviour	1	2	3	4	5
d) Self-abusive behaviour	1	2	3	4	5

	ALMOST NEVER	RARELY	SOMETIMES	OFTEN	ALMOST ALWAYS
e) Extreme anxiety	1	2	3	4	5
f) Extreme mood swings	1	2	3	4	5
g) Aggressive behaviour	1	2	3	4	5
h) Coprolalia (obscene words)	1	2	3	4	5
i) Running away from home	1	2	3	4	5
j) Lying and/or stealing	1	2	3	4	5

HOW OFTEN HAS EACH OF THE FOLLOWING BEEN A PROBLEM FOR THE PERSON WITH TOURETTE?

	ALMOST NEVER	RARELY	SOMETIMES	OFTEN	ALMOST ALWAYS
a) Problems getting to sleep	1	2	3	4	5
b) Problems staying awake	1	2	3	4	5
c) Sleepwalking	1	2	3	4	5
d) Bad dreams or night terrors	1	2	3	4	5
e) Bedwetting	1	2	3	4	5
f) Loss of bladder control during waking hours	1	2	3	4	5

BIRTH AND DEVELOPMENT HISTORY

MOTHER'S AGE AT BIRTH OF PATIENT: _____

PREGNANCY (insert appropriate number for each parent): _____ _____

Mother   Father

0 Unknown	2 Accidental	4 Desired, accidental
1 Desired	3 Undesired	5 Undesired, accidental

6 Adopted, Agency
7 Adopted, Non-agency

MOTHER'S ILLNESSES, INJURIES, FEVERS, MISCARRIAGES, ET CETERA, DURING PREGNANCIES.
(e.g., German measles the first trimester)

Occurrence	Trimester 1	2	3

DID ANY OF THESE OR OTHER ILLNESSES HAPPEN DURING THE PREGNANCY?

1____Overweight (more than 20 pound gain in weight)

2____Water or swelling in the ankles and legs

3____High blood pressure

4____Diabetes (high sugar in the blood or in the urine)

5____Kidney disease

6____Bladder disease

7____Low iron or not enough red blood cells

8____Tuberculosis

9____Vomiting beyond third month of pregnancy

10____Bleeding from below at any time during pregnancy

11____Surgery that used gas to put you to sleep

12____Other _____

WERE THERE ANY DRUGS OR MEDICATIONS TAKEN DURING YOUR PREGNANCY? PLEASE CHECK BELOW AND ADD ANY YOU MAY HAVE TAKEN THAT ARE NOT ON THE LIST.

1____Iron supplement

2____Pills that make more urine and reduce swelling

3____Vitamins

4____Reducing drug to lose weight

5____Pills to sleep better

6____Tranquilizers (pills to calm you down)

7____Other _____

IS THERE ON EITHER SIDE OF THE FAMILY A HISTORY OF:

	MOTHER'S SIDE	FATHER'S SIDE
1____Diabetes (sugar in urine)	_____	_____
2____Tuberculosis	_____	_____

	MOTHER'S SIDE	FATHER'S SIDE
3____Epilepsy (seizures)	_____	_____
4____High blood pressure	_____	_____
5____Other neurological disorders	_____	_____
6____Other (specify)_____	_____	_____

WHAT WAS THE LENGTH OF THE PREGNANCY?

1____Less than 28 weeks (7 months)

2____28 weeks (7 months)

3____32 weeks (8 months)

4____36 weeks (9 months)

5____40 weeks (10 months)

DELIVERY:

0____Unknown

1____Spontaneous

2____Cesarean Section

3____Breech birth

4____Forceps   COMMENT: _____

5____Other (specify) _____

ANAESTHESIA:

0____Unknown

1____None

2____Spinal

3____Twilight

4____Gas, Ether, Other _____

DID YOU HAVE ANY PROBLEMS WITH THE PATIENT'S BIRTH SUCH AS:

1____Labour stopped or was irregular

2____Bad position of baby

3____Other (specify) _____

WEIGHT OF PATIENT AT BIRTH: _____pounds _____ounces

WAS PATIENT SEEN BY MOTHER DURING FIRST 24 HOURS AFTER BIRTH? ____Yes ____No

DID THE PATIENT HAVE ANY PROBLEMS DURING BIRTH SUCH AS:

1____Trouble in breathing

2____Birth injury (hurt by the delivery)

3____Too much bleeding

4____Infection

5____Heart stopped

6____Too sleepy

WAS MOTHER DEPRESSED WITHIN FIRST THREE WEEKS POST-PARTUM? ____Yes ____No

COMMENT:_____

FEEDING:

Poor sucking or eating? ____Yes ____No   ____Breast ____Bottle

Colic: ____ Lasted ____months. Weaned to cup at ____months.

DEVELOPMENT AND TRAINING:

Sat without support ____months.

Crawled ____months.

Walked alone ____months.

Made sentences ____months.

Self-feeding ____months.

Self-dressing _____ months.

Started bowel training _____ months. Completed at _____ months.

Did a struggle develop with bowel training? _____ Yes _____ No

Nature of struggle: _____

When was night bladder training completed? _____ Years _____ Months

MEDICAL HISTORY

SIGNIFICANT CHILDHOOD DISEASES:

Disease	Age	Accompanied by high fever and/or coma; other comments
1. _____	____	_____
2. _____	____	_____
3. _____	____	_____
4. Encephalitis	____	_____
5. Undiagnosed high fever	____	_____

SIGNIFICANT SURGICAL PROCEDURES:

Age	Part of Body	Patient's Reaction	Parent's Reaction
____	_____	_____	_____
____	_____	_____	_____
____	_____	_____	_____

SIGNIFICANT ACCIDENTS:

Age	Event	Patient's Reaction	Parent's Reaction
____	_____	_____	_____
____	_____	_____	_____
____	_____	_____	_____

ALLERGIES:

Please describe allergy symptoms: _____

_____

Insert check(s) in appropriate box(es):

	Pre 2	2 - 4	4 - 6	Past 6
Mild				
Moderate				
Severe				

Is patient now on antihistamines? ____Yes ____No

If so, what kind? _____

Has patient ever been on antihistamines? ____Yes ____No

If yes, what kind? _____

Is the patient hyperactive? ____Yes ____No

If no, has he/she ever been hyperactive? ____Yes ____No

If yes: a) At home   ____Yes ____No

        b) At school   ____Yes ____No

        c) What time of day? _____

        d) Will he sit and play a game? ____Yes ____No

        e) Will he sit and watch TV? ____Yes ____No

Is the patient on any medication for hyperactivity? ____Yes ____No

What kind? _____

Has he/she ever been on medication for hyperactivity? ____Yes ____No

What kind? _____

Comment: _____

_____

EDUCATION

School: _____

Address: _____

School Division: _____ Phone #: _____ Grade: _____

Principal: _____ Teacher: _____

Counsellor: _____

Did he/she attend nursery school? _____ How long? _____

At what age did he/she attend Kindergarten? _____

Did he/she like school? _____ Does he/she like his teacher? _____

Are any school subjects, including physical education, difficult for him/her?

_____

Has he/she ever failed or skipped a grade? _____ Which ones? _____

What are his/her best subjects? _____

_____

Have you discussed his/her problems with his/her teacher? _____

Has the patient been in any Special Education classes (e.g., Resource Room, Precision Learning Centre, MBI class, LD class, et cetera?) _____ If yes, give dates and type of placement:

_____

_____

_____

HAS YOUR CHILD EVER BEEN EVALUATED BY:

Yes/No If yes, specify when, where, clinician's name and results if assessed.

_____ Neurologist _____

_____

_____ Occupational Therapist _____

_____

_____ Physiotherapist _____

_____

_____ Psychologist _____

_____

_____ Reading Clinician _____

_____

_____ School Nurse _____

_____

_____ Social Worker _____

_____

_____ Speech & Hearing Clinician _____

_____

What was the reason for the assessment?

_____

_____

What were the results of the assessment?

_____

_____

When was the assessment done? _____

By whom? _____

Agency: _____

HOW DOES THE TEACHER DESCRIBE THE PATIENT'S BEHAVIOUR IN SCHOOL?

_____ Poor work habits

_____ Does not pay attention

_____ Does not use time and materials effectively

_____ Does not listen

_____ Written work careless

_____ Does not discipline himself

_____ Other _____

WHAT KIND OF MARKS DOES/DID THE PATIENT RECEIVE?

_____A's   _____C's & D's   _____Inconsistent grades

_____A's & B's   _____D's   (Describe): _____

_____B's & C's   _____D's & F's   _____

_____C's   _____F's   _____

SOCIAL

IS THE PATIENT INVOLVED IN ANY OF THE FOLLOWING?

_____Yes   _____No   Organized sports in the community

_____Yes   _____No   Organized sports at school

_____Yes   _____No   Church groups

_____Yes   _____No   YMCA, YWCA, YMHA

_____Yes   _____No   Other community clubs or organizations
                       (Describe): _____

_____Yes   _____No   Special interests or hobbies but not a member of a group
                       (Describe): _____

DOES THE PATIENT HAVE:

_____1. No friends

_____2. Some friends, but no close friends

_____3. Some friends, including one or two close friends

_____4. More than three close friends

ARE THE FRIENDS OF THE PATIENT MOSTLY:

_____1. Older

_____2. Younger

_____3. Same age

ARE FRIENDS MOSTLY:

_____1. Same sex

_____2. Opposite sex

ARE THERE ANY SPECIAL CONCERNS ABOUT SEXUALITY?

_____1. Yes (Describe): _____

_____2. No

WHAT ARE THE PATIENT'S STRENGTHS?

1. _____

2. _____

3. _____

4. _____

THE MOST IMPORTANT RELATIONSHIPS TO THE PATIENT ARE:

Name of Person   Relationship to Patient
                                        (ie friend, brother, mother,
                                            father, et cetera)

1. _____   _____

2. _____   _____

3. _____   _____

FAMILY

What effect has the patient's symptoms had on the family? (Has it been hard
to cope with the symptoms? Are family members ever embarrassed by the symptoms?
Have disagreements about how to cope caused stress in the family? et cetera)

_____

_____

_____

_____

_____

_____

Have there been any changes in the family unit (due to birth, death, separation, divorce, remarriage) in the last two years? _____Yes _____ No

Please describe:

_____

_____

_____

_____

_____

_____

How did this affect the patient?

_____

_____

_____

_____

Did the symptoms:

_____Remain the same

_____Get worse

_____Improve

Does the patient appear to have

_____more

_____fewer

_____the same

symptoms when stressed (by events at home, school, work, et cetera).

PARENTS

MOTHER'S Name: _____

     Birthdate: _____

     Occupation: _____

     Education Level: _____

FATHER'S Name: _____

     Birthdate: _____

     Occupation: _____

     Education Level: _____

IF PARENTS IN HOME ARE NOT NATURAL PARENTS, PLEASE SPECIFY RELATIONSHIP TO TS PATIENT:

Mother: Adoptive _____   Father: Adoptive _____

       Step   _____   Step   _____

SIBLINGS

Please indicate any problems in the following areas:

NAME	BIRTHDATE	EMOTIONAL	ACADEMIC	SOCIAL	MEDICAL

Please specify the nature of the above problem(s):

_____

_____

_____

_____

Do you have any comments, or is there other information that you think is important for us to know? (Continue on back of sheet if necessary).

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

Appendix C

TOURETTE SYNDROME GLOBAL SCALE

TOURETTE SYNDROME GLOBAL SCALE

The Tourette Syndrome Global Scale will provide information about the severity of your child's tics, and about how well he/she is doing in terms of social functioning. The first two pages are the scale itself. Pages 3 to 6 provide guidelines and examples for filling out the scale.

If your child is 16 or under, fill out all sections except "Work and Occupation Problems". If your child is over 16 and still in school, fill out all sections except "Work and Occupation Problems". If your child is 17 or older and not in school, fill out all scales except "School and Learning Problems". For children 17 and older, who are not in school, fill out "Work and Occupational Problems" even if he or she is not currently employed.

Please fill out this form to reflect your child's overall level of functioning over the past few months.

Thank you for your time and effort in completing this scale.

TOURETTE SYNDROME GLOBAL SCALE (TSGS)

NAME: _____ DATE: _____ RATER: _____

CODE FOR FREQUENCY

1 = 1 or less in 5 min

2 = 1 in 2-4.9 min

3 = from 1 in 1.9 min to 4 in 1 min

4 = 5 or more in 1 min

5 = virtually uncountable

	FREQUENCY (F)						DISRUPTION (D)						OFFICE USE ONLY
	None	Rarely	Occasionally	Frequently	Almost Always	Always		Camouflaged	Audible or Visible No Problem	Some Problem	Impaired Functioning	Cannot Function	
SIMPLE MOTOR (SM): Nonpurposeful, tics, jerks and/or movements	0	1	2	3	4	5		1	2	3	4	5	FXD = ____
COMPLEX MOTOR (CM): Purposeful, thoughtful actions (systematic actions), rituals, touching self, others or objects	0	1	2	3	4	5		1	2	3	4	5	FXD = ____
SIMPLE PHONIC (SP): Nonpurposeful noises, throat clearing, coughing	0	1	2	3	4	5		1	2	3	4	5	FXD = ____
COMPLEX PHONIC (CP): Purposeful, insults, coprolalia, words, distinguishable speech	0	1	2	3	4	5		1	2	3	4	5	FXD = ____

BEHAVIOUR (B) (Conduct)

0   No problem

5   Subtle problems normal peer, school, and family relations

10 Some problems, at least one relationship area impaired

15 Clear impairment in more than one area

20 Serious impairment, affects all areas

25 Unacceptable social behaviour, constant supervision

RATING _____

MOTOR RESTLESSNESS (MR)

0   Normal movement

5   Adventitial movements, visible no problem

10 Increased motor restlessness, clearly visible, some problem

15 Clear motor restlessness, moderate problem

20 Mostly in motion but occasionally stops, impaired functioning

25 Nonstop motion, clearly cannot function

RATING _____

SCHOOL AND LEARNING PROBLEMS

0   No problem

5   Low grades

10 Should be or in some special classes, or repeated

15 All special classes

20 Special school

25 Unable to remain in school, home bound

RATING _____

WORK AND OCCUPATION PROBLEMS

0   No problem

5   Stable job, some difficulty

10 Serious problems

15 Lost lots of jobs

20 Almost never employed

25 Unemployed

RATING _____

(See following pages for Rating Guidelines)

THE FOLLOWING ARE GUIDELINES FOR RATING THE FREQUENCY AND DISRUPTIVENESS OF PHONIC AND MOTOR TICS.

CODE FOR FREQUENCY:

1. 1 or less in 5 min

2. 1 in 2 to 4.9 min

3. 1 to 4 in 1 to 1.9 min

4. 5 or more in 1 min

5. Virtually uncountable

CODE FOR DISRUPTION:

1. Camouflaged: Some tics, but untrained person would not recognize (Example: tossing hair back).

2. Audible/visible no problem: Recognizable but does not interfere (Example: picking at hair, throat clearing).

3. Some problem: Significant problem but functioning continues (Example: interrupted speech, head-jerks, interruptions while reading).

4. Impaired function: Symptom definitely a problem (Example: prolonged complex movements, series of nonstop tics).

5. Cannot function: Cannot do anything when symptom is present.

DESCRIPTION OF MOTOR SYMPTOMS:

Simple Motor Tics:

Rapid, Darting, "Meaningless": Eyeblinking, grimacing, nose twitching, lip pouting, shoulder shrugs, arm jerks, head jerks, abdominal tensing, rapid kicks, finger movements, jaw snaps, tooth clicking, frowning, rapid jerking of any part of body

Complex Motor Tics:

Slower, "Purposeful": Hopping, clapping, touching objects or others or self, throwing, arranging, gyrating and bending, "dystonic" postures, biting mouth, lip, arm, headbanging, thrusting arms, striking out, picking scabs, writhing movements, rolling eyes to the ceiling, holding funny expressions, sticking out the tongue, kissing, pinching, writing over-and-over the same letter or word, pulling back on pencil while writing, tearing paper or books

Copropraxia:

"Giving the finger" - cursing through gestures

DESCRIPTION OF PHONIC SYMPTOMS:

<u>Simple Phonic Symptoms</u>: Fast, "Meaningless" Sounds

Whistling, coughing, sniffling, spitting, screeching, barking, grunting, gurgling, clacking, hawking, hissing, sucking, uh-uh, eeee, ah-uh, ah, and innumerable other sounds

<u>Complex Phonic Symptoms</u>: Language

Words, Phrases, Statements: Shut up, stop that, OK, I've got to, I'm going to better - right? Right. What makes me do this. How about it. Now you've seen it, all right, oh boy.

<u>Rituals</u>:

Counting rituals. Repeating a phrase until it is "just right".

<u>Speech Atypicalities</u>:

Unusual rhythms, tone, accents, intensity of speech.

<u>Coprolalia</u>:

Obscene and aggressive words and statements.

THE FOLLOWING ARE GUIDELINES FOR RATING SOCIAL FUNCTIONING.

I. **BEHAVIOUR**: Provocative, argumentative, poor frustration tolerance, temper fits (with three main areas of interaction; peers, school or authority figures and family relations)

    0: No problems, normal relationships
1-4: Somewhat more than normal behaviour problems
  5: Subtle problems, no particular relationship threatened
6-9: Strained relationships
 10: Visible problem, at least one relationship impaired
11-14: Degree of impairment (Example: If OK relationship in school and peers but not with family, 14)
 15: Clear impairment in more than one area
16-19: Degree of impairment
 20: Serious impairment affects all areas, occasional interactions
21-24: Degree and number of social interactions (Example: Older brother and patient have good relationship)
 25: Unacceptable social behaviour, no attempt at good social interaction. Cannot be trusted, constant supervision.

II. **MOTOR RESTLESSNESS**: Increased motor activity, more than normal movement for task

    0: Normal movement for task - good concentration
1-4: Something more than normal
  5: Adventitial, occasional, increased movement, mostly fine motor, visible, but no problem
6-9: More frequent but still no problem
 10: Increased motor restlessness, clearly visible (Example: e.g., shaking, fidgety, would be trouble at dinner table or movies), mild interference
11-14: Greater degree of interference
 15: Clear motor restlessness, fidgetting, hyperactive, some impairment (intervention)
16-19: Greater degree of impairment
 20: Mostly in motion but occasionally stops, impaired direction, difficulty with structure, functioning greatly impaired
21-24: Fewer stops, greater impairment
 25: Nonstop motion, impaired concentration, unable to sit still, always in motion, clearly cannot function

III. (A) SCHOOL AND LEARNING PROBLEMS:

    0: No problem, at grade level, doing at least average work
1-4: Degree of borderline grades (Example: 4 Cs)
  5: Low grades: Cs + Ds - not working up to potential
6-9: Degree of failing (Example: 2Fs might be an 8)
 10: Should be or is in some special classes, special teacher, learning laboratory, tutor, or repeated grade
11-14: Degree of special help (Example: Special class for 2 subjects might be 12)
 15: All special classes or repeated more than one grade

III.   (A) SCHOOL AND LEARNING PROBLEMS (continued)

16-19: Degree of learning (Example: If very little learning, 19)
   20: Special school
21-24: Having trouble in special school
   25: Unable to remain in school, home bound, unable to learn.

III.   (B) WORK AND OCCUPATIONAL PROBLEMS

    0: Have job, no problems.
  1-4: Occasional problem
    5: Has held down job for at least 6 months, some problems doing work,
       getting along with co-workers, or taking orders
  6-9: Shorter duration and/or degree of problems
   10: Poor functioning, changed jobs a few times in the past year.
       Serious problems (2 or 3 jobs)
11-14: Number of jobs or seriousness of problems
   15: Cannot hold a job for long, lost lots of jobs
16-19: Number of jobs, or seriousness of problems
   20: Almost never employed, sporatic employment, out of work 2-3
       months
21-24: Number of months out of work
   25: Unemployed - did not work for 6 or more months.

Appendix D

STRUCTURAL SUMMARY FOR THE EXNER COMPREHENSIVE SYSTEM

STRUCTURAL SUMMARY

R =   Zf =   ZSum =   P =   (2) =   Fr + rF =

Location Features

W =

D =

Dd =

S =

Determinants (Blends First)

Contents

H	=	Bl	=
(H)	=	Bt	=
Hd	=	Cg	=
(Hd)	=	Cl	=
A	=	Ex	=
(A)	=	Fi	=
Ad	=	Fd	=
(Ad)	=	Ge	=
Ab	=	Hh	=
Al	=	Ls	=
An	=	Na	=
Art	=	Sc	=
Ay	=	Sx	=
		Xy	=

Contents (Idiographic)

_____ =

_____ =

_____ =

_____ =

_____ =

_____ =

Single Determinants

M	=
FM	=
m	=
C	=
Cn	=
CF	=
FC	=
C'	=
C'F	=
FC'	=
T	=
TF	=
FT	=
V	=
VF	=
FV	=
Y	=
YF	=
FY	=
rF	=
Fr	=
FD	=
F	=

DQ

+ =

v/+ =

o =

v =

M Quality

+ =

o =

u =

− =

NO FORM =

Form Quality

FQx		FQf	
+	=	+	=
o	=	o	=
u	=	u	=
−	=	−	=
NO FORM =			

S-CONSTELLATION (Adult)

___ FV + VF + V + FD > 2

___ Col-Shd Bl > 0

___ 3r + (2)/R ≤ .30 or ≥ .45

___ MOR > 3

___ Zd > ±3.5

___ es > EA

___ CF + C > FC

___ X + % < .70

___ S > 3

___ P < 3 or > 8

___ Pure H < 2

___ R < 17

_____ TOTAL

Special Scorings

DV	=
INCOM	=
DR	=
FABCOM	=
ALOG	=
CONTAM	=
WSUM6	=
AG	=
CONFAB	=
CP	=
MOR	=
PER	=
PSV	=

RATIOS, PERCENTAGES, AND DERIVATIONS

ZSum-Zest =

Zd =

EB =   EA =

eb =   es =   } D =

(FM =   m =   T =   C' =   V =   Y =   )

a:p =   Adj D =

Ma:Mp =   Depi =

S-Con =   Sczi =

FC:CF + C =   W:M =

Pure C =   W:D =

Afr =   Isolate:R =

3r + (2)/R =   Ab + Art =

L =   An + Xy =

Blends:R =   H + (H):Hd + (Hd) =

X + % =   Pure H =

X − % =   (H) + (Hd):(A) + (Ad) =

F + % =   H + A:Hd + Ad =

Appendix E

PIERS-HARRIS CHILDREN'S SELF-CONCEPT SCALE

"THE WAY I FEEL ABOUT MYSELF"

The Piers-Harris Children's Self-Concept Scale

Ellen V. Piers, Ph.D. and Dale B. Harris, Ph.D.

Name: _____ Today's Date: _____

Age: _____ Sex (circle one): Girl Boy Grade: _____

School: _____ Teacher's Name (optional): _____

Directions: Here are a set of statements that tell how some people feel about themselves. Read each statement and decide whether or not it describes the way you feel about yourself. If it is *true or mostly true* for you, circle the word "yes" next to the statement. If it is *false or mostly false* for you, circle the word "no." Answer every question, even if some are hard to decide. Do not circle both "yes" and "no" for the same statement.

Remember that there are no right or wrong answers. Only you can tell us how you feel about yourself, so we hope you will mark the way you really feel inside.

TOTAL SCORE: Raw Score_____ Percentile_____ Stanine_____

CLUSTERS: I_____ II_____ III_____ IV_____ V_____ VI_____

W-180A

1. My classmates make fun of me ....yes no

2. I am a happy person ....yes no

3. It is hard for me to make friends ....yes no

4. I am often sad ....yes no

5. I am smart ....yes no

6. I am shy ....yes no

7. I get nervous when the teacher calls on me ....yes no

8. My looks bother me ....yes no

9. When I grow up, I will be an important person ....yes no

10. I get worried when we have tests in school ....yes no

11. I am unpopular ....yes no

12. I am well behaved in school ....yes no

13. It is usually my fault when something goes wrong .....yes no

14. I cause trouble to my family.............................yes no

15. I am strong ....yes no

16. I have good ideas ....yes no

17. I am an important member of my family ....yes no

18. I usually want my own way ....yes no

19. I am good at making things with my hands ....yes no

20. I give up easily ....yes no

21. I am good in my school work ....yes no

22. I do many bad things ....yes no

23. I can draw well ....yes no

24. I am good in music...yes no

25. I behave badly at home ....yes no

26. I am slow in finishing my school work ....yes no

27. I am an important member of my class ....yes no

28. I am nervous ....yes no

29. I have pretty eyes ....yes no

30. I can give a good report in front of the class ....yes no

31. In school I am a dreamer ....yes no

32. I pick on my brother(s) and sister(s) ....yes no

33. My friends like my ideas...................................yes no

34. I often get into trouble ....yes no

35. I am obedient at home ....yes no

36. I am lucky ....yes no

37. I worry a lot...yes no

38. My parents expect too much of me.....................yes no

39. I like being the way I am ....yes no

40. I feel left out of things ....yes no

41. I have nice hair ....yes   no

42. I often volunteer in school ....yes   no

43. I wish I were different ....yes   no

44. I sleep well at night ....yes   no

45. I hate school ....yes   no

46. I am among the last to be chosen for games ....yes   no

47. I am sick a lot ....yes   no

48. I am often mean to other people ....yes   no

49. My classmates in school think I have good ideas ......yes   no

50. I am unhappy ....yes   no

51. I have many friends ....yes   no

52. I am cheerful ....yes   no

53. I am dumb about most things ....yes   no

54. I am good-looking ....yes   no

55. I have lots of pep ....yes   no

56. I get into a lot of fights ....yes   no

57. I am popular with boys ....yes   no

58. People pick on me ....yes   no

59. My family is disappointed in me ....yes   no

60. I have a pleasant face ....yes   no

61. When I try to make something, everything seems to
    go wrong ....yes   no

62. I am picked on at home ....yes   no

63. I am a leader in games and sports ....yes   no

64. I am clumsy ....yes   no

65. In games and sports, I watch instead of play ....yes   no

66. I forget what I learn ....yes   no

67. I am easy to get along with ....yes   no

68. I lose my temper easily ....yes   no

69. I am popular with girls ....yes   no

70. I am a good reader ....yes   no

71. I would rather work alone than with a group ....yes   no

72. I like my brother (sister) ....yes   no

73. I have a good figure ....yes   no

74. I am often afraid ....yes   no

75. I am always dropping or breaking things ....yes   no

76. I can be trusted ....yes   no

77. I am different from other people ....yes   no

78. I think bad thoughts ....yes   no

79. I cry easily ....yes   no

80. I am a good person ....yes   no

Percentile	T	I Behavior	II Intellectual and School Status	III Physical Appearance and Attributes	IV Anxiety	V Popularity	VI Happiness and Satisfaction	Total Score	T	Percentile
	85								85	
	80							80	80	
								78-79		
								77		
	75							76	75	
99				14						99
98	70		17	13	14	12		75	70	96
								74		
								73		
								72		
93	65	16		12				71	65	93
								70		
			15		13		10	69		
						11		68		
84	60	15	15	11	12			67	60	84
								66		
								65		
								64		
								63		
69	55	14	14	10	11	10	9	61-62	55	69
								60		
			13	9	10	9	8	59		
								57-58		
								56		
								54-55		
50	50	13	12	8	9			53	50	50
								52		
		12	11	7	8	8	7	50-51		
31	45	11	10					49	45	31
						7		47-48		
		10	9	6	7		6	45-46		
								44		
								42-43		
16	40	9	8	5	6	6		40-41	40	16
		8				5		39		
			7	4	5			37-38		
			6					35-36		
								33-34		
		7				4	5	31-32		
7	35	6	5	3	4	3		30	35	7
		5						28-29		
						2	4	26-27		
								24-25		
2	30	4	4	2	3	1	3	23	30	2
								21-22		
1		3	3	1	2		2	20		1
	25	2	2	0		0	1	19	25	
								18		
								0-17		
	20		1		0		0		20	
	15								15	
		0-1	0							
	10								10	

Raw Scores

Figure 1
Sample Profile

9

Appendix F

CHILD BEHAVIOR CHECKLIST

CHILD BEHAVIOR CHECKLIST FOR AGES 4–16

For office use only
ID #

CHILD'S NAME

SEX ☐ Boy ☐ Girl

AGE

ETHNIC GROUP OR RACE

TODAY'S DATE

Mo._____ Day_____ Yr._____

CHILD'S BIRTHDATE

Mo._____ Day_____ Yr._____

GRADE IN SCHOOL

PARENT'S TYPE OF WORK (Please be specific—for example, auto mechanic, high school teacher, homemaker, laborer, lathe operator, shoe salesman, army sergeant, even if parent does not live with child.)

FATHER'S TYPE OF WORK:_____

MOTHER'S TYPE OF WORK:_____

THIS FORM FILLED OUT BY:

☐ Mother (name):_____

☐ Father (name):_____

☐ Other—name & relationship to child:_____

I. Please list the sports your child most likes to take part in. For example: swimming, baseball, skating, skate boarding, bike riding, fishing, etc.

☐ None

	Compared to other children of the same age, about how much time does he/she spend in each?				Compared to other children of the same age, how well does he/she do each one?			
	Don't Know	Less Than Average	Average	More Than Average	Don't Know	Below Average	Average	Above Average
a. _____	☐	☐	☐	☐	☐	☐	☐	☐
b. _____	☐	☐	☐	☐	☐	☐	☐	☐
c. _____	☐	☐	☐	☐	☐	☐	☐	☐

II. Please list your child's favorite hobbies, activities, and games, other than sports. For example: stamps, dolls, books, piano, crafts, singing, etc. (Do not include T.V.)

☐ None

	Compared to other children of the same age, about how much time does he/she spend in each?				Compared to other children of the same age, how well does he/she do each one?			
	Don't Know	Less Than Average	Average	More Than Average	Don't Know	Below Average	Average	Above Average
a. _____	☐	☐	☐	☐	☐	☐	☐	☐
b. _____	☐	☐	☐	☐	☐	☐	☐	☐
c. _____	☐	☐	☐	☐	☐	☐	☐	☐

III. Please list any organizations, clubs, teams, or groups your child belongs to.

☐ None

	Compared to other children of the same age, how active is he/she in each?			
	Don't Know	Less Active	Average	More Active
a. _____	☐	☐	☐	☐
b. _____	☐	☐	☐	☐
c. _____	☐	☐	☐	☐

IV. Please list any jobs or chores your child has. For example: paper route, babysitting, making bed, etc.

☐ None

	Compared to other children of the same age, how well does he/she carry them out?			
	Don't Know	Below Average	Average	Above Average
a. _____	☐	☐	☐	☐
b. _____	☐	☐	☐	☐
c. _____	☐	☐	☐	☐

3-81 Edition

V. **1.** About how many close friends does your child have? ☐ None ☐ 1 ☐ 2 or 3 ☐ 4 or more

2. About how many times a week does your child do things with them? ☐ less than 1 ☐ 1 or 2 ☐ 3 or more

---

VI. Compared to other children of his/her age, how well does your child:

	Worse	About the same	Better
a. Get along with his/her brothers & sisters?	☐	☐	☐
b. Get along with other children?	☐	☐	☐
c. Behave with his/her parents?	☐	☐	☐
d. Play and work by himself/herself?	☐	☐	☐

VII. **1.** Current school performance—for children aged 6 and older:

☐ Does not go to school

	Failing	Below average	Average	Above average
a. Reading or English	☐	☐	☐	☐
b. Writing	☐	☐	☐	☐
c. Arithmetic or Math	☐	☐	☐	☐
d. Spelling	☐	☐	☐	☐

Other academic subjects—for example: history, science, foreign language, geography.

	Failing	Below average	Average	Above average
e. _____	☐	☐	☐	☐
f. _____	☐	☐	☐	☐
g. _____	☐	☐	☐	☐

2. Is your child in a special class?

☐ No ☐ Yes—what kind?

---

3. Has your child ever repeated a grade?

☐ No ☐ Yes—grade and reason

---

4. Has your child had any academic or other problems in school?

☐ No ☐ Yes—please describe

When did these problems start?

Have these problems ended?

☐ No ☐ Yes—when?

VIII. Below is a list of items that describe children. For each item that describes your child **now or within the past 6 months**, please circle the **2** if the item is **very true or often true** of your child. Circle the **1** if the item is **somewhat or sometimes true** of your child. If the item is **not true** of your child, circle the **0**. Please answer all items as well as you can, even if some do not seem to apply to your child.

0 = Not True (as far as you know)   1 = Somewhat or Sometimes True   2 = Very True or Often True

0 1 2	1.	Acts too young for his/her age 16
0 1 2	2.	Allergy (describe): _____
		_____
0 1 2	3.	Argues a lot
0 1 2	4.	Asthma
0 1 2	5.	Behaves like opposite sex 20
0 1 2	6.	Bowel movements outside toilet
0 1 2	7.	Bragging, boasting
0 1 2	8.	Can't concentrate, can't pay attention for long
0 1 2	9.	Can't get his/her mind off certain thoughts; obsessions (describe): _____
0 1 2	10.	Can't sit still, restless, or hyperactive 25
0 1 2	11.	Clings to adults or too dependent
0 1 2	12.	Complains of loneliness
0 1 2	13.	Confused or seems to be in a fog
0 1 2	14.	Cries a lot
0 1 2	15.	Cruel to animals 30
0 1 2	16.	Cruelty, bullying, or meanness to others
0 1 2	17.	Day-dreams or gets lost in his/her thoughts
0 1 2	18.	Deliberately harms self or attempts suicide
0 1 2	19.	Demands a lot of attention
0 1 2	20.	Destroys his/her own things 35
0 1 2	21.	Destroys things belonging to his/her family or other children
0 1 2	22.	Disobedient at home
0 1 2	23.	Disobedient at school
0 1 2	24.	Doesn't eat well
0 1 2	25.	Doesn't get along with other children 40
0 1 2	26.	Doesn't seem to feel guilty after misbehaving
0 1 2	27.	Easily jealous
0 1 2	28.	Eats or drinks things that are not food (describe): _____
		_____
0 1 2	29.	Fears certain animals, situations, or places, other than school (describe): _____
		_____
0 1 2	30.	Fears going to school 45

0 1 2	31.	Fears he/she might think or do something bad
0 1 2	32.	Feels he/she has to be perfect
0 1 2	33.	Feels or complains that no one loves him/her
0 1 2	34.	Feels others are out to get him/her
0 1 2	35.	Feels worthless or inferior 50
0 1 2	36.	Gets hurt a lot, accident-prone
0 1 2	37.	Gets in many fights
0 1 2	38.	Gets teased a lot
0 1 2	39.	Hangs around with children who get in trouble
0 1 2	40.	Hears things that aren't there (describe): _____
		_____ 55
0 1 2	41.	Impulsive or acts without thinking
0 1 2	42.	Likes to be alone
0 1 2	43.	Lying or cheating
0 1 2	44.	Bites fingernails
0 1 2	45.	Nervous, highstrung, or tense 60
0 1 2	46.	Nervous movements or twitching (describe): _____
		_____
0 1 2	47.	Nightmares
0 1 2	48.	Not liked by other children
0 1 2	49.	Constipated, doesn't move bowels
0 1 2	50.	Too fearful or anxious 65
0 1 2	51.	Feels dizzy
0 1 2	52.	Feels too guilty
0 1 2	53.	Overeating
0 1 2	54.	Overtired
0 1 2	55.	Overweight 70
	56.	Physical problems without known medical cause:
0 1 2	a.	Aches or pains
0 1 2	b.	Headaches
0 1 2	c.	Nausea, feels sick
0 1 2	d.	Problems with eyes (describe):
0 1 2	e.	Rashes or other skin problems 75
0 1 2	f.	Stomachaches or cramps
0 1 2	g.	Vomiting, throwing up
0 1 2	h.	Other (describe): _____
		_____

Please see other side

0 1 2	57.	Physically attacks people		
0 1 2	58.	Picks nose, skin, or other parts of body (describe): _____		
				80
0 1 2	59.	Plays with own sex parts in public		16
0 1 2	60.	Plays with own sex parts too much		
0 1 2	61.	Poor school work		
0 1 2	62.	Poorly coordinated or clumsy		
0 1 2	63.	Prefers playing with older children		20
0 1 2	64.	Prefers playing with younger children		
0 1 2	65.	Refuses to talk		
0 1 2	66.	Repeats certain acts over and over; compulsions (describe): _____		
0 1 2	67.	Runs away from home		
0 1 2	68.	Screams a lot		25
0 1 2	69.	Secretive, keeps things to self		
0 1 2	70.	Sees things that aren't there (describe): _____		
0 1 2	71.	Self-conscious or easily embarrassed		
0 1 2	72.	Sets fires		
0 1 2	73.	Sexual problems (describe): _____		
				30
0 1 2	74.	Showing off or clowning		
0 1 2	75.	Shy or timid		
0 1 2	76.	Sleeps less than most children		
0 1 2	77.	Sleeps more than most children during day and/or night (describe): _____		
0 1 2	78.	Smears or plays with bowel movements		35
0 1 2	79.	Speech problem (describe): _____		
0 1 2	80.	Stares blankly		
0 1 2	81.	Steals at home		
0 1 2	82.	Steals outside the home		
0 1 2	83.	Stores up things he/she doesn't need (describe):		
				40

0 1 2	84.	Strange behavior (describe): _____		
0 1 2	85.	Strange ideas (describe): _____		
0 1 2	86.	Stubborn, sullen, or irritable		
0 1 2	87.	Sudden changes in mood or feelings		
0 1 2	88.	Sulks a lot		45
0 1 2	89.	Suspicious		
0 1 2	90.	Swearing or obscene language		
0 1 2	91.	Talks about killing self		
0 1 2	92.	Talks or walks in sleep (describe): _____		
0 1 2	93.	Talks too much		50
0 1 2	94.	Teases a lot		
0 1 2	95.	Temper tantrums or hot temper		
0 1 2	96.	Thinks about sex too much		
0 1 2	97.	Threatens people		
0 1 2	98.	Thumb-sucking		55
0 1 2	99.	Too concerned with neatness or cleanliness		
0 1 2	100.	Trouble sleeping (describe): _____		
0 1 2	101.	Truancy, skips school		
0 1 2	102.	Underactive, slow moving, or lacks energy		
0 1 2	103.	Unhappy, sad, or depressed		60
0 1 2	104.	Unusually loud		
0 1 2	105.	Uses alcohol or drugs (describe): _____		
0 1 2	106.	Vandalism		
0 1 2	107.	Wets self during the day		
0 1 2	108.	Wets the bed		65
0 1 2	109.	Whining		
0 1 2	110.	Wishes to be of opposite sex		
0 1 2	111.	Withdrawn, doesn't get involved with others		
0 1 2	112.	Worrying		
0 1 2	113.	Please write in any problems your child has that were not listed above:		
0 1 2		_____		70
0 1 2		_____		
0 1 2		_____		

PLEASE BE SURE YOU HAVE ANSWERED ALL ITEMS.   PAGE 4   **UNDERLINE ANY YOU ARE CONCERNED ABOUT.**

Appendix G

FAMILY ADAPTABILITY AND COHESION SCALES III

COMPLETED BY: ____MOTHER

                ____FATHER

                ____STEPMOTHER

                ____STEPFATHER

FAMILY QUESTIONNAIRE I

1	2	3	4	5
ALMOST NEVER	ONCE IN A WHILE	SOMETIMES	FREQUENTLY	ALMOST ALWAYS

DESCRIBE YOUR FAMILY NOW:

____ 1. Family members ask each other for help.

____ 2. In solving problems, the children's suggestions are followed.

____ 3. We approve of each other's friends.

____ 4. Children have a say in their discipline.

____ 5. We like to do things with just our immediate family.

____ 6. Different persons act as leaders in our family.

____ 7. Family members feel closer to other family members than to people outside the family.

____ 8. Our family changes its way of handling tasks.

____ 9. Family members like to spend free time with each other.

____ 10. Parent(s) and children discuss punishment together.

____ 11. Family members feel very close to each other.

____ 12. The children make the decisions in our family.

____ 13. When our family gets together for activities, everybody is present.

____ 14. Rules change in our family.

____ 15. We can easily think of things to do together as a family.

____ 16. We shift household responsibilities from person to person.

____ 17. Family members consult other family members on their decisions.

____ 18. It is hard to identify the leader(s) in our family.

____ 19. Family togetherness is very important.

____ 20. It is hard to tell who does which household chores.

David H. Olson, Joyce Portner, and Yoav Lavee.

Appendix H

FAMILY SATISFACTION

COMPLETED BY: ____MOTHER

____FATHER

____STEPMOTHER

____STEPFATHER

FAMILY QUESTIONNAIRE III

RESPONSE SCALE

1	2	3	4	5
DISSATISFIED	SOMEWHAT DISSATISFIED	GENERALLY SATISFIED	VERY SATISFIED	EXTREMELY SATISFIED

HOW SATISFIED ARE YOU:

____ 1. With how close you feel to the rest of your family?

____ 2. With your ability to say what you want in your family?

____ 3. With your family's ability to try new things?

____ 4. With how often parents make decision in your family?

____ 5. With how much mother and father argue with each other?

____ 6. With how fair the criticism is in your family?

____ 7. With the amount of time you spend with your family?

____ 8. With the way you talk together to solve family problems?

____ 9. With your freedom to be alone when you want to?

____ 10. With how strictly you stay with who does what chores in your family?

____ 11. With your family's acceptance of your friends?

____ 12. With how clear is it what your family expects of you?

____ 13. With how often you make decisions as a family, rather than individually?

____ 14. With the number of fun things your family does together?

David H. Olson and Marc Wilson (1982).

Appendix I

FAMILY CRISIS ORIENTED PERSONAL EVALUATION SCALE

COMPLETED BY: ____MOTHER

____FATHER

____STEPMOTHER

____STEPFATHER

FAMILY QUESTIONNAIRE II

WHEN WE FACE PROBLEMS OR DIFFICULTIES IN OUR FAMILY, WE RESPOND BY:	STRONGLY DISAGREE	MODERATELY DISAGREE	NEITHER AGREE NOR DISAGREE	MODERATELY AGREE	STRONGLY AGREE
1 Sharing our difficulties with relatives	1	2	3	4	5
2 Seeking encouragement and support from friends	1	2	3	4	5
3 Knowing we have the power to solve major problems	1	2	3	4	5
4 Seeking information and advice from persons in other families who have faced the same or similar problems	1	2	3	4	5
5 Seeking advice from relatives (grandparents, etc.)	1	2	3	4	5
6 Seeking assistance from community agencies and pro- grams designed to help families in our situation	1	2	3	4	5
7 Knowing that we have the strength within our own family to solve our problems	1	2	3	4	5
8 Receiving gifts and favors from neighbors (e.g., food)	1	2	3	4	5
9 Seeking information and advice from the family doctor	1	2	3	4	5
10 Asking neighbors for favors and assistance	1	2	3	4	5
11 Facing the problems "head-on" and trying to get solu- tion right away	1	2	3	4	5
12 Watching television	1	2	3	4	5
13 Showing that we are strong	1	2	3	4	5
14 Attending church services	1	2	3	4	5

WHEN WE FACE PROBLEMS OR DIFFICULTIES IN OUR FAMILY, WE RESPOND BY:	STRONGLY DISAGREE	MODERATELY DISAGREE	NEITHER AGREE NOR DISAGREE	MODERATELY AGREE	STRONGLY AGREE
15 Accepting stressful events as a fact of life	1	2	3	4	5
16 Sharing concerns with close friends	1	2	3	4	5
17 Knowing luck plays a big part in how well we are able to solve family problems	1	2	3	4	5
18 Exercising with friends to stay fit and reduce tension	1	2	3	4	5
19 Accepting that difficulties occur unexpectedly	1	2	3	4	5
20 Doing things with relatives (get-togethers, dinners, etc.)	1	2	3	4	5
21 Seeking professional counselling and help for family difficulties	1	2	3	4	5
22 Believing we can handle our own problems	1	2	3	4	5
23 Participating in church activities	1	2	3	4	5
24 Defining the family problem in a more positive way so that we do not become too discouraged	1	2	3	4	5
25 Asking relatives how they feel about problems we face	1	2	3	4	5
26 Feeling that no matter what we do to prepare, we will have difficulty handling problems	1	2	3	4	5
27 Seeking advice from a minister	1	2	3	4	5
28 Believing that if we wait long enough, the problem will go away	1	2	3	4	5
29 Sharing problems with neighbors	1	2	3	4	5
30 Having faith in God	1	2	3	4	5

Appendix J

FAMILY STRENGTHS

COMPLETED BY: ____MOTHER

                 ____FATHER

                 ____STEPMOTHER

                 ____STEPFATHER

FAMILY QUESTIONNAIRE IV

RESPONSE SCALE

1	2	3	4	5
STRONGLY DISAGREE	MODERATELY DISAGREE	NEITHER AGREE NOR DISAGREE	MODERATELY AGREE	STRONGLY AGREE

____ 1. We can express our feelings

____ 2. We tend to worry about many things.

____ 3. We really do trust and confide in each other.

____ 4. We have the same problems over and over.

____ 5. Family members feel loyal to the family.

____ 6. Accomplishing what we want to do seems difficult for us.

____ 7. We are critical of each other.

____ 8. We share similar values and beliefs as a family.

____ 9. Things work out well for us as a family.

____ 10. Family members respect one another.

____ 11. There are many conflicts in our family.

____ 12. We are proud of our family.