SPEECH FLUENCY DEMONSTRATED BY CHILDREN WITH TOURETTE SYNDROME

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ABSTRACT
Speech Fluency Demonstrated By Children With Tourette Syndrome
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Children with Tourette Syndrome (CWTS) frequently exhibit a high prevalence of disfluent speech behaviors which are often labeled stuttering. The present study analyzed the fluency characteristics of CWTS, in comparison to children who stutter (CWS) and typically developing peers (TDP). It was predicted that CWTS would be less fluent than TDP but more fluent than CWS. A related purpose was to explore whether differences existed in the pattern of disfluencies demonstrated by these groups. To this end, it was predicted that CWTS would demonstrate significantly lower proportions of stuttering-like disfluencies than CWS and significantly higher proportions of stuttering-like disfluencies than TDP.

Participants included eight CWTS, eight CWS and eight TDP. Speech samples, collected during a narrative story telling task, were analyzed to determine whether significant differences in the type and frequency of disfluencies were evident between the groups. Results revealed that CWTS were significantly more fluent than CWS and that CWTS produced significantly lower proportions of stuttering-like disfluencies than CWS. Although not statistically significant, CWTS were twice as disfluent as TDP and CWTS produced significantly higher proportions of stuttering-like disfluencies than TDP.

These findings confirmed that CWTS present with an atypical disfluency pattern which can be differentiated from that of CWS and TDP based on the total disfluency level and the proportion of stuttering-like disfluencies.

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As I look back on the journey, the starting point has become a memory, the route has gotten foggy, and the experiences have blurred together. All that remain are the lessons learned and the memories of the mentors who have helped along the way. To them I am eternally grateful and look forward to future adventures together!

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CHAPTER 1
OVERVIEW OF TOURETTE SYNDROME

Background

Tourette Syndrome (TS) is a neurological disorder with childhood onset. The primary characteristics of the disorder include recurrent motor, phonic and mental tics, complex vocalizations and behavioral complications that usually appear by seven years of age and are episodic and situational in nature (Bruun & Bruun, 1994; Pauls, Leckman, & Cohen, 1993; Richard & Hoge, 1999). Tics are repetitive, rapid, involuntary movements or vocalizations that are thought to be secondary to abnormal metabolism of the neurotransmitter dopamine (Bruun & Bruun, 1994). Tics can vary in presentation, severity, duration and complexity over time (Bruun & Bruun, 1994; Pauls, et al., 1993). Tics may be simple, involving only one muscle group, or complex in nature, involving multiple muscle groups. They can be either motor or phonic in nature. Motor tics include involuntary movements of the face, limbs or body (Richard & Hoge, 1999). Common facial tics include facial grimaces, eye-blinking, shrugging, head-jerking, face-rubbing and lip-licking.

Current literature has adopted the term phonic tic to describe any involuntary vocalization. This term is preferred over the term vocal tic since many of the vocalizations do not involve the vocal folds (Jankovic, 1997). Common phonic tics include throat-clearing, snorting, grunting, sniffing, repeating words, barking and impulsive remarks (Pennsylvania Tourette Syndrome Association [PTSA], 1990). The term mental tic is used to describe phenomena such as intrusive words/images, rituals, or a fascination with morbid or perverse topics (PTSA, 1990). Complex motor acts such as

compulsively pulling on clothing or thrusting of the arms, groin or upper torso are also seen (PTSA, 1990).

According to the Diagnostic and Statistical Manual IV (American Psychiatric Association [APA], 1994) criteria for TS are based on:

- The presence of both multiple motor and one or more vocal (i.e. phonic) tics.
- Tics occurring multiple times per day, usually in spurts but nearly every day or intermittently over a period lasting more than one year.
- Marked distress or significant impairment in social, occupational or other important areas of functioning.
- Onset before 18-years of age.
- The disturbance is not due to the direct physiologic effects of a substance or general medical condition.

Coexisting Diagnoses

Besides the prerequisite tic presentation, individuals with TS frequently present with other diagnoses. In fact, many people frequently discuss TS as a continuum of behaviors which encompasses traits of several other diagnoses. Although originally thought to represent divergent expressions of a shared genetic etiology, it is now commonly held that regardless of the high comorbidity between TS and certain diagnoses, they are distinct, unrelated disorders (Pauls, et al., 1993). In fact, these diagnoses do not figure into the diagnostic criteria of TS as per the Diagnostic and Statistical Manual IV (APA, 1994). These commonly co-occurring disorders can include

Obsessive Compulsive Disorder (OCD), Attention Deficit Hyperactivity Disorder (ADHD), Anxiety Disorder and Learning Disabilities.

OCD is characterized by recurrent obsessive thoughts or a compulsive urge to perform certain behaviors which cause marked distress or significant impairment to the individual (APA, 1994). The behaviors can include: checking rituals, arithromania or counting routines, compulsively touching oneself or other things, habitual balancing or evening up, compulsive hygiene rituals and an overwhelming need to repeat an action until the individual experiences a sense of relief. It is estimated that OCD affects at least half of children with TS (Comings, 1995) as opposed to 2.5% of the general population (APA, 1994).

ADHD is the most commonly diagnosed co-occurring diagnosis seen in children with TS (Pauls, et al., 1993). Estimates suggest that as many as 70% of children with TS additionally meet the diagnostic criteria for ADHD. Learning Disabilities affect approximately 20-40% of children with TS, and anxiety disorders or phobias affect roughly 30% of these children (Brunn & Brunn, 1994; Coffey, Biederman, Smoller, Geller, Sarin, Schwartz, & Kim, 2000; Comings & Comings, 1994; Fenichel, 1997; Pauls, et al., 1993). Additionally, a survey of 431 individuals with TS found that 65-75% of the participants reported problems with temper control, mood swings, rage and aggression (Stefl, 1984). Additional symptoms associated with TS can include: behavioral concerns, recurrent negativism, impulse control difficulties, depression, tantrums, and overreaction, reduced focusing/attention, hyperactivity, persistent obsessive thoughts, reduced visuomotor or visuospatial competence, impaired written mathematical and computation skills, reduced reading comprehension skills, and limited

cognitive/linguistic functioning (Brunn & Brunn, 1994; Coffey et al, 2000; Comings & Comings, 1994; Fenichel, 1997; Packer, 1997; Pauls, et al., 1993; PTSA, 1990). However, it should be noted that given the difficulty involved in differentiating which behaviors are characteristics of TS versus which are secondary to the coexisting diagnoses, the comorbidty between these disorders may be drastically underestimated.

Speech and Language Concerns

Families of children presenting with TS frequently report the coexistence of speech and language issues. Often, the difficulties involving speech production overshadow the impact of the core TS symptoms. Pauls, Leckman, & Cohen (1993) investigated the relationship between TS and a history of speech and language deficits in 85 individuals diagnosed with TS between the ages of 7 and 62 years (mean = 20.7 years, SD 1.3 years). They compiled their data using prestructured interviews which included probes on a variety of speech and language issues. They found that 24.7% of the participants reported a significant history of speech and/or language difficulties. Specific areas of speech and language impairment associated with TS have included: pragmatic/socialization concerns, the use of frequent fillers, delayed expressive language scores, reading and writing problems, limited vocabulary, vocal abuse, atypical suprasegmentals, word finding problems, spasmodic dysphonia and increased speech rate (Richard & Hoge, 1999; Shapiro, 1999).

O'Quinn and Thompson (1980) reported on a case study of five children diagnosed with TS. Clinical reports demonstrated that all of the children presented with disfluencies which appeared related to their organization of language. Four of the

children presented with significant histories of developmental speech and language issues. The most common concerns related to the organization and formulation of language, and word finding abilities to a lesser extent. Unfortunately this study was based on clinical findings from a small sample and did not follow a rigorous protocol utilizing standardized measures. Thus, it is difficult to generalize the finding beyond the sample.

Ludlow, Polinsky, Caine, Bassich and Ebert (1982) used spontaneous speech samples and The Neurosensory Center Comprehensive Examination of Aphasia (Spreen & Benton, 1977) to gauge the functional language abilities of 54 individuals with TS. The group was split with 13 participants between the ages of 5 and 11 years, 26 participants between 12 and 18 years and 15 participants older than 19 years. The results indicated that the participants with TS demonstrated significantly lower scores on tasks of language expression. These differences were noted in each of the three age groups with the greatest difference demonstrated by the group between 5 and 11 years of age. Furthermore, it was found that participants with TS spoke for significantly less time than controls and required increased prompting to engage in speaking. No significant differences were found on tasks involving comprehension or receptive language abilities.

Legg, Penn, Temlett, and Sonnenberg (2005) employed The Test of Language Competence (Wiig & Secord, 1989) and a fable interpretation task to measure the higher-level language processing and discourse abilities of 10 adolescents (7 male and 3 female) diagnosed with TS. The results indicated that half of the participants demonstrated no deficits in understanding or using abstract concepts or in their ability to organize and formulate language. However, in accordance with the findings mentioned above, a

significant portion of the sample demonstrated expressive language deficits involving the organization and expression of high-level and/or abstract language.

Several complex vocal tics have become virtually synonymous with the disorder. For example, TS is commonly considered a disorder where the individual demonstrates coprolalia or the uncontrollable, blurting out of obscene and/or forbidden words and noises. Although this vocal behavior can certainly result in significant social, educational and occupational distress, it is demonstrated by less than 40% of individuals with TS (Brunn, Cohen & Leckman, 1997) and does not factor into the diagnostic criteria (APA, 1994). Nevertheless, due to the common media distortion, this behavior has become a trade-mark characteristic of TS. Echolalia, repeating others' words, and palilalia, repeating one's own words, are also common behaviors demonstrated by individuals with TS. It is interesting to note that palilalia, which often manifests as the repeating of the last sound, word or phrase in an utterance, has received considerable attention as of late. It has been discussed as an atypical disfluent behavior and has been linked with a variety of neurologically based disorders including: Asperger's Syndrome, Attention Deficit Hyperactivity Disorder and Apraxia.

The speech-related profile of an individual with TS often includes: the use of interjections and fillers ("um," "well"), a high frequency of disfluencies, stuttering, vocal abuse and misuse concerns, atypical suprasegmentals, reduced intelligibility, spasmodic dysphonia and an increased rate of speech characterized by minimal pause time (DeNil, Sasisekaren, Van Lieshout, & Sandor, 2005; Donaher, 2005; Richard & Hoge, 1999; Tetnowski & Donaher, 2003; Van Borsel, Goethals & Vanryckeghem, 2004; Van Borsel & Vanryckeghem, 2000). Comings and Comings (1987), in a study of 246 patients with

TS, found that the TS sample had significantly higher levels of stuttering when compared to the general population. Pauls, Leckman, and Cohen (1993), in the study cited earlier, found that 15.3% of all respondents with TS reported a significant history of stuttering. Other studies incorporating retrospective, self-reports or survey methodologies, have estimated as high as 31% of patients with TS stutter or have stuttered in the past (Comings & Comings, 1994; Pauls, et al., 1993). Unfortunately, the use of self-reports makes it difficult to determine what each individual considered stuttering. Since no formal definitions were included in these studies, it is possible that everything from short developmental periods of disfluency to confirmed stuttering were included under the stuttering label.

CHAPTER 2
LITERATURE REVIEW

Fluency and Stuttering

During language acquisition or periods of increased pressure to perform linguistically, many children struggle with language formulation and organization. This is especially true between the ages of 2 ½ and 5 years when language skills are still emerging. These periods of uncertainty are frequently marked by disruptions or disfluencies in the child's speech. There is a great deal of variability and fluctuation during these periods of developmental or normal disfluency (Starkweather, Gottwald, & Halfond, 1990). Parents often comment that they only occur in certain situations or with certain individuals. For example, disfluencies are more frequent when the child is tired, competing for attention with siblings, or attempting to express complex thoughts and ideas.

The most common types of disfluencies demonstrated by nonstuttering children include effortless and rhythmic repetition of whole words and phrases (Shapiro, 1999; Starkweather, 1987). Examples of this include: "Can, can, can we go" or "My dad is, my dad is big." Other common disfluencies include pauses, hesitations, interjections (e.g. e.g. "um," "well," "you know"), and sentence revisions (e.g. "I want, I need to go"). Manning (2001) refers to these commonly produced disfluencies as formulative fluency breaks. These formulative fluency breaks: (a) occur between words, phrases or larger syntactic units, (b) lack tension or struggle, (c) are interjections between whole words or larger syntactic units (Manning, 2001). As children mature and begin to use more elaborate linguistic structures, they actually may increase the frequency of these formulative fluency breaks until they have mastered the language (Ambrose & Yairi,

1999; Starkweather, 1987; Yaruss, Newman, & Flora, 1999). Typically, a nonstuttering speaker will achieve an adult-like level of fluency by adolescence once the neurological and psycholinguistic systems have reached full maturity (Manning, 2001). It is important to note that developing children who do not stutter also produce disfluencies that are more like those in a stuttering disorder. However, these are infrequent and diminish to less than 1% of syllables by 3-4 years of age (Ambrose & Yairi, 1999; Pellowski & Conture, 2002).

Close to onset, children who stutter often look much like nonstuttering children (Starkweather, 1987) but over time, the two groups can be differentiated by the type and frequency of behaviors and the individual's reactions to the disfluencies. Children who stutter tend to present formulative fluency breaks in addition to high levels of motoric fluency breaks (Manning, 2001). Motoric fluency breaks: (a) occur between syllables within a word, (b) contain tension or struggling, (c) inhibit the ability to initiate airflow to support speech, or (d) are prolongations of sounds or syllables (Manning, 2001). Children who stutter tend to have increasingly more motoric fluency breaks as they develop but reduce the number of formulative fluency breaks throughout childhood up to adolescence. This is the opposite trend from nonstuttering children who produce relatively few motoric fluency breaks and more formulative fluency breaks which can increase in frequency until adolescence (Manning, 2001). Bloodstein (1995) reported on 418 children who stutter between the ages of 2 and 16 years of age. His findings revealed that the majority of school-aged children who stutter were exhibiting increased stuttering in specific situations or on specific words and sounds. They produced substitutions in an effort to avoid the frustration that commonly accompanied their stuttering and they were

beginning to develop negative speech-related attitudes. He found that many children from the age of 10 years experienced fearful anticipation of stuttering and used frequent avoidance behaviors. These emotional reactions coupled with the stuttering behaviors significantly impaired the social relationships and interactions of children who stutter.

Van Riper (1971) delineated motoric fluency breaks into core and secondary behaviors. Core behaviors are the basic, involuntary behaviors of stuttering. They include repetitions, prolongations and blockages. Repetitions can be in the form of word repetitions ("my, my, my house") or part-word repetitions ("da da da daddy"). Prolongations, which appear developmentally after repetitions and involve more effort on the part of the speaker, are characterized by a continuation of airflow without movement of the articulators ("mmmmmmy name"). Blockages, which usually appear last in development and are indicative of excessive tension and effort on the part of the speaker, involve a total stoppage of the airflow without movement of the articulators. Secondary behaviors are learned as a means to escape from or avoid the stuttering event. Examples of secondary behaviors can include eye-blinking, head-twitching or inserting extra units of sound to get the stuttered word out ("I, um, um want").

Guitar (1998) suggested that the stuttering child can be differentiated from the nonstuttering child by a frequency of stuttering equaling less than 10% of a nonstuttering child's words and more than 10% of a stuttering child's words. Zebrowski (1995) compared a number of studies examining the frequency of stuttering behaviors and found that children who stutter were twice as disfluent as children who do not stutter, and the children who stutter consistently stuttered on more than 10% of words produced. However, it would be unwise to use frequency as the sole criterion of whether a child is

stuttering. Several studies have indicated that although the majority of nonstuttering children fall under the 10% benchmark, a percentage of nonstuttering children show more than 10 disfluencies per 100 words (Yairi, 1993). For example, Yairi and Ambrose (1999) identified a subgroup of disfluent children who demonstrate high levels of formulative and motoric disfluencies prior to age three. However, these children tended to quickly "recover," regardless of the severity of their behaviors and fall below the 10% level.

Any definition of stuttering must include more than these basic, behavioral descriptions. Van Riper (1971) defined stuttering as, "a word improperly patterned in time and the speaker's reaction thereto" (p. 12). The second part of Van Riper's definition is what sets stuttering apart from normal disfluency. Although a person who stutters experiences a relatively high frequency of disfluency, typically motoric in nature, and has stuttering behaviors with longer durations than those produced by nonstutterers, it is the reactions to the disfluencies that set stuttering apart. Andrews and Harris (1964) described stuttering as a pattern of disruption where the frequency and abnormality of the behavior must be significant enough to "cause distress" to the speaker or the intended audience. Nonstuttering children, even when their disfluencies are apparent to others, are generally unaware of the fluency breaks in their own speech and rarely react with distress or attempt to hide them (Guitar, 1998). Children who stutter are typically aware of the fluency breaks in their own speech and react to them. At first they may feel frustrated and embarrassed and believe that their stuttering generates negative reactions from conversational partners (Bloodstein, 1987; Starkweather, 2002). As their stuttering persists they can develop a deeper aversion to speaking and begin to use escape or

avoidance or secondary behaviors, when they anticipate stuttering (Guitar, 1998). These avoidance behaviors may include limiting eye contact, switching words, avoiding certain sounds and words, using shorter sentences, or avoiding any speaking situation where they anticipate stuttering. Other common secondary behaviors that may develop are: facial grimacing, increased tension in the mouth, face or neck, head-twitching, body-stiffening, taking deep gulps of air before speaking, hand/foot tapping and pitch rises. Guitar (2006) separated normal disfluency, borderline stuttering and beginning stuttering according to the factors presented in Table 1.

Table 1. Cross comparison of Normal Disfluency, Borderline Stuttering and Beginning Stuttering (Guitar, 2006).

Normal Dislfluency	**Borderline Stuttering**	**Beginning Stuttering**
Disfluencies account for less than 10% of syllables or words	Disfluencies account for more than 10% of syllables or words	Disfluencies account for more than 10% of syllables or words
Majority of disfluencies are formulative in nature	Majority of disfluencies are Stuttering-like and include repetitions, blocks and prolongations	Stuttering-like behaviors may become rapid and abrupt with increased tension and speeding up
Repetitions consist of 1-2 extra units in duration (b-b-baseball)	May have duration greater than 2 extra units	Patterning may include pitch rises and difficulty starting airflow or phonation
Limited awareness of the behaviors	Disfluencies may cluster together	Frustration leading to secondary behaviors
No outward reactions		Occasional avoidances of words or situations

Although average speakers do not continually monitor their speech for fluency, speakers can increase their attention for a specific speech parameter in an effort to prevent a potentially disruptive event (Levelt, 1993). This frequently occurs when speakers monitor their speech for syntactic, pragmatic, and linguistic appropriateness. When speakers detect that an error has occurred, they typically interrupt the discourse immediately following the troublesome word or within one or two words (Levelt, 1993). It is not typical for a speaker to interrupt themselves during or prior-to the production of the troublesome word. Following the interruption, the speaker employs a systematic repair in an effort to maximize comprehension for the listener. Speakers can also attempt to repair an utterance without the typical stop and start procedure. However, this type of operation often sacrifices the syntactic integrity of the utterance in consideration of time or to uphold the overall fluency level (Levelt, 1993).

Given that people who stutter typically consider their stuttering to be a negative trait, it is not uncommon for them to want to hide it from the listener. In an effort to accomplish this, people who stutter frequently monitor their speech for the presence of stuttering behaviors and enact repair techniques during and/or before the production of the disfluent unit. Following the detection of a troubling event, the person who stutters uses a variety of techniques to hide the behaviors that are deemed negative. These techniques, which frequently include circumlocutions, increased tension and word substitutions, are an effort to avoid the impending frustration and embarrassment that accompany the stuttering (Bloodstein, 1995). This is in stark contrast to the repairs employed by non-stuttering speakers in an effort to increase the listener's ability to follow the intended utterance. The person who stutters often inserts behaviors extraneous

to the intended utterance in an attempt to uphold the overall fluency level without consideration for the listener's ability to infer the intended message.

Stuttering and Tourette Syndrome

The phenomenological profiles of TS and stuttering share many commonalities (Comings & Comings, 1994; Donaher, 2002; Tetnowski & Donaher, 2003; Maguire, Riley, Franklin, & Gottschalk, 2000; Pauls, et al., 1993; Van Borsel & Vanryckeghem, 2000; Wu, Maguire, Riley, Lee, Keator, Tang, Fallon, & Najafi, 1997). For example, both disorders have higher concordance rates within monozygotic twin pairs than within dizygotic twin pairs, suggesting a primary genetic transference. Twin studies have also demonstrated that both disorders demonstrate a large environmental component, because a number of identical twin pairs were discordant. Both TS and stuttering are situational and episodic in nature and both occur in boys more than girls by a ratio of roughly 3-5:1 (Maguire et al., 2000; Pauls et al., 1993; Singer & Walkup, 1991; Wu et al., 1997). Both disorders have symptoms occurring during childhood and are exacerbated by stress, increase in severity over time, and can be managed or controlled temporarily with a great deal of effort (Bloodstein, 1995; Fenichel, 1997; Menkes, 1997; Shapiro, 1999; Singer & Walkup, 1991).

These similarities led researchers to question whether stuttering and TS share a corresponding neuropathology possibly involving the circuitry of the basal ganglia (Alm, 2005; Caruso, 1991; Maguire et al., 2000; Wu et al., 1997). The basal ganglia**,** due to its location, functioning and interconnection with a variety of neural areas**,** plays a significant role in the regulating of motor behaviors, emotions and cognition. One of its

primary roles is to provide the internal timing cues for the submovements underlying complex or well-learned motor sequences (Alm, 2005; Caruso, 1991). If a breakdown were to occur in the basal ganglia circuitry, the internal timing cues for such tasks could be affected. Kent (1994) proposed that stuttering is a disorder of timing and occurs when the brain is unable to properly time the sequential movements of speech. He supported this theory with the well known phenomena of providing external temporal cues, such as reading in chorus, speaking with a metronome or singing, to reduce the frequency of stuttering.

It is interesting to note that people who stutter have shown deficits in both speech and non-speech motor movements. For example, studies have shown reduced accuracy and speed on tasks involving the coordination of laryngeal musculature, lip closing, finger and hand tapping and reaction times for people who stutter (Caruso, 1991; Max & Gracco, 2005). While this line of research is still in the early stages, it does raise the possibility that stuttering may be the result of a centralized motor deficit and may not be purely speech specific. If this were true, it may further support the idea that the underlying deficit in stuttering may be related to the basal ganglia circuitry.

Abwender and colleagues (1998) hypothesized that if stuttering and TS are pathogenetically related, people who stutter would demonstrate other manifestations of basal ganglia dysfunction including abnormal movements such as tics or dystonia. They hypothesized that a large proportion of people who stutter may represent undiagnosed cases of TS where the speech disorder is the most troublesome characteristic warranting intervention. To investigate this possibility, they conducted a study of 22 people who stutter attending a residential camp for stuttering. The participants included ten children

(1 girl and 9 boys) between 8 and 16 years of age, and 12 adults (2 women and 10 men) between 17 and 48 years of age. Only one of the stuttering children initially presented with a comorbid diagnosis of TS. Case history interviews and a battery of psychometric tests were administered to investigate a variety of neuropsychiatric features commonly seen in TS. The investigators also reported on any observable movements that may be indicative of basal ganglia dysfunction. Results indicated that 40% of the children who stutter and 58% of the adults who stutter displayed definite or probable motor tics.

Mulligan, Anderson, Jones, Williams and Donaldson (2003) compared 300 word spontaneous speech samples and 300 word reading samples from 16 adults who stutter and 16 controls matched for age and gender. Their study analyzed the involuntary movements commonly employed by people who stutter. The results indicated that the adults who stuttered produced significantly more involuntary movements, predominantly simple and complex motor tics. Furthermore, the results demonstrated that complex motor tics were more frequent than simple motor tics for the participants who stuttered but not for the controls. They found that these behaviors, which typically involved the facial muscles, included: repetitive eye blinking, extended eye closure, eyebrow raising, and extraneous jaw movements. Thus, they concluded that secondary stuttering behaviors could be classified as either simple or complex motor tics.

Unfortunately, neither Abwender et al. (1998) nor Mulligan et al. (2003), differentiated between motor tics and secondary or associated stuttering behaviors, which are specific reactions used to hide or escape from stuttering episodes. The authors could have addressed this problem by indicating whether these tics occurred specifically during speech tasks, as would be the case with secondary stuttering behaviors, or whether they

occurred during non-speech tasks, as would be the case with true motor tics. Alm (2005) further argued against the classification of secondary stuttering behaviors as tics by suggesting that people who stutter do not generally feel a "premonitory craving" prior to stuttering as experienced by individuals with tic disorders. In addition, people who stutter rarely report a sense of physical soothing after releasing a stuttering behavior as reported by people with tic disorders. Unfortunately, neither study mentioned above nor the existing stuttering literature have explored this possibility.

It is commonly accepted that TS involves a hyper-dopaminergic state in the basal ganglia circuitry which results in decreased inhibition of motor behaviors (Bruun & Bruun, 1994). This has been supported by the effectiveness of certain medications, dopamine D2 receptor blockers in particular, in the reduction of tic severity in patients with TS. It has also been suggested that increased levels of dopamine may be responsible or at least contributory in stuttering. Studies have reported significantly higher levels of dopamine and/or higher levels of dopamine uptake in individuals who stutter when compared to non-stuttering controls (Wu et al., 1997). This led to studies exploring the effectiveness of dopamine receptor antagonists, especially those commonly used in the treatment of TS, on stuttering (Brady, 1991; Brunn & Brunn, 1994; Maguire et al., 2000; Wu et al., 1997). Many of these early medications were effective in reducing the severity of stuttering behaviors but the unwanted side effects significantly outweighed the benefits and the trials were discontinued (Brady, 1991). In addition, considering that both stuttering and TS are developmental disorders, the long-term implications of using such medications with children prevented their wide spread use.

Speech Literature on Stuttering and Tourette Syndrome

Studies cited previously from the neurology literature suggested a higher prevalence of stuttering in individuals with TS based on epidemiological similarities. However, several recent studies from the speech and language literature have not reached the same conclusions. In fact, these studies have suggested that while individuals with TS present with higher than would be expected levels of disfluent speech, the pattern of disfluency is not indicative of true stuttering (DeNil, Sasisekaren, Van Lieshout, & Sandor, 2005; Donaher, 2002; Van Borsel, Goethals & Vanryckeghem, 2004; Van Borsel & Vanryckeghem, 2000).

Van Borsel and Vanryckeghem (2000) conducted a study of an 18 year-old male with TS who presented with disfluent behaviors. It should be noted that the disfluencies reportedly began when the participant was 14 years of age. Furthermore, the participant demonstrated low-normal intelligence with a significant gap between his verbal and performance IQs (WAIS VIQ = 82, PIQ = 94). The participant was hospitalized for aggressive behavior and amphetamine, alcohol and cocaine abuse and was subsequently referred during that stay for a speech and language assessment. Speech samples were collected in a variety of speech modalities including automatic speech, word/sentence repetition, oral reading and conversational speech. The authors found that only 15% of the participant's disfluent behaviors pretreatment could be considered actual stuttering. These behaviors included part-word repetitions and monosyllabic word repetitions. However, 85% of his disfluent behaviors were consistent with disfluencies commonly produced by nonstutterers. These consisted of multisyllabic word repetitions, interjections, sentence revisions and incomplete phrases.

Following seven 30-minute therapy sessions over a period of three weeks, which focused on reduced speech rate and increased awareness of tics, Van Borsel and Vanryckeghem (2000) reported a significant reduction in overall disfluencies (32.3% to 7.3%). However, the distribution of types and frequency post therapy was the same as that before therapy. Interjections and phrase repetitions accounted for 69% of total disfluencies while part-word and monosyllabic word repetitions accounted for less than 15% of the total disfluencies. The authors suggested that their participant may have been demonstrating behaviors consistent with cluttering; a disorder which affects both speech and language functioning and has been described as the verbal manifestation of a central language disorder. Cluttering rarely appears by itself and frequently occurs with stuttering. Its primary characteristics include: rapid, dysrhythmic, unorganized and frequently inarticulate speech which goes largely unnoticed by the individual (Daly, 1993).

Donaher (2002) conducted a pilot study of 10 children diagnosed with TS between the ages of 8 years, 3 months to 15 years, 3 months. During routine visits to the Neurology Department at a children's hospital in a large urban area, all of the children had complained of stuttering or stuttering-like behaviors. A standard set of conversational prompts about school performance, family issues and current speech functioning were used to elicit spontaneous utterances. Results showed that only three children demonstrated stuttering-like behaviors (i.e. single-syllable word repetitions, part-word repetitions, sound prolongations or blocks) during the evaluation. However, eight of the ten produced typical disfluencies (i.e. interjections, phrase repetitions and sentence revisions) which significantly interfered with the forward flow of speech. It is also worth

noting that nine of the children presented with decreased eye contact and a rapid rate while speaking. In accordance with the findings of Van Borsel and Vanryckeghem (2000), this pilot study suggested that the disfluencies demonstrated by children with TS are not indicative of a true stuttering disorder but may be consistent with disfluencies commonly produced as part of a cluttering disorder.

Van Borsel, Goethals and Vanryckeghem (2004) described the speech fluency patterning of two Dutch speaking boys (9;11 and 12;7 years) and one Dutch speaking girl (12;2 years) with TS who additionally presented with fluency concerns requiring speech therapy. One male participant was currently medicated but the specific medication was not named. Neither of the other participants were medicated at the time of the study. Samples were collected during a variety of speech tasks including: automized speech, word/sentence repetition, final-word repetition, reading, conversation, monologue, picture naming, and singing. In addition, the Communication Attitude Test (CAT) (Brutten & Vanryckeghem, 2003) was administered to each participant. All three participants presented with increased disfluencies during conversation, monologue and picturing naming tasks. None of the participants presented with disfluencies during singing or final word repetition and only one participant presented with disfluencies during the automatized speech task. The vast majority of the disfluencies demonstrated by each participant (71.5%, 69.7% and 67.5%) were between-word disfluencies (multi-syllable whole word repetitions, phrase repetitions, interjections, revisions and incomplete phrases). Interjections, repetitions and revisions were the most predominant type of disfluency for all participants and prolongations and blocks were not demonstrated by any of the participants. The authors also reported that speech patterning

of their participants did not conform to the characteristic behaviors of palilalia which has been typically reported regarding individuals with TS. Results from the CAT indicated that although two of the participants demonstrated a negative speech related attitude, only one scored above the mean average for people who stutter. The authors, in conclusion, suggested that the speech patterning of individuals with TS is a unique disfluency pattern which warrants further investigation.

DeNil, Sasisekaren, Van Lieshout, and Sandor (2005) analyzed the frequency and type of disfluencies in 69 children with TS ranging in age from 4 to 18 years (mean = 12.49, SD = 3.32). All of the children were diagnosed and followed at a TS clinic at a large metropolitan hospital. Eighty-two percent of the children with TS were taking a variety of medications including neuroleptics, stimulants, and/or anticonvulsants at the time of the study. Additionally, only 10% of this group presented with pure TS whereas 12% presented with TS and OCD, 28% presented with TS and ADHD and 45% presented with TS, OCD and ADHD. No statistically significant differences were found between the children with TS only and children with TS plus co-existing disorders on speech fluency measures. Therefore, all of the children with TS were pooled into a single group. Twenty-nine control participants were recruited from the general medical clinic at the same hospital. Their ages ranged from 6 to 17 years of age (Mean = 10.9, SD = 3.08). Results revealed no significant differences in less typical disfluencies (sound repetitions, prolongations and blocks) during reading or conversation between the groups. Children with TS did demonstrate significantly higher frequencies of more typical disfluencies (word repetitions, filled/unfilled pauses, hesitations, and interjections) during conversation. No significant differences were found in more typical disfluencies during

reading between the groups. The authors suggested that their finding support the idea that although TS and stuttering do not share a common etiology, both disorders may have similar impacts on the overall stability of the speech fluency system.

There are several issues which must be addressed in the above mentioned studies prior to generalizing the results to the entire TS population. First, all of the participants mentioned in these studies, with the exception of those from the DeNil et al. (2005) study, initially presented with stuttering behaviors or fluency concerns which resulted in them seeking an evaluation and/or therapy by a speech language pathologist. Thus, these participants may represent a subgroup of children with TS who happen to demonstrate co-exiting stuttering. Given the incidence of stuttering, it would stand to reason that some children with TS would also present with stuttering behaviors. This should not necessarily suggest that the two conditions are related. Second, three of the studies were retrospective case reviews with sample sizes of one, three and ten participants with no matched references. The nature of this sort of investigation does not provide the rigorous control or generalizability of a systematic, prospective study. Third, all of the studies recruited their participants from specialized clinics, tertiary medical centers or support groups for people with TS. These facilities and groups may not be available universally or be accessible to all individuals with TS which again raises the question of generalizability. Fourth, none of the studies controlled for comorbid conditions or for the impact of medications which may significantly affect an individual's ability to produce speech fluently.

Statement of Purpose and Research Questions

A significant body of literature has explored the relationship between TS and a variety of behavioral, psychological, learning and neuropsychological disorders. Most notably, TS has been linked with Obsessive Compulsive Disorder, Attention Deficit Hyperactivity Disorder, Learning Disabilities, Dyslexia, Anxiety Disorders and Depression. In contrast, the literature regarding the speech characteristics of children with TS is limited at best. However, given the clinical, genetic, behavioral, and neurological similarities between stuttering and TS, a systematic analysis of the speech fluency of children with TS is timely. By defining the clinical speech symptomatology of TS, the study would add insight into possible intervention strategies and etiological considerations for the disfluencies associated with this multidimensional, neurological disorder. Such a study could also contribute to the knowledge base of stuttering by suggesting avenues for future research which support or refute theories of stuttering based on existing knowledge of TS.

The present study represents an initial step in gathering normative data on the fluency characteristics commonly demonstrated by children diagnosed with TS. The goal of the study is a descriptive analysis of the fluency characteristics of children diagnosed with TS, in comparison to children who stutter and to typically developing peers, to determine if children with TS demonstrate higher levels of disfluency than children in the other two groups and to examine whether their patterning resembles stuttering. Specifically, the study analyzed speech samples in an effort to characterize the groups according to the frequency and characteristics of any disfluent behaviors. The following research questions were explored in this study:

Is there a significant difference in the frequency of disfluency in the speech of children with TS when compared to children who stutter and typically developing peers?

Is there a significant difference in the pattern of the disfluency demonstrated by children with TS when compared to children who stutter and typically developing peers?

Hypothesis

The null hypothesis states that there are no differences in the frequency or types of disfluency demonstrated by children with TS, children who stutter and typically developing peers. However, it was expected that the null hypothesis would not be upheld. It was predicted that children with TS would demonstrate patterns of disfluency which were qualitatively and quantifiably different from those demonstrated by children who stutter or those demonstrated by typically developing peers. In particular, it was hypothesized that children with TS would demonstrate significantly higher total disfluency levels when compared to typically developing peers but significantly lower total disfluency levels when compared to children who stutter. Additionally, it was hypothesized that children with TS would demonstrate significantly lower proportions of stuttering-like disfluencies when compared to children who stutter and significantly higher proportions of stuttering-like disfluencies when compared to typically developing peers.

CHAPTER 3
METHODS

Participants

Participants in this study included eight children, clinically diagnosed by a pediatric neurologist as meeting the Diagnostic and Statistical Manual IV (DSM-IV) criteria for TS, eight children who stutter with no diagnosis of TS and eight typically developing children who do not meet the criteria for either stuttering or TS. The children ranged in age from 7 to 14 years (mean = 10.79, SD = 2.06). In an effort to compare groups in terms of age, an ANOVA was computed to ensure equivalency. The results yielded no statistically significant difference (F (2,21) = .695, p = .510) in age between groups. In addition, effect sizes were calculated using Cohen's d statistic to describe the magnitude of any group differences in age. A small effect size between children with TS and children who stutter (d = .10) and a medium effect size (d = .5) between children with TS and typically developing peers were found. All participants were native English speakers with English being the only language spoken in the home. All participants presented with no significant history of psychological, audiological, neurological or medical problems other than those already associated with the diagnostic features of TS.

Participants from all ethnic and racial groups were recruited from a pediatric neurologist specializing in TS, speech-language pathologists specializing in stuttering and related fluency disorders and from an elementary school in the Philadelphia metropolitan area. The various professionals distributed materials to the parents of children who met the inclusion criteria, describing the study and requesting participation. Parents who agreed to participate were asked to sign informed consent for their child's participation,

and the children were asked to sign assent once the project had been described in detail to them by the investigator.

Inclusion Criteria

Children With Tourette Syndrome

Participants with TS were recruited from a single neurologist specializing in TS. In order to be included in the TS group, each participant had to have a current diagnosis of TS based on the DSM-IV (APA, 1994) criteria and had to be referred by the diagnosing neurologist. In an attempt to control for the possible effects of medication, only children with TS who were not currently taking medications were included. Participants with a significant family history of speech-language disorders, learning disability, social disorders, Obsessive-Compulsive Disorder or Attention Deficit-Hyperactivity Disorder were not excluded from the study because these diagnoses frequently co-occur with TS.

The mean age of the TS participants was 10.5 years (SD = 2.32). There were six males and two females included in the sample. As for coexisting diagnoses: one participant presented with Attention Deficit-Hyperactivity Disorder, one with Obsessive-Compulsive Disorder, one with a diagnosed Learning Disability and one with Attention Deficit-Hyperactivity Disorder and Obsessive-Compulsive Disorder. One participant had a history of speech and language therapy aimed at increasing his expressive and receptive language abilities. No participants had been treated for stuttering or fluency related concerns. None of the participants were currently taking medication, although seven had taken medication in the past to address tic severity. None of the parents nor any of the

participants themselves, reported side effects regarding speech production. Participants' age, gender and accompanying diagnoses are summarized in Table 2.

Children Who Stutter

Participants who stutter were recruited from the clinical caseloads of speech language pathologists specializing in stuttering and fluency related disorders. In order to be included in the group of children who stutter, participants were free from any co-existing neurological, cognitive-linguistic, or emotional conditions not typically coexisting with stuttering. Additionally, stuttering participants needed to demonstrate a stuttering severity rating of Moderate or greater according to the diagnostic criteria of The Stuttering Severity Instrument –3 (Riley, 1994) during their initial evaluation. This tool bases severity on a combination of factors including: the frequency of stuttering-like behaviors, average duration of the stuttering behaviors and any physical or concomitant behaviors. Children with mild stuttering were excluded from this study given the difficulty in properly diagnosing mild stuttering and the increased risk that disfluent but not truly stuttering children would be included in the sample.

The mean age of the children who stutter was 10.25 years (SD = 2.43). There were six males and two females included in the sample. None of the participants presented with coexisting diagnoses. All of the participants were either currently enrolled or had been enrolled in speech therapy to address their stuttering. No

Table 2. Participants' age, gender and accompanying diagnoses

Participant Number	Group	Gender	Age	Accompanying Diagnoses
1	TS	Male	12 Years	ADHD
2	TS	Male	10 Years	
3	TS	Male	14 Years	LD
4	TS	Male	10 years	
5	TS	Female	9 Years	OCD, ADHD
6	TS	Male	7 Years	OCD
7	TS	Female	13 Years	
8	TS	Male	9 Years	
9	CWS	Male	12 Years	Moderate Stuttering
10	CWS	Male	10 Years	Moderate Stuttering
11	CWS	Male	14 Years	Severe Stuttering
12	CWS	Male	10 years	Moderate Stuttering
13	CWS	Female	10 Years	Severe Stuttering
14	CWS	Male	8 Years	Moderate Stuttering
15	CWS	Female	12 Years	Very Severe Stuttering
16	CWS	Male	7 Years	Very Severe Stuttering
17	TDP	Male	13 Years	
18	TDP	Female	11 Years	
19	TDP	Male	14 Years	
20	TDP	Male	12 years	
21	TDP	Female	11 Years	
22	TDP	Male	9 Years	
23	TDP	Female	12 Years	
24	TDP	Male	10 Years	

Tourette Syndrome (TS), Children who stutter (CWS), Typically developing peers (TDP). Obsessive Compulsive Disorder (OCD), Attention Deficit Hyperactivity Disorder (ADHD), Anxiety Disorder, Learning Disabilities (LD). Stuttering behaviors classified according to The *Stuttering Severity Instrument –3* (Riley, 1994)

participants were currently taking medication. Participants' age, gender and accompanying diagnoses are summarized in Table 2.

Typically Developing Peers

Participants for the peer control group were recruited from a grade school in the Philadelphia vicinity which boasts a diverse student population with representation from many cultures, ethnicities and levels of socio-economic status. In order to be included in the peer group, participants were: (a) free from any neurological, cognitive-linguistic, academic or emotional diagnoses, (b) free of parental or teacher concerns regarding their ability to clearly and fluently produce speech, (c) categorized as nonstuttering during the initial interaction with the evaluator, and 3) free of any significant family history of stuttering or TS.

The mean age of the peer group was 11.62 years (SD = 1.68). There were five males and three females included in the sample. None of the participants presented with coexisting diagnoses nor were any currently enrolled in speech therapy. No participants were currently taking medications of any sort. Participants' age, gender and accompanying diagnoses are summarized in Table 2.

Instrumentation

Data collection was conducted in a standard hospital examination room with each participant seated across the table from the clinician. Sessions lasted approximately 15 minutes. All participants were recorded using a JVC Digital Camcorder GRDVL320 focused on the child's head and upper body. To enhance audio quality a small

microphone, Sony TCS-30D, was placed approximately 3 feet from the participant's mouth. A table-top microphone was chosen over a body-worn microphone to avoid the possibility of physical movements interfering with the audio signal.

Procedures

Data Collection

The clinician orally reviewed the intake questionnaire with the parents of each participant at the start of the research appointment. Specific emphasis was placed on demographic information, medical history concerning concomitant disorders/diagnoses and academic performance. The information collected during this process was used to assess criteria for inclusion and to ensure group comparability. No participants were deemed as not meeting the criteria for inclusion, thus none were excused prior to their participation.

To determine fluency levels, speech samples were collected during a narrative task prompted by the wordless picture book, A Boy, A Dog, and A Frog (Mayer, 1967). The book's plot centers around a boy and his dog attempting to catch a frog that is sitting in a pond. After several unsuccessful attempts, the boy decides to leave with his dog and return home. The frog then follows the boy home, enters into the house and joins the boy and the dog who are playing in the bath. Each participant was read the following directions:

> Thank you for agreeing to participate in the study. The study is designed to look at how people communicate. Our goal is to identify a pattern so that we will be better able to help children in the future. These forms (assent and consent) go into more detail about the study and describe your rights as a participant. I want to emphasize that everything that we do is confidential and you have the right to stop

participating at any time. I want you to read it over carefully and sign it if you agree to participate. Any questions?

We are going to have you complete one simple task. It involves creating a story from a set of pictures that I will show you. I want to remind you that there are no right or wrong answers and you are not being graded on your responses. So, no matter what you do, you are doing great! Before we begin, do you have any questions?

This is a picture book with no words. I would like you to make up the story. You will look at each page and describe what you see. When you are done with a page, you can either look up at me or tell me to turn to the next page. Ok lets begin. The story starts with, Once upon a time there was a boy and a dog. Now lets make up the rest of the story. (the researcher now shows the child each of the 29 pages of the book in sequence, turning the page when signaled by the child).

The clinician was not required to provide any prompting and/or cueing to stimulate lengthier speech samples since all participants produced well over 100 syllables. Speech samples ranged from 192 syllables to 666 syllables with a mean of 360.25 syllables (SD = 126.4). In an effort to compare groups in terms of sample length, an ANOVA was computed to ensure equivalency. The results yielded no statistically significant difference (F (2,21) = .230, p = .797) in sample length between groups. Additionally, there were no group differences in mean length of utterance in words (F (2,21) = 2.96, p = .074) or number of different words (F (2,21) = .650, p = .532). Table 3 summarizes measures of syllables per sample, mean length of utterance in words and number of different words.

Table 3. Group means for syllables per sample, mean length of utterance in words and number of different words.

	Syllables per sample		MLU-W		NDW	
Group	M	SD	M	SD	M	SD
TS	367.37	85.59	13.02	2.0	101.50	17.68
CWS	378	158.62	12.30	3.65	90.87	25.09
TDP	335.37	137.80	10.05	1.44	88.87	27.53

Tourette Syndrome (TS), Children who stutter (CWS), Typically developing peers (TDP).

Transcription of Speech Sample

Each of the samples was viewed and all utterances, including all disfluencies, were orthographically transcribed verbatim into a Microsoft Word database. Extraneous behaviors that were visible but inaudible, like motor tics, were described at the locations where they occurred. An utterance was defined as a series of words or clauses that (a) communicated an idea, (b) were set apart by pauses and (c) were bound by a single intonation contour (Logan and LaSalle, 1999; Meyers and Freeman, 1985). It has been demonstrated that native speakers of a language can reliably identify the intended utterance of a speaker from the actual utterance produced (Levelt, 1993). The transcriber was permitted to listen to a segment up to three times for clarity of transcription. Unintelligible utterances or those that did not meet the above mentioned criteria were excluded from the sample.

Each disfluency was then coded at the point where it occurred based on the classification system established by Yairi and Ambrose (1992). This system differentiates between stuttering-like disfluencies and other disfluencies. This measure has been shown to be sensitive to fluctuations in stuttering over time and highly

correlated with measures of severity (Yairi, Ambrose & Nierman, 1993). Although the Yairi and Ambrose (1993) protocol includes both prolongations and blocks under the category titled disrythmic phonations, the current study chose to distinguish between these two behaviors. A summary of this classification system and the specific codes used in the current study are summarized in Table 4.

Table 4. Classification system (Yairi and Ambrose, 1992) for types of disfluency, examples and codes from the present study.

Measure	Specific Type	Example	Code
Stuttering - Like Disfluencies	Part-Word Repetitions	b-b-boys are loud	PWR
	Single Syllable Word Repetitions	My, my, my red hair	SSWR
		I, I, I like soup	
	Prolongations	Weeee will go home	PRO
	Blocks	My b-(pause) aby doll	BLO
Other Disfluencies	Interjections	I like (um) spicy food	INT
	Revisions	I want some, I need, give me some candy	REV
	Multi-syllable Word Repetitions	Summer, Summer is hot	MSR
	Phrase Repetitions	My name is, my name is Bob	PHR

Total Disfluency Level

Total disfluency levels were calculated by dividing the total number of disfluencies in each sample by the total number of syllables in the sample. When a syllable contained more than one disfluency, each event in the cluster was counted as a separate disfluency. For example, the sentence *W-W-W-------- hen are we going,* contains 1 disfluent syllable (W-W-W-------- hen) and 4 fluent syllables (are we going). However, it contains 2 stuttering events, a part-word repetition (W-W) and a prolongation (W---). Thus, that sentence would obtain a total disfluency level of .4 or 40%.

Frequency Per Type of Disfluency

Each disfluency was coded to account for the specific type of disfluency produced. The number of each disfluency type produced in the sample was divided by the total number of syllables in the sample to tabulate an overall frequency for each specific type of disfluency. When a syllable contained more than one disfluency type, each event in the cluster was counted as a separate disfluency.

Proportion of Stuttering-Like Disfluencies

All disfluencies were categorized as either stuttering-like or other disfluencies based on the classification system discussed earlier. The number stuttering-like disfluencies was divided by the total number of disfluencies produced in that sample to tabulate the proportion of stuttering-like disfluency.

Reliability

Three months after the initial analyses were completed, the author randomly chose and reanalyzed one complete video sample from each group (12.5%) to assess intrajudge reliability. Interjudge reliability was obtained using a second reviewer, also a certified speech-language pathologist specializing in stuttering and fluency related disorders. The second reviewer was trained by the author in the calculation methods and categorization procedures. Video samples and transcripts which included all utterances, including any behaviors extraneous to the intended message, were provided. The second reviewer then analyzed the same 12.5% of the samples, one from each group, for total disfluency level and frequency for each specific type of disfluency.

Reliability for total disfluency level was calculated by determining the percentage of agreement between raters and between consecutive ratings by the author. For interjudge reliability, this was accomplished by dividing the smaller score obtained by one rater by the larger score obtained by the second rater. This ratio was then multiplied by 100 to obtain an interjudge score of agreement of 94.2%. For intrajudge reliability, this was accomplished by dividing the smaller score obtained by the author by the larger score obtained during a consecutive review by the author. This ratio was then multiplied by 100 to obtain an intrajudge score of agreement of 90.3.

Reliability for each specific type of disfluency was calculated by determining the percentage of agreement on a point by point basis. Interjudge reliability was calculated by examining each disfluent event to see whether the raters agreed or disagreed as to the type of disfluency. Then the total number of agreements were divided by the total number of disagreements plus the total number of agreements. This ratio was then

multiplied by 100 to yield an interjudge score of agreement of 85.6%. Intrajudge reliability was calculated by examining each disfluent event to see whether the author agreed or disagreed as to the type of disfluency on consecutive reviews. Then the total number of agreements were divided by the total number of disagreements plus the total number of agreements. This ratio was then multiplied by 100 to yield an intrajudge score of agreement of 88.3%.

It should be noted that previous studies utilizing similar protocols have demonstrated low interjudge and intrajudge reliability, especially when calculating agreement for specific types of disfluencies (Einarsdottir & Ingham, 2005). Thus, it is not surprising that reliability calculations for total disfluency level in the present study were higher than reliability calculations for specific types of disfluencies.

Statistical Analysis

Mean scores were compared and a series of univariate ANOVAs were conducted using SPSS for Windows, Version 11.0 to determine if significant differences in the type and frequency of disfluencies were evident between children with TS, children who stutter and typically developing peers. The dependent variables included: (1) total disluency level, (2) frequency per type of disfluency, and (3) proportion of stuttering-like disfluencies. Given that a series of ANOVAS were performed, there was an increased risk for Type 1 errors. To control for type 1 errors, the alpha level of $\underline{p} = 0.05$ was divided by the number of tests performed (8) to calculate an adjusted alpha level of $\underline{p} = .006$. When variances were deemed significantly different, post hoc testing was completed using the Fisher LSD (Least Significant Difference) Test with alpha set at $\underline{p} =$

.05 to determine between which groups the differences exist. It should be noted that since prolongations and blocks were not demonstrated by any of the children with TS or the typically developing peers, statistical procedures were not calculated regarding the frequency of these specific types of disfluency. However, these disfluencies were factored into the analysis of total disfluency level since they represent a major behavior demonstrated by individuals who stutter.

In addition to calculating statistical significance, effect sizes were calculated using Cohen's d statistic to describe the magnitude of any group differences. The general convention for the interpreting effect sizes, according to Cohen (1988), were used and included: small effect (.2 to .49), medium effect (.5 to .79), and large effect (greater than .8).

CHAPTER 4
RESULTS

This study compared the disfluencies of children with TS, children who stutter and a control group of typically developing peers. Two experimental questions were proposed in the study. The first question explored whether there is a significant difference in the frequency of disfluencies in the speech of children with TS when compared to children who stutter and typically developing peers. The second question was to determine whether there was a significant difference in the types of disfluencies demonstrated by groups. The null hypotheses stated that there are no differences in the frequency or types of disfluency demonstrated by children with TS, children who stutter and typically developing peers. However, it was expected that the null hypotheses would not be upheld. It was predicted that children with TS would demonstrate patterns of disfluency which were qualitatively and quantifiably different from those demonstrated by children who stutter and those demonstrated by typically developing peers. In an effort to answer these questions, group means were compared for total disfluency level, frequency per type of disfluency, and proportion of stuttering-like disfluencies. Group means, standard deviations and significance levels are shown in Table 5.

Table 5. Means, standard deviations and significance levels per group for total disluency level, frequency per type of disfluency, and proportion of stuttering-like disfluencies.

	Group			Sig. Level
	TS (N = 8)	CWS (N = 8)	TDP (N = 8)	p Value
Total Disfluency Level	4.10(2.98)	8.22 (4.17)	2.08 (.82)	.002*
Frequency Per Type of Disfluency				
Part-Word Repetitions	3.57 (2.82)	7.0 (3.82)	1.33 (.57)	.040
Prolongations	-	8.4 (8.17)	-	-
Blocks	-	2.0 (.00)	-	-
Single-Syllable-Word Repetitions	2.5 (2.13)	5.12 (5.54)	.75 (1.75)	.070
Interjections	6.0 (7.3)	10.5 (13.18)	4.42 (2.76)	.457
Revisions	3.14 (2.11)	3.66 (3.88)	1.71 (1.11)	.371
Multi-syllable Word Repetitions	.12 (.35)	.37 (.51)	.25 (.71)	.662
Phrase Repetitions	4.0 (3.93)	4.85 (7.33)	1.5 (.71)	.783
Proportion of Stuttering-Like Disfluencies	37.81 (16.79)	65.25 (23.22)	15.83 (21.88)	.000*

Note: None of the children with TS or the typically developing peers demonstrated prolongations or blocks. * indicates statistical significance with an adjusted alpha level of $p < .006$.

Total Disfluency Level

The first question was to determine whether children with TS demonstrated high levels of disfluency in their speech when compared to children who stutter and typically developing peers. Group means are presented in Table 5. As was expected, ANOVAs computed to compare total disfluency levels between groups yielded a statistically significant difference ($F(2,21) = 8.66$, $p = .002$). Post hoc analysis of significant

subgroup effects revealed that children with TS produced significantly fewer total disfluencies than children who stutter (p = .012, d = 1.13). No significant difference in total disfluency level was found between children with TS and typically developing peers (p = .393). However, children with TS were twice as disfluent as their typically developing peers and a large effect size of .92 was obtained. Not surprisingly, children who stutter were significantly more disfluent than the typically developing peers (p = .001, d = 2.04).

Disfluency Types

The second question was to determine whether significant differences exist in the specific types of disfluency employed by children with TS when compared to children who stutter and typically developing peers. Analyses were conducted on the frequency of the total sample occupied by each type of disfluency and on the proportion of total disfluencies categorized as stuttering-like behaviors.

Frequency Per Type of Disfluency

ANOVAs were computed to compare the percentage of the total sample occupied per type of disfluency between groups. Group means are presented in Table 5. The results yielded no statistically significant differences. Although not statistically significant, trends were evident between the groups. For example, children with TS produced approximately half as many part-word repetitions (p = .059, d = 1.02) and single-syllable-word repetitions as children who stutter (p = .157, d = .62). Additionally, children with TS produced approximately twice as many part-word repetitions (p = .316,

d = 1.10), phrase repetitions (p = .623, d = .89) and sentence revisions (p = .307, d = .85) as their typically developing peers. Moderate to large effect sizes were obtained for all of these comparisons. It should also be noted that blocks and prolongations were not demonstrated by any of the children with TS or the typically developing peers.

Proportion of Stuttering-Like Disfluencies

All disfluencies were categorized as either stuttering-like or other disfluencies based on the classification system discussed in the previous chapter. The number of disfluencies in each category was then divided by the total number of disfluencies produced in that sample to tabulate the proportion of total disfluency occupied by each category. Group means are presented in Table 5. The results yielded a statistically significant difference ($F(2,21) = 11.31$, $p = .000$). As was predicted, children with TS produced significantly lower proportions of stuttering-like disfluencies than children who stutter ($p = .015$). A Cohen's d of 1.35 was obtained, indicating a large effect size. Additionally, children with TS produced significantly higher proportions of stuttering-like disfluencies than their typically developing peers ($p = .047$). A Cohen's d of 1.12 was obtained, again indicating a large effect size. As would be expected, children who stutter presented with significantly higher proportions of stuttering-like disfluencies than their typically developing peers ($p = .000$). A Cohen's d of 2.19 was obtained, again indicating a large effect size.

CHAPTER 5
DISCUSSION

Given the continued debate regarding a possible relationship between stuttering and TS, the purpose of this study was to provide a descriptive analysis to better understand the speech fluency patterning of children with TS. Specifically, the goal was to determine whether children with TS demonstrated higher than would be expected total disfluency levels and to examine whether their patterning resembled stuttering. Such an investigation could significantly contribute to our understanding of TS by further defining the clinical symptomatology that comprises the disorder and possibly offering insight into further research and intervention endeavors.

At the start of this project, it was hypothesized that children with TS would present with a significantly higher total disfluency level than typically developing peers but a significantly lower total disfluency level than children who stutter. The results of the study revealed that the children with TS were, in fact, half as disfluent as participants who stuttered. The children with TS presented with disfluencies ranging from 1.7% to 9.5% of the speech sample. On average, they were approximately 4% disfluent. This is significantly less than the children who stutter who presented with disfluencies ranging from 2.6% to 13% with an average disfluency level of 8%. These findings are in line with the findings from Zebrowski (1995) which showed that children who stutter are twice as disfluent as children who do not stutter. Although not statistically significant, children with TS were twice as disfluent as the typically developing peers. The typically developing peer group presented with disfluencies ranging from .9% to 3% with an average disfluency level of 2%.

The second hypothesis predicted that children with TS would present with qualitative differences in their pattern of disfluencies. To that end, children with TS were expected to demonstrate significantly lower proportions of stuttering-like disfluencies when compared to children who stutter. This hypothesis was supported as the children with TS demonstrated 38% stuttering-like disfluencies versus 65% for the children who stutter. Interestingly, while 75% of the children who stutter exceeded the 50% threshold, only one participant in the TS group presented with proportions of stuttering-like disfluencies which exceeded that mark. That same child, who presented with 68% stuttering-like disfluencies, had a total disfluency level of 7.4% which is just under the mean for the group of children who stutter. It is quite possible that in addition to TS, that child was truly stuttering which may have skewed the overall numbers for the TS group. As mentioned earlier, given that approximately, 1% of the general population stutters, it should not be surprising that a small percentage of children with TS demonstrate co-existing stuttering. The typically developing peer group demonstrated 16% stuttering-like disfluencies and none of the participants exceeded the 50% threshold. Interestingly, blocks and sound prolongations, which are a hallmark feature of stuttering, were nonexistent in the TS and typically developing peer groups whereas 65% of the children who stutter demonstrated prolongations and 25% presented with blocks.

In summary, the results of this study suggest that children with TS present with an atypical disfluency pattern which may be a fundamental factor in the cluster of symptoms that comprise the disorder. Furthermore, the findings suggest that the speech fluency pattern of children with TS can be differentiated from that of children who stutter and

typically developing peers based on the total disfluency level and the proportion of stuttering-like disfluencies.

Integration with the Current Literature

A significant debate has continued over the years regarding a possible relationship between TS and stuttering. Much of the discussion has been fueled by the many clinical, neurological and epidemiological similarities between the two disorders. While it has been commonly believed that children with TS often present with higher frequencies of stuttering than would be expected in the general population, the literature base has failed to substantiate this belief. As discussed earlier in the literature review, two studies appear to be at the center of this notion. The first conducted by Pauls, Leckman and Cohen (1993) reported on a sample of 85 individuals diagnosed with TS between the ages of 7 and 62 years using a survey methodology and patient report on a variety of speech and language issues. The second study, conducted by Comings and Comings (1994), also utilized a retrospective, survey methodology to determine the prevalence of stuttering in the TS population. Taken together, the results suggested that between 15 and 31% of individuals with TS stutter or have stuttered in the past.

Contrary to the findings of Pauls et al. (1993) and Comings and Comings (1994), the results of the current study support more recent findings which have suggested that the pattern of disfluency seen in individuals with TS is not indicative of true stuttering (DeNil, Sasisekaren, Van Lieshout, & Sandor, 2005; Donaher, 2002; Van Borsel, Goethals & Vanryckeghem, 2004; Van Borsel & Vanryckeghem, 2000). Furthermore, the results support the idea that children who do not stutter can be reliably differentiated

from children who stutter based on total disfluency levels and the proportion of stuttering-like behaviors (Pellowski & Conture, 2002). To this end, both children with TS and typically developing peers in the present study significantly differed from the group of children who stutter based on these factors.

If the atypical speech fluency pattern demonstrated by children with TS is not indicative of true stuttering, then speculation regarding a possible etiology is warranted. The first hypothesis would implicate the circuitry of the basal ganglia and the neurotransmitter dopamine. As mentioned previously, the basal ganglia provides timing cues for automatic, well-learned, sequential motor acts. This process is vulnerable to fluctuation in dopamine levels. Low levels of dopamine are thought to inhibit the release of cortical impulses generated by the substructures of the basal ganglia to the pre-motor areas.

Conversely, high levels of dopamine are thought to disinhibit the release of cortical impulses generated within the basal ganglia and are theorized to be the basic mechanism responsible for motor tics. If a series of neural cues were released simultaneously, they would have the potential to flood a specific motor system, which could result in the system freezing momentarily. If multiple timing cues for a specific speech motor plan were released simultaneously, they could have the potential to freeze the pre-motor speech system momentarily. This could result in a gap in the speech motor plan and, as explained by Howell and Au-Yeung (2002), could cause the system to stall in an effort for the incomplete plan to catch up. One way that the system can stall, is to retrieve the plan of the previous sound, word or phrase and re-execute it until the system is ready to move forward (Howell & Au-Yeung, 2002).

The notion that disfluent behaviors can result from momentary freezing due to defective execution of the motor sequences for speech has been explored in Parkinson's disease. In fact, Benke, Hohenstein, Poewe, and Butterworth (2000) investigated the disfluencies demonstrated by 53 patients with Parkinson's disease. Their findings revealed that 48% of the disfluencies demonstrated by their sample could be described as repetitive speech phenomena characterized by 1-2 well articulated iterations of syllables, words or phrases produced with a constant rate and loudness. The authors proposed that these disfluencies were the result of "a malfunction at the level of motor speech, probably resulting from a disintegration of subcortical interplay, whereby the basal ganglia stimulate speech activity in the cortex, in an uncontrollable manner."

This motor-based theory is intriguing in light of the results from the current study. Children with TS demonstrated significantly higher proportions of repetitive speech behaviors when compared to typically developing peers. On average, 59% of the disfluencies demonstrated by the children with TS were repetitive iterations of sounds, words or short phrases. In addition, 50% of these children presented with repetitive behaviors which accounted for more than 73% of their total disfluencies. In contrast, 21% of the disfluencies demonstrated by the typically developing peers were repetitive speech events. However, it should be noted that only two individuals in the peer group presented with repetitive speech events which exceeded the 15% threshold.

Descriptively, these repetitive behaviors: 1) consisted of a single iteration of sounds, words or phrases, 2) ranged between 1 and 3 syllables in length, 3) occurred in various positions throughout the sentence, 4) contained no apparent change in rate or effort, 5) lacked pausing between repeated units, and 5) incurred no apparent reaction

from the speaker. Consequently, the repetitive speech behaviors demonstrated by the children with TS in the current study were similar to tics which as discussed in chapter one, are repetitive, rapid, involuntary movements or vocalizations.

An alternative hypothesis would suggest that the atypical speech patterning of children with TS may be secondary to an underlying cognitive-linguistic impairment. Similar to the gaps mentioned above which were considered secondary to an incomplete motor plan, impairments in an individual's ability to quickly organize and formulate language could also result in the system using a strategy to gain extra time to formulate the linguistic plan (Howell & Au-Yeung, 2002; Postma & Kolk, 1993). Interestingly, research has identified that various types of language breakdown can significantly influence whether an utterance will be produced fluently and the type of disfluency that will be produced (Scott Trautman, Healey & Norris, 2001). Specifically, it has been shown that when an individual struggles with formulating concepts or syntactic complexity, they are more likely to produce phrase repetitions. When an individual struggles with word finding, they are more likely to produce whole word repetitions, and part word repetitions have been associated with phonological impairment (Guo, Tomblin, & Samelson, 2008; Postma & Kolk, 1993; Howell & Au-Yeung, 2002).

This hypothesis could be supported by 2 findings mentioned earlier. The first relates to the relationship between TS and expressive language deficits. Even though there is a paucity of research in this area, several studies mentioned earlier have demonstrated that children with TS present with expressive language deficits involving the organization, formulation and expression of higher level language (Legg, Penn, Temlett, & Sonnenberg, 2005; Ludlow, Polinsky, Caine, Bassich & Ebert, 1982;

O'Quinn & Thompson, 1980). Furthermore, it has been documented that language impairment is present in the majority of children with ADHD (Giddam, 1991) which is the most frequently coexisting diagnosis with TS. Research has demonstrated an inverse relationship between language impairment and disfluencies resulting in children with lower language abilities demonstrating more disfluencies (Guo, Tomblin, & Samelson, 2008).

The second finding which implicates cognitive-linguistic ability is the high incidence of learning disability in children with TS. Learning disabilities can be characterized by a variety of symptoms which represent a marked disparity between measured cognitive ability and academic achievement (Healey, Reid, & Donaher, 2005). Language problems associated with children with learning disabilities can include difficulties with phonological awareness, delayed speech development, difficulty with grammar and syntax, vocabulary deficiencies, and difficulty comprehending spoken language (Healey, Reid, & Donaher, 2005; Lerner, 2000). Unfortunately, documentation of the disfluency characteristics of children with learning disabilities in the literature is limited. However, given the strong correlation with a variety of language impairments, it would not be surprising to find increased disfluencies in children with learning disabilities.

A third hypothesis would suggest that the atypical fluency pattern of children with TS may be secondary to a combination of factors which vary greatly for each individual. These factors could include an individual's speech motor capacities, expressive and receptive language abilities, cognitive functioning, attention and focusing skills, temperamental profile and various other factors which have been considered part of the

behavioral spectrum which defines TS. In addition, the number of factors may multiply depending on whether the individual demonstrates coexisting diagnoses like ADHD, OCD or learning disability. These factors, either individually or in combination, could affect the integrity of the speech motor system which could result in an increased probability of fluency breakdowns. Smith (1999), in her multifactorial model, suggested that the stability of the speech motor system fluctuates over time and can be directly affected by a number of factors such as sentence length and linguistic complexity. As a result of this individual variability, it may be difficult to describe a specific speech pattern for all individuals with TS. Thus, differential diagnosis which takes into account all areas of functioning for children with TS is optimal for effective management strategies to be designed and enacted.

Clinical Implications

From a clinical perspective, the identification of coexisting diagnoses in children with TS is vitally important because these subgroups of children will require a different type of treatment than those who demonstrate TS alone. This suggestion is particularly relevant considering the findings of Freeman, Fast, Burd, Kerbeshian, Robertson, and Sandor (2000) which indicated that the presence of significant disfluencies, questionably labeled as stuttering by the authors, was twice as likely in TS patients with comorbid conditions than in patients with TS alone. The authors speculated that presence of significant disfluencies may be indicative of "more widespread neurodevelopmental pathology." For this reason, before designing intervention plans, speech language pathologists should consider the impact of each diagnosis on the child's functional ability

to communicate. Clinicians can then determine where to best steer their intervention efforts based on the specific deficits being experienced by the individual.

Gaining a better understanding of how a child's ability to communicate is affected by various factors begins with a detailed case history including medical, social and developmental information. Clinicians would benefit from coordinating reports and suggestions from other professionals who have worked closely with the family. These individuals include classroom teachers, special education providers, neurologists, social workers and pediatricians. In addition, psycho-educational testing can provide invaluable information on a child's overall cognitive abilities while providing insight on ways to promote increased learning and suggestions for minimizing specific areas of weakness. Following that, a well-designed, thorough speech and language assessment is essential. This should include a combination of standardized and non-standardized testing protocols including various speech modalities and levels of difficulty. If possible, it should include multiple speech samples with a variety of communicative partners. Once the clinician has identified the child's unique characteristics, it is time to design a treatment program which addresses the individual needs of the child and their family. Most children significantly benefit when there is a structured intervention plan which has been coordinated with their family, classroom teacher and entire academic and medical team. This allows everyone to use similar words when discussing things, to coordinate rules, regulations and expectations and to better understand how various factors interact and effect performance in other areas.

Specific goals that speech language pathologists often need to address with children with TS include: (1) To assist the family and classroom teacher in creating an optimal environment to encourage the child's fluent speech, (2) To increase language formulation and organizational skills through functional activities which promote more active learning, (3) To address basic attention and focusing skills which underlie language learning and include topic maintenance skills and retention of information presented verbally, (4) To increase the child's ability to effectively monitor their speech, and (5) To teach timing cues like pausing and clustering which may promote decreased disfluencies while allowing increased time for planning.

Healey, Reid, and Donaher (2005) offered a variety of strategies for addressing fluency concerns in the presence of coexisting learning, behavioral, and cognitive challenges. While not all strategies will be pertinent for all children with TS, they provide a starting point for clinicians. The strategies suggest that intervention should incorporate systematic and gradual approximations of behaviors which eventually extend to outside environments and provide opportunities to master strategies in a controlled fashion. It is recommended that the therapist explain the goals on the child's level with clear and concise directions. In addition to using concrete examples and visual and tactile cues, the therapist should initially address the child's awareness of when and how speech is being interrupted and the individual's ability to self-monitor during running speech. Motivation can be increased with the use of a consistent reward system but the therapist must ensure that the child fully understands why they are being rewarded. Finally, the clinician should attempt to include topics that are interesting and motivating for the child to keep them engaged in therapy.

Future Studies

This study should be replicated with a larger sample of individuals with TS prior to generalizing any results to the entire population. The primary limitation of the current study related to the limited sample size which, due to the low incidence of TS, is a common concern with research on this topic. Given that only 8 participants were included in each group, statistical power was limited. For this reason, effect sizes, which are not generally affected by sample size, were reported. However, the major restriction with the sample size involved not being able to stratify the TS group based on a variety of factors.

With a larger sample, future studies could analyze subgroups of children with TS based on the presence of comorbid diagnoses including: ADHD, OCD, LD and a variety of conditions which frequently co-exist with TS. By segregating the TS group according to concomitant diagnoses, it may be possible to determine whether the findings of the current study relate to the presence of TS or are secondary to one or more comorbid conditions. This suggestion is particularly relevant considering the findings of Freeman, Fast, Burd, Kerbeshian, Robertson, and Sandor (2000) which were mentioned earlier and indicated that the presence of significant disfluencies may be indicative of a more significant neurodevelopmental pathology.

From a clinical management perspective, the effects of medication on the fluency pattern of individuals with TS needs further investigation. The current study excluded children who were taking medications but considering that a variety of pharmacological agents have been determined to affect fluency, it would be prudent to investigate this topic more in the future. Additionally, pharmacological treatments addressing the

various symptoms of TS remain the first line of defense and need to be systematically evaluated for possible speech-based side effects. Of particular interest would be an exploration of psychostimulant medications which remain the most effective single intervention for children with ADHD. Given the high comorbidity between TS and ADHD, plus the research linking stimulant medications with an increased occurrence of motor tics in children with no history of tics and an exacerbation of motor tics in children with tic disorders (Packer, 1997), this study could significantly aid in the management of TS. The lack of well-controlled studies describing the effects of psychostimulant medications on speech fluency make it difficult to draw any conclusions regarding a possible relationship. However, several clinical studies have shown that the use of stimulant medications significantly increased disfluency levels in some individuals (Burd & Kerbeshian, 1991; Lavid, Franklin and Maguire, 1999; Riley and Riley, 2000).

In the future, longitudinal designs may assist with tracking possible changes throughout the lifespan. The present study included a narrow age range centering on school-aged children with TS. By limiting the participant pool to this age range, it was impossible to comment on the possible relationship between TS symptoms and the development of speech and language functioning. This may be especially important given the high degree of learning disability, expressive language delay and variability across time of TS symptoms. Furthermore, considering that the fluency patterns of typically developing children change over time, it is plausible to suggest that fluency patterns of children with TS would also change over time.

Further exploration of the findings relating to the high proportion of repetitive speech events in individuals with TS is warranted. Particularly, the speech patterns of

individuals with TS should be compared to those of individuals with other neurologically based disorders, especially those like Parkinson's disease which also involve the neurotransmitter dopamine and the circuitry of the basal ganglia. Although comparative studies based on perceptual accounts of speech fluency would be informative, future studies should also incorporate neuroimaging techniques in an attempt to explore the neurobiological basis of the various disfluent behaviors which have been linked with these disorders.

Given that significantly more boys present with TS than girls, it may be interesting to explore the role of gender in the speech patterning of children with TS. The current study, by including more females in the typically developing peer group, may have unwittingly biased the results. Future studies, especially those employing larger sample sizes, should investigate whether differences exist between boys and girls who present with TS.

Finally, the current study recruited children with TS from a specialty TS clinic at a large, metropolitan children's hospital by a neurologist who specializes in the disorder. Considering that the average child with TS is not managed in this sort of specialized environment, it is possible that the caseload represents individuals with the most severe TS symptoms. Additionally, children who stutter were recruited from a specialty care center that specializes in fluency and fluency related disorders. Again, it is possible that the caseload of that clinic is not truly representative of the average child who stutters. Evidence of this, may be derived from the fact that all of the stuttering participants demonstrated moderate or greater stuttering severity according to the Stuttering Severity Instrument –3 (Riley, 1994). These recruitment practices may have inadvertently

resulted in a sampling bias which may restrict the generalizability of the findings. Future studies should employ efforts to recruit a more heterogeneous sample of children.

REFERENCES

Abwender, D.A., Trinidad, K.S., Jones, K.R., Como, P.G., Hymes, E., & Kurlan, R. (1998). Features resembling Tourette's Syndrome in developmental stutterers. *Brain and Language, 62,* 455-64.

Alm, P.A. (2005). On the Causal Mechanisms of Stuttering. Lund University, Sweeden, 2005.

Ambrose, N.G., & Yairi, E. (1999). Normative disfluency data for early childhood stuttering. *Journal of Speech, Language, and Hearing Research, 42*, 895-909.

American Psychiatric Association. (1994). Diagnostic and statistical manual of mental disorders (4th ed.). Washington, D.C.: Author.

Andrews, G., & Harris, M. (1964). The Syndrome of Stuttering. London: Spastics Society Medical Education and Information unit in association with W. Heinemann Medical Books

Benke, T., Hohenstein, C., Poewe, W., & Butterworth, B. (2000). Repetitive speech phenomena in Parkinson's disease. *Journal of Neurology, Neurosurgery, and Psychiatry.* Sep; 69 (3): 319-24.

Bloodstein, O. (1995). A Handbook on Stuttering. (5th ed.). San Diego: Singular.

Brady, J. P. (1991). The pharmacology of stuttering: A critical review. *American Journal of Psychiatry, 148*, 1309 – 1316.

Brutten, G.J., & Dunham, S. (1989). The Communication Attitude Test: A Normative Study of Grade School Children. *Journal of Fluency Disorders, 14*, 371-377.

Bruun, R.D., & Bruun, B. (1994). A mind of its own. Tourette's Syndrome: A story and a guide. New York. NY: Oxford University Press.

Caruso, A.J. (1991). Neuromotor Processes Underlying Stuttering. In H.F.M. Peters, W. Hulstijn, and C.W. Starkweather (Eds.), Speech motor control and stuttering. Amsterdam: Elsevier. Pg. 27-42.

Coffey, B.J., Biederman, J., Smoller, J.W., Geller, D.A., Sarin, P., Schwartz, S., & Kim, G.S. (2000). Anxiety disorders and tic severity in juveniles with Tourette's Disorder. *Journal of American Academy of Child and Adolescent Psychiatry, 39*(5), 562-8.

Cohen, J. (1988). Statistical power analysis for the behavioral sciences (2nd ed.). Hillsdale, NJ: Lawrence Earlbaum Associates

Comings, D.E. (1995). Tourette's Syndrome. A behavioral spectrum disorder. In W.J. Weiner and A.E. Lang (Eds.). Behavioral Neurology of Movement Disorders (Vol. 65.) [Advances in Neurology Series]. New York, NY: Raven Press.

Comings, D.E., & Comings, B.G. (1994). TS, learning and speech problems. *Journal of the American Academy of Childhood Adolescent Psychiatry, (33)* 429.

Daly, D. A. (1993). Cluttering: Another fluency syndrome. In R.F. Curlee (Ed.) Stuttering and Related Disorders of Fluency. New York, NY: Thieme Medical Publishers.

De Nil, L.F., Sasisekaran, J., Van Lieshout, P.H., & Sandor, P. (2005). Speech disfluencies in individuals with Tourette Syndrome. *Journal of Psychosocial Research.* Jan; 58(1):97-102.

Donaher, J.G. (2006). Tourette Syndrome and Stuttering. Perspectives on Fluency and Fluency Disorders, Vol. 16. 2, 5-6.

Donaher, J.G. (2002). Fluency and Tourette Syndrome. Unpublished pilot data analysis.

Einarsdottir, J., & Ingham, R. J. (2005). Have disfluency-type measures contributed to the understanding and treatment of developmental stuttering? *American Journal of Speech-Language Pathology, 14* 260-273.

Fenichel, G.M. (1997). A signs and symptoms approach. Clinical Pediatric Neurology. (3rd ed.).

Freeman, R., Fast, D.K., Burd, L., Kerbeshian, J., Robertson, M., & Sandor, P. (2000). Tourette Syndrome International Database Consortium. An international perspective on Tourette Syndrome: selected findings from 3500 cases in 22 countries. *Dev Med Child Neurol* 42: 436–447.

Giddan, J. (1991). School Children with emotional problems and communicative deficits: Implications for speech-language pathologists. *Language, Speech, and Hearing in the Schools, 22*, 291-295.

Guitar, B. (1998). Stuttering: An Integrated Approach to Its Nature and Treatment, 2nd Edition. Baltimore, MD: Williams and Wilkins.

Guitar, B. (2006). Stuttering: An Integrated Approach to Its Nature and Treatment, 3rd Edition. Baltimore, MD: Williams and Wilkins.

Guo, L., Tomblin, J.B., & Samelso, V. (2008). Speech Disruptions in the Narratives of English-Speaking Children with Specific Language Impairment. *Journal of Speech, Language, and Hearing Research, 51*: 722-738.

Healey, E.C., Reid, R., & Donaher, J. (2005). Treatment of the child who stutters with co-existing learning, behavioral, and cognitive challenges. In R. Lees and C. Stark (eds.) Treatment of the school-age child who stutters. London: Whurr Publishers Ltd.

Howell, P., & Au-Yeung, J. (2002). The EXPLAN theory of fluency control and the diagnosis of stuttering. Pathology and therapy of speech disorders. In Fava, E. (ed). Current issues in linguistic theory series. Amsterdam: John Benjamins. pp. 75–94.

Jankovic, J., (1997). Neurologic Clinics: TS. Volume 15, Number 2. Philadelphia, PA: W.B. Saunders Company.

Kent, R. D., (1984). Stuttering as a temporal programming disorder. In R.F. Curlee, and W.H. Perkins (Eds), Nature and Treatment of Stuttering: New Directions. San Diego, College-Hill Press, pg. 283-301.

Legg, C., Penn, C., Temlett,.J., & Sonnenberg, B. (2005). Language skills of adolescents with tourette syndrome. *Clinical Linguistics and Phonetics. 19*(1):15–33

Levelt, W.J.M. (1998). Speaking: From intention to articulation. Cambridge, MA: MIT Press.

Ludlow, C., Polinsky, R., Caine, E., Bassich, C., & Ebert, M. (1982). Language and speech abnormalities in Tourette Syndrome. *Advances in Neurology. 35*:351–361

Maguire, G.A., Riley, G.D., Franklin, D.L., & Gottschalk, L.A. (2000). Risperidone for the treatment of stuttering. *Journal of Clinical Psychopharmacology*, Aug; 20(4):479-82.

Maguire, G.A., Riley, G.D., Franklin, D.L., Maquire, M.E., Nguyen, C.T., & Brojeni, P.H. (2004). Olanzapine in the treatment of developmental stuttering: a double-blind, placebo-controlled trial. *Annals of Clinical Psychiatry*, Apr-Jun;16(2):63-67.

Manning, W.H. (2001). Clinical Decision Making in Fluency Disorders (ed 2). San Diego: Singular.

Max, L., & Gracco, V. (2005). Coordination of oral and laryngeal movements in the perceptually fluent speech of adults who stutter. *Journal of Speech, Language, and Hearing Research, 48*, 524-542.

Mayer, M. (1967). A boy, a dog, and a frog. New York: Dial.

Menkes, J. (1997). Textbook of Child Neurology (5th ed).

Mulligan, H.F., Anderson, T.J., Jones, R.D., Williams, M.J., & Donaldson, I.M. (2003). Tics and developmental stuttering. *Parkinsonism Related Disorders, 9*(5), 281-289.

O'Quinn, A.N., & Thompson, R.J. (1980). Tourette's syndrome: an expanded view. *Pediatrics., 66*(3), 420-4.

Packer, L.E. (1997). Social and educational resources for patients with Tourette Syndrome. *Neurologic Clinics of North America, 15*(2), 457-471.

Pauls, D.L., Leckman, J.F., & Cohen, D.J. (1993). Familial relationships between Gilles de la Tourettes Syndrome, Attention Deficit Disorder, Learning Disabilities, Speech Disorders, and Stuttering. *Journal of the American Academy of Child Adolescent Psychiatry. 32*(5), 1044-1050.

Pellowski, M.W., & Conture, E.G. (2002). Characteristics of speech dysfluency and stuttering behaviors in 3- and 4-year-old children. *Journal of Speech, Language and Hearing Research, 45*, 20-23.

Pennsylvania Tourette Syndrome Association. (1990). Tourette Syndrome: Common symptoms. [checklist]. Gettysburg, PA: Pennsylvania Tourette Syndrome Association.

Postma, A., & Kolk, H. (1993). The covert repair hypothesis: Prearticulatory repair processes in normal and stuttered disfluencies. *Journal of Speech, Language, and Hearing Research, 36*, 472-487.

Richard, G.J., & Hoge, D.R. (1999). The Source for Syndromes. Moline, IL: LinguiSystems.

Riley, G. (1994). The Stuttering Severity Instrument for Children and Adults (3rd ed.). Austin, Texas: Pro-Ed.

Scott-Trautman, L. A., Healey, E. C., & Norris, J. (2001). Effects of contextualization on Fluency in three groups of children. *Journal of Speech, Language, and Hearing Research, 44*, 564-576.

Shapiro, D.A. (1999). Stuttering Intervention: A Collaborative Journey to Fluency Freedom. Austin, TX: Pro-Ed.

Singer, H.S., & Walkup, J.T. (1991). TS and other tic disorders: Diagnosis, pathophysiology, and treatment. *Medicine. 70*, 15.

Sisskin, V. (2006). Speech disfluency in Asperger Syndrome: Two cases of interest. *Perspectives on Fluency and Fluency Disorders, 16*, 12-14.

Smith, A. (1999). Stuttering: A unified approach to a multifactorial, dynamic disorder. In Research and Treatment of Fluency Disorders: Bridging the Gap. N. Ratner and C. Healey (Eds). Mahwah, NJ: Erlbaum, p. 27-44.

Spreen, O., & Benton, A.L. (1977). Neurosensory Center Comprehensive Examination for Aphasia. Victoria, BC: Neuropsychology Laboratory.

Starkweather, C.W. (1987). Fluency and stuttering. Englewood Cliffs, NJ: Prentice Hall.

Starkweather, C.W., Gottwald, S.R., & Halfond, M.H. (1990). Stuttering Prevention: A Clinical Method. Englewood Cliffs, NJ: Prentice Hall.

Starkweather, C.W. (2002). The epigenesis of stuttering. *Journal of fluency Disorders, 27*, 269-288.

Starkweather, C.W. (1987). Fluency and Stuttering. Englewood Cliffs: Prentice Hall.

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

Tetnowski, J. & Donaher, J. G. (2003). Disfluency associated with Tourette Syndrome: Two case studies. International Stuttering Awareness Day 2003 Online Conference. Mankato State University. Stutteringhomepage.com.

Van Borsel, J. & Vanryckeghem, M. (2000). Dysfluency and phonic tics in Tourette Syndrome: A case report. *Journal of Communication Disorders. 33*, 227-240.

Van Borsel, J., Goethals, L & Vanryckeghem, M. (2004). Disfluency in Tourette syndrome. *Folia Phoniatrica et Logopaedica, 56*, 358-366.

Wu, J.C., Maguire, G., Riley, G., Lee, A., Keator, D., Tang, C., Fallon, J., & Najafi, A. (1997). Increased dopamine activity associated with stuttering. *Neuroreport, 8,* 767-770.

Yairi, E. (1993). Epidemiologic and other considerations in treatment efficacy research with preschool age children who stutter. *Journal of Fluency Disorders, 18*, 197-219.

Yairi, E., & Ambrose, N. (1992). A longitudinal study of stuttering in children: A preliminary report. *Journal of Speech, Language, and Hearing Research, 35*, 755-760.

Yairi, E., & Ambrose, N.G. (1999). Early childhood stuttering I: Persistency and recovery rates. *Journal of Speech, Language, and Hearing Research, 42*, 1097-1112.

Yairi, E., Ambrose, N., & Niermann, R. (1993). The early months of stuttering: A developmental study. *Journal of Speech, Language, and Hearing Research, 36*, 521-528.

Yaruss, J.S., Newman, R., & Flora, T. (1999). Language and disfluency in nonstuttering children's conversational speech. *Journal of Fluency Disorders, 24,* 185-207.

Zebrowski, P. M. (1995). The topography of beginning stuttering. *Journal of Communication Disorders, 28*, 75-91.