

**YOU SAY LEMON, I SAY LEMONADE: THE IMPACT  
OF ATTITUDE WHEN DEALING WITH DISORDER**

by

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A research project  
presented to the University of Waterloo  
in fulfillment of the  
research project requirement for the degree of  
Master of Applied Science  
in  
Psychology

Waterloo, Ontario, Canada, 1998

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## ***Abstract***

49 children (M age 12.6) with Tourette's syndrome (TS), their parents, and their non-affected siblings were interviewed to establish whether each viewed TS as unchangeable, uncontrollable, and beyond the responsibility of the individual with TS (i.e. whether they were entity theorists), or whether they saw their disorder as malleable, manageable, and as something the individual with TS should be accountable for (i.e. whether they were incremental theorists). It was found that TS'ers with an entity view suffered from lower self-esteem, felt more helpless, and had worse sibling interactions than did those who held incremental views towards TS. All family members agreed that TS'ers should be held less responsible for tics than for associated symptoms (anger/rage, impulsivity), but overall there was little agreement on attitude towards TS. Disorder severity did not predict attitude towards the disorder, and one's own attitude towards the disorder predicted fathers' relationship satisfaction, but not mothers'.

### *Acknowledgements*

I would like to thank the Tourette Syndrome Foundation of Canada and all of its members for supporting this project. In particular, the Toronto chapter membership actively solicited participants from its ranks for this endeavor, and the TSFC Main Office provided photocopying resources for questionnaires, and interview materials.

I would also like to thank Michael Ross, my advisor. Although Tourette's syndrome is far from his primary interest area, like a true scholar he found the ideas intriguing, graciously accepted me as a student, and encouraged my ideas. I would not be at the University of Waterloo never mind publishing this project was it not for him, and I am grateful.

## Dedication

***I write this for every person who has ever endured this quirky and insufferable, yet wondrous and marvelously complex disorder. My friends, Tourette's syndrome is disinhibiting in every sense: negative and positive, an asset and a liability. As with any quality, should you look for the advantages in it you will eventually come to realize that your worst enemy was never the “disorder”, but rather your own attitude, assumptions, and approach.***

## **Introduction**

Tourette's syndrome (TS) is now considered to be an inherited, neurological disorder (Robertson, 1989) characterized by involuntary tics/twitches (American Psychiatric Association (APA), 1994) which typically manifest between the ages of seven and ten (Comings, 1990). Georges Gilles de la Tourette, a French neurologist at Salpêtrière Hospital in Paris, first recognized the syndrome in 1885 (Kirshner, 1997). He described nine patients "who exhibited a combination of motor and phonic tics, spectacular and vulgar verbal outbursts, echolalia, obsessive thoughts, and repetitive behaviors" (Scalhill, Ort, & Hardin, 1993). At that time, "maladie de tic à Gilles de la Tourette", (also the name of the paper written jointly by Tourette and Charcot), was seen as a psychological degeneration, and was interpreted within psychoanalytic terms. Together, Tourette and Charcot believed that, should the tics be made to disappear, hysteria or even worse psychoses would result (Kirshner, 1997). TS all but disappeared from research literature in the 1900's, until Shapiro, Shapiro, Bruun and Sweet (1978) "rescued" Tourette's original article, and officially established TS as a disorder in 1978 (Kirshner, 1997). The Shapiros et al. were influential in steering views of etiology from the psychoanalytical to the neurobiological: they saw TS as an organic pathology of the central nervous system (Kirshner, 1997).

The most recent Diagnostic and Statistical Manual of the American Psychiatric Association (DSM-IV) defines TS as the presence of both motor and vocal tics (not necessarily concurrently) at least intermittently for more than one year. Tic-free periods cannot be longer than three consecutive months, and the disturbance must cause marked distress or impairment in important areas of functioning (APA, 1994). Chronic Tic Disorders require only motor or vocal tics (but not both), and Transient Tic Disorders do not last for

longer than 12 months (APA, 1994). A tic has been defined as “a sudden, rapid, recurrent, nonrhythmic motor movement or vocalization” by the DSM-IV. Tics are often precipitated by uncomfortable sensations, which the tic helps to relieve (Kurlan, 1989) and include such things as eyeblinking, mouth opening, head throwing, hopping, lip-licking, and arm extending in the motor domain, and throat clearing, barking, snorting, spitting, and humming in the vocal domain (Comings, 1990). While tics are ultimately irresistible, they can be suppressed for varying lengths of time (Bruun, Cohen, & Leckman, 1998). Considerable conscious effort is necessary to inhibit the tics, and an inevitable “rebound” effect follows the effort (Scalhill, Ort, & Hardin, 1993).

The prevalence of TS was once considered to be quite rare (.0005 percent of the population, Robertson, 1989, .0000046 percent, Robertson, 1994), however contemporary estimates are considerably higher. The current official stance of the Tourette Syndrome Foundation of Canada (TSFC) is that .05 percent of people have TS (Bruun et. al., 1998), however this continues to be a topic for debate. Comings, Himes, and Comings (1990) believe that TS may be as prevalent as 12 percent in special-education populations, and Freeman and Fast (1998) report that the most recent estimates of the disorder are closer to 3 percent of the population. Simply having some tics is common in children; in a community sample, Scalhill, Schwab-Stone, Leckman and Muller (1997) reported seeing tics in 12 percent of children between six and eleven year old. Confusions between different types of tic disorders may be partially responsible for varied prevalence estimates.

There is also considerable variability reported in the ratio of males to females, according to Freeman and Fast. The generally accepted figure is 4:1 (Freeman & Fast, 1998), but figures of 2:1 (Rutter and Hemming, 1970), 9.3:1 (Burd, Kerbeshian, Wikenheiser & Fisher, 1986)

and 1.7:1 (Apter et. al., in press) have been reported. The CATS (Canadian-American TS) Database, which includes over 900 cases from North America, UK, Norway, Australia, and Japan, reports that prevalence estimates vary by site from 19 to 1, to 2 to 1 (Freeman & Fast, 1998). No doubt these varying estimates depend upon, “at least in part, the definition, the type of ascertainment method and the type of epidemiological investigation undertaken” (Robertson, 1994). Consider that, in 1997 Van Ameringen, Mancini and Oakman argued that trichotillomania, the recurrent pulling out of one’s hair in an “unvoluntary tic” (Van Ameringen et. al., 1997), is responsive to the same treatments as TS, and could be fitted within the TS spectrum. As trichotillomania is much more prevalent in women than men (Cohen et. al., 1995), prevalence estimates for TS could be considerably altered if those with a comorbid diagnosis of TS and trichotillomania are included in the sample or not.

Evidence has accumulated to consider many other symptoms as being associated with TS as well. Attention-Deficit-Hyperactivity Disorder (ADHD) is considered by some to be the most common symptom in TS after tics (Comings, 1990). Prevalence rates vary from 50-60% (Comings & Comings I, 1987) to as high as 94% (Sverd, Curley, Jandorf & Volke, 1988). Obsessions and compulsions (O/C’s) exist in anywhere from 46-71% of TS patients (Comings & Comings IV, 1987, Frankel et. al., 1986, Pauls et. al., 1986), as well as other anxiety disorders (Coffey, Frazier, & Chen, 1992, Comings & Comings, 1987 III). Also implicated in TS are impulsivity, rage, self-injurious behaviours, depression, learning difficulties, conduct disorder, sleep problems, and substance abuse (Bruun et. al., 1998; Comings, 1990; Sverd et. al., 1988; Wand et. al., 1993). Comings suggests that these various disorders all share in common the problem of disinhibition (Comings, 1990).

The suggestion by Comings that these various disorders may be different manifestations of a similar underlying process begs for studies to determine whether a common etiology exists. Traditionally, TS is believed to be the result of an autosomal single dominant gene (for a review see Robertson, 1989), however it seems that the majority of the research supporting this claim looks at tics alone. Comings analyzed the pedigrees of over 200 TS families. He determined that members of the TS child's immediate and extended families exhibited *both* tics and behaviours associated with TS (ADHD, O/C's, etc.) significantly more so than in controls (Comings, 1990). He suggests that the gene is neither fully dominant (as not all parents had symptoms) nor fully recessive (as many parents and relatives of the parents did exhibit tics and/or associated behaviours), but rather semi-dominant, semi-recessive. Further, Comings proposed that most cases of TS are homozygous since a significant number of families demonstrate tics and/or associated behaviours in both parents and both their families (Comings, 1990).

The vast majority of articles on TS are neurobiological: Medline contained over 600 articles on TS in 1996 (Kirshner, 1997). Early work in EEG studies were summarized by Cohen, Bruun, and Leckman in 1988; they concluded that most researchers agree that minor, nonspecific abnormalities on the EEG record occur more frequently among TS'ers than within the normal population. EEG abnormalities in TS are considered rare (Weate et al., 1993), and neither EEG readings nor ER findings are considered helpful in the diagnosis or therapy of TS (Krumholz, 1983). Of latest interest are abnormalities in Contingent Negative Variations (CNV's) in TS. CNV's are slow, negative, brain potentials, which occur after a particular stimulus in anticipation of a second stimulus associated with the first. CNV's are thought to reflect the levels of the neurotransmitter dopamine in the central areas of the brain.

The study of CNV's in TS was prompted by the fact that dopamine has been implicated in TS (Comings, 1990). Researchers have found that the CNV amplitude was significantly higher ( $p < .01$ ), and that Post-Imperative Negative Variation (PINV – a potential occurring after the second stimulus) was significantly more often present among TS patients than among controls (Weate et. al., 1993). These findings are similar to those associated with individuals suffering from depression, schizophrenia, and Parkinson's patients undergoing dopamine replacement therapy (Weate et. al., 1993). Frontal PINV has also been associated with obsessive thoughts, and distractibility problems – problems previously cited to be prevalent in TS (Tecce & Cattanach).

Neurobiological research on TS implicates the frontal-subcortical circuits in both tic disorders, and obsessive-compulsive spectrum disorders in general (Van Ameringen et. al, 1997). The basal ganglia, part of this circuit, is responsible for storing patterns of motor movement (Amen, 1998). The frontal lobes appear to be inhibitors, selectively orchestrating motor scripts (Kolb & Whishaw, 1990). The caudate, putamen, and striatum are all components of the basal ganglia, but there is some debate as to which structure is actually associated with TS: evidence has been found to implicate abnormalities in each (Comings, 1990; Peterson et al., 1993; Singer et. al., 1993; Weinberger, 1996). Since the caudate has been closely associated with OCD (Baxter et al., 1990; Luxenberg et al., 1988; Rubin et al., 1992), and, as mentioned, OCD is quite common in TS patients, difficulties obtaining a pure TS sample may be distorting results somewhat.

In the search for biochemical abnormalities, most early attention was focused on the neurotransmitter dopamine, largely because dopamine antagonists such as Haloperidol (Haldol) and Pimozide (Orap) seemed most effective in eliminating tics, and are the

“[pharmacological] treatment of choice” (Robertson, 1994; Sandor, 1995). The frontal-subcortical circuits, as mentioned above, also are typically rich in dopamine, and dopamine is integral in the execution of complex muscle movements (Comings, 1990). Initially researchers could not determine whether the problem was one of too much dopamine, or oversensitive receptors. However Butler et al. (1979) discovered that HVA<sup>1</sup> (homovanillic acid) levels in TS patients were significantly lower than in controls, which seemed to support the receptor oversensitivity theory. Indeed, when Weinberger and his colleagues studied five sets of identical twins in 1996 he determined that the D2 dopamine receptors in the caudate were highly sensitive in the severe TS twin compared to his/her mildly affected sibling. As dopamine typically serves to inhibit the frontal cortex and energize the basal ganglia, and inhibition difficulties of the basal ganglia are definitive of TS, it would seem that the problem in TS is really twofold. Overactive dopamine receptors in the basal ganglia may cause incidentally associated motor movements to become stored rituals, while too little dopamine to the frontal lobes results in poor inhibition of those stored collections of movements. To illustrate, suppose an individual with TS happens to walk under a doorway simultaneous to blinking his/her eyes. According to this theory, and based on Hebbian Rules<sup>2</sup> the basal ganglia would connect these two events to an inordinate degree. Hence, the next time (s)he were to walk under that same doorway the urge to also blink would be stronger than it would be in an individual without these basal ganglia overactivities. Moreover, because of poor frontal lobe impulse control the individual with TS may actually succumb to the urge to blink, thus further strengthening the ritual. And so on. Given enough time, enough “incidental associations”, and enough generalizations of these incidental associations, these movements

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<sup>1</sup> Homovanillic acid is a metabolite of dopamine, of which elevated levels may be tested in urine and/or cerebrospinal fluids.

would eventually appear to be random, nonsensical, “tics”. If true, this incidental associations theory would explain a great many things reported by individuals with TS such as why tics tend to be frequent overlearned movements, why tic repertoires are unique to each individual, why tics involved an “urge” for completion, and why the longer one has a tic, the harder that tic is to lose.

As mentioned, Haldol and Orap are considered the first and best options for treating TS pharmaceutically. There are many other medications for the treatment of TS besides dopamine antagonists, but none, including the dopamine antagonists, are a panacea. Others largely focus on decreasing the level of norepinephrine in the brain (Clonidine), and/or increasing brain serotonin levels (Clomipramine, Fluoxetine) (Bruun et al., 1998).

Although tics may decrease or disappear altogether when children with TS reach adulthood (Comings, 1990), generally the prognosis is lifelong. As well, it is possible that the associated symptoms of TS increase as one enters into adulthood (Bruun & Budman, 1992; Comings, 1990). A study by Goetz et al. in 1992 followed the course of TS in 58 adults diagnosed as children, and found that while only 24% of them had moderate to severe tics, all 58 of them nevertheless still had tics. Goetz et al. found that worst functioning for these adults occurred in adolescence (13 years), however more recent evidence suggests that just prior to adolescence may be on average the time of greatest severity (Freeman, 1998).

Impacts of this disorder are far-reaching and not always intuitive. Beyond the tics and twitches and even the multiple associated problems stemming from the numerous associated conditions are considerable social, academic, and emotional difficulties. Not surprisingly, this translates into many problems in the home, school, and with peers. Considerable anger is seen

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<sup>2</sup> Synapses which are active when a post-synaptic membrane is depolarized are incremented (Hebb, 1949).

in these patients: violent tendencies have been reported to be as high as 42% (Robertson, 1989) and 65% (Stefl, 1984). This could be due to such factors as painful tics, feelings of "why me", the inability of these children to adequately communicate their difficulties and needs, and the subsequent assumptions made of them, and the fact that monumental effort on their parts to do well and restrain themselves may be minimized or seen as insufficient. Comings & Comings (1985) quoted discipline problems as being the most prevalent theme in TS families whom they treat. More than forty percent of 210 people with TS who completed a survey experienced problems in dating, and making and keeping friends (Champion, Fulton & Shady, 1988). Unfortunately Champion et al.'s design did not include a control sample, so statements about the severity of these problems when compared to the normal population are impossible. However, a survey study conducted in Halifax by Stokes et al. provides some support for believing that relationships are difficult for people with TS. They found that 35% of children with TS were ranked the least popular in their classes by their peers (Stokes et al., 1991). Depression (18.5%), family problems (four percent) unemployment difficulties (approximately two percent), medication side-effects (approximately two percent), and lack of knowledge of the syndrome in both the public and among professionals (20.2%) were all reported by some to be the most disabling aspects of TS (Wand et al., 1993). Again, the lack of a proper comparison group makes the interpretation of these results difficult.

Hubka et al. wrote an excellent review of problems typically faced within the family dynamic which interferes with normal functioning (Hubka et al., 1988). In it, they discussed how it is difficult for parents to learn how to teach the child to channel his/her impulses into forms acceptable to society. Enormous energy and time are invested in this and other chores when TS is present in the family. This can make families less spontaneous and flexible.

Parents need to mourn the loss of their expectations for parenthood, and have a “vent” for their anger, shame, and guilt. Non-affected siblings may get more attention, making the TS child feel unloved. Conversely, if the child with TS is high-needs, the sibling may feel left out, or guilty that (s)he is “normal”. Finally, the parents’ marriage can be stressed (seen in 21% of the cases of a TS family-only study, Hubka et al., 1988) if blame is assigned, or home duties are vastly asymmetrical.

Intuitively, one might think that all of these additional problems might be associated with severity of the disorder. Some researchers have indeed claimed this (Comings & Comings I, 1987; Wilson, Garron, Tanner & Klawens, 1982). Singer and Rosenberg were the first to explore the relationship between severity of behaviour problems and tics after noting that Gilles de la Tourette’s own original notes tended to suggest this relationship (Gilles de la Tourette, 1885, 1899). They found that tic severity was *not* a significant predictor of behavioural problems in a sample of 78 males aged 6-16 (Singer & Rosenberg, 1988). In a later paper, Rosenberg modified this claim, suggesting that there is a positive correlation between behaviour problems and tic severity, but only in the moderate range of severity. Those at the highest severity seemed to have the least behaviour problems, and were coined resilient, and those at the lowest severity seemed to have the most behaviour problems. This group was coined vulnerable (Rosenberg et al., 1995). Why some children are vulnerable and others are resistant, and how to prevent vulnerability and develop resiliency are the paramount questions of these researchers. They suggest that environmental factors probably play important roles in these patterns. For example, Rosenberg et al. review literature which suggests that resilient children are those who have access to a supportive network. Although

the causal direction between behaviour problems and degree of support has not been determined, it is at least a place to start.

Researchers are confused by the constant and unpredictable shifts and changes in the evolution of this disorder within individuals. Why do some symptoms that appear early not become chronic, and why do symptoms change and/or spontaneously appear and disappear all throughout life (Bruun & Budman, 1992)? An analysis of the neurology of the disorder does not appear to provide all of the answers, but factors such as holidays and seasons have been observed to be involved (Bruun et al., 1998).

Maybe to capture this entire disorder, one needs to consider environmental contributions as well as neurobiological factors. Recall the above study by Wand et al. (1993) which chronicled the most disabling aspects of TS in 422 parents of TS'ers or in the TS'ers themselves. The most disabling problem reported by the highest proportion of people was not actual symptomatology (tics were considered most disabling by 36.5% of the sample), but social isolation and embarrassment (42.5%). This is most intriguing; it suggests that those "suffering from TS" may in fact be suffering from problems more subjective and environmentally determined than the actual physiological symptoms themselves. Along these lines, it is common to hear exhausted parents of TS children proclaim that the tics are the least of their problems. Finally, many children (for example, Scahill et al., 1993) are diagnosed with TS only after they have been referred to a specialty clinic for behaviour problems rather than their tics.

So what specific environmental factors have been found to influence the presentation of TS? To date, most research has focused on physical and psychological stresses. Everything from menstruation, to visits to the doctor's office, to orgasm has been studied in connection

with the exacerbation or disappearance of symptoms (Bruun & Budman, 1992; Robertson, 1989; Silva, Munoz, Barickman & Friedhoff, 1995). The most recent neurological research has begun to consider the impact of the environment as a potential trigger to dormant genes. Specifically, the Group A  $\beta$ -hemolytic streptococcal infection (strep throat) has been linked to TS (Kiessling et al., 1993). Children with this infection develop antibodies which autoimmunize areas in the basal ganglia, perhaps leading to or worsening TS symptoms.

No doubt there are other factors, both environmental and psychological, which contribute to each Tourettic individual's unique presentation. Consideration of what some of these may be was the impetus for this current study.

Research by Carol Dweck has suggested that individuals can possess one of two implicit theories about intelligence and moral character. Those who have an “entity” approach towards a particular aspect view it as stable and unmalleable, while those with an “incremental” theory view it as more dynamic and changeable (Dweck, Chiu & Hong, 1995). Dweck has argued that the entity approach can lead to helpless patterns of behaviour in response to personal setbacks (Dweck et al., 1995). For example, obstacles are confronted with increased negative cognitions and emotions, as well as avoidance. This occurs because entity theorists are concerned with what Dweck describes as “performance goals”, or the tendency to document or demonstrate the “fixed” level of their attribute (Burhans & Dweck, 1995). In contrast, an incremental approach, or the belief in attribute improvement and ability development, leads to learning goals. Incremental theorists tend to relish challenge, remain optimistic in the face of challenge, and in general demonstrate more “mastery-oriented” responses (Burhans & Dweck, 1995).

In my dealings with various TS families I have observed this mastery versus helpless pattern of response. One young mother of an eight-year-old TS boy once said to me, “There are two kinds of Touretters – winners, and whiners”. Hence, I believe that the entity-incremental distinction posed by Dweck may be important to understanding people’s reactions to TS. In the present study I examine whether TS’ers and their family members would demonstrate these two forms of implicit theory when perceiving the impact of Tourette’s Syndrome (TS) on their lives.

Assuming that the entity/incremental distinction does extend to perspectives on disorders such as TS, what would these entity and incremental theorists look like? I expect to find family members to fit one of two profiles. I suggest that if TS’ers and family members have an entity view, they will see their condition as both unchangeable and uncontrollable. Thus, they will also see themselves as absolved of responsibility for *trying* to change. On the other hand, I expect that those who fit an incremental profile will see their TS as a challenge to be overcome and controlled; growth and change can still be achieved by learning to cope with, strategize against, and ultimately to minimize the disorder. Finally, these individuals will continue to hold themselves responsible for their TS symptoms.

If it can be established that individuals with TS and their family members do fit one of these two profiles, I will measure any differences in coping abilities between profiles. Dweck’s predicted patterns of helplessness included negative cognitions towards oneself and feelings of low control. Hence I will look at self-esteem and degree of helplessness in the present sample. I will also consider behaviour problems, and poor sibling relationships, since both of these are prominent difficulties with TS’ers, and could be worse in entity theorists who feel that the disorder renders them blameless for their behaviours and family conduct. In

summary, I expect that after holding constant the severity of the disorder, children who hold an entity view of disorder will experience more feelings of helplessness, have lower esteem, and have more behavioural problems and poorer sibling relationships than those children who hold an incremental view of disorder (Hypothesis 1a). Since, regardless of theory, TS is a difficult burden to deal with as a child, I also hypothesize that children with TS will score worse on all of these measures than unaffected siblings (Hypothesis 1b).

TS is a very complex, wide-ranging disorder. It seems logical that if these entity/incremental theories do apply to interpretations of TS, a person might have different theories for different aspects of the disorder. One might be apt to see symptomatology definitive of the disorder as unchangeable, given that the label is life-long, and real physiological abnormalities associated with these symptoms have been catalogued. Since currently only tics are essential features of the disorder, and many families do not in fact even realize that there are many associated conditions which affect their children with TS (Harper, 1992), associated symptoms such as anger/rage and impulsivity may escape this entity perspective. Hence, for our second hypothesis we predict that an entity theory will be stronger for the tics and twitches relative to the behavioural (and currently less diagnostically definitive) aspects of the disorder (Hypothesis 2).

All children, not just those in this study, first learn who they are and how the world is through their family. Thus, parents would be thought to be crucial in how these children first learn to interpret their disorder (Harper, 1992). It would make sense then to expect an association between the parents' interpretation, and the childrens' interpretation of the disorder, in that children would tend to view the disorder in the same way that their parents do (Hypothesis 3). Should it also be found that the implicit theory one holds is associated with

success of coping, parents will have been found to play a vital role in how well their children deal with their disorder.

Research was presented earlier which suggested a link between severity of disorder and being resilient or vulnerable to behaviour problems (Rosenberg et al., 1995). It has also been hypothesized that different theories of disorder could lead to different levels of behaviour problems. Perhaps then there is also a link between severity of disorder and theory towards disorder (Hypothesis 4). One's interpretation of disorder may be a result of severity – perhaps only those with a mild case of TS can find effective coping strategies, and thus become incremental theorists. Those with a more severe manifestation of the disorder find it overwhelming, and adopt an entity view. Another possibility is that one's implicit theory actually affects disorder severity in a sort of self-fulfilling prophecy. An individual believes that they cannot do anything about their disorder, so they do not try. Only those who believe that they can minimize their condition through strategies attempt the effort, and succeed.

Finally, as indicated in Hubka et al. (1988), over 20% of parents of children with TS can find their relationships strained. Hubka et al. cited spousal blame, and asymmetrical home duties as reasons, however there may be even more factors involved. In an attempt to broaden the understanding of why marital tensions can arise in families dealing with disorder, we predict that both the severity of the disorder and the interpretation of the disorder will be related to parental measures of marital satisfaction. We expect greater severity of the tics and behavioural problems and the adoption of an entity theory to be associated with lower ratings of satisfaction (Hypothesis 5).

## **Method**

### **Participants**

This sample consisted of 41 families with children between the ages of 9 and 20 who fulfilled criteria for Tourette's disorder. Table 1 displays the sample by geographical distribution. Although the original age criterion was 10 to 14, this was later relaxed. Potential volunteers were notified of this change via the TSFC, and word-of-mouth. Forty-nine children with TS (42 males, seven females, M age = 12.8, SD = 2.59), and 18 non-affected siblings (8 males and 10 females, M age = 12.3, SD = 2.93) participated from the 41 families; 41 mothers and 33 fathers also participated. Table 2 breaks both children with TS and their non-affected siblings down by age and sex. Of the 18 families in which a non-affected sibling was interviewed, a child with TS could be compared to a non-affected sibling in 17 of them: in one case, the sibling, but not the child with TS, was interviewed. In eight of these families the non-affected sibling was the elder child; in 10, the child with TS was the elder.

### **Procedure**

282 families holding membership with the Tourette Syndrome Foundation of Canada (TSFC) were mailed solicitation packages consisting of introduction letters from the researchers and the TSFC, a Background Questionnaire which collected family information, and a return-addressed, postage-paid envelope for returning the questionnaire (see Appendix A). Families were chosen by computer based only on their postal code; every TSFC member within each region was selected to receive the mailing. To ensure the confidentiality of its membership, the TSFC conducted all mailings. "Tic-Talk", a

newsletter circulated to the Toronto chapter membership, also included in its winter edition a brief request for volunteers.

Families were asked to complete and return the Background Questionnaire, and the attached Consent to be Contacted form. Fifty-six replies were received (approximately a 20% return rate). Four responses were from doctors and other professionals in the Tourette's field. Four were from families who could not participate because their child was not willing to be involved in the process. Four families could not be used, as they did not fulfill the age criterion (a wonderful letter of support was received from a 76-year-old man with TS), two responses were from relatives of Touretters other than immediate family, and one family did not have an official diagnosis of TS. This left the 41 families used in the study.

Once chosen, families were contacted via telephone to arrange a mutually convenient time for the researcher to meet with the family. Sessions took place in-home. The researcher conducted all sessions. Sessions lasted approximately 1.5 hours; the researcher remained in the home long enough to conduct all interviews, and to assist the children in completing their questionnaires. Appendix B contains materials used in the sessions. If the children were old enough (15 or older) they were given the option of completing their questionnaires alone. Parents and older children were asked to work on their questionnaires while other family members were being interviewed. Parents typically were able to finish their own questionnaires by the time the researcher left the home: those who were not were provided with a return-addressed, stamped envelope in which to mail any questionnaires not yet complete.

In six cases, there was more than one TS child interviewed in the family. In these cases, parents completed all scales for each child with TS. These six cases were only included in

those analyses of sibling interactions which did not employ siblings as a comparison group to children with TS.

At the conclusion of the interviews and questionnaires, families were thanked for their participation, and told that upon completion of the study they would receive feedback letters in the mail. These letters would provide them with a summary of the results (see Appendix C).

## **Measures**

### **Interview.**

The purpose of the interview was to measure the degree that one held an “entity” view of disorder versus an “incremental” one. There were two parts to the interview. This was done to decrease method variance, and to tap the content in slightly different ways. Interviews were conducted individually.

Six statements comprised Part One, two that addressed tics, two that addressed anger/rage, and two that addressed impulsivity (see Table 3). One statement in each of these subsections asked participants to gauge how controllable they believed that particular aspect of TS to be, and the other asked participants to gauge how responsible they believed a person with TS is for that behaviour. After hearing each sentence, participants were required to either agree or disagree with it. They were then further prompted to say whether they really (dis)agreed with the statement, (dis)agreed with the statement just a little, or were in the middle or middle of the road. Hence there were six possible responses to each question. A “1” indicated the most extreme entity view and a “6” symbolized the most extreme incremental viewpoint.

Agreement and disagreement with statements was counterbalanced with endorsement of an

entity versus and incremental viewpoint, and all items were worded positively so as to avoid confusion.

Part Two consisted of three subsections, again addressing tics, anger/rage, and impulsivity. While Part One was designed to elicit entity or incremental attitudes towards control and responsibility for symptoms, Part Two focused on the factors participants saw as involved in the severity of the disorder. Factors were considered “entity” if they implied fixedness (such as biological makeup, or inborn traits) and “incremental” if they suggested pliancy (transient environmental factors such as stress level).

In each section participants were to first indicate what they believed the child with TS’s severity level to be. Each was given a Likert-type scale in the form of five pictures, with the following descriptive statements accompanying them (for examples of the pictures used, see Appendix B):

- 1) Symptoms are present **all** of the time, they are **always** there.
- 2) Symptoms are present **most** of the time, they are **usually** there.
- 3) Symptoms are present **some** of the time, they are **sometimes** there.
- 4) Symptoms are **rarely** present, they are **hardly ever** there.
- 5) Symptoms are **never** present, they **aren’t ever** there.

Participants were then asked three additional questions in each subsection (see Table 4). Responses were considered to be incremental, entity, “other”, or “no information”. Examples of typical responses are provided in Appendix D. If the participant seemed confused by a question, restatements (i.e., “what do you think might be different between people with lots of tics and people that hardly ever tic”) were provided. Once interviewing began, it became obvious that an additional category, a combination entity/incremental response, was required

for those participants who included in their responses both entity and incremental factors.

Any interviews conducted before this change was made were re-scored.

Order of family member interviews was left to the discretion of each family – family schedules, and varying family make-ups, made a strict counterbalancing scheme infeasible. Since parents and older children completed their questionnaires independently while others were being interviewed, the order in which they chose to be interviewed also determined whether they received the interview or the questionnaires first. Since younger children completed both the interview and the questionnaires with the researcher, I flipped a coin to establish which they would receive first.

#### **Yale Global Tic Severity Scale (YGTSS).**

Parents were asked to complete the YGTSS together for each child with TS involved in the study, as they have presented in the last week. Part One is a checklist of various Motor Tics, Phonic Tics, and Tic Combinations that the child has been experiencing. Part Two consists of five Severity dimensions (Number, Frequency, Intensity, Complexity, and Interference) and are scored separately for motor and phonic tics. Part Three assesses degree of impairment (Tic Impairment, and Overall Impression) without specification of tic type. Total Motor Tic scores and Total Phonic Tic scores may be obtained by adding up individual severity scores, and a Total Tic score is obtained by combining these scores (Scahill & Leckman, 1995). Finally, the Tic Impairment score can be added to the Total Tic score to obtain a composite score. For the purposes of this study average severity scores were obtained from the Total Motor, Total Phonic, and composite scores. Finally, the Overall Impression score was analyzed separately.

Leckman et al. tested this scale on 105 participants between the ages of five and 51 (Leckman et. al., 1989). Internal consistency of the YGTSS was good – Leckman et al. found that items correlated between .78 and .90 with their respective subscale scores. As well, the YGTSS correlated well with other instruments for assessing TS, namely the Tourette Syndrome Global Scale (TSGS), The Shapiro TS Severity Scale (STSSS), and the TS – Global Clinical Impression scale (TS – GCI-S) (Leckman et. al., 1989). The YGTSS was chosen over these and other measures of tic disorders because of its unrivaled comprehensiveness in measuring purely tic behaviours. The STSSS confounds social disabilities in its tic severity rankings, and both the STSSS and the TSGS fail to assess important tic characteristics such as number and complexity of tics (Kompolti & Goetz, 1997). Also, the YGTSS assesses both severity and impact of tics, while the TS – GCI-S assesses only the impact of TS on daily functioning (Kompolti & Goetz, 1997). Finally, the psychometric properties of the YGTSS are superior to any other TS scale to date (Kompolti & Goetz, 1997).

### **Child Behavior Checklist (CBCL).**

Parents were asked to complete the Parent Form of the CBCL, which assesses a child's activities, social and school life, and behaviours. Eight behaviour subscales (Withdrawn, Somatic Complaints, Anxious/Depressed, Social Problems, Thought Problems, Attention Problems, Delinquent Behavior, Aggressive Behavior) are scored, and grouped into three composite scores: Internalizing (Withdrawn, Somatic Complaints, and Anxious/Depressed), Externalizing (Delinquent and Aggressive Behavior), and Total (all eight subscores). Scores are identified as being in the normal, borderline, or clinical ranges. The CBCL, which is a

well-documented and utilized tool for assessing behavioural problems, was chosen for this study primarily because of its demonstrated use and effectiveness on other TS samples (Rosenberg, Harris & Singer, 1984).

### **Coopersmith Self-Esteem Inventory (SEI).**

The SEI has been administered to over 40,000 individuals aged nine to adult, and comes in three forms: A consists of 58 items (50 items and 8 defensive, Lie reaction items), and five subscales. B is a 25-item short-form, with no subscales, and C is a 25-item scale adapted for adults. Split-half reliability on the full form has been previously reported at .87 (Fullerton, 1972) and .9 (Taylor & Reitz, 1968). The SEI short-form has correlated .6 with the Rosenberg self-esteem scale; the longer form correlates .63 with the Soares scale, .45 with the CPI self-acceptance scale, and .46 with the Bill's scale of self-esteem (Coopersmith, 1975). All final scores are out of 100: scores on Form A are multiplied by two, and scores on forms B and C are multiplied by 4 to achieve this. The SEI was the best measure of self-esteem to use as it permitted us to use almost identical measures on both children and adults in the family. Forms B and C, used in this study, were developed through item analysis of Form A. They have been found to correlate .86 and .8 with the longer version, respectively (Coopersmith, 1975). The mean score for preadolescents (9-15) in Coopersmith's sample was 70.1 for females, and 72.2 for males. Young adults from 16-23 obtained a mean score of 76.1.

It should be noted that the present researchers slightly altered the wording of four items in the parent form (Form C). Specifically, the phrase "my family" was replaced with, "my spouse" each time it appeared. This was done so that data from the Coopersmith could

potentially be aggregated with marital satisfaction data. In the present study the Coopersmith was administered to all family members. Parents were asked to complete the forms individually, while children under 15 were administered the questionnaire by the researcher.

**Thinking About My Relationship (see Appendix B).**

A brief (eight-item) satisfaction and commitment scale developed within the psychology department at the University of Waterloo was employed (Murray, Holmes, MacDonald, & Ellsworth, in press). Parents were asked to complete this form individually to assess their relationship with one another. Each item (for example, "I am perfectly satisfied in my relationship") could be rated from 1 ("not at all true") to 9 ("extremely true"). As there were eight items, final scores were simply the sum of all responses out of 72.

**Sibling Interaction Questionnaire (see Appendix B).**

This scale was originally developed within the developmental division at the University of Waterloo (Ross, Woody, Smith & Lollis, manuscript submitted for publication). Testing of this scale at the University of Waterloo revealed a single-factor solution. Test-retest reliability was found to be good for adults (.63), however children aged 6-7 had good test-retest reliability only when appraising themselves as a sibling (.82); when appraising their siblings test-retest reliability was poor (.5). Alphas were good for both adults (.7) and children aged 6-7 (.84) (Ross et al.). Two versions of this questionnaire – a parent form and a child form -- were used in the present study. Each questionnaire consisted of eight questions for each child interviewed. Each item on each questionnaire was on a four-point scale, but the parent and child forms were laid out slightly differently. The items on the parent form were

split into positive and negative appraisals of the particular aspect being questioned, with two possible degrees within each. For example, question three (“Rate how often your children want to be around their sister/brother”) required the parents to first choose “wants to be around” or “doesn’t want to be around”, and then specify within their choice “a lot” or “a little” for each child. On the child form the children were read two sentences, and prompted to choose the one which best described them. For example, question five required the child to choose either, “I want to be around my sister/brother”, or “I don’t want to be around my sister/brother”. They were then asked if the chosen sentence described them A LOT or A LITTLE.

The original parent scale was adapted for the current study simply by replacing the descriptives “Younger” and “Older” (sibling) with “TS Child” and “Sibling”. Some parents mistakenly circled two numbers for an item rather than one – in these instances the average between both responses was taken. When the child with TS was the only child in the family, neither scale was given. If the child with TS had siblings who were not old enough to participate in the study, the parents and TS child still completed the questionnaires. Two final numbers were derived from each questionnaire (a total score for each child), with larger numbers indicating better interactions.

### **The Nowicki-Strickland Internal-External Control Scale for Children.**

The original scale consisted of 40 items, and was normed on 1017 children ranging from third to twelfth grade (Lefcourt, 1991). Two short forms exist, one for grades 3-6 (consisting of 19 items) and the other for grades 7-12 (consisting of 21 items). Both short forms were derived from items in the original scale, and both were used for the present study. Spearman-

Brown split-half reliability estimates for the full form ranged from .63 for grades 3-5 to .81 for grade 12. (Nowicki & Duke, 1983), and test-retest reliabilities vary from .63 to .76 (Nowicki & Rountree, 1971). Correlations of this instrument with the Intellectual Achievement Responsibility Questionnaire and the Bialer-Cromwell Scale were modest (Crandall, Katkovsky & Crandall, 1965; Nowicki & Strickland, 1973). Lefcourt believes it “to be one of the better measures of locus of control as a generalized expectancy presently available for children” (Lefcourt, 1991). In the present study, the researcher administered the Nowicki-Strickland to most children – only some older children were permitted to self-administer. Total scores were derived for each child, with higher numbers indicating greater externalization (i.e. greater feelings of helplessness).

## Results

Responses to Parts One and Two of the interview were analyzed separately. For Part One, varimax rotated factor analyses on all subjects, just parents, and just children were performed. Although the ‘n’ was quite low for the second and third analyses, we were somewhat concerned that by grouping both adults and children together in the first factor analysis we may misrepresent one or more groups. Both the second and third analyses yielded a structure comparable to the first. In other words, while family members may have differed in their entity versus incremental viewpoints for Part One, the particular aspects of the disorder that were considered *as* either entity or incremental were the same for each family member. Hence the first solution was used for all participants.

Three factors were identified as being shared by all family members (see Table 5). The first factor, consisting of questions 1, 3, and 6 were all questions concerning degree of control over TS symptoms. Hence factor one was labeled **Perceived Control over Symptoms**. Higher scores in this factor would indicate that the participant sees the individual as having more control. Factor two, comprised of questions 2 and 4, dealt with degree of personal fault or blame for the associated symptoms of TS (rage/anger, and impulsivity). It was thus named **Perceived Fault/Blame for Symptoms**, with increasing scores indicating more responsibility placed on the individual with TS. Question 5 concerned perceived responsibility for tics, and stood alone as a third factor (**Perceived Responsibility for Tics**). The more responsible the participant perceived a Tourette individual to be for his/her tics, the higher the obtained score.

Part One of the interview was designed to elicit views of two aspects of disorder, namely controllability and responsibility. Part Two was intended to address one issue: that of factors

contributing towards the severity of disorder. Depending upon the factors cited (i.e. traits or situational factors), a person made implicit conclusions regarding the changeability of the disorder. Out of the nine open-ended responses possible by each participant, the number of entity responses, incremental responses and entity/incremental (known herein as “combination”) responses were summed for each individual. The total number of entity responses was multiplied by one, the number of combination responses was multiplied by two, and the number of incremental responses was multiplied by three. These products were then summed, and divided by the total number of responses given to arrive at a mean attitude number between 1 and 3. This general score was labeled **Factors Contributing To Severity**. Higher numbers indicated increasingly incremental views; that is, the more malleable the participant viewed their symptom severities, the higher a score they received.

For the remainder of this section, analyses and results will be presented by hypothesis. Unless otherwise noted, an alpha level of .05 was used for all tests of significance.

**Hypothesis #1a: Holding constant the severity of the disorder, children who hold an entity view of disorder are expected to experience more feelings of helplessness, have lower esteem, and have more behavioural problems and poorer sibling relationships than those children who hold an incremental view of disorder.**

This was our major hypothesis. The chief aim of this thesis project was to determine possible implications of holding an entity versus an incremental view towards disorder. In these analyses, all 49 TS children interviewed were included. Although eight of these children were siblings of other TS’ers in the sample, and so had the same parents, the only data supplied by the parents in these analyses were tic and behaviour severity scores. The

reader will recall that individual and independent CBCL's and YGTSS's were completed for each child.

Five stepwise multiple regressions were performed on the children with TS. The total t-score on the CBCL, two sibling interaction test scores as measured by Myself As A Sibling, Coopersmith SEI scores, and Nowicki-Strickland IEC scores of the TS child were alternated as the dependent variables. The four attitude scores derived from Parts One and Two of the TS child's interview, his/her age, and severity of disorder as measured by the tic composite score from the YGTSS were all entered into each regression as independent variables. Results can be seen in Table 6. Seeing oneself as beyond responsibility for associated symptoms of TS significantly predicted low self-esteem, and poor evaluation of one's sibling interactions. Holding an entity view towards the factors contributing to TS was predictive of low feelings of control. Finally, increasing tic severity predicted both poor sibling interaction evaluation by the children with TS, and more behaviour problems. None of the independent variables predicted how children with TS would evaluate themselves as a sibling.

In summary, an entity view held by the children with TS towards some aspect of TS was predictive of low self-esteem, feelings of helplessness, and poor sibling relations in the children with TS. Behaviour problems and self-evaluations as a sibling were not predicted by attitude towards TS disorder.

**Hypothesis 1b: Children with TS are expected to score worse on measures of esteem, helplessness, behaviour, and sibling interactions than unaffected siblings.**

In general, this hypothesis was supported for all measures except sibling interactions.

We performed independent sample t-tests on scores on both sibling interaction scales, the Coopersmith SEI, and the Nowicki-Strickland IEC.

We found that parental rating of both children as siblings (with all TS sibling interactions removed from the sample) were not significantly different from one another. In addition, self-evaluations by each child as a sibling were not significantly different, nor were evaluations of the other child as a sibling.

Non-TS sibling scores on the Coopersmith SEI ( $M = 74.25$ ,  $SD = 20.18$ ) were significantly higher than the children with TS scores ( $M = 58.25$ ,  $SD = 19.9$ ). In other words TS children were of significantly lower self-esteem than their non-TS siblings ( $t = -2.76$ ,  $p = .011$ ).

Finally, the non-TS sibling scores on the Nowicki-Strickland IEC ( $M = 6.33$ ,  $SD = 3.83$ ) were significantly lower than the children with TS scores ( $M = 8.76$ ,  $SD = 3.48$ ). To wit, TS children tended to externalize (i.e. experience more feelings of general helplessness) significantly more than their non-TS siblings ( $t = 2.18$ ,  $p = .04$ ).

With respect to differences in behavioural problems, TS children could not be tested against their non-TS siblings. Parents were not asked to complete the CBCL on both children in our study. However the TS children in the present sample were found to have significantly more behavioural problems than a normal population sample. This analysis was conducted in the following manner: the mean t score for total CBCL behaviour problems in our sample was 71.65. The mean t-score within the CBCL normative data for a non-clinical normative sample aged 12-16, collected from the CBCL manual, was 50.8. Through backward conversion using charts supplied by the CBCL (Appendix A, Achenbach & Edelbrock, 1983), we can ascertain that the mean raw score from our sample is 67 (total). The mean raw score

from the normative data (Appendix D, Achenbach & Edelbrock, 1983) is 17.5. Using the standard error of measurement (SEM) for the CBCL normative sample (10.4: also in Appendix D), a 95% confidence interval of 7.1 – 27.9 can be calculated around the normative mean behaviour problem score. As the present sample score falls well outside this confidence interval, the behaviour problems experienced by the present TS sample are significantly higher than those of the CBCL clinical sample are. Thus, the TS children on average were experiencing general behaviour problems significantly greater than those of non-clinical children. Non-TS siblings could not be compared to the CBCL non-clinical normative sample, again because CBCL forms were not completed on them.

**Hypothesis #2: An Entity theory will be stronger for the tics and twitches relative to the behavioural (and currently less diagnostically definitive) aspects of the disorder.**

Testing of this hypothesis was limited to Part One of the Interview (control, and responsibility of symptoms), as scores for Part Two of the interview collapsed across symptoms. A four (family members) by three (question) Between/Within MANOVA was conducted to determine whether responses to the three responsibility questions posed in Part One of the Interview (responsibility for tics, anger/rage, and impulsivity) were significantly different, and/or different between family members. The Hotellings F test was used for tests of significance. The main effect for question was highly significant  $F(2,144) = 285.532$ ,  $p = .00$ . Analysis of the means, provided in Table 7, reveals that all family members saw people with TS as being significantly less to blame for their tics than for other behaviours associated with TS. The main effect of family members was nonsignificant [ $F(3, 145) = .5$ ,  $p = .68$ ], however a significant interaction of family members and question [ $F(6,290) = 2.27$ ,  $p = .04$ ]

was found. Both parents, when asked about tics, saw the child with TS as significantly less responsible for this symptom than did the children. This interaction does not qualify the finding that all family members still saw children with TS as being much less responsible for their tics than for other associated behaviours – despite differences within the factor, all family members are still quite entity-driven in their responses (see Table 7).

In summary, an entity theory is much stronger for tics relative to the associated symptoms for all family members when responsibility for, but not control of, symptoms is discussed.

**Hypothesis #3: Holding constant the severity of the disorder, we expect an association between the parents' interpretation, and the disordered childrens' interpretation of the disorder. Children will tend to view the disorder in the same way that their parents do.**

Family members were compared to each other on each of the three factors developed from Part One of the Interview, and on the Factors Contributing to Severity score developed from Part Two. In general, correlations between family members by each of these four numbers showed no agreement, as Table 6 displays. As can also be seen in Table 8, nonsignificant correlations exist between the children with TS and the non-TS siblings, and between fathers and non-TS siblings. It is suspected that more of these would have reached significance had the sample included more unaffected siblings (n=18). Non-TS siblings were interviewed in only 12 families where fathers were also interviewed and 14 families where children with TS were also interviewed.

Since agreement was so low, a further set of analyses was performed to determine whether the response patterns of family members were significantly different. In these analyses, the eight extra children with TS were removed from the sample to prevent the responses of some mothers and fathers being represented twice or more in the sample.

First we performed a four (family members) by three (Part One Interview factors) MANOVA to ascertain whether the differences in responses between family members, the between-subjects variable, were significant for Part One of the Interview. They were, using the Hotellings F test of significance [ $F(9,359) = 2.64, p > .01$ ]. As the overall F was significant, individual univariates were examined. We found that the pattern of responding for Perceived Control over Symptoms was significantly different across family members [ $F(3,123) = 3.13, p = .03$ ], as was Perceived Responsibility for Tics [ $F(3,123) = 3.97, p = .01$ ].

To further analyze response differences between family members on the Perceived Control factor, a simple post-hoc contrast was conducted. Mothers saw the children with TS as being capable of significantly more control than did the TS children [ $t(123) = 2.78, p > .01$ ]; fathers and non-TS siblings did not. An independent t-test was also conducted comparing mothers and fathers on the control factor. This comparison showed that mothers and fathers differed significantly in how much control they perceive their TS children as having [ $t(66) = 2.18, p = .03$ ], with fathers seeing their TS children as more helpless than did mothers.

Regarding response differences between family members within the Tic Responsibility factor, an independent t-test comparing children to parents revealed a significant difference in perception of tic responsibility [ $t(61) = -2.89, p > .01$ ]. Both children saw the child with TS as more responsible than parents did for his/her tics.

To analyze Part Two of the interview a simple ANOVA was conducted on the Factors Contributing to Severity score derivative. There were no differences in response patterns between family members [ $F(3,124) = 1.33, p = .27$ ].

Contrary to Hypothesis 3, mothers' and fathers' responses to both parts of the interview were not predictive of either of the childrens' responses. In other words, children do not

adopt the views of their parents towards TS. Although the responses of fathers were consistently more predictive of their children's than were those of the mothers, they were not significantly so. At mean levels, mothers saw the children with TS as having more control for all symptoms than did fathers or children with TS, but both mothers and fathers saw the children with TS as less responsible for their tics than did both children.

**Hypothesis #4: The more severe the disorder, the more likely family members are to adopt an entity theory of the disorder.**

We correlated two scores from the YGTSS (the tic severity composite score, and the overall tic impact score) and the CBC total t-score to the four attitude scores obtained in Parts One and Two of the interview. To support the hypothesis, we would expect significant negative correlations between the severity measures and attitude scores (an increase in severity would correlate with a decrease in the attitude scores, or an increasingly entity view, and vice versa). Table 9 displays the results, which indicate that severity of both tics and behaviour problems has little effect on anyone's attitude concerning the disorder. Only five of 48 correlations were significant (10%). However 33 (69%) were in the right direction, suggesting that a more fine-grained analysis (perhaps looking at the relationship between attitude and tics alone, or attitude and behaviour problems alone) might yield significant results. Interestingly, however, tic severity and behaviour problem severities were positively correlated. Total behaviour problems correlated with average tic severity ( $r = .29$ ,  $p=.04$ ), and with overall tic impact ( $r = .32$ ,  $p=.03$ ).

**Hypothesis #5: Both the severity of the disorder and the interpretation of the disorder will be related to parental measures of marital satisfaction. We expect greater severity of the tics and behavioural problems and the adoption of an entity theory to be associated with lower ratings of satisfaction.**

All four scores derived from the interview were correlated to parents' relationship satisfaction scores. As well, each family member's Coopersmith score was correlated to parental relationship satisfaction.

Hypothesis 5 was in general not well supported. Tic severity and behaviour problem severity did not seem to predict relationship satisfaction in either parent (see Table 10). Next, while fathers' attitudes predicted their own relationship satisfaction, it was not in the anticipated direction. The less fathers saw the children with TS as being responsible for their tics (i.e. the more entity-driven they were), the more satisfied they were ( $r = -.33$ ,  $p = .05$ ). Also, the less changeable fathers saw the factors contributing towards the disorder to be, the happier they were in their relationships ( $r = -.32$ ,  $p > .06$ ). Only when discussing responsibility for behaviours associated with TS (anger/rage, impulsivity) was an incremental view connected to higher satisfaction ratings in fathers; more responsibility equaled more satisfaction ( $r = .38$ ,  $p = .02$ ). Mothers did not display any associations between attitude and relationship satisfaction (see Table 11).

Looking at the "Sibling" portion of both 6 a and b, the non-TS siblings' attitude towards responsibility for disorder was strongly correlated to both parents' relationships. The relationship satisfaction of both parents improved if the non-TS sibling perceived the TS child as being responsible for the associated symptoms ( $r = .54$ ,  $p = .04$  for mothers,  $r = .76$ ,  $p > .01$  for fathers).

Overall, disorder severity was unrelated to marital satisfaction. One's own attitude towards TS also did not seem to be related to marital satisfaction for mothers, however it was for fathers. The attitude of non-TS children towards the disorder was related to the marital satisfaction of both parents.

## **Discussion**

Participants on the whole participated enthusiastically; I was welcomed into most homes with warmth and the offer of refreshment. Family members seemed to enjoy the chance to “vent”, and often went into detail far surpassing the requirements of the study. One mother commented that an opportunity for family members to have their views heard by professionals was both rare and sorely needed.

An interesting difference between the current study and similar work in attitudes towards intelligence done by Dweck is that we differentiated various entity/incremental domains (Control, Responsibility, Factors Contributing to Severity) within TS disorder, rather than simply measuring an overall entity/incremental attitude. As seen in the regression results from hypothesis four (see Table 9), an entity view in one domain might be related to one measure, but not another. For example, an entity view towards Perceived Fault/Blame was associated with low self-esteem in children with TS, but an entity view towards Factors Contributing to Severity was associated with feelings of helplessness. It makes intuitive sense that if people believe there is something inside of them making them abusive towards others and causing them to act without thinking, this is primarily going to lead to self-esteem problems. Other problems, such as feeling powerless and ineffective in one’s life would be more likely to stem from a different belief, namely judging one’s disorder to be permanent despite all efforts to change it. In an author’s response (Dweck, Chiu, & Hong 1995), Dweck et al. comment that although the entity and incremental perspectives are polar opposites many people do hold both. They explain this finding as an example of the many cognitive inconsistencies seen in people. It may also be, however, that intelligence, like disorder, can

be differentiated into various domains, each with its own implicit theory. Further study of this differentiation process could be quite interesting. For example, is this a developmental process, and how does it progress? Although this could not be tested in the present study, younger children (below nine) may start out with a more general, all-encompassing entity or incremental view of disorder. Perhaps as individuals grow, their general perspective on disorder begins to differentiate, with different attitudes in different domains. There is research that suggests that this process does occur over the course of trait concept development in children (Heyman, Dweck & Cain, 1992). Is this differentiation process a function of general maturity, or of such factors as time since diagnosis, or amount of education on TS?

In this study there seemed to be good support for believing that different people differentiate their perspectives on disorder in the same way. Each family member showed the same patterns in the factor analyses. Would extended family members also demonstrate this pattern? Or teachers? Peers? Are there other domains in which a person may hold an entity or incremental view towards disorder, such as the ability to be autonomous when diagnosed with a disorder, or to be capable of performing particular tasks? Entity theorists may see themselves as people who require others to always do things for them, and as unable to take certain jobs. In contrast, incremental theorists may see others as useful for teaching them how to do things for themselves, and see themselves as growing by tackling a form of work that challenges them to improve their weaknesses.

In the initial factor analysis it was found that questions of controllability comprised one factor, and questions of responsibility tapped two other factors –tic responsibility, and

perceived blame for associated symptoms. This is somewhat disturbing; since this means that there was little relationship between one's attitude towards control, and one's attitude towards responsibility, this makes possible two unfortunate scenarios. First, a child could learn that although he may have some control over his symptoms, he won't be held responsible for that control, or lack of. This communicates permission to use one's disorder as an excuse. Second, a child may learn that although he is not in control of his disorder, he is to be held accountable for it. This perceived powerlessness over the rewards or punishments one will receive is a classic pattern for depression.

This study replicated Dweck's findings that entity theorists will demonstrate more negative self-evaluations of themselves, and more feelings of helplessness (Erdley & Dweck, 1993). Believing that people with TS should not be responsible for the associated symptoms of TS (anger/rage, impulsivity) was significantly predictive of poor self-esteem. Beliefs that TS is caused by stable, unchanging factors was significantly predictive of feelings of low internal control. In addition to Dweck's traditional findings, this study revealed that having an entity theory towards disorder could affect evaluations made of others. Children with TS rated interactions with their siblings significantly lower if the children with TS believed that they are not responsible for their associated symptoms.

Multiple regression does not allow for statements of causality, hence it cannot be definitively stated based on this study alone that it is attitude towards disorder which is contributing to low self-esteem, feelings of helplessness, and poor sibling relations in children with TS. It may be that having poor scores or good scores on these different measures causes

one to adopt an entity or incremental approach to disorder, respectively. We believe the former scenario, in which attitude causes the scores on these measures, to be more persuasive.

As described in the introduction of this paper, the impact of TS and its associated symptoms encompasses virtually every aspect of one's life. They encroach upon and interfere with peer relations, academic successes, home life, extra-curricular activities, and intimate relationships. The initial view that one has towards the disorder would undoubtedly affect how children with TS face these aspects of their lives as they develop general views about themselves, others, and the world. In addition, studies in other realms have shown that attitude can lead to helpless behaviour. For example, the types of feedback a child receives from adults about his/her work affected not only how the children then began to judge themselves, but also their subsequent performances, even generalizing to new situations (Dweck et al., 1995; Dweck & Leggett, 1988; Heyman et al., 1992).

Once an implicit theory has been developed, it is possible that the relationships between attitude and self-esteem, feelings of control, and sibling relations become reciprocal. To illustrate, suppose a child with TS adopts an entity approach to his disorder, because no matter how hard he tries, he cannot stop his tics. Since he feels powerless to stop the disorder, he is miserable, and a good target for peers to tease him when he tics. Ostracized by his peers, and believing that the situation cannot change since he cannot stop his disorder, he develops low self-esteem. Now that he has low self-esteem, he avoids the other children at recess. He misses out on so much peer involvement that he becomes delayed in his development of interaction skills, reading social cues, and so on. He assumes that any new children he meets will also ridicule him for his tics, and does not make an effort to befriend them. In short, he acts in a very helpless manner, which would result in the child continuing to have few

friendships. Since neither the disorder nor his lack of friends has changed, this “proves” to the child that nothing can change because of the disorder, thus strengthening his entity view.

When children with TS describe themselves as a sibling, age, severity of disorder, and attitude towards disorder seemed not to matter. However, when the same children appraise their sisters or brothers as siblings, greater disorder severity and a greater perception of responsibility for anger/rage and impulsivity were associated with higher sibling evaluations. It is easy to see how non-TS siblings’ behaviours could be affected by severity of tics, and whether the children with TS believe that they can throw rages or act impulsively without being at fault for the consequences. It is also easy to see how the TS siblings’ behaviours could be affected by these two factors though. Hence, a question remains as to whether these two factors really had no impact on the children with TS as siblings, or whether the children with TS were just more accurate in their ratings of others than they were for themselves (a not uncommon finding; for a review see Kenny, 1994).

In hypothesis 1b we found, as we had anticipated, that children with TS have lower esteem and more externalizing behaviours than do their nonaffected siblings. Further, children with TS experienced significantly more behavioural problems than a non-clinical sample. One rating that was not significantly different between children with TS and their non-TS siblings was that of sibling interactions. This is perhaps not surprising. Within a family network, it is probably very difficult if not impossible to have good relations with a sibling who is not able to have, or is not interested in having, good relationships in return. In essence, “it takes two to tango”. Perhaps a more logical approach would be to compare sibling networks within families dealing with TS to families who are not.

It was predicted in hypothesis two that tics and twitches would be seen in a more “entity” way than other associated symptoms of TS. This was tested and found in the Responsibility domain. Some symptoms were considered within an entity perspective, while others weren’t. The basis for this hypothesis was that this was a sample of people dealing with TS and TS is primarily defined as uncontrollable tics and twitches. The associated problems are not considered core to the disorder; because of this, and/or perhaps because these symptoms are not seen as being neurobiological in nature for any number of reasons, this population may view these symptoms more incrementally. Yet these very associated problems, although not central (at least yet) to the diagnosis of TS, ARE the primary symptoms for other disorders (such as ADHD and Intermittent Explosive Disorder) that can be seen as neurobiological. Is it the case that samples of ADHD and IED populations would view impulsivity and rage respectively from an entity perspective, and perhaps tics less so? Some support for believing that this may be so comes from Freeman, Johnston, and Barth (1997), who studied the reactions of mothers and fathers to scenarios involving various behaviours of their ADHD children. Both parents saw their child’s inattentive-overactive (primary aspects of ADHD) as less controllable by the child than oppositional-defiant and prosocial behaviours (Freeman, Johnston, & Barth, 1997). Further research in this area would be most interesting to see if indeed labels based on imperfect and continually changing diagnostic criteria can actually effect how different behaviours are viewed, and what factors (such as literature, or professionals espousing medical models) can cause that process to occur.

Addressing hypothesis three, we found that parent attitudes do not seem to predict those of their children. Why? The almost random pattern of responding seen within the family units may be signaling different things. The sheer volume of information available on TS, and

consumed by these parents, is fraught with contradiction, unknowns, and very complicated discussions of topics such as neuroanatomy, statistics, and genetics. Perhaps attempting to glean all of this knowledge only serves to confuse. There is no doubt that many do exert this effort. One question in the initial solicitation package asked parents to list the various means through which they have learned about TS. Many parents filled the abundant space provided, plus the back of the page in an exhaustive (just to look at!) list of resources they have used.

In some cases the opposite may be true – little education on TS in the family, and few resources used by the family could lead to confused and differing opinions among family members. These families are in all likelihood underrepresented in the present sample however, since subjects were chosen from the membership bank of an organization committed to educating its members.

A third possibility is posed by Harris, 1995. She found in her study that parents have no important long-term effects on the development of their child's personality (Harris, 1995). She suggests that peer group processes modify children's personality characteristics to a far greater degree than do parents.

What seems abundantly clear from the results of hypothesis three is that attitudes towards TS are not consistent, even between family members. Hypothesis one confirmed that esteem, feelings of control, and sibling interactions are associated with attitude towards disorder. Should future studies prove that one's attitude causes success of coping, it will become important to determine how to make each family members' attitude incremental. Further studies should also focus on whether an individual's spoken beliefs actually match his/her actions – it is possible that while each family member has a different reported attitude towards

disorder, a study which ascertained attitude through behaviour might reveal that parental attitudes do influence those of their children.

Hypothesis four, namely that theory towards disorder would be associated with severity of disorder, was not supported. It was thought that as severity increased, so might a sense of helplessness as to how to handle these symptoms. However, as there is literature which demonstrates that entity and incremental views in domains other than disorder occur at all levels of the particular trait in question, it is perhaps not a surprise that in the present study tic severity was not related to the attitude held. For instance, Goetz and Dweck (1980) gave 130 boys and girls in fourth and fifth grades four imaginary situations in which they were socially rejected, and then asked them why this might happen to them. The children were to differentially endorse five explanations. Four were “incremental” in that the explanation implied that the situation could be changed (i.e. misunderstandings, unfortunate circumstances, rejector attributions, or incompatibility between rejector and rejectee). One was “entity” in that the explanation implied that the person was helpless to change the situation (i.e. personal incompetence). Goetz and Dweck found that there were students who endorsed an “entity” explanation, and acted in a helpless fashion across all popularity levels. In another study, Hong and Dweck (1993, Study 2) manipulated the feedback that college students received on a conceptual ability test that they had completed. Both entity and incremental theorists were found within the same performance levels. Hence there did not seem to be any evidence before this study supporting the notion that a particular theory is associated with a particular competency in the social or intelligence domains. The present study extends this finding to the disorder realm.

The results of the final hypothesis revealed that the attitude of most family members did not predict the marital or relationship satisfaction of either the mothers or fathers in this sample. The one exception to this was the attitude of fathers, which correlated significantly with their own happiness. Interesting to note is that while an incremental view on one aspect of disorder (Perceived Fault/Blame) was associated with increased relationship satisfaction, an entity theory on two other aspects of disorder (Responsibility for Tics, and Factors Contributing To Severity) were associated with increased relationship satisfaction. Although these results would need to be replicated before taken too seriously, Dweck does provide some rationale for why being an entity theorist can be advantageous. In a 1995 paper, she suggests that the appropriate times include when a real limitation does exist, and when your ability is unquestionably high. Accepting for the moment that the results of hypothesis five are replicable, we could safely assume that the second situation is not relevant in this discussion, since it is definitive of disorder that a problem does exist. This leaves us to consider the situation where a limitation is very real, as could easily be pictured within a family that does not know how to deal with TS. When living with a disorder that is particularly distressing for all family members, the belief that you may have somehow contributed to that distress would be very disturbing. Indeed, past studies have identified the tremendous guilt that many parents feel once they learn that their child must grow up with a genetic disorder, and a diagnosis is made (Hubka et al., 1988). If you are a father unfamiliar with any coping strategies for either yourself or your child, believing that nothing can be changed, that “what’s done is done”, could be a way of coping with this guilt.

Also note that while fathers’ relationship satisfaction increased if they saw behaviours primary to TS (tics and twitches) within an entity perspective, it also increased if they saw

associated behaviours within an incremental one. Recall from hypothesis two that tics and twitches, primary to the diagnosis of TS, were viewed within an entity approach significantly more often than were other associated symptoms of TS. This pattern could be an attempt by fathers to define the boundaries of the disorder, to further minimize the guilt they experience. In the context of the present results, fathers then have drawn the entity “line in the sand” after tics; any other symptoms are considered in an incremental light. It is possible that by making definitive judgements regarding what is disorder and what is not, they are then able to easily decide which behaviours could be looked at in an entity way (to reduce guilt for passing this disorder onto their child, and to avoid accidentally punishing a child for something that (s)he cannot help.), and which in an incremental one (to avoid guilt for being permissive parents). This exercise would help to “contain” the disorder, and not allow the impact of a disorder to touch every aspect of their child. In other words, it is easier to deal with the guilt of a particular problem in your child and to isolate that problem than to consider the possibility that this disorder affects the child globally, whether it actually does or not. Again, replication of these unexpected results would be necessary before definitive statements could be made.

In summary, this research suggests that the Dweck entity/incremental model of implicit theories extends well to individuals dealing with Tourette’s syndrome. Although there was little familial verbal agreement on implicit theories in particular domains, overall all family members saw tics and twitches within an entity framework. Further, children with TS holding an entity perspective scored worse on measures of self-esteem, perceptions of control, and sibling interactions than did those holding an incremental view after controlling for severity of tics. Severity of disorder was surprisingly unimportant not only in what attitude one held

towards one's disorder, but also on one's level of self-esteem, and feelings of control when dealing with TS.

Although an incremental theory is associated with a higher level of coping, it is easy to see how our society may inadvertently promote the entity position in individuals with TS, even while explicitly espousing an incremental position. The best strategies for handling TS are not intuitive; in fact, fighting to directly suppress the impulses and movements directly results in symptom worsening after the effort is expended (Bruun et al., 1998). Unfortunately the initial (and logical) response of most people is to simply urge the person to stop. When the individual is met with repeated failed attempts to do so, classic helplessness, defined as a learned noncontingency between behaviour and outcome, results (Seligman, Maier, & Solomon, 1971). Note that this learned noncontingency was developed only because *that particular strategy* endorsed and suggested by society did not change the outcome (did not reduce the tics). Other behaviours can result in the reduction of symptoms. The problem is that if children are not taught these strategies at an early age, an entity theory will develop. Self-negativism, reduced effort, and avoidance of situations in which they may fail again (Burhans & Dweck, 1995) are all associated with the adoption of an entity view, and might interfere with any later attempts to train these children to control their disorder.

As mentioned in the discussion of hypothesis three, it may be the case that while reported attitudes towards disorder are different between parents and children, actual behaviour is more similar. Hence, although parents may purport to be incremental theorists, they may communicate entity messages implicitly. To illustrate, a parent or teacher may see a child with TS as being capable of learning to succeed (i.e. an incremental theory), yet have the attitude that having a disorder means that you have to work harder to achieve. This

communication of a relationship between degree of disorder and effort necessary to achieve implicitly communicates the message, “the more ability that I have (i.e. the less disorder I have) the less effort I need to invest to do well”. This mode of thinking is correlated to the entity theory framework, and contrary to the incrementalist position, which emphasizes that effort and ability are complementary (Dweck & Leggett, 1988; Dweck et al., 1995). A child faced with this feedback, when brute force of will does not cause the symptoms to go away, could develop an entity theory despite being surrounded by “incremental theorists”.

The logical continuation of this work (assuming that attitude does cause these problems) would be to test whether an individual’s implicit theory can be changed or not, and to see whether this shift in attitude results in corresponding increases in self-esteem, feelings of control, good behaviours, and good familial relationships. I speculate that one route to accomplishing this might be through simple demystification of children with TS. From my own experience, even when parents, teachers, and doctors are well informed, sometimes the children with TS themselves have not been demystified. Simply handing children a label without a subsequent explanation of what that label means or how the labeled disorder affects them could potentially cripple their ability to empower themselves and to recognize an ability to change through, for example, development of strategies. It can be a challenging and time-consuming role to educate children on their disorder(s), as the information must be presented in a fashion in which they can absorb it, and I’ve found that children can sometimes find diagnosis threatening and resist attempts to discuss it. Nevertheless, in my opinion the importance of teaching a child about his/her disorder cannot be emphasized enough. It can provide them with the necessary vocabulary to verbalize their experiences, decreasing

misinterpretations and misperceptions of their actions, the frustration of not being able to communicate their needs, and the anxiety of hiding something that they do not understand. It can allow them to see particular symptoms and difficulties in themselves, rather than seeing themselves as globally bad or incompetent. Global dispositional inferences have been found to be associated with entity theorists (Dweck, Hong & Chiu, 1993), and dispelling them may help in shifting children into an incrementalist role. Children who understand how this disorder works have an opportunity to develop effective strategies on their own, possibly increasing their sense of self-efficacy, and lessening the feeling that the way they are is stable and will not change, which could increase self-esteem. Finally, a better understanding of what TS is can help children to see how some positive aspects of themselves have resulted from the disorder. This could make having a disorder less threatening to consider. It could also help a child to conclude that while the neurobiological aspects of the disorder (i.e. the disinhibition) is a constant, how it manifests itself (i.e. how one handles impulsivity, rages and tics) is malleable, thus representing yet another possible way of differentiating aspects of disorder.

In 1975 Dweck provided some evidence that it is possible to alter implicit theories. 12 children were selected to participate in one of two procedures designed to train them to not have extreme helpless reactions in the face of failure. One procedure ensured only successes, while the other taught the children to reattribute their failures to lack of effort. Dweck found that the Success Only group, when tested for failure reactions at the end of the sessions continued to display increasingly helpless patterns. In contrast, the Attribution Retraining group actually increased their performance, and was more likely after training to explain their failures as motivational problems rather than stable negative dispositional traits (Dweck,

1975). Further evidence comes from Matthews et al., 1985. They found that children with TS coped best when the children were expected to function normally, and where the tics were not expected to be controlled.

It is the hunch of this researcher that success in training an incrementalist perspective will generalize to those with TS as well. As an individual with Tourette's syndrome myself, I take considerable pride in the fact that I have evolved from a lost, miserable soul beaten by my disorder to someone happy, efficacious, and capable of completing this report you now hold in your hand. Many of these children also have the potential to achieve great heights – the tragic irony is that their own attitudes towards their disorder may be holding them back. It is up to us to educate and encourage these children, so that they may realize the pinnacles they can achieve.

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