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THE RELATIONSHIP BETWEEN VOCAL TICS AND MOTOR SPEECH CONTROL IN TOURETTE SYNDROME

By

Roya Sayadi

A DISSERTATION

Submitted to
Michigan State University
in partial fulfillment of the requirements
for the degree of

DOCTOR OF PHILOSOPHY

Department of Audiology and Speech Sciences

1992

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ABSTRACT

THE RELATIONSHIP BETWEEN VOCAL TICS IN TOURETTE SYNDROME AND MOTOR SPEECH CONTROL

By

Roya Sayadi

The purpose of this study was to investigate the relationship between the vocal tics in Tourette syndrome and motor speech control. Specifically, habitual utterances were compared with their paired experimental utterances which involved variations in pitch, loudness, rate and sentence prosody. To investigate the role of motor complexity, these variations were studied separately at the vowel and the sentence level.

Ten adults with Tourette syndrome voluntarily participated in this study. The experiment required repeating 26 utterance each three times in a systematic random order, producing a sample of 78 utterances for each subject. Pitch tasks required the subjects to produce the vowel /a/ with habitual, high, low, glide up, and glide down pitches. An 11 word sentence was also produced with subjects' habitual, high, and low pitches. Loudness tasks required the subject to produce both the vowel and the sentence stimuli with habitual, loud, soft, and whisper levels of loudness. Rate tasks required the subjects to repeat the vowel and the sentence with their habitual, fast,

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and slow rates. Lastly, prosody tasks required the subjects to repeat the sentence with a statement, question, happy, and angry intention.

Compared to the habitual vowel utterances, the glide up, glide down and the loud conditions were found to significantly increase the vocal tics. Compared to the habitual sentence, the soft and whispered sentences significantly decreased the vocal tics. Various rate and prosody conditions did not affect the vocal tics. Compared to the vowel productions, sentence productions significantly increased the vocal tics. Finally, a significant correlation between the severity of Tourette syndrome and performance on the experimental tasks was observed.

It was concluded that vocal tics appear to increase as the motor speech complexity increases. It appears that a relationship exists between the laryngeal adjustments for the rapid changes of pitch and laryngeal and respiratory adjustments for loudness. It is suggested that vocal tics may result from an intermittent lack of inhibition of the cortical voluntary systems over the reflexive systems during speech.

For their love and support in multiple ways, this work is dedicated to my mother, Mrs. F. Zia Sayadi, and my two daughters, Tara and Nika Makhmali.

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LIST (

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INTRO

METHO

TABLE OF CONTENT

SUBJE

RESUL

DISCU

SUMMA

APPEN

REFER

TABL

1

2

3

4

5

6

7

8

9

10

11

12

13

14

LIST OF TABLES

Figu

1

LIST OF FIGURES

Figure **Page**

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INTRODUCTION

History

The first descriptions of individuals with possible symptoms of Tourette syndrome can be found in the book Malleus Maleficarum (Hammer of the Witches). This book was a textbook of inquisition for identification and punishment of witches published by two Dominican priests, Sprenger and Heinrich Kramer, in the late fifteenth century (Shapiro and Shapiro, 1982). The authors described a priest who used to thrust his tongue out or cry out anytime that he kneeled before the Virgin Mary. Stevens (1971) reported a seventeenth century French nobleman Prince de Conde who stuffed his mouth to suppress an involuntary bark in the court of Louis XII. Generally, it was believed that these individuals were possessed by the devil and exorcism was used as treatment.

Another compelling example of a historical description of an individual with possible Tourette syndrome is Dr. Samuel Johnson. Samuel Johnson lived from 1709-1784 and was famous as a great poet, playwright, and biographer. Historians recorded his abnormal involuntary motor and verba

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verbal tics and his compulsive behavior. Recently, Murray (1982) studied recorded descriptions provided by friends and acquaintances of Dr. Johnson and suggested that he had been suffering from Gilles de la Tourette syndrome.

According to Gilles de la Tourette himself, the first clinical document of this disorder was reported by Jean-Marc-Gaspard Itard in the late nineteenth century. Although Itard is well known for his work with wild children and the education of deaf children, the first clear description of Tourette syndrome was written by him. Itard (1825) described the disorder in a French noblewoman, Marquise de Dampierre, who lived until 1884. She was also examined by Charcot, and her affliction was diagnosed as chorea with laryngeal and diaphragmatic involvement.

George Albert Edouard Brutus Gilles de la Tourette (1857-1904) was a 28-year-old student of Charcot at La Salpatriere, the "Mecca of Neurology." Tourette's assignment as the chief of the clinic was to reclassify movement disorders which at the time were grouped under the condition of chorea. It was during this period that Gilles de la Tourette found the opportunity to observe six cases whom he later described in his classic paper (1885) as having a disorder other than chorea (Goetz and Klawans, 1982).

Tourette extensively described a distinct disorder seen in nine cases. Six of them were personally observed by Tourette himself, and three of them were reported previously by ot

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by other physicians. Based on his careful clinical observations, Tourette accurately concluded that tics are completely a different class of abnormal movements and should be differentiated from chorea.

One year later, in 1886, Charcot honored his student by naming the described condition Gilles de la Tourette syndrome. The brilliant discovery of Tourette provided a clinical understanding for this affliction which is still acceptable in the twentieth century. The following quotation reflects Gilles de la Tourette's clear understanding of the disorder (Goetz and Klawans, 1982, pg. 10):

> Let us recall first some of the fundamental symptoms: 1) this illness is hereditary; it is characterized by motor incoordination in the form of abrupt muscular jerks that are often severe enough to make the patient jump; 2) the incoordination can be accompanied by articulated or inarticulated sounds. When articulated, the words are often repetitions of words which the patient may have just heard. Such vocal imitation (echolalia) may have a physical corollary whereby the subject imitates an act or gesture that he has just seen; 3) among the expression which the patient may repeatedly utter during one of his convulsions, some have the special character of being obscene (coprolalia); 4) the physical and mental health of these patients are otherwise basically normal. The condition seems incurable and life long, with onset in childhood.

However, in his paper, Tourette failed to differentiate the condition observed in his own six patients and those three described by other physicians: Latah (O'Brian, 1883), Jumping Frenchman (Beard (1880), and Miyrachit (Hommond, 1884)

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1884). In fact, he concluded that these three conditions were consistent with the disorder which he had described. This account is completely rejected today. These three conditions represent separate entities, and none of them present tic disorders in their symptomatology. The primary characteristic of these conditions is the unusual exaggerated startle response. In addition, tics in Tourette syndrome are spontaneous, a condition which is not characteristic of any of these other three disorders. Similarly, Tourette's emphasis on echolalia as the major feature of the Tourette syndrome and his proposition that vocalizations occur simultaneously with a motor tic elsewhere in the body are not currently accepted.

Tourette clearly emphasized that tics are an entirely separate class of abnormal movements which must be differentiated from chorea. This contribution was a major advance in the development of the classification of the movement disorders. Aside from some aspects of Tourette's descriptions which are revised today, his contribution still has its clinical value as a distinct disorder.

In early twentieth century Kinnier Wilson (1907) translated a monograph by Meige and Feindel's named Les Tics et leur Traitement. The monograph had a significant effect on the insight about the etiology of the Tourette syndrome. In this book, it was reassured that tics occur in emotionally immature and neurotic persons. An assertion which

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which was partly due to the lack of evidence for any neuroanatomical involvement. This view was strengthened with the dominant influence of the psychoanalytic theories. The syndrome was recognized as hysteria by psychoanalysts, and the concept of acknowledging the symptoms as a neurological disorder diminished (Shapiro and Shapiro, 1982). The syndrome was recognized as bad habits and was explained in the same way for the patients and their families.

Interest in Tourette syndrome dramatically increased with its successful pharmacological treatments in the mid-twentieth century. In 1961, Seignot in France and Caprini and Melotti in Italy independently reported the beneficial effect of treatment with haloperidol in patients with Tourette syndrome. Such success, once again, strengthened the notion of an organic etiology for the syndrome. In addition, Shapiro and co-workers (1978) reintroduced the disorder to the medical and health community with the strong emphasis on its physical bases. Also, the notion of organic etiology for Tourette syndrome became strengthened from the findings of pharmacological treatments for disorders other than Tourette syndrome. Sacks (1969) used the L-Dopa drug for treatment of patients with post-encephalitis lethargica. However, the drug resulted in a Tourette-like syndrome in patients who received it. Such observation further suggested the idea of an organic cause for Tourette syndrome.

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Gradually, the new scientific findings began to increase the public awareness of Tourette syndrome. Sacks (1971) published an article titled "Tics" in the Washington Post which aided in publicity of the disorder (Sacks, 1985). Eventually, in 1974, the new scientific achievements coupled with the increased public awareness led to the establishment of the American Tourette Syndrome Association.

Today, the studies of Tourette syndrome are based on controlled data collection and replication. Recent studies define Tourette syndrome based on relationships among behavioral, neurochemical, and genetic factors through multidisciplinary approaches. The disorder is recognized as a neuropsychiatric syndrome related to CNS involvement with a familial basis, childhood onset, fluctuation of symptoms, a life-long course, a male-to-female ratio of 3:1. Tourette syndrome has been suggested as a neuropsychiatric model for the relationship between emotions and motor control in human beings (Shapiro, Shapiro, Young, and Feinberg, 1988).

Prevalence

The prevalence of Tourette syndrome has been underestimated for a long time. The underdiagnosis of Tourette syndrome has been partly due to the lack of awareness and partly due to inadequate description of the disorde

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disorder. For a long time, the diagnosis of Tourette syndrome required the presence of associated symptoms of echolalia, coprolalia, and mental deterioration (Shapiro, Shapiro, Young, and Feinberg, 1988). Recent studies demonstrated increased prevalence of Tourette syndrome. DSM-III-R (Diagnostic and Statistical Manual, third edition, revised, American Psychiatric Association, 1987) estimates the lifetime prevalence rate of Tourette syndrome to be at least 0.5 per thousand. Based on its membership, the Tourette Syndrome Association estimates that there are 100,000 patients with Tourette syndrome in the U.S, about 0.046% of the U.S. population. The disorder occurs more frequently in males than in females, in an approximate 3,4:1 ratio (Lees, Robertson, Trimble, and Murray, 1984). Nevertheless, researchers believe that because patients with mild symptoms of Tourette syndrome tend not to seek help, epidemiological figures are only approximate because of the large number of undiagnosed cases.

The incidence of Tourette syndrome has been reported throughout the world, and it exists among various races and ethnic groups including western and northern Europeans, eastern European Ashkenazi Jews, Blacks, Hispanics of Mediterranean origin, and Orientals (Golden, 1982; Lieh-Mak, Chung, and Chen, 1982). Furthermore, Tourette syndrome does not belong to a particular social class, and it afflicts individuals with various socio-economic backgrounds (Asam, 1982 a

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Frequent references have been made to the similarity between Tourette syndrome and other tic disorders consisting of chronic motor tic disorder and transient tic disorders (Leckman and Cohen, 1988). However, consensus is that Tourette syndrome is a separate entity (DSM-III, 1980; and DSM-III-R, 1987). Chronic motor tic disorder is the most common tic disorder and is characterized with either motor or vocal tics but not both. Transient tic disorder is a childhood disorder characterized by one or more simple motor tics that are usually mild and has a duration of less than one year.

Symptomatology

Tics are often more easily recognized than precisely defined. Tics are divided into those that consist of involuntary movements (motor tics) and those that consist of involuntary sounds (vocal tics). Motor tics are sudden, rapid, repetitive, stereotyped motor movements (Leckman and Cohen, 1988). They can be characterized by their anatomical location, number, frequency, duration, intensity, and complexity. When motor tics involve the vocal apparatus to cause the production of sounds, the resulting noises are called vocal tics (Fahn and Erenberg, 1988). Vocal tics require

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require the same criteria as motor tics except that they are characterized as sounds, words, phrases, or sentences.

Although tics are defined as involuntary, they may be preceded by sensory urges causing them to occur (Shapiro, Shapiro, Young, and Feinberg, 1988). With self-observation, these sensory urges can be recognized and the tics can be controlled or substituted temporarily (Bliss, 1980). The diagnosis of Tourette syndrome can be determined when the vocal tics appear after the onset of motor tics (Fahn and Erenberg, 1988).

Age at onset for Tourette syndrome is before 21 with the mean age of 7 and the majority before age 14 (DSM-III-R, 1987). Abuzzahab and Anderson (1976) reported 15 patients whose initial symptoms were after the age of 20. The variability of age at onset is a source of a methodological problem in the study of Tourette syndrome, since it is difficult to find homogenous groups of Tourette syndrome subjects along this parameter.

The general progression of symptoms is cephalocaudal (Nomura and Segawa, 1982), although many exceptions are found. The first symptoms are usually eyelid blinking (Commings and Commings, 1984), facial twitching (Lieh-Mak et al., 1982), and neck movements (Lees et al., 1984). The occurrence of coprolalia as the initial symptom is rare (Shapiro et al., 1978). The movements are brief and explosive. Clearly, because of the wide variation of the

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initial symptoms among the afflicted individuals, the diagnosis of Tourette syndrome based on the initial symptoms is not appropriate.

Three consistent phases exist in the development of this syndrome. The first phase consists of simple motor tics, usually of the eye, face, neck, or shoulders. Frequently, the motor tics are misinterpreted as "nervous habits" or as childhood tics (Lacey, 1986). During the second phase, the motor tics are accompanied by non-speech vocal tics (Leckman and Cohen, 1988). Vocal tics often include throat clearing, sniffing, coughing, or barking sounds; and their presence is necessary for the diagnosis of Tourette syndrome (DSM-III, 1980, 1980; DSM-III-R, 1987). In addition, in some individuals the vocal tics may extend to coprolalia and less often echolalia in the later stage. Coprolalia is present in only 50% of patients with Tourette syndrome and is the most socially troublesome aspect of Tourette syndrome. Extended duration of the syndrome does not result in mental and neurological deteriorations nor is the outcome a psychotic one (Caine, 1985).

The frequency with which the symptoms of Tourette syndrome appear varies considerably. At times of high stress, fatigue, anger, and excitation, symptoms become worse (Leckman et al., 1988). In contrast, except in a minority of cases with Tourette syndrome (Burds and Kerbeshian, 1988), relaxation and sleep improve the symptoms

(DSM-III, 1980; Fahn, and Erenberg, 1988). The marked decrease of tics during sleep indicates that complete cortical activities are necessary for the full expression of tics (Shapiro et al., 1988).

Spontaneous changes in severity of symptoms characterize the syndrome (Shapiro et al., 1978), and there is no periodicity in this variation (Brunn, 1988). The disorder is lifelong with periods of remission lasting from weeks to years (DSM-III-R, 1987). Although the course is usually lifelong, arrest may occur at any stage (Lieh-Mak et al., 1982). Spontaneous, complete remission has also been reported (Shapiro et al., 1978).

Etiology

Currently, existence of an organic etiology is accepted and supported for Tourette syndrome. However, the disorder is referred to as a syndrome because its exact etiology is unknown and the diagnosis is based on its signs and symptoms.

Research findings have documented that factors such as childhood diseases, allergy, or psychological events are not related to the etiology of Tourette syndrome. Shapiro and co-workers (1988) reported no significant relation between any childhood illnesses and the onset of Tourette syndrome.

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They reported that the average age for maturational milestones for children with Tourette syndrome was within the normal expectations. However, informal observations indicated a slightly higher frequency of left handedness in patients with Tourette syndrome compared with the general population.

Some authors have speculated allergic bases for the etiology of Tourette syndrome. However, research findings of gender differences do not support this speculation for a number of reasons: (1) The predominance of males in Tourette syndrome is in contrast to the predominance of females with allergies (Lanier, 1985); (2) drugs that are useful with allergies (i.e., antihistamines) are not effective for the treatment of Tourette syndrome; and (3) drugs that are useful for the treatment of the Tourette syndrome (i.e., haloperidol or pimozide) are not helpful for allergies.

Traumatic psychological events, although found in a minority of cases (23%), have not been found to be a precipitating factor in Tourette syndrome (Shapiro et al., 1988). Examples of psychogenic factors include death of a family member, separation of a family member, birth of a sibling, and father's hospitalization.

Recent studies have shown that Tourette syndrome is a familial disorder in half of the afflicted individuals (Comings, Comings, Devor, and Cloninger, 1984). In addition, studies of identical (monozygotic) and fraternal (dizygotic)

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(Price, Kidd, Cohen, Pauls, and Leckman, 1985). Shapiro and co-workers (1988) reported that approximately 85% of identical twins with Tourette syndrome transmit the disorder to their children.

In 1961, the successful use of haloperidol therapy by Seignot resulted in studying Tourette syndrome from a pharmacological perspective. It is now well accepted that drugs that block postsynaptic dopamine receptors (e.g. haloperidol) tend to relieve symptoms, whereas drugs that increase dopaminergic activity (e.g., levadopa) acutely exacerbate symptoms (Devinskey, 1983; Sacks, 1985).

In conclusion, the exact organic etiology of Tourette syndrome remains unclear. Recent models of the etiology of Tourette syndrome incorporate genetic and pharmacological findings and suggest an interaction between these two variables. Pharmacological data indicate that Tourette syndrome is a disorder in which postsynaptic dopaminergic receptors are hypersensitive. Such findings suggest that abnormalities in the central dopaminergic system predispose genetically vulnerable individuals to expression of the disorder (Leckman et al., 1988).

Neurology

Many patients with Tourette syndrome show subtle neurological abnormalities, e.g., impairment of rapid alternative movements and hyperflexia (Sweet, Solomon, Wayne, Shapiro, and Shapiro, 1974; Lees et al., 1984). However, neurological findings are nonfocal, nonlocalizing, and do not indicate a consistent pattern among patients with Tourette syndrome. Therefore, although Tourette syndrome is an organic syndrome of the CNS, it is not primarily associated with a specific neurological sequelae (Shapiro et al., 1988). In addition, because of some similarities between Tourette syndrome and some other specific diseases of the CNS, differential diagnosis should include consideration of chorea (Shapiro et al., 1988), encephalitis lethargic (Sacks, 1982), Wilson's disease (Shapiro et al., 1988), head trauma (Fahn, 1982) and schizophrenia (Shapiro, Shapiro, Wayne, and Clark, 1972).

A minority of patients with Tourette syndrome present nonspecific EEG irregularities (Krumholz, Singer, Niedermeyer, Burnite, and Harris, 1983). However, short latency evoked potentials appear to be normal. Domino, Piggot, Demetrioue, and Culbert (1982) and Krumholz and associates (1983) demonstrated that visual and auditory evoked potentials in patients with Tourette syndrome do not differ from normals. On the contrary, long latency

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event-related potentials appear to be abnormal. A study of Contingent Negative Variation (CNV is a long latency event-related potential) in 18 patients with Tourette syndrome showed underactivity of right parietal involvement (Van Woerkom, Fortgens, Van de Wetering, and Martens, 1988).

Lees, Robertson, Trimble, and Murray (1984) reported normal CAT scan results in 53 patients with Tourette syndrome. However, a positive emission tomography (PET) study of five drug-free patients showed 16% increase of glucose utilization in the basal ganglia but no difference in overall cerebral glucose metabolism (Chase, Foster, Fedio, Brooks, Mansi, Kessler, and DiChiro, 1984).

Only three autopsies have been reported. Bing (1925) reported gross neuroanatomical findings characterized by meningitis thickening and adhesions in the region of the facial nerve. Bogaert (1940, cited in Shapiro, et al., 1988) performed a postmortem examination on a patient with Tourette syndrome. No evidence of gross or microscopic pathology was found. Finally, another patient was reported on two separate occasions by Balthasar (1954 and 1956, cited in Shapiro et al., 1988). This patient had neuronal loss in the third and fifth cortical layers and in the thalamus. The main findings were abnormalities in the caudate and putaman nuclei of the basal ganglia.

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In short, the basal ganglia and the substantia nigra are suggested to be the neuroanatomical regions associated with Tourette syndrome. Also, Devinsky (1983) suggested that Tourette syndrome is the result of altered dopaminergic function in the midbrain. The substantia nigra and ventral tegmental area (VTA) are adjacent midbrain sites rich in dopamine neurons.

Psychopathology

A normal IQ distribution among patients with Tourette syndrome has been reported (Shapiro et al., 1978). However, based on some neuropsychological examinations such as WAIS, WISC, and Bender-Gestalt results Shapiro and colleagues (1978) suggested that a right hemisphere dysfunction may exist in Tourette syndrome. Moreover, based on deficits in memory and copying of visually presented nonverbal material and reduced verbal fluency, Sutherland, Kolb, Schoel, Whishow, and Davies (1982) concluded that the right temporal, orbital frontal, and parietal lobe functions all show evidence of impairment.

The personal characteristics of patients with Tourette syndrome are expectedly diffuse. However, a review of literature indicates that patients with Tourette syndrome have a greater incidence of behavioral problems than the normal population. The most common behaviors frequently reported associated with Tourette syndrome include attentional disorders, hyperactivity, and obsessive-compulsive symptoms. Attentional problems and hyperactivity occur in about half of the patients with Tourette syndrome (Comings and Comings, 1988). Data regarding obsessive-compulsive disorder are controversial. While Shapiro and associates (1988) argued that the incidence of obsessive-compulsive disorder in Tourette syndrome was within the normal range, Frankel, Cummings, Robertson, Trimble, Hill, and Benson (1986) claimed that 51% of their patients with Tourette syndrome exhibited obsessive compulsive disorder.

Treatment

Both pharmacological and behavioral treatments have been employed in Tourette syndrome. However, the pharmacological therapy is the most effective treatment for this disorder. Medication which is used in pharmacological treatment of Tourette syndrome include haloperidol (Haldol), pimozide (Orap), and clonidine (Catapress). The treatment of choice is haloperidol, which results in marked improvement in the majority of patients with Tourette syndrome (Shapiro et al., 1978). The function of Haloperidol is to selectively block dopamine receptors in CNS. Clonidine is an antihypertension drug which has shown effectiveness in some individuals with Tourette syndrome (Brunn, 1984). Clonidine functions in reducing the tension and increasing the attention span. These effects assist in decreasing motor and vocal tics in some individuals with Tourette syndrome. Pimozide is another dopaminergics receptor blocker which also aids in the suppression of motor and vocal tics (Bruun, 1986). Pimozide is a therapeutic alternative for patients who do not respond well to haloperidol or clonidine.

It must be noted that the available medications only reduce the symptoms of the disorder rather than eliminating them (Bruun, 1986). All the medications used in the treatment of Tourette syndrome have the potential to cause side effects in some individuals. The side effects most often encountered are tardive dyskinesia (Riddle, Hardin, Towbin, Leckman, and Cohen, 1988), akinesia (Shapiro and Shapiro, 1988), and parkinsonism (Shapiro and Shapiro, 1988). In addition, cognitive impairments including drowsiness, impaired attention, reduced motivation, memory difficulties, and slow reaction time are limitations of the use of haloperidol especially in children (Shapiro and Shapiro, 1988). Furthermore, depression, dysphoria, and phobia may occur as side effects of very low dosages in susceptible individuals.

Another treatment approach that has been tried and found to be somewhat successful is behavioral therapy. Consensus is that Tourette syndrome is influenced greatly by psychological factors. The ability to suppress movements voluntarily for a limited amount of time reflects the effect of such factors (Bliss, 1984). Behavioral techniques that have been applied for controlling tics include massed negative practice such as forceful, repeated execution of tics (Storms, 1985); contingency management including positive and/or negative reinforcement (Canavan and Powell, 1981); relaxation training (Turpin and Powel, 1984); self-monitoring including self-observation and self-awareness (Billings, 1978); and habit reversal, a procedure including awareness training, self-monitoring, contingency management, and relaxation (Azrin, Nunn, and Frants, 1980). All the behavior therapy procedures mentioned are found to be somewhat beneficial in reducing the frequency of tics in Tourette syndrome. Behavioral therapy is recommended as the treatment of choice to Tourette syndrome patients who are affected by the side effects of drugs or who do not respond to drugs. Furthermore, behavior therapy is recommended to many patients with Tourette syndrome who, despite taking medications, still exhibit its symptoms or prefer to decrease the dosage levels of medication.

Speech and Language Characteristics

Months after the earliest motor symptoms, involuntary and unintended vocal tics appear (Bruun, 1989). They may occur so frequently and intensely that normal speech becomes impaired or unintelligible. Vocal tics fluctuate over time and respond to the same medications that are effective for motor tics (Bruun, 1989). Vocal tics are classified as simple vocal tics and complex vocal tics. Table 1, adopted from Cohen, Leckman, and Shaywitz (1985), demonstrates an example of simple and complex vocal tics.

TABLE 1. Vocal Symptoms Associated With Tourette Syndrome

Simple Vocal Tics
    Whistling, coughing, sniffling, spitting, barking, grunting, gurgling, hissing, sucking, uh-uh, eeee, and innumerable other sounds

Complex Vocal Tics
    Words, Phrases, and Statements
    Rituals (counting rituals, repeating a phrase until it is "just right"

Speech Atypicalities
    Unusual rhythms, tone, accents, intensity of speech

Coprolalia: obscene and aggressive words and statements

Simple vocal tics refer to fast and meaningless production of sounds or noises. The first frequently appearing simple vocal tics are throat clearing, grunts, sniffing, coughing, screams, snorting, and high-pitched sounds such as squeals, shrieks, and yelps. Also, irregular word accentuation has been reported by some authors (Bruun, 1988; Shapiro et al., 1988). Word accentuation is characterized by sudden increase in the loudness and/or pitch changes in a word or a phrase during normal speaking.

Complex vocal tics (verbal tics) refer to the involuntary interjections of syllables, words, or phrases. Complex vocal tics are quite diverse and include echolalia, palilalia, and coprolalia. Echolalia refers to involuntary repetition or echoing of sounds, words, phrases, and sentences. Echolalia is a form of complex vocal tics that may appear later in the course of the syndrome in some patients with Tourette syndrome. The most frequent characteristic of echolalia in Tourette syndrome is repeating the last word spoken by another person (Shapiro et al., 1988). Lees and associates (1984) reported a patient with Tourette syndrome who echoed the accents of other speakers. Some patients also echo sounds heard in the environment such as animal and automobile noises. A less frequent type of complex vocal tic is palilalia. Palilalia refers to involuntary repetition of one's own last sounds, words, phrases, or sentences. The last verbal symptoms which appears in some individuals with Tourette syndrome is coprolalia. Coprolalia refers to the explosive utterance of obscenities, most often 'shit' and 'fuck' (Abuzzahab and Anderson, 1974).

Similar to motor tics, vocal tics have periods of waxing and waning (Bruun, 1989); however, there is no regular pattern to this variation. Some individuals with Tourette syndrome can anticipate their tics. Often, the individual will suddenly develop a new vocal tic after months of stability which may disappear suddenly or gradually. Most individuals have few long-lasting familiar vocal tics while other tics come and go.

The daily variations of vocal tic severity are usually more predictable (Shapiro et al., 1988). Excitement, anxiety, and impatient anticipation will cause an increase, whereas concentration on an absorbing activity, such as playing a musical instrument, produces a decrease (Bruun, 1989). Tics generally increase when the individuals with Tourette syndrome reduce their self control (Bruun, 1989). This may be when they are alone or when they relax with their family and friends. Watching television is frequently mentioned as a time when symptoms increase, perhaps because it induces relaxation without involvement.

Vocal tics in Tourette syndrome are different from the utterances that appear during ordinary speech, especially from brief expressions of a person who is surprised, frustrated, or emotionally upset. Nuwer (1982) compared the type of obscenities uttered by 30 normals and 12 individuals with Tourette syndrome. The recorded words indicated that coprolalia in Tourette syndrome involves mainly certain words rather than all obscenities and profanities typically used. Normal subjects generally use religious profanities and/or obscenities related to physical acts or organs. However, coprolalia in Tourette syndrome patients is characterized by physical obscenities but not religious profanities. Nuwer further hypothesized that physical obscenities are shorter words and might be easily produced by "short circuits" in the brain.

Verbal tics have been found not to be related to the syntactic structures per se; however, syntactic boundaries have been suggested to be related to verbal tics. Martindale (1977) analyzed the syntactic correlation of coprolalia with the structure of the sentence in a spontaneous speech sample of one subject with Tourette syndrome. The speech sample was divided into sentences according to content, intonational contour, and grammatical structure. The subject produced 693 sentences during which he had 345 occasions of coprolalia. Coprolalic words were produced more rapidly and between the words but never while a word was being articulated. Sentences containing coprolalia were almost twice as long as undisrupted sentences in the number of words (23.51 versus 11.57). Examination of sentences yielded that 16.3% of 693 sentences produced by the subject were associated with coprolalia, which were distributed at syntactic boundaries. Tics most occurred at the beginning (8.5%) or end (5.3%) of sentences. Those which occurred in the middle (2% of all sentences) fell between phrases. Only one of the 345 tics occurred within a phrase between a modifying adjective and its noun. When coprolalia occurred in the middle of the sentence, 52% preceded function words (e.g., "and," "if," "but," because," and "that"), 28% preceded pronouns, and 20% preceded content words. The author suggested that necessity for grammatical coordination elicits the tic as the result of overactivity of subcortical centers mediating this operation.

Ludlow, Polinsky, Caine, Bassich, and Ebert (1982) found that vocal tics are not related to receptive or expressive language performance in Tourette syndrome. The authors studied 54 subjects with Tourette syndrome to determine whether any language abnormalities are associated with Tourette syndrome. Uniform prescribed conversation and picture description tasks were used to elicit speech samples. The Neurosensory Center Comprehensive Examination for Aphasia was employed to assess language functions. Results indicated an impairment in the areas of language expression and written expression in subjects with Tourette syndrome. They presented lower scores on language expression, written expression, and copying tasks. In contrast to language expression, language comprehension, verbal repetitions and verbal memory skills were found to be intact.

The examination of vocal tic type revealed no homogeneity among subjects with Tourette syndrome (Ludlow et al., 1982). The authors classified the vocal tics in 11 categories. Simple vocal tics were classified based on the anatomical sites where they occurred. Following were the vocal tic categories: (1) lingual tics; (2) laryngeal tics, phonations made on exhalations; (3) inhalations (gasps and forced inhalations); (4) nasal tics, e.g., snorts and sniffs; (5) labial tics, e.g., lip smacking, spitting, raspberry noises, and bronx cheers; (6) coprolalia; (7) palilalia; (8) echolalia; (9) jargon (single or strings of meaningless syllables); (10) word tics, e.g., any meaningful word interjections which are not part of the communication; and (11) sterotypic phrases or words, e.g., "you know," "oh yeah." Although all the subjects with Tourette syndrome produced vocal tics (with an interjudge reliability of $r = .81$), a great variability in the type of vocal tics was found. 40% of the subjects produced laryngeal tics and 35% subjects produced verbal tics including, jargon, coprolalia, word production, stereotypic phrases and palilalia. Importantly, the severity and type of vocal tics in subjects with Tourette syndrome did not correlate with their severity of language expression impairments. The Pearson correlation coefficient of $r= -0.07$; $p = 0.31$ indicated no relationship between these two types of symptoms associated with Tourette syndrome.

Ludlow et al. (1982) discussed that vocal tics are related to the planning and sequencing of speech as a motor behavior. Their origin might be related to speech planning which initiates at the cortex and is further regulated and coordinated at the basal ganglia and cerebellum. Because vocal tics occur at the beginnings and the ends of phrases in speech, the authors postulated that the cortical systems fail to check the automatic stereotypic speech and motor behaviors at these boundaries.

Few studies have examined the motor speech status in Tourette syndrome. Darley, Aronson, and Brown (1975) categorized the nature of motor speech disorder in Tourette syndrome as hyperkinetic dysarthria. Dysarthria refers to a group of speech disorders involving one or all of the basic motor speech processes (respiration, phonation, resonance, articulation, and prosody) resulting from disturbances in the muscular control because of the damage to the central or peripheral nervous system. Hyperkinetic dysarthria results from the damage to the extrapyramidal system of the motor organization consisting of the basal ganglia, subthalamic nuclei and the upper brain stem. General characteristics of hyperkinetic dysarthrias are rapid and discrete individual movements. Rapid abnormal involuntary movements are either unsustained or sustained only briefly. They are random in occurrence, generally unpatterned, and usually do not occur repetitively in the same muscle. Some other examples of hyperkinetic dysarthrias include chorea, hemiballismus, and myoclonic jerks.

Recent investigators have demonstrated a relationship between prosodic abnormalities and the nature of motor speech disorders in individuals with dysarthrias (Kent and Rosenbek, 1982). Sayadi and Cooke (1991) suggested that vocal tics might be influenced by the physiological changes required for the prosodic aspects of speech. The authors studied a 34-year-old subject with an 18 year history of Tourette syndrome. The subject produced the following samples: spontaneous speech, reading, counterbalanced repetitions of sentences with implied emotional (angry and happy) and propositional (declarative and interrogative) prosodies, singing, syllable repetitions, and maximum vowel prolongations. Vocal tic episodes were counted by the examiner, and, further, the frequency of individual vocal tics within each episode were tallied. Vocal tic episodes were defined as one single vocal tic or multiple vocal tics occurring in strings. Thus, the occurrence of one vocal tic was counted as one vocal tic episode, and the occurrence of multiple vocal tics at one point in the linguistic string was also counted as one episode.

All the speech samples collected in different conditions were interrupted by vocal tics. During the spontaneous speech, vocal tics interfered with the communication on the average of every 10 words. All the vocal tics were laryngeal tics characterized by sudden forceful phonations on exhalation or prolongations. A few phonemic repetitions and one production of jargon also occurred. The oral reading sample presented similar frequency and type of vocal tics found in the spontaneous speech. No jargon occurred during reading. The authors concluded that oral reading of a standard paragraph can be a useful procedure for estimating the vocal tic frequency. Singing was dysprosodic and subject's voice quality resembled spastic dysphonia characterized by an effortful, hoarse voice. Spastic dysphonia associated with Tourette syndrome has been reported in the literature. Lang and Marsden (1983) described a patient with Tourette syndrome who developed spastic dysphonia while he was being treated with haloperidol. His voice was characterized by a hoarse, strained voice of low volume and high pitch accompanied by tensing of the jaw and larynx during phonation. His spastic dysphonia did not improve despite cessation of neuroleptic therapy. The authors cautiously suggested that spastic dysphonia might be another phonatory complication in Tourette syndrome.

An analysis of sentence repetitions (Sayadi and Cooke, 1991) concluded that laryngeal tics increased during emotional and interrogative prosody but not during the declarative prosody. The 10 emotional sentence repetitions were associated with 25 vocal tics, whereas only 9 vocal tics occurred during the 10 linguistic prosody repetitions. Only the interrogative prosody repetitions were associated with vocal tics. That is, the subject did not produce any vocal tics during the repetitions of declarative sentences. When the emotional and linguistic prosody repetitions were compared for the number of vocal tic episodes, a small difference was found (9 vocal tic episodes in emotional prosody repetitions versus 6 episodes in the linguistic prosody repetitions). The authors concluded that when the vocal tics increase, they increase within the vocal tic episodes.

Finally, laryngeal tics occurred at syntactic boundaries and at pause times, consistent with Martindale (1979) and Ludlow et al. (1982). Syntactic boundaries are marked by the "fall-rise" patterns of fundamental frequency (Cooper and Sorensen, 1981), and fundamental frequency variations are features of prosody which are controlled and regulated by motor speech centers. Thus, Sayadi and Cooke (1991) hypothesized that laryngeal tics might be related to the control of prosodic patterns in speech as the result of motor control involvement. In addition, reduced maximum vowel duration, decreased syllable repetition rate, slow speaking rate, and dysphonic, dysprosodic singing found in this subject further supported motor speech involvement suggested by Darley et al. (1975) and Ludlow et al. (1982).

Prosody And Its Implications

Sayadi and Cooke (1991) suggested that vocal tics might be related to actualizing features of prosody which mark syntactic boundaries. Prosodic features refer to pitch, loudness, rate, and prosodic contour. Pitch relates to the average frequency of vocal fold vibrations. Loudness relates to the amplitude of vocal fold vibrations, and rate relates to the timing patterns of speech utterances affecting the overall rhythm of speech. These features play distinctive roles in human verbal communication. First, they function as a linguistic device to indicate points of syllabic stress and the sentence mode (e.g., declarative versus interrogative). Second, prosodic contours (patterns of pitch and loudness over time) correspond to major syntactic boundaries (Cooper and Sorensen, 1981). Another function of prosodic contour is to express and communicate emotions. For instance, Lieberman (1967) suggested that emotion is expressed in a prosodic contour lasting about the length of a breath.

Emotional expression in speech is mainly executed via prosodic features (Scherer, 1986). Studies have taken a set of utterances, eliminated some of the prosodic features and investigated whether these degraded utterances could express emotion. In general, no matter how an utterance is degraded, the loss of prosodic features seems to impair recognition of emotion (Burns and Beier, 1973; Ross, Duffy, Cooker, and Sargeant, 1973). Scherer (1986) suggested that anger, happiness, and fear are signalled by increases in pitch, loudness, and a fast rate; grief has a low pitch and a slow rate.

Stimuli can be validated as expressing a particular emotion by asking the speaker to express an emotion, by inducing the emotion in the speaker before speaking, or by having judges rate the stimulus as expressing the emotion. No differences between the three methods have been noted (Williams and Stevens, 1972). Further, portrayals by actors are similar (although not always identical) to productions by people in real situations.

Physiologically, there are several muscular controls for prosodic features (Zemlin, 1989). Increase in tension and decrease in the mass of the intrinsic laryngeal muscles are responsible for pitch raising. However, in order to produce tones near the extreme ends of the pitch range and to facilitate rapid changes in pitch, extrinsic muscles also play a role. Intensity increases with a rise in subglottal pressure and amplitude of vibratory movements of the vocal folds. Also, the duration of the closed phase of the vibratory cycle increases with intensity.

Increased muscle tension is a correlate of some emotions. Huttar (1968) postulated that increased tension in the respiratory muscles would lead to louder speech and that increased tension in laryngeal muscles would lead to higher pitched speech. Increased muscle tension is one aspect of sympathetic arousal (Zemlin, 1989) which also increases rate of respiration perhaps resulting in faster speech. These various physiological responses would lead to a correlation between various emotions and prosodic features of pitch, loudness, and rate.

An assessment of the prosodic features in motor speech disorders should include evaluation of pitch, loudness, rate, and intonational contour. Barnes (1983) suggested that prosodic features such as pitch and loudness must be examined independently because these features are primarily dependent on the laryngeal and respiratory functions. However, few comprehensive methods for assessment of prosodic features of speech have been developed (Barnes, 1983). Both perceptual and acoustic analyses have been employed to examine the nature of prosodic disturbances in the dysarthric population (Goodglass and Kaplan, 1972; Ludlow and Bassich, 1983). For example, the rating of speech characteristics in the Boston Diagnostic Aphasia Examination (Goodglass and Kaplan, 1972) requires a perceptual judgment of the melodic line in speech; melodic line corresponds to the prosodic contour.

Ludlow and Bassich (1983) argued that although the acoustic analysis of dysarthric speech provides objective results, it may not assess the patient's communication impairment that listeners' perceptual ratings do. Therefore, they designed a study to determine whether acoustic and perceptual analyses of the dysarthric speech differentiate the pattern of dysarthric speech from the normal speech in the same way. Matched with age and sex, seven subjects with Shy-Drager syndrome and 7 subjects with Parkinson disease were compared with normals. Both Shy-Drager and Parkinson disease involve the basal ganglia and are characterized by hypokinetic dysarthria. The speech tasks included imitation of vowels, syllables, and sentences with various rate, pitch, loudness, and prosodic contours. Two different assessment methods were employed: (1) perceptual rating and (2) acoustic analysis. Three graduate speech-language pathology students participated as listeners. The perceptual ratings were based on 19 speech attributes reported by Darley et al. (1975) to be associated with the basal ganglia diseases of the hypokinetic nature. The acoustic analysis included 14 measures including intensity level, fundamental frequency, and time. Results indicated that both assessment systems differentiated between normals and dysarthric groups (interjudge reliability of 85% was reached for the perceptual ratings). Furthermore, both the acoustic and perceptual assessment systems were capable of accurately discriminating between the two types of dysarthria and variations in rate or time, intensity level or loudness, fundamental frequency or pitch, and reduced stress.

Focusing on prosodic aspects of speech, Ludlow, Bassich, and Connor (1985) developed speech tasks and acoustic measures for the identification and assessment of the speech problems associated with neurologic disorders. The system assesses the prosodic aspects of speech including the adequacy of phonatory, vocal intensity, fundamental frequency control, and the use of these variables in speech. Vowels, syllables, and sentences are used in tasks which assess the speech production rather than nonspeech oral movements. Using clinician's modeling, subjects are required to imitate various task categories consisting of (1) maximum phonation time; (2) vowel, syllable, and sentence repetitions with various rates; (3) rapid initiation of various loudness levels; and (7) imitation of stress contrasts. The system employs acoustic measures and provides normative data on these measures including, age and sex effects. A scoring system provides measures of the degree of impairment relative to normal. Finally, the system allows to determine the pattern of motor speech problem by identifying the impaired and unimpaired aspects of speech. Previous studies have shown 100% construct validity and 85% interjudge reliability of this system (Ludlow and Bassich, 1983).

In conclusion, the literature review indicates that the occurrence of vocal tics impairs the communication of individuals with Tourette syndrome. Vocal tics call attention to themselves, interfere with communication, and cause both the speaker and the listener to be distressed. The available data suggest that vocal tics in Tourette syndrome are associated with intermittent motor speech control impairments involving interactions among respiratory, laryngeal, and supralaryngeal systems. Control of prosody is an important function of the above systems which provide linguistic meaning and emotional color to the speech. Therefore, an examination of the relationship between vocal tics and the prosodic features of speech may prove to be important for the occurrence of vocal tics in the speech of individuals with Tourette syndrome.

Purpose of The Study

Sayadi and Cooke (1991) speculated that the occurrence of vocal tics might be related to the production of prosodic features. While their study suggested a connection between vocal tics and prosodic control, it contained two important limitations. First, their study relied on only one subject. Second, it did not permit independent examination of the major carriers of prosodic information (pitch, loudness, rate, and contour). The purpose of the present study was to examine, in a larger sample of subjects (n = 10), whether vocal tics are related to the control of pitch, loudness, rate, and prosodic contour in the speech of individuals with Tourette syndrome.

This study examined the relationship between motor speech control for prosody and the occurrence of vocal tics. Thus, it may provide some beneficial information for physiological explaination of the vocal tics in Tourette syndrome. Clinically, the results of this study may be particularly useful. They may provide an insight to whether modifying speech features could be an adjunct to current pharmaceutical and behavioral therapies for the control of vocal tics. Specifically, the aspects of prosody which influence the occurrence of vocal tics as well as those not influencing could be employed for treatment planning. Particular questions of the present study were as follows:

(1) Are vocal tics related to pitch changes in speech, compared to the habitual pitch?

(2) Are vocal tics related to loudness changes in speech, compared to the habitual loudness?

(3) Are vocal tics related to rate changes in speech, compared to the habitual rate?

(4) Are vocal tics related to the overall linguistic and emotional prosody?

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METHODS

Subjects

Ten adult subjects with Tourette syndrome volunteered to participate in this study. All the subjects were diagnosed as having Tourette syndrome based on DSM-III-R (1987) criteria. Subjects were recruited through the Detroit Area Chapter of the Tourette Syndrome Association, and all of them were native American English speakers. Based on a self-report questionnaire (Sayadi and Cooke, 1991), none of the subjects reported a history of other neurological disorders or head injuries. In addition, all the subjects successfully passed the auditory testing procedures for the frequencies of 500, 1000, 2000, and 4000 Hz in both ears. The samples auditory threshold ranged between -10 to 20 dB SPL-HL. Finally, using the self report questionnaire (Sayadi and Cooke, 1991) further information regarding demographic and clinical data was collected. Sayadi and Cooke's questionnaire was developed based on the detailed and well-documented study reported by Shapiro et al. (1988). This information assisted in explaining subjects' variability.

Pretesting Tools

Subjects were assessed for the severity of Tourette syndrome, motor speech, and language performance. This information about the subjects was completed in order to get a broad based information about the subjects so that general patterns or outlier information could be obtainded.

The Yale Global Tic Severity Scale (YGTSS) (Leckman, Riddle, Hardin, Ort, Swartz, Stevenson, and Cohen, 1989) was used to rate the severity of Tourette syndrome in each subject. The YGTSS uses a semi-structured interview to elicit information concerning the specific character and anatomical distribution of tics observed during the course of a one week interval before the assessment. During the interview, this information is recorded on a prepared "Tic Inventory" form. Following the completion of the interview and the Tic Inventory, the severity of motor tics, vocal tics, and the overall Tourette syndrome impairment are rated based on a six-point ordinal scale. The rating addresses the various aspects of tics consisting of number, frequency, intensity, complexity, and interferences. The overall Tourette syndrome impairment estimates the impact of the disorder on self-esteem, family life, social, acceptance, and school or job functioning. The YGTSS is based on a scale ranging from none, to minimal, moderate, marked, and severe, with descriptive statements and relevant examples describing each scale. The same ordinal scales are used to rate motor and vocal tics. YGTSS has a interjudge reliability of 0.80 for motor tics and 0.90 for vocal tics which is significant at $p < 0.001$ (Leckman et al., 1989).

The Frenchay Dysarthria Battery (FDA) (Enderby, 1983) was employed to assess the motor speech functions in each subject. The Frenchay system is aimed at determining which components of the speech production system are affected (e.g., respiration, phonation, velopharyngeal function, articulation, and prosody). The battery is divided into 8 sections: reflex, respiration, lips, jaw, palate, larynx, tongue, and intelligibility. It requires a set of tasks for the subject to undertake and describes certain observations for the clinician to rate. The ratings are scored on a nine point scale. The battery also includes rating of non-linguistic but commonly related areas of function, for example, tongue, lip, and jaw movements during nonspeech tasks. Abnormal functioning in these areas may have discriminatory and therapeutic implications in subjects with dysarthria. The results are presented on a profile which represents the performance level on each of oral, facial, and speech tasks. Profiles of each dysarthric group with the specific underlying pathologies are also provided. These profiles are similar to descriptions provided by the Mayo Clinic dysarthria study (Darley et al., 1975). The FDA has a high interjudge reliability of $r = 0.92$ (Enderby, 1983).

The Western Aphasia Battery (WAB) (Kertesz, 1982) was employed to assess the language performance in each subject. The WAB assesses seven areas of functioning: spontaneous speech, auditory comprehension, repetition, naming, reading, writing, praxis, and construction. The spontaneous speech subtest rates both the fluency and information content of the subject's speech in question-answer and picture description contexts. The auditory comprehension subtest assesses understanding of yes-no questions, identifications of objects and pictures in 10 categories, and execution of commands increasing in complexity. Repetition of words, phrases, and sentences compose the repetition subtest. Naming is assessed using tasks of object naming, word fluency, sentence completion, and responsive speech. The sum of weighted subtests scores results in the Aphasia Quotient (AQ). The AQ is a functional measure of the severity of the spoken language deficit in aphasia with 100, a perfect score, easily attainable by normal speaking adults. The original population on which the standardization of the WAB was based consisted of a sample of three groups: normals, aphasics, and mixed brain-damaged patients (e.g., diffusely brain-damages, subcortical damaged). The WAB has a intrajudge reliability of $r = 0.98$ ($p < 0.001$), interjudge reliability of $r = 0.99$ ($p < 0.001$), and construct validity $r = 0.91$, ($p < 0.01$) (Kertesz 1982).

Experimental Procedures

Each subject completed the experimental tasks during one session lasting approximately half an hour. Subjects were asked to remain relaxed and perform the tasks as naturally as possible. All the speech samples were recorded on videotape, with a camera approximately six feet infront of each subject.

For each experimental task, the subjects were given instructions followed by demonstrations and one practice trial. If the subject misunderstood the instructions or was unable to perform the task, the instructions were repeated and if needed, another practice trial was provided. Then, the subject was asked to try again, and the second attempt was recorded for the data collection. To assist subjects with the tasks, the stimuli and the required conditions for their productions were printed on an index card and placed in front of them during the experiment.

Twenty-six experimental tasks, with three repetitions of each task (seventy-eight total repetitions), were completed. All the tasks were systematically randomized (across subjects and tasks) in such a way that no single task order was repeated. One vowel and one sentence were employed as the stimuli for all experimental tasks. This allowed the examiner to determine the degree to which the vocal tics were related to phonation only or to when supralaryngeal articulation was also required to be coordinated with the phonation for the production of the sentence (Ludlow, Bassich, Connor, 1985).

Stimuli

The use of vowels as stimuli for the assessment of motor speech disorders has been validated in the literature (Darley et al., 1975). In the present study the vowel /a/ was selected because it is a "lax" vowel and does not demand the elevation of the larynx (Ludlow, Bassich, and Connor 1985). A sentence repetition task was employed to examine the research questions. This task allowed the subject to produce the sentence with typical conversational English (Cooper and Sorensen, 1981). Other authors have also employed sentence repetition tasks to assess the dysarthric speech (e.g., Ludlow, Bassich, and Connor, 1985; Yorkston and Beukelman, 1981). Ludlow and co-workers (1985) used sentence repetitions to assess rate, loudness, and intonational contour in speech. Finally, Colsher, Cooper, and Graff-Radford (1987) used a sentence repetition tasks in order to examine the emotional and linguistic prosody in subjects with right-hemisphere damage. Across sentences the subjects were required to convey three emotions (happy, sad, and angry) and two linguistic forms (statement and question).

To maintain a constant linguistic environment, only one sentence from Yorkston and Beukelman (1981) was employed for the tasks which required sentence repetitions: " This can be the cheapest way to ship them long distance." Th selected sentence did not contain clause boundaries. To control the possible effect of long words, the sentence consisted of 11 short words (nine were one syllable long and two contained two syllables). Phonemes within the sentence required labiodental, alveolar, palatal, and bilabial valvings.

Experimental Tasks

In the present study, the experimental tasks were adopted and adapted from the assessment system developed by Ludlow et al. (1985). The following summarizes the tasks implemented to examine each of the research questions in the present study.

Pitch

Eight tasks were employed to test the relationship between pitch regulation and vocal tics. These tasks indicated the relationship between holding, lowering, and raising pitch and vocal tics during the prolongation of the vowel /a/, during the sentence production, and during pitch glides on the vowel /a/. The following introductory instructions were used for each task:

Task 1. " Say an /a/ sound with your own regular pitch. Like this (demonstrate: aaa...)."

Task 2. " Say an /a/ sound, but this time with a high pitch. Like this (demonstrate aaa...)."

Task 3. " Say an /a/ sound, but this time with a low pitch. Like this (demonstrate aaa...)."

Task 4. " Say an /a/ sound, but this time glide up the scale as high as you can go on a single /a/ sound. Start from low. Like this (demonstrate)."

Task 5. " Say an /a/ sound, but this time glide down the scale as low as you can go on a single /a/ sound. Start from high. Like this (demonstrate)."

Task 6. " Say this sentence with your own regular pitch. Like this (demonstrate)."

Task 7. " Say this sentence again, but this time with a high pitch. Like this (demonstrate)."

Task 8. " Say this sentence again, but this time with a low pitch. Like this (demonstrate)."

Loudness

Eight tasks were used to test the relationship between vocal tics and loudness regulation. These tasks indicated the relationship between vocal tics and various vocal efforts for whisper, soft, regular, and loud voice for the production of vowel /a/ and the sentence. The following introductory instructions were used for each task:

Task 1. " Whisper /a/. Like this (demonstrate)."

Task 2. " Say /a/, but this time with your own natural loudness. Like this (demonstrate)."

Task 3. " Say /a/ again, but this time say it with a soft voice. Like this (demonstrate)."

Task 4. " Say /a/ again, but this time say it with a loud voice. Like this (demonstrate)."

Task 5. " Whisper this sentence. Like this (demonstrate)."

Task 6. " Say this sentence again, but this time say it with your own natural loudness. Like this (demonstrate)."

Task 7. " Say this sentence again, but this time say it with a soft voice. Like this (demonstrate)."

Task 8. " Say this sentence again, but this time say it with a loud voice. Like this (demonstrate)."

Rate

Six tasks were employed to test the relationship between vocal tics and the temporal control of speech mechanisms. The tasks included vowel and sentence repetitions with different rates of slow, regular, and fast. The following introductory instructions were used:

Task 1. " Say /a/ again, but this time instead of a long /a/ sound, say a series of /a/ sounds. Like this (demonstrate)."

Task 2. " Say a series of /a/ again but this time say it slowly. Like this (demonstrate)."

Task 3. " Say a series of /a/ again but this time say it as fast as you can. Like this (demonstrate)."

Task 4. " Say this sentence with your own regular speed. Like this (demonstrate)."

Task 5. " Say this sentence, but this time say it as fast as you can. Like this (demonstrate)."

Task 6. " Say this sentence slowly. Like this (demonstrate)."

Emotional and Linguistic Prosody

Four tasks were completed to assess the relationship between the vocal tics and regulation of prosodic contours during emotional and linguistic prosody. The following introductory instructions were used for each task:

Task 1. " Say this sentence as if you are making a statement. Like this (demonstrate)."

Task 2. " Say this sentence, but this time as if you are questioning. Like this (demonstrate)."

Task 3. " Say this sentence again, but this time say it with anger. Like this (demonstrate)."

Task 4. " Say this sentence again, but this time say it with a happy voice. Like this (demonstrate)."

Data Treatment

The dependent variable of the present study was the number of tic-filled utterances in a condition. Tic-filled utterances were defined as those productions which were accompanied with one or more vocal tics immediately before, within, or after the utterance. This method of data treatment had two important benefits. First, it secured judgment reliability by determining presence or absence of vocal tics. Second, it eliminated skewing the data in cases of vocal tic bouts where multiple tics existed in either discrete or continuous patterns. For example, if a subject produced a vocal tic bout which contained several discrete tics, that observation was counted as one tic-filled production.

The vocal tics were defined based on the DSM-III-R (1987). The criteria require the vocal tics to be involuntary, rapid, brief, sudden, repetitive, stereotypic, and/or nonrhythmic. In addition, tics are purposeless, inappropriate, and irresistible. Tics may be single or multiple and simple or complex.

The dependent variable was scored by examining the audio-video recordings. For each subject the frequency of tic-filled utterances for the various experimental tasks was recorded on a scoring sheet. Thus, a profile of performance by task for each subject was created. The subjects' total tic-filled utterances yielded the data that were submitted to statistical analysis.

Reliability

Inter-and intra-judge reliability were computed for 100% of the data from two randomly selected subjects (20% of the group data). A second judge, a certified speech-language pathologist, assisted the examiner for the inter-judge reliability. The judges viewed the video tapes and tallied the frequency of the tic-filled utterances without the knowledge of the previously recorded data. A

100% intra-judge and 95% interjudge reliability of the measured dependent variable were obtained.

Statistical Analysis

In this study, each subject completed a total of twenty-six (26) tasks, repeated three times each in a systematically random order. Thus, the group of ten subjects produced thirty (30) utterances of each task, for a total of 780 productions.

A series of two-by-two chi-square tests were used to test the difference between each of the habitual (utterances produced with subjects' own natural pitch, loudness, and rate) and their respective atypical (utterances which subjects' were instructed to deviate from their habitual productions, e.g., whisper, speak with a high pitch or a fast rate) conditions. For example, in the loudness tasks, each of the loud, soft, and whispered utterances were separately compared against their habitual loud trials. The two-by-two chi-squares were used because each of these tasks demand different and specific motor patterns. The data were analyzed using the chi-square test for the following reasons: (1) the dependent variable was based on a nominal scale (presence or absence of vocal tics); and (2) the distribution function of the sample population was not normal (Kirk, 1982). In order to correct for the effect of multiple chi-square tests the decision rule for rejecting the null hypothesis was set at p or bellow < 0.02. However, because of the exploratory nature of the experiment and the small sample size an alpha < 0.05 was accepted as suggestive. The following illustrates a general two-by-two chi-square table where each observation is classified into either class 1 (tic-free) or class 2 (tic-filled).

	Class 1 Tic-free	Class 2 Tic-filled	Total
Habitual condition	O_{11}	O_{12}	n_1
Atypical condition	O_{21}	O_{22}	n_2
			$N = n_1 + n_2$

The following general chi-square formula was used (Kirk 1982):

$$X^2 = \frac{N(O_{11}O_{22} - O_{12}O_{21})}{n_1 n_2 (O_{11} + O_{21})(O_{12} + O_{22})}$$

Where

O = Number of Observations

$n_1 = O_{11} + O_{12}$ (30 for this study)

$n_2 = O_{21} + O_{22}$ (30 for this study)

SUBJECTS' BACKGROUND INFORMATION

Ten adult native American subjects with Tourette syndrome (eight males and two females) volunteered to participate in this study. Prior to the initiation of the experimental tasks, information regarding subjects' demographic and clinical background, severity of Tourette syndrome, and communication abilities was collected.

This information was collected to illustrate the diversity of subjects in this sample regarding their wide variety of clinical characteristics and concomitant behaviors. Although, the validity and reliability of the employed questionnaire has not been systematically tested prior to this study, the collected data support the characteristics published in the literature for this population. Nevertheless, this information must be noted cautiously. Finally, this information was not used to test the main questions of this investigation.

Demographic Data

Using the self-report questionnaire (Sayadi and Cooke, 1991), information regarding subjects' age, gender, handedness, education, occupation, and marital status were collected.

The mean age for the present sample was 36 (SD = 11 and range 21 to 49) with the median of 38 years. A right hand dominance was reported in the present sample. All the subjects rated themselves as strongly right-handed for writing, throwing, cutting, drawing, brushing hair, and using a spoon.

The educational status in this sample ranged from the completion of the eleventh grade (20%) to graduate level (20%). The mean year of education was 14.5 (SD = 3.53 with a range from 11 to 22) with the median of 13. The majority of the sample were free of difficulties in expressive writing, reading disability, mathematical disability, and learning disability. Only one subject who completed the eleventh grade in high school had been diagnosed as having a learning disability. Also, one female subject with an undergraduate degree in English literature reported having mathematical difficulties.

The occupation status varied among the individuals in the present sample. 40% of the sample were unemployed, 30% had full-time employment, 20% were employed part-time, and 10% were full-time college students. Those who were employed expressed difficulties about having Tourette syndrome at the work place. Those who were unemployed did not have any professional training which would facilitate finding jobs. The two female subjects were unemployed.

The marital status included 90% who were single and 10%

who were currently married. Two of the single subjects were previously married and were divorced at the time of the present study and 70% were never married. The two female subjects were both single, but one had been previously married. Subjects who had gone through a divorce found Tourette syndrome as a major variable influencing the failure of their marriages. 30% of the singles lived with a significant other, 40% lived with their family, and the remaining 20% lived independently. All those who were single reported difficulty communicating with initiating, establishing, and maintaining intimate relationships with the members of the opposite gender as the result of having Tourette syndrome.

A family history of Tourette syndrome and other tic disorders was reported by 30% of the subjects. One subject had a biological sister and father with Tourette syndrome and another had a brother with Tourette syndrome. In addition, one subject reported a family history of tic disorders other than Tourette's. Subjects' demographic data is summarized in Table 2.

Table 2. Summary of Subjects' Demographic Data

ID	Gender	Age	Education (in years)	Marital Status	Employment
1	Male	36	11	Divorced	Unemployed
2	Male	27	12	Single	Part-time
3	Male	24	13	Single	Student
4	Male	23	13	Single	Part-time
5	Male	40	16	Single	Full-time
6	Male	21	11	Single	Unemployed
7	Male	41	18	Single	Full-time
8	Female	48	16	Single	Unemployed
9	Female	49	12	Divorced	Unemployed
10	Male	49	22	Married	Full-time

Clinical Symptoms

Using the self-report questionnaire (Sayadi and Cooke, 1990), the following clinical Tourette syndrome information was collected for each subject: age at onset, symptoms fluctuations, medical and psychological management of the disorder, associated neurological and psychological problems, and external and internal stimuli affecting tics.

Age at Onset

The mean age at onset for Tourette syndrome in the present sample was 9 (SD = 5) with a range of 3 - 18 years. The mean age at diagnosis was 25 (SD = 12) with a range of

10 - 40 years. The average difference between age at onset and age at diagnosis was 16 (SD = 11) and the range was 3 to 36.

Inspection of the data on age at onset and age at diagnosis indicated that the older subjects were diagnosed later in the course of their disorder. The mean onset age for motor tics was 8 (SD = 4.80) with the range of 3 - 18. The mean onset age for vocal tics was 9 (SD = 4.77) with a range of 3 - 18. The mean duration of the Tourette syndrome was 27 (SD = 11) with a range of 16 - 43 years. Table 3 shows the age at onset of the variables and the duration of Tourette syndrome in the present sample.

Table 3. Subjects' Tourette Syndrome Age at Onset Variables

Variable	Mean	SD	Range
Onset age	9	5	3 - 18
Age at diagnosis	25	12	10 - 41
Difference	16	11	3 - 36
Onset age for motor tics	8	5	3 - 18
Onset age for vocal tics	9	5	3 - 18
Duration of TS symptoms	27	11	16 - 43

Symptoms Fluctuations

Fluctuations of type and severity of Tourette syndrome symptoms were reported in 100% of the sample. These variations generally existed for one week to years. Also, two subjects reported spontaneous remissions lasting from 1 to 6 years. The first subject with the onset age at 8 had a complete remission at age 12 through age 18. The second subject with onset at age 18 had a one year remission of the symptoms at the age of 36.

Tourette Syndrome Management

Five subjects currently used medications for controlling Tourette syndrome symptoms (haloperidoal and pimozide). Two of them also took medications for controlling their depression. Only one subject found the medication very effective in diminishing most of his motor and vocal tics. The rest of the subjects who were on medications for Tourette syndrome found it effective to some degree in decreasing their symptoms, but both motor and vocal tics were still present. All the subjects who took medication complained of its side effects, including drowsiness, cognitive slowness, and weight gain. None of the subjects had behavioral approaches or psychotherapy in management of their Tourette syndrome. Two subjects with compulsive drinking symptoms and history of alcoholism participated in AA meetings directed by health care specialists. Although indirect, both subjects found the meetings helpful in reducing the overall effect of Tourette syndrome on their functioning as an individividual. Subjects reported that the meetings helped them in resolving psychological conflicts resulting from Tourette syndrome.

Associated Neurological Problems

Attention deficit disorder and hyperactivity also were reported by a number of subjects. Four of the subjects were diagnosed as having attention deficit disorder. Three of these subjects were males less than 26 years of age and the fourth one was a 48 year old female. Overall, 80% of the sample reported short attention span and 70% complained from being inattentive and easily distracted. Hyperactivity was also a common problem among the subjects. 60% rated themselves as restless and overactive. Other associated neurological problems were migraine (10%) and stuttering (10%).

Psychopathology and Behavioral Problems

Depression and obsessive-compulsive-like symptoms were reported by the subjects. 30% of the subjects (two males and one female) were diagnosed as having chronic depression. Two of three subjects with depression were taking medications for their depression. The first subject (male) had a history of hospitalization as the result of having depression. The second subject (female) had recently gone through a divorce. Finally, the third subject (female) had a family history of Tourette syndrome. Also 10% of the rest of the sample (one subject) had experienced suicidal thoughts in the period of one week prior to the present study. Regarding obsessive-compulsive-like symptoms, 90% of the subjects had repetitive thoughts which they could not turn off (such as worries and fears), 60% of the subjects reported counting numbers in their minds, 60% had repetitive touching of objects and people, and 20% compulsive drinking. All the subjects with the diagnosis of attentional deficit disorder (30% of the sample) also suffered from the occurrence of repetitive thoughts.

Other behavioral problems included anger and moodiness: 60% temper outbursts, 60% explosive anger, 50% quick and drastic mood changes, 40% difficulty expressing anger, 40% difficulty expressing emotions, 30% destructive acts, and 10% destructive acts in their thoughts. Table 4 summarizes the associated neurological, psychological, and behavioral disorders reported by the subjects in the present sample.

Table 4. Subjects' Associated Neurological and Psychological Problems

Description	Percentage
Associated Neurological Problems	
Attention deficit disorder	40%
Short attention span	80%
Inattentive, easily distracted	70%
Hyperactivity	60%
Others	
Migraine	10%
Stuttering	10%
Associated Psychological Problems	
Depression	30%
Obsessive-compulsive-like symptoms	
Repetitive thoughts	90%
Counting numbers in their minds	60%
Touching objects and people	60%
Compulsive drinking	20%
Thoughts of destructive acts	10%
Other Behavioral Problems	
Temper outbursts	60%
Explosive anger	50%
Difficulty expressing anger	40%
Difficulty expressing emotions	40%
Destructive acts	30%

External and Internal Stimuli Affecting Tics

Various times of the day affected the tics differently. All the subjects reported that their symptoms remained unchanged (50%) or decreased (50%) in the morning. Symptoms increased for 40% of the subjects in the afternoon and remained unchanged for 60% of the subjects. 80% of the subjects reported an increase of their symptoms in the evening. 20% remaining reported no change in the severity of their tics in the evening. Sleeping also decreased the symptoms in 90% of the sample; and the tics were absent for the remaining subject.

Personal moods affected the severity of symptoms differently. The occurrence of tics noticeably increased with anxiety (100%) and anger (90%). One subject reported decrease of his symptoms during anger. Joy increased the symptoms in 50% and decreased in the rest 50% of the sample. Pleasurable anticipation decreased the symptoms in 40%, increased in 30%, and did not effect the remaining 30% of the sample. Sadness increased both motor and vocal tics in 60% of the sample. However, one subject reported an increase of motor tics and a decrease of vocal tics in relation to sadness. In addition, 20% reported a decrease and 20% remained unchanged during sadness.

External stimuli influenced the occurrence of the symptoms in the present sample. Interpersonal relationships influenced the symptoms differently. When with strangers 50% reported an increase in their symptoms, 40% reported a decrease and 10% remained unchanged. When with family members, 30% reported an increase of their symptoms, 30% reported a decrease and 40% remained unchanged. School, work, or classroom situations increased the symptoms in 40% of the sample, kept the symptoms unchanged for 50%, and decreased in 10%. Fifty percent of the sample had an increase in their symptoms at church activities, 30% had a decrease, and 20% remained unchanged. Involvement in passive activities increased the tics differently. Watching movies and television increased the tics in 50%, decreased in 20%, and 30% rest remained unchanged. 100% of the sample reported that full concentration in activity (e.g., working on cars or playing music) strongly reduced their symptoms. Table 5 summarizes the effect of internal and external stimuli on subjects' tics.

Table 5. The Effect of External And Internal Stimuli on Subjects' Tics

Variable	Decreased	Unchanged	Increased
Day Progression Effect			
Morning	50%	50%	
Afternoon	60%	40%	
Evening		20%	80%
Sleeping *	90%		
Personal Mood			
Anxiety			100%
Anger	10%		90%
Joy	50%		50%
Pleasurable anticipation	40%	30%	30%
Sadness	20%	20%	60%
Interpersonal relationships			
Strangers	40%	10%	50%
Family	30%	40%	30%
Situations			
School or work	10%	50%	40%
Church	30%	20%	50%
Passive activities			
Movies	20%	30%	50%
TV	20%	30%	50%
Absorption in an activity	100%		

* One subject reported an absence of tics while sleeping.

Severity of Tourette Syndrome

The Yale Global Tic Severity Scale (YGTSS) (Leckman, Riddle, Hardin, Ort, Swartz, Stevenson, and Cohen, 1989) was used to rate the global severity of Tourette syndrome in each subject. Using a semi-structured interview, three steps were completed. Firstly, the motor and vocal tic types which occurred during the week prior to the study were determined. Secondly, the severity of both motor and vocal tics were determined based on number, frequency, intensity, complexity, and interference components. Finally, the subjects' overall Tourette syndrome impairments of social functioning was scored. The motor and vocal tics and the overall Tourette syndrome impairment were summed into an overall global score. The scale ranged from 0 (no symptoms) to 100 (severe Tourette syndrome).

Motor Tics

Both simple and complex motor tics were present (Table 6). The most frequent simple tic was eye blinking (100%) following by head jerks (90%), eye movements (70%), mouth movements (70%), facial grimaces (70%), shoulder shrugs (70%), leg/foot/toe movements (70%), arm movements (50%), abdominal tensing (40%), nose movements (20%), and tongue movements (20%). The most frequent complex behavior included tic-related compulsive behavior, such as, touching (70%), grooming (40%), finger tapping (10%) and straightening (10%). Fourty percent of the sample reported self-abusive behavior including picking and tearing off toenails, hitting things with their bare hands, biting inside the mouth, and hitting the mouth with their hand. Copropraxia such as finger showing (30%) and touching genitals (10%) were reported in 40% of the sample.

Table 6. Subjects' Simple and Complex Motor Tics

Description	Percentage
Simple motor tics	
Eye blinking	100%
Head jerks	90%
Eye movements	70%
Mouth movements	70%
Facial grimaces	70%
Shoulder shrugs	70%
Leg or foot or toe movements	70%
Hand movements	60%
Arm movements	50%
Abdominal tensing	40%
Nose movements	20%
Others	
Sticking tongue out	10%
Tongue twitches	10%
Complex motor tics	
Tic related compulsive behavior	
Touching	70%
Grooming	40%
Finger tapping	10%
Straightening	10%
Others	
Putting food on head while eating	10%
Copropraxia	40%
Self abusive behavior	40%

Vocal Tics

Simple and complex vocal tics were present among all the subjects of the present sample (Table 7). The simple vocal tics in a descending order of frequency were throat clearing (60%), coughing (50%), sniffing (40%), animal noises (30%) such as barking, bird noises, and monkey noises. Other simple vocal tics included blowing (10%), and hiccupping (10%). The most frequent complex vocal tic was copropraxia (50%) followed by palilalia (40%), words (30%) such as "Hi", echolalia (30%), syllables (20%), blocking (20%), sentences (10%) such as " I'm telling you Jack," stuttering (10%), and jargon (10%).

Table 7. Subjects Simple And Complex Vocal Tics

Description	percentage
Simple vocal tics	
Throat clearing	60%
Sniffing	50%
Animal noises	30%
Barking	20%
Bird noises	10%
Monkey noises	10%
Others	
Blowing	10%
Hiccups	10%
Complex vocal tics	
Coprolalia	50%
Palilalia	40%
Words	30%
Echolalia	30%
Syllables	20%
Blocking	20%
Sentences	10%
Stuttering	10%
Jargon	10%
Others	
Speaking with a tense stomach	10%

Global Tourette Syndrome Severity

The overall Tourette syndrome impairment represents the third component of the global severity score in the Yale Global Tic Severity Scale and examines the effect of Tourette syndrome on the individual's self esteem, family life, social acceptance,and job or school functioning. A maximum of 50 points can be obtained and it represents 50% of the total severity score.

The global severity score in the Yale Global Tic Severity Scale is comprised of ratings for the motor tic severity, the vocal tic severity, and the overall Tourette syndrome impairment. These scores are presented in Table 8 for the subjects in this study. The mean for global severity of Tourette syndrome was 55 (SD = 19.78) with the range of 28 to 96. The maximum possible score was 100 indicating the most severe Tourette syndrome case. At the present time, the Yale Global Tourette Syndrome Severity Scale does not provide descriptive categories for the global severity scores.

Table 8. Subjects' Tourette Syndrome Severity Based on The Yale Global Tic Severity Scale

ID	Global Severity	Motor Tics Severity *	Vocal Tics Severity *	Overall TS Impairment
1	96	23	23	50
2	76	18	18	40
3	56	18	18	20
4	52	16	16	20
5	45	17	18	10
6	43	11	17	10
7	28	8	10	10
8	40	10	10	20
9	67	13	14	40
10	48	14	14	20
Mean	55.10	14.80	15.80	24.00
SD	19.78	4.49	3.96	14.29
Range	28-9	8-23	10-23	10-50

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* Both the motor and vocal tic severity scores are the sum of their respective subcomponents comprising of number, frequency, intensity, complexity, and interference.

Communication Abilities

The underlying communication functions including hearing, language, and motor speech control were examined in each subject. A portable audiometer was used for the auditory screening. The Western Aphasia Battery (Kertesz, 1982) was employed for assessing the language functions. The Frenchay Dysarthria Battery (Enderby, 1983) was used to examine the motor speech control.

Language

The Western Aphasia Battery (WAB) (Kertesz, 1982) was employed to assess the language performance in each subject. All the subjects acquired the highest possible scores on spontaneous speech (content and fluency), comprehension (yes/no questions, auditory word recognition, and sequential commands), and naming subtests of object naming, sentence completion, and responsive speech. However, the Aphasia Quotient (AQ) computed for each subject in the present sample showed that 60% of the sample failed to acquire the cut-off score of 100. The mean AQ for the sample was 99.4 with (SD = 0.7) with the range of 98.2 - 100. All the subjects who failed to achieve the maximum AQ of 100 had difficulty in the word fluency subtest of the naming section of the WAB. The word fluency subtest requires the subject to orally produce as many animal names as possible during a one minute period. The mean for the word fluency task was 17.3 (SD = 3.3) and the range of 11 to 20. The highest possible score for the word fluency task is 20 which indicates an adequate ability for this language function. It is noteworthy to mention that the lower scores in the animal naming task was not as the result of tic occurrences.

Motor Speech

The Frenchay Dysarthria Battery (FDA) (Enderby, 1983) was employed to assess the motor speech functions in each subject. In general, subjects performed normally on the FDA's tasks. Nevertheless, some minimal difficulties were observed in 60% of the sample. The most common problems were observed for the following FDA sections: reflexes (60%), larynx (50%), palate (40%), respiration (10%), and lips (10%) (Table 9). The reflex section contains subtests of cough, swallow, and dribble/drool. Sixty percent of the sample reported occasional coughing or choking when eating. In one case, the subject reported that he had to take particular care while eating or drinking. Thirty percent of the sample reported swallowing problems i.e., food going down the respiratory passageway. Two cases reported that they avoid solid foods such as steaks. One subject reported that tongue tics result in chocking while eating. He added that in such cases he must throw the food out of his mouth. The third case reported that he must eat a little bit at a time, otherwise he would have stomach tics which would cause him vomit. Finally, 30% of the sample reported occasional dribbling.

Laryngeal problems occurred in 50% of the sample. The observed problems were as follows: difficulty in holding the vowel /a/ in tic free productions for 15 seconds (30%), pitch cracks (10%), difficulty in production of tic free glides (10%), difficulty controlling the volume of the speech (10%), and in 20% of the sample the laryngeal tasks triggered tics. Forty percent of the sample had problems with the palatal subtests. Thirty percent of this sample reported occasional difficulty with fluids coming down the nose when drinking, and 10% (one case) showed imbalanced nasal resonance during speech. Lastly, one subject had difficulty with adequately maintaining pressure by sealing the lips. This task required the subject to blow air into the cheeks and maintain it for 15 seconds.

Table 9. Subjects' Dysarthria Assessment Results

Mechanism	**Percentage**
Reflex	60%
Occasional coughing and chocking	60%
Swallowing problems	30%
Dribbling / drooling	30%
Larynx	50%
Difficulty holding /a/ for 15 sec.	30%
Pitch cracks	10%
Difficulty producing glides	10%
Difficulty controlling volume	10%
Trigger of tics as the result of all the laryngeal tasks	10%
Palate	40%
Fluid coming down the nose	30%
Imbalance resonance during speech	10%
Lips: difficulty sealing for 15 sec.	10%

RESULTS

The tasks were grouped according to four categories: pitch, loudness, rate, and prosody. The first three conditions included habitual and atypical production conditions at the vowel and sentence levels. The habitual productions were those utterances which subjects were instructed to produce the stimuli with their own natural pitch, loudness, and rate. The atypical productions were those utterances which subjects were instructed to deviate from their habitual productions, e.g., whisper, speak with a high pitch, or speak with a fast rate. The forth category, prosody, used only sentence utterances and was composed of four different intonational patterns (two linguistic and two emotional) for one sentence. A total of twenty six (26) tasks, repeated three times in a systematically random order (no single task order was repeated), comprised the experimental component of this investigation. Thus, the group of ten subjects produced thirty (30) utterances of each task, for a total of 780 productions.

The dependent variable of the present study was the number of tic-filled utterances (productions accompanied with one or more vocal tics) associated with each task. This study examined how different laryngeal and vocal tract modifications affected vocal tics compared to the habitual productions. A series of two-by-two chi-square tests were used to test the differences between the habitual condition and their respective atypical conditions.

Table 10 shows the number of tic-filled utterances (out of seventy eight utterances) for each subject. Overall seventy four percent of the sample (579 utterances) were tic-free and 26% were tic-filled. Three subjects produced most of the tic-filled utterances ranging from 41 to 46. Four subjects produced tic-filled utterances ranging from 11 to 16, and the three remaining subjects produced minimal number of tic-filled utterances ranging from 2 to 4. The total number of tic-filled utterances produced by each subject for each experimental task (consisting of three trials) is provided in the appendix of this document.

To determine whether the variability of the number of tic-filled utterances across subjects was related to their Tourette syndrome severity, the total number of tic-filled utterances for all subjects were correlated with their Vocal Tic Number component of the Yale Global Tic Severity Scale. The Spearman rank order correlation value was found to be 0.750 ($p < 0.02$).

Table 10. Subjects' Tic-Filled Utterances Recorded For All The Experimental Tasks

Subject	Tic-filled
1	41
2	4
3	46
4	42
5	16
6	11
7	2
8	2
9	14
10	13
Total Utterances	**201**

In order to assure that the three trials for each task did not have any adaptation effect on the subjects' performance, the number of tic-filled utterances associated with each trial within each task were compared. Insignificant chi-square ($p > 0.05$) test statistics values were obtained for all the experimental tasks.

Pitch

Compared to the habitual pitch vowel productions, tic-filled utterances increased in all of the atypical pitch tasks (Table 11). The number of tic-filled utterances associated with the habitual, high, low, glide up, and glide down were 2, 7, 5, 10, and 8 respectively. Compared to the habitual pitch vowel, tic-filled utterances significantly increased with glide-up ($X^2 = 6.67$, df = 1, and $p < 0.05$) and glide-down ($X^2 = 4.32$, df = 1, and $p < 0.01$) conditions.

An almost similar number of tic-filled utterances were associated with the habitual (10), high (9), and low (12) pitch sentences. Insignificant chi-square values ($p > 0.05$) were obtained when the number of tic-filled utterances associated with high and low pitch sentences were each compared with the habitual sentence.

Table 11. Tic-Filled, Tic-Free, And Chi-Square Values Associated With Pitch Tasks

Stimuli	Tic-free	Tic-filled	Chi-Square
Vowel			
Habitual	28	2	
High	23	7	3.27
Low	25	5	1.45
Glide up	20	10	6.67 **
Glide down	22	8	4.32 *
Sentence			
Habitual	20	10	
High	21	9	0.07
Low	18	12	0.38

NOTE: Each of the nonhabitual tasks were compared with their respective habitual tasks.

* denotes $p < 0.05$

** denotes $p < 0.01$

Loudness

Compared to the habitual loudness vowel productions, tic-filled utterances increased with the loud voice and stayed about the same with the soft and whispered productions (Table 12). The number of tic-filled utterances associated with the habitual, loud, soft, and whispered vowels were 3, 11, 2, and 4. Compared to the habitual loudness vowel,tic-filled utterances significantly increased with the loud vowel ($X^2 = 5.96$, df = 1, and $p < 0.05$).

The number of tic-filled productions associated with the habitual loudness and loud sentences were relatively similar. However, compared to the habitual loudness sentence productions, tic-filled utterances decreased with the soft and whispered sentences (Table 12). The number of tic-filled utterances associated with the habitual, loud, soft, and whispered sentences were 13, 10, 5, and 2. Compared to the habitual loudness sentence, the number of tic-filled utterances significantly <u>decreased</u> with the soft ($X^2 = 5.08$, df = 1, and $p < 0.05$) and whispered ($X^2 = 10.76$, df = 1, and $p < 0.005$) sentences.

Table 12. Tic-Filled, Tic-Free, And Chi-Square Values Associated With Loudness Tasks

Stimuli	Tic-free	Tic-filled	Chi-Square
Vowel			
Habitual	27	3	
Loud	19	11	5.96 *
Soft	28	2	0.21
Whisper	26	4	0.16
Sentence			
Habitual	17	13	
Loud	20	10	0.63
Soft	25	5	5.08 *
Whisper	28	2	10.76 ***

NOTE. Each of the nonhabitual tasks were compared with their respective habitual tasks.

* denotes $p < 0.05$

*** denotes $p < 0.005$

Rate

Compared to the habitual rate of vowel repetitions, tic-filled productions slightly increased with the fast repetitions and remained the same for the slow repetitions (Table 13). The number of tic-filled utterances associated with the habitual, fast, and slow rate vowel repetitions were 8, 11, and 8. The chi-square values were found to be insignificant ($p > 0.05$).

Compared to the habitual sentence rate, tic-filled productions slightly decreased with the fast and slow sentence rate (Table 13). The number of tic-filled utterances associated with the habitual, fast, and slow rate sentences were 9, 5, and 7. No significant differences were obtained ($P > 0.05$). A summary of the pitch, loudness, and rate data for the vowel and sentence stimuli respectively is illustrated in Figure 1.

Table 13. Tic-Free, Tic-Filled, And Chi-Square Values Associated With Rate Tasks

Stimuli	Tic-free	Tic-filled	Chi-Square
Vowel			
Habitual	22	8	
Fast	19	11	0.69
Slow	22	8	0.00
Sentence			
Habitual	21	9	
Fast	25	5	1.49
Slow	23	7	0.34

NOTE: Each of the nonhabitual tasks were compared with their respective habitual tasks.

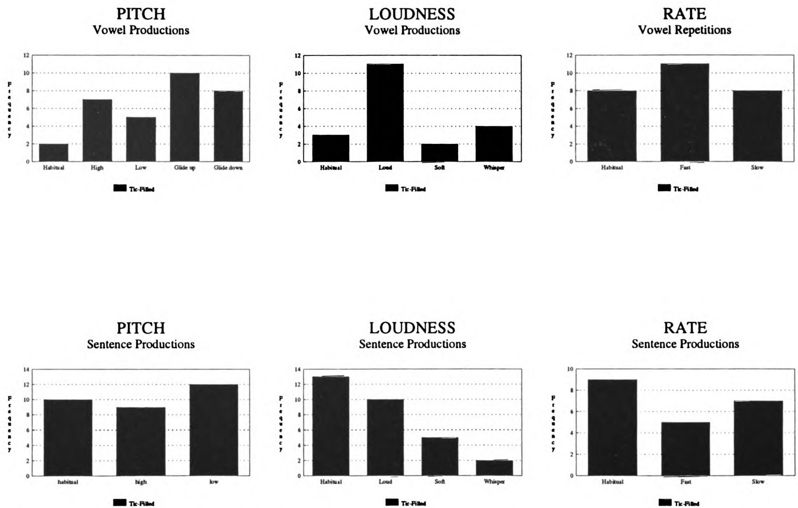

FIGURE 1. A Summary of the Frequency of Tic-filled Utterances Associated with the Pitch, Loudness, and Rate Data for the Vowel and Sentence Stimuli.

PROSODY

The exact same number of tic-filled utterances (10) were associated with statement, question, happy, and angry prosodic productions which clearly resulted in no significant differences (Table 14).

TABLE 14. Tic-Filled, Tic-Free, Associated With Prosody Tasks

Stimuli	Tic-free	Tic-filled
Linguistic		
Statement	20	10
Question	20	10
Emotional		
Happy	20	10
Angry	20	10

Stimulus Level Effect

Inspection of the data indicated that there were more tic-filled sentence productions than tic-filled vowel productions. Thus, a chi-square analysis was employed to determine whether a statistical difference existed. For this analysis, only the data from the pitch and loudness tasks were used. The data from the rate tasks were excluded because the vowel stimuli required repetitions of the vowel /a/ as opposed to only one vowel production in the pitch and loudness tasks.

Each habitual vowel and sentence productions comprised of six trials (three trials of the habitual pitch and three trials of the habitual loudness). Thus, a sample of 60 utterances was analyzed for each stimulus level. Among the 60 habitual vowel productions, five utterances were tic-filled. However, among 60 habitual sentence productions 23 utterances were tic-filled (Table 15). The tic filled utterances were significantly associated with the sentence productions compared to vowel utterances ($X^2 = 5.35$, $p < 0.05$).

Table 15. Tic-Free, Tic-Filled, And Chi-Square Value Associated With The Vowel And Sentence Productions

Production	Tic-free	Tic-filled	Chi-square
Habitual vowel	55	5	
Habitual sentence	37	23	5.35 *

* denotes $p < 0.05$

Vocal Tic Positions in The Sentences

When the vocal tic positions were inspected, tics almost never interrupted the vowels and appeared either prior to or after the vowel productions. For the sentence productions, vocal tics appeared mostly before (57) and after (67) the sentence. Occasionally, vocal tics interrupted the sentences, mostly in more severe subjects. In fact, mainly two subjects produced almost 90% of the tics (22) which occurred within the sentence utterances.

Friedman two-way analysis of variance was employed to determine whether the subjects tended to produce higher frequencies of vocal tics at any of the three sentence positions. Friedman's test was used because the data consisted of one class of observations (the position of vocal tics).Friedman's test is a nonparametric test of ranked data and is an analogous to the randomized block analysis of variance design (Kirk, 1968, Pg. 498). In this design each subject is considered as a block. Friedman test can be used when repeated measures on the same subjects are obtained (Kirk, 1968). Friedman's computational formula is

$$\chi^2 = \frac{12}{nK(K+1)}\left[\sum_{i=1}^{k}\left(\sum_{j=1}^{n} R_{ij}\right)^2\right] - 3n(k+1)$$

df = K -1

where n = number of subjects,

K = categories

R = ranked data

There was a significant difference in the frequency of vocal tics across the three positions (X = 5.850, df = 2, and $p < 0.07$). Table 16 shows sums of the ranks for the vocal tic positions occurred before (21.5), within (14.0), and after (24.5) the sentence productions.

TABLE 16. Rank Sums Associated with The Position of Vocal Tics During Sentence Productions

Position	**Rank Sum**
Before	21.5
Within	14.0
After	24.5

A series of Sign tests were performed to determine which pairs of positions were significantly different. In the Sign test each subject is assigned a positive (+) or a negative (-) sign, depending on whether the frequency of which position (e.g., before or within) is higher. If there is no differences the subject is assigned a zero. For example, if a subject's vocal tic position frequency is higher before the sentence starts he/she is assigned a positive (+) sign, if his/her vocal tic position frequency is higher within the sentence, he/she is assigned a (-) sign. If these frequencies are the same he/she is assigned a zero. The total number of positive signs are compared with the total number of signs (positive and negative) to determine the probability of positive signs occurring by chance (Silverman, 1985, Pg. 290-291). Both before and after the sentence tic positions were significantly different from those which occurred within the sentences (sign test = 7, $p < 0.05$). However, a comparison between the before and after positions was insignificant ($p > 0.05$).

DISCUSSION

SUBJECTS' BACKGROUND INFORMATION

The purpose of the present study was to investigate the relationship between motor speech control for prosody and the occurrence of vocal tics in individuals diagnosed with Tourette syndrome. The experimental tasks included repetition of one vowel and one sentence with different pitch, loudness, and rate levels. The sentence was also repeated with two linguistic and two emotional prosodies. Each task was repeated three times in a systematic random order. For each subject, the number of tic-filled utterances associated with the conditions were measured.

Ten volunteer subjects with Tourette syndrome participated in this study. Prior to the experimental tasks, background information, Tourette syndrome variables, severity of the disorder, language performance, and motor speech status were investigated in each subject.

Demographic Data

Although all the subjects who participated in the present study had been medically diagnosed as having Tourette syndrome, they were diverse with respect to their individual characteristics. The following provides a brief discussion about these findings.

The male-to-female ratio in the present study was 4:1 which reflects the demographic studies suggesting the higher frequency of Tourette syndrome among males (Shapiro et al., 1988). The gender variable has been found to be unrelated to the severity and characteristics of the symptoms (Shapiro et al., 1988). Subjects were variable regarding their age. Bruun (1988) and Kerbeshian (1991) found that each decade after age 20 contains a higher percentage of mild cases, a finding which was consistent with the sample of this study. The wide variability in education and occupation background found in this sample agrees with the epidemiological studies postulating that Tourette syndrome is unrelated to the individual's social class (Asam, 1982; Golden, 1982). Regardless of the education level, present subjects found that Tourette syndrome interferred with their school work. Hagin and Kugler (1988) also proposed that Tourette syndrome implicates school activities directly (e.g., taking notes and public speaking are interfered by tics) and indirectly (e.g., concentration on holding tics interferes with the learning processes). The unemployed subjects in the present sample reported an excessive difficulty in finding jobs. Stefl (1983) and Bruun (1988) reported that individuals with Tourette syndrome are often subjected to employment difficulties and job discrimination. Finally, subjects were concerned that social isolation resulted from their symptoms. The individual's life situation has a direct implications on the global severity of Tourette syndrome. Shapiro and colleagues (1988) stated that although Tourette syndrome is an organic disorder, variables affecting an individual's psychological functioning contribute to the characteristics of this disorder.

Clinical Symptoms

Subjects were also heterogenous regarding their Tourette syndrome clinical symptoms. Variables of age at onset, remission, medication, associated neurological problems, psychopathology, and other behavioral problems were sources of variation in the present sample. A large range of age at onset was observed for the subjects in this study. The motor tics occurred earlier than the vocal tics in all subjects. All subjects reported symptom fluctuations and severity changes; and two subjects reported temporary remissions in the course of their disorder. External and internal stimuli influenced the tic occurrences. Half of the sample took medications for Tourette syndrome at the time of the present study; none of the subjects had behavioral therapy. Finally, Tourette syndrome was associated with neurological, psychological, and behavioral problems. The following provides a brief discussion about these findings.

The lowest reported age at onset was three, whereas the highest was 18 years of age in this sample. DSM-III-R (1987) requires the age at onset of below 21 year for Tourette syndrome. The elevation of the mean age at onset for vocal tics supports the previous literature stating that chronologically the initial phase of the syndrome is characterized by motor tics and that vocal tics occur later in the course of the syndrome (Cohen et al., 1985). The large difference between the age at onset and age at diagnosis indicated that compared to younger subjects, the older ones were diagnosed later in the course of their disorder. Shapiro et al. (1988) stated that the decreased age at diagnosis demonstrates the increased recognition of the disorder among the health care professionals. Symptom fluctuations and severity changes reported in the present study correspond with the DSM-III-R (1987) criteria for Tourette syndrome and have also been reported by other investigators (Lieh-Mak et al., 1982; Leckman et al., 1985; and Shapiro et al., 1988). The medication intake was the individual's choice for the subjects in this sample. While the medication decreased, but did not eliminate, the tics, it also had disturbing side-effects.

Other authors also reported that the external and internal stimuli affect the symptoms in Tourette syndrome (Bruun, 1989; Shapiro et al., 1988; Sacks, 1987; Sacks 1992). In the present sample, fatigue and unpleasant emotionality (e.g., anxiety and anger) increased the tics.

However, pleasant feelings, unless excessive, decreased the symptoms. Increased attention had an extreme influence on decreasing the tics for all the subjects. Different mechanisms might explain the stimuli role in various situations. It seems that fatigue and extreme excitations (positive and negative) lessen the volitional ability to control the tics and the desire to consciously inhibit them. On the other hand, active attention assists both the spontaneous and volitional inhibition of the tics. In addition, control mechanisms in Tourette syndrome may be closely related to inter-intra-personal communication skills, coping strategies, attitudes, and social adjustments in an individual with Tourette syndrome.

Subjects in this study had various concomitant behaviors including, attention deficit disorders (40%), hyperactivity (60%), secondary depression (30), obsessive-compulsive-like symptoms (90%), and behavioral problems (60%). Such problems associated with Tourette syndrome have been documented for a subgroup of subjects in the literature. Some researchers maintain that Tourette syndrome is more severe when it is accompanied by attention deficit disorders and hyperactivity (Leckman et al., 1985; Comings and Comings, 1988; Shapiro et al., 1988). Depression is not intrinsic to Tourette syndrome (Shapiro et al., 1988), however, it may be developed as a secondary reaction to Tourette syndrome (Leckman and Cohen, 1988; Silver, 1988). Despite the controversy about the linkage between obsessive compulsive disorders and Tourette syndrome, authors have reported the presence of obsessive-compulsive-like symptoms in this population (Lees, et al., 1984; Leonard, 1992). A higher rate of behavioral problems among the individuals with Tourette syndrome has been reported by others (Stefl, 1983). Some authors attribute these behavioral problems to the severity of Tourette syndrome (Robertson, 1991), whereas others suggest them to be an indication of poor coping and adjustment strategies (Asam, 1982).

Tourette Syndrome Severity

Subjects were variable regarding their motor and vocal tic type, severity, and overall functional impairment resulting from Tourette syndrome indicated by the Yale Global Tic Severity Scale (Leckman et al., 1989). Symptom variability in Tourette syndrome has been well-documented in the literature (Caine, Polinsky, Lodlow, Ebert, and Nee, 1982). In the present sample, the most common simple motor tics were eye-blinking and head jerks and the most common complex motor tics were touching, grooming, and copropraxia. Similar findings have been reported by other authors (Comings and Comings,1988; Lieh-Mak et al., 1982; Lees et al., 1984; and Robertson, 1991). Furthermore, the most common simple tics were laryngeal tics and the most common complex vocal tics were coprolalia and palilalia in the present sample. Other authors have reported similar findings (Ludlow et al., 1982; Lees et al., 1984). In the present study, Tourette syndrome was generally milder in subjects over 40 years of age. Subjects were variable regarding the effect of Tourette syndrome and their psychological state. It was noted that subjects who had less severe Tourette syndrome had more positive self-esteem. Also, subjects who were better educated, employed and had emotional support rated themselves as being less affected by their symptoms.

Language

Subjects seemed to be competent in their linguistic abilities. Their spontaneous speech and picture descriptions were adequate in the form, content, and use components of the language. Furthermore, their language comprehension and production were normal based on the Western Aphasia Battery (WAB) (Kertesz, 1982) results. Although the subjects did not present language disturbances based on the the WAB cut-off score, the scores were slightly suppressed for more than half of the sample (60%). The Western Aphasia Battery is a simple enough test which normal subjects regularly achieve the maximum score of 100 (Kertesz, 1982). The word fluency subtest of oral naming section of the WAB was the unanimous source of suppression for the six subjects. The low scores achieved on the word fluency task were not related to the vocal tic frequency. Subjects adequately performed on the object naming, responsive naming, and sentence completion tasks. In addition, subjects' lexicon was functionally and sufficiently reflected in their spontaneous speech.

Previous studies, however, have reported the presence of language difficulties in subjects with Tourette syndrome. Ludlow et al. (1982) found a reduction of oral language in their sample of subjects with Tourette syndrome. The oral language impairment was not related to the overall severity of vocal tics. The present study, however, showed a slight reduction only in the word fluency subtest. Two factors may explain the difference found between the two studies: (1) the sensitivity of the language assessment tools and (2) the presence of subgroups within the Tourette syndrome population. In fact, in a recent study of language characteristics, Brookshire (1991) compared a group of the school-aged children with Tourette syndrome with their normal sibling controls. Although no significant differences were found between the two groups, the author noted the presence of subgroups who performed poorly on both language and fluency measures. The reduced word fluency in Tourette syndrome has been reported by other authors, too. In a neuropsychological assessment of 32 adults and children subjects with Tourette syndrome, Sutherland, Kolb, Schoel, Whishaw, and Davies (1982) asked each subject (1) to name as many objects and then animals as possible in one minute respectively and (2) to alternate between giving colors and bird names as many as possible in one minute. Compared to control subjects, individuals with Tourette syndrome achieved significantly lower scores in the word fluency tasks. However, the authors did not report the medical status of their subjects.

In the present study, subjects who showed the word fluency difficulty were those who had one or more of the following: greater severity,on medication, and having a learning disability. Therefore, based on these findings the source of the word fluency problem in the present sample may be attributed to these variables and not a language impairment.

Motor Speech Control

Based on the Frenchay Dysarthria Battery (Enderby, 1983), subjects appeared to have a competent motor speech control for the speech tasks. On the other hand, subjects demonstrated some minor difficulties in some non-speech activities. The striking characteristics evidenced by the motor speech assessment was the swallowing problem reported by more than half of the sample (70%). For some, this problem was complicated by minimal reduction of laryngeal and respiratory functions for the non-speech tasks (e.g., decreased maximum phonation time unrelated to the vocal tics). For other subjects, the laryngeal non-speech tasks (e.g., glides and vowel repetitions) induced vocal tics. Enderby (1983) provided a motor speech profile for individuals with extrapyramidal impairments. In her profile, however, subjects were more impaired in speech tasks compared to the non-speech ones. Contrary to her findings, in the present sample the difficulties were mainly associated with the non-speech tasks. Two explanations for this contradiction may be offered. One is that Enderby (1983) observed a large variability in her findings resulted from having a sample with diverse extrapyramidal disorders (e.g., Parkinson disease and chorea). Alternatively, it is highly possible that her sample did not include subjects with Tourette syndrome.

Darley et al. (1975) maintained that strength, speed, range, accuracy, and motor steadiness of contractions are salient features of neuromuscular functions. Impairments in these functions affect various aspects of speech. A breakdown in motor steadiness results in one of the two deviations: tremor or random involuntary movements. In general, the results of this study suggest that excluding the motor steadiness, other salient neuromuscular features (strength, speed, and accuracy) of speech movements appear to be intact in Tourette syndrome.

Moreover, it was interesting to note the high rate of a swallowing reflex problems in this sample. Sweet et al. (1973) reported hyperflexia in some cases with Tourette syndrome. The fact that both the laryngeal tics and swallowing reflex problem are the most common symptoms in this sample is noteworthy. The swallowing reflex is triggered when the pharyngeal stage of the swallow begins (Logemann, 1983). The swallowing reflex results in the elevation and closure of the larynx at all three spaces (epiglottis, false vocal folds, and true vocal folds) (Zemlin, 1988). The swallowing control center is located in the medullary reticular formation within the brainstem (Zemlin, 1988). It might be hypothesized that the swallowing reflex problem in this sample results from an impaired reflexive control mechanism for the larynx. Furthermore, this hypothesis might explain the frequent occurrence of laryngeal tics in this population.

EXPERIMENTAL TASKS

The purpose of the experimental tasks was to study the relationship between the vocal tics and motor speech control. The specific questions of this study were as follow: (1) are vocal tics related to pitch changes in speech, compared to the habitual pitch?; (2) are vocal tics related to loudness changes in speech, compared to the habitual loudness?; (3) are vocal tics related to rate changes in speech, compared to the habitual rate?; and (4) are vocal tics related to the overall prosodic contour during linguistic and emotional prosody? The experimental tasks included a range of habitual and atypical speech conditions which required respiratory, laryngeal, and articulatory performances at the vowel and sentence level. The sentence stimuli contained short frequently used words (nine one-syllable words and two two-syllable words) and could be produced with one breath, with no required pauses within the sentence. The underlying assumption for this design was that it would allow for the comparrison of the motor complexity effect of each atypical task with its corresponding habitual pairs. For each task the number of vocal tic-filled utterances were determined. The associations between vocal tic-filled utterances and pitch, loudness, rate, and overall sentence prosody were studied.

The findings of the experimental tasks and their physiological and clinical implications will be discussed.

Pitch

Vocal tics increased with high and low pitches at the vowel and the sentence levels. However, such increases were not significant for the group. On the other hand, glide utterances produced a different pattern. As a group, subjects had a significant increase in vocal tics with rapid ascending and descending pitch changes. Also, further inspection of the data suggests that high and low pitch conditions increased the tics in a subgroup of subjects, specifically in those who had higher Tourette syndrome severity scores. Thus, it is suspected that the present pattern of results might be influenced by the heterogeneity of the sample.

Pitch is the psychological term for the voice fundamental frequency which is the average rate of cycle-to-cycle vocal fold vibrations per second. A direct relationship exists between the rate of vocal fold vibratory control and various pitches. The high pitch requires a faster rate of vocal fold vibrations whereas the low pitch demands a slower rate. The glottal cycles are shorter during the high pitch and longer during the low pitch.

The primary vocal fold adjustments required in pitch changes are tension/length and mass modifications (Zemlin, 1988). An increase in tension and a decrease in mass increases the pitch, whereas a decrease in tension and/or increase in mass of the vocal folds decreases the pitch. Rapid laryngeal changes for length, tension, and mass are regulated by laryngeal reflexes (Kirshner, 1985).

Compared to a steady maintained habitual pitch in the vowel production, the motoric complexity of the task is increased with the glide conditions. Rapid changes in pitch during this condition demands increased and rapid reflexive laryngeal adjustments. It is hypothesized that the increased vocal tics associated with the rapid pitch changes is related to the increased motoric complexity of the task.

Loudness

Speaking loudly significantly increased the vocal tics in vowels. The loud sentences, however, did not significantly differ from the habitual loudness sentences regarding their associations with the vocal tics. On the other hand, whispered sentences significantly decreased the vocal tics.

Loudness is the psychological term for the acoustic intensity of the voice which is predominantly regulated with the subglottal pressure and the laryngeal resistance. These regulations occur below and within the larynx (Zemlin, 1988). Below the larynx, the subglottal pressure is regulated by the respiratory reflexes. Within the larynx, the loudness is regulated by varying the vocal fold resistance. The laryngeal resistance determines the time period which the vocal folds are closed during this cycle. The longer they are closed, the more resistance to the subglottal pressure they produce. Above the larynx, articulatory movements influence the laryngeal resistance for the voiced and voiceless sounds.

A direct relationship exists among the subglottic pressure, the laryngeal resistance and the voice intensity, i.e., the voice intensity increases as the subglottal pressure and the laryngeal resistance increase. Compared to the habitual voice, the soft voice is characterized by minimum subglottal pressure and laryngeal activity, whereas the loud voice increases both of these parameters. Whisper, however, requires minimum resistance demands (Zemlin, 1988). During whisper minimal laryngeal resistance to the subglottal pressure is required and the subglottal pressure is adjusted by reflexive respiratory regulations. In addition, whispered speech has no periodic vibrations, fundamental frequency, or harmonics (Zemlin, 1988).

The decrease of vocal tics with the decrease of the loudness level might be explained by the physiology of phonation. The present results suggest that vocal tics might be related to the regulatory demands of the loudness produced. As the demand for the loudness control decreases, the respiratory and phonatory muscular activities decrease and in turn fewer involuntary vocalizations are evoked. During whisper, the vocal tics, particularly in sentence context, are minimum, when the respiratory activity and laryngeal resistance require simplified motor activities.

Rate

No significant group differences were found for the rate tasks at the vowel and sentence levels. The fast and slow rates change the time interval factor in the vowel repetitions and sentence productions. For the vowel repetition, this time factor is related to the timing coordination between the respiratory and laryngeal activities. For the sentence production the articulatory adjustments are added to the respiratory and laryngeal control. These results indicate that changing the time interval was not associated with the vocal tics.

However, it is important to consider two other relevant findings. First, vocal tics significantly increased with the habitual sentence productions compared to the habitual vowel productions for the rate and loudness conditions. However,

this was not the case for the rate condition. That is, not much difference was observed between the frequency of vocal tic utterances associated with the habitual vowels and sentences. This suggests that the rapid on-off laryngeal activities necessary for the vowel repetitions are associated with increased vocal tics.

Second, compared to one time vowel utterances for the habitual pitch and loudness tasks, the vowel repetitions largely increased the vocal tics. Note that the 60 habitual pitch and loudness vowel utterances were associated with five tic-filled utterances, whereas the 30 habitual vowel repetitions were associated with eight tic-filled utterances. Compared to the habitual pitch vowel, a statisticaly significant (chi-square value = 4.32, $p = 0.05$) vocal tic increase is associated with the habitual vowel repetition task. This observation also suggests that a relationship exists between the rapid on-off laryngeal activities and the vocal tics. Furthermore, this finding agrees with Sayadi and Cooke (1991) who observed similar characteristics in their single subject with Tourette syndrome.

Prosody

The two types of linguistic and emotional prosody did not present different levels of difficulty for the subjects as a group. These data contradict with Sayadi and Cooke's (1991) observations of a single subject with Tourette syndrome. The authors reported one subject whose vocal tics increased with interrogative and emotional prosody, especially with the angry utterances. Although the authors relied on a single subject, methodological differences might also contribute to this contradiction. For example, their study involved a larger sample of utterances. In addition, similar to the results of the other experimental conditions, a subgrouping factor might also play a role in the shape of the present data.

Nevertheless, the finding that vocal tics did not increase with the emotional prosody contradicts other well-known clinical reports (Sacks, 1997, 1992; Shapiro, et al., 1998) and the subjects' own perceptions. The questionnaire data of this study yielded responses which suggested generally tics increase when associated with anger. However, this was not verified by the experimental tasks. One reason for such results may be that the experimental tasks were influenced by the subject's compliancy. The majority of subjects found it difficult to mimic the actual prosody produced in the real emotional situation. Thus, the fact that they had to role play the emotional prosody may be a methodological reason for the present results.

Vowels Versus Sentences

Compared to vowel productions, sentence productions were associated with a significant increase in the number of vocal tics. This finding suggests that the utterance motoric complexity appears to affect the frequency of vocal tics. Recall that all subjects easily passed the sentence formulation tasks on the Western Aphasia Battery (e.g., sentence completion and sentence repetition). This indicates that the sentence repetitions used in the present study did not present any excessive linguistic difficulties for the subjects.

On the other hand, the sentence task allowed the subjects to speak for a longer time compared to their matched vowel productions. This longer period of time might explain why more tics occurred with the sentences during the pitch and loudness tasks. However, this explanation is inconsistent with the rate data of this study. If time length of vocalizations is considered to be related to the activation of vocal tics, then the vocal tics should increase with the slow sentences compared to the habitual rate sentence. Figure 1, however, shows a slight reduction of tics instead of the expected increase. Thus, it is hypothesized that a relationship exists between the frequency of vocal tics and the motoric complexity. Nevertheless, the time factor role should not be disregarded quickly, because when the time period was compresses for the fast sentences, the vocal tics decreased. However, one can not build a hypothesis based on this observation because no significant differences were obtained. Therefore, future research is required to investigate the relationship between the timing role and vocal tics.

Other authors have suggested that vocal tics might be associated with an impaired motor programming (Ludlow, 1982). Based on the results of the present study the significant increase in the number of vocal tics during the sentence productions may be as the result of complex motor control and reflexive adjustments for respiratory, phonatory, and articulatory systems. During the vowel productions the demand for all these three factors is reduced. Furthermore, communicationally, vowel production tasks carry the lowest amount of information and emotion, and require a minimum psychosocial demand regarding the speaker-listener interactions.

Position of Vocal Tics in Sentence Productions

The vocal tic position showed a particular pattern in association with sentences. Compared to the medial position, the vocal tics significantly increased at the initial and final positions. This was noteworthy because such a pattern was observed across all the sentence productions regardless of the motoric focus of the task. This evidence confirms other observations reported in the literature (Martindale, 1975; Ludlow et al., 1982; Sayadi and Cooke, 1991). Martindale (1977) found a similar coprolalia positioning in a single subject with Tourette syndrome. Ludlow and colleagues (1982) noted that vocal tics did not occur randomly in the spontaneous speech of their subjects and suggested that cortical motor programming for speech is complicated in the speech of individuals with Tourette syndrome. In view of the other findings of this study, it is possible to propose that in addition to an impaired cortical motor programming for speech, the entire integrity of motor speech control is involved in Tourette syndrome. More specifically, the vocal tic presence at the beginning and the end of the sentence indicates an involuntary and inappropriate spatial-temporal activation of the vocal tract system, suggesting an impairment of the feedback mechanisms in Tourette syndrome.

Relationship Between Experimental Tasks And Severity

The significant correlation between the experimental tasks and the Vocal Tic Number component of the Yale Global Tic Severity Scale indicated that there is a relationship between the subjects' performances on the experimental tasks and the severity found by the scale. This correlation suggests that both systems were capable of discriminating the subjects regarding their tics. It is suggested that both these assessment tools are capable of providing information regarding different aspects of vocal tics. For each individual, the Yale Global Tic Severity Scale yields particular data regarding the Tourette syndrome severity - the type of vocal and motor tics, their number, frequency, intensity, complexity, interference aspects, and their influence on the individual's psycho-social domains. Diagnostically, the additional advantage of the experimental tasks used in the present study is that they provide specific information about vocal tics in relationship to various functions of the speech apparatus and its subsystems. This allows the clinician to observe particular functions which are involved for any given client with Tourette syndrome. A subgrouping factor was observed in this study. For example, although rate did not prove to be an area of difficulty for the subjects as a group, some subjects had their greatest difficulty with the rate conditions. Clinically, this information offers the speech-language pathologist data for constructing compensatory therapy plans and follow-up evaluations.

Physiological Implications

The results of the present study demonstrate that respiratory, phonatory, and articulatory functions in speech are related to the vocal tics in Tourette syndrome. As to which system plays a key role in Tourette syndrome speech, it appears that the present results implicate the laryngeal functions. Other clinical studies have repeatedly reported that laryngeal vocal tics are proportionally more common among individuals with Tourette syndrome (Ludlow et al., 1982; Lees et al., 1984). The data collected for the present study essentially yielded similar findings. In addition, the experimental conditions evidenced a key relationship between the laryngeal activities and vocal tics: (1) vocal tics increased with the rapid changes for pitch; (2) vocal tics increased with the laryngeal on-off activity; whereas (3) whisper, which reduces the laryngeal activity, significantly reduced the vocal tics. Nevertheless, it should be immediately mentioned that the respiratory, laryngeal, and articulatory systems do not function independently in speech. During speech, subglottal pressure requires constant adjustments of the entire vocal tract in place, length, and rate. Adjustments occurring in one system result in changes and adjustments in the other systems.

The findings of the present study agree with Ludlow et al.'s (1982), suggesting that vocal tics have their origin in normal speech acts. Thus in order to explain the vocal tics, the normal motor speech control and its interaction with the emotional and attentional variables should be considered. Speech is a goal-directed motor act. The goal to be achieved has a temporal-spatial nature for the precise phonetic coding for articulation, fluency, and tone (Netsell, 1983 and 1984). Once the goal is established, the system controls for the initiation and the coordination of the movements and checks for errors. Two different but interrelated control levels regulate the speech acts: voluntary and automatic/reflexive (Darley et al., 1975; Netsell, 1983 and 1984). The volitional level is initiated at the cortical level by specifying and sequencing the exact motor speech targets. This volitional plan is executed via the pyramidal and extrapyramidal pathways to activate the lower motor neuron for the muscle movement. Mistakes are monitored by feedback from the same centers.

Automatic aspects of the movement are mediated by inherent reflexes which are automatic motor responses to a particular stimulus. Once the stimulus occurs, the motor response appears immediately. Reflexes range from less complex (spinal cord and the brainstem) to complex cortical levels. These reflexes are integrated with each other and mediate cortically controlled automatic movements. Automaticity in speech is achieved by less involvement of the cortical structures.

The motor signals are transferred to muscles via pyramidal (direct system) and extrapyramidal (indirect system) pathways (Darley et al., 1975). The pyramidal system, initiating in the cortex, is excitatory and mediates volitional movements. The extrapyramidal system, initiating in the basal ganglia, is both excitatory and inhibitory and mediates various reflexive responses. These two systems function in a harmony to produce smooth movements.

Based on the present results and the neurology of speech, the following explanation for the vocal tics in Tourette syndrome is suggested. The increased presence of the vocal tics during speech of the individual with Tourette syndrome suggests that the existing neurotransmitter hyperactivities in Tourette syndrome make the system especially unstable when the speech pathways are activated. The decreased vocal tics during whisper suggest that when the reflexes were minimized, the vocal tics decreased. It is hypothesized that Tourette syndrome implicates the motor speech control at the reflexive, nonpurposive, and involuntary levels. Indeed, DSM-III-R (1987) criteria define the tics as reflexive, nonpurposive, and involuntary.

Literature (Shapiro, et al., 1988; Graybiel, 1991) suggests that the basal ganglia are involved in Tourette syndrome. The basal ganglia are a part of the extrapyramidal system and are capable of activating reflexive pathways independent of cortical structures. In addition, prior to the execution of the voluntary movement, the cortical plans are transmitted to the basal ganglia and cerebellum for feedback. This feedback then is transmitted to the cortical system and movement is executed. The basal ganglia involvement might explain why tics occur prior to the initiation and termination of speech and why the speech systems are activated unrelated to the linguistic and communication needs.

Clinical Implications

The changes observed during the experimental tasks in the present study suggest that behavioral intervention for reducing the vocal tics in Tourette syndrome is possible. If the hypothesis of impaired motor integration between automatic and involuntary movement is taken, then therapies which enhance such an integration may reduce the vocal tics in speech. Nevertheless, it should also be noted that Tourette syndrome is an organic disorder with an unknown etiology. Thus, the therapy objective can only focus on the compensation but not on the cure of the symptoms.

Furthermore, the present study and reports in the previous literature have indicated the presence of a subgroup of subjects with language and motor speech control involvements. A comprehensive evaluation of an individual with Tourette syndrome requires assessment of these areas, too. The relationship between the vocal tics and respiratory, phonatory, articulatory functions, and rate and rhythm provides specific information about the vocal tics in a given client with Tourette syndrome.

Finally, Sacks (1992) referred to Tourette syndrome as a neuro-psycho-social impairment. On the basis of the Yale Global Tic Severity Scale findings, the present study also found a strong relationship between the overall functional impairment resulting from Tourette syndrome and other variables such as subjects' attitudes and personal and social conditions. Therefore, it is proposed that the evaluation and treatment of an individual with Tourette syndrome should include other factors influencing the symptoms. Factors to be considered may involve personal, educational, and occupational background; the clinical history of Tourette syndrome; associated neurological and psychological problems; attitudes toward Tourette syndrome; prior treatments effects; situations which affect the symptoms including adaptive and adjustment skills; and support systems. Such influencing factors may assist the therapist in prognosis and deciding therapy strategies. Further, the maintenance and carryover of the therapy objectives are closely related to these variables.

SUMMARY AND CONCLUSIONS

INTRODUCTION

Background

Gilles de la Tourette is a neuropsychiatric movement disorder of an unknown etiology which manifests itself primarily with motor and vocal tics. Few studies have studied the vocal tics in this disorder. Darley et al. (1975) asserted that the spontanous contraction of the speech muscles results in the vocal tics. Martindale (1975) and Ludlow et al. (1982) observed that vocal tics occurred at syntactic boundaries in speech. Ludlow et al. (1982) also reported that vocal tics are more common during speech rather than during silence and that laryngeal tics are the most common tic type. Finally, Sayadi and Cooke (1991) reported a single subject with Tourette syndrome whose vocal tics increased during the vowel repetition and emotional prosody. The authors hypothesized a relationship between the vocal tics and the control of prosodic aspects of the speech.

Purpose

The primary purpose of the present study was to investigate the relationship between the vocal tics and the motor control in speech. Specifically, habitual utterances were compared with their paired experimental utterances which involved atypical variations in pitch, loudness, rate, and sentence prosody. In order to investigate the role of motor complexity, these variations were separately studied at the vowel and sentence levels.

The assumption for this design was that when the speech motor control is imposed by various speech demands, these measures may provide insights about the relationship between vocal tics and motor speech control. Physiologically, such an insight may assist in understanding the neuroanatomical and neurophysiological origins of the vocal tics in Tourette syndrome. Clinically, it may provide a base of information with which the clinician can approach behavioral management of vocal tics in this disorder.

EXPERIMENTAL DESIGN

Subjects

Ten adult subjects, two females and eight males, with Tourette syndrome who were members of <u>Tourette Syndrome</u>

Association, Detroit Chapter volunteered to participate in the present study. Prior to the initiation of the study, information regarding the following categories were collected: demographic background, clinical data, severity of Tourette syndrome, hearing, motor speech control, and language performance.

Procedures

Each subject completed the experimental tasks during one session lasting approximately two hours. They were asked to remain relaxed and perform as natural as possible. All the speech samples were recorded on videotape. For each experimental task, the subjects were given instructions and demonstrations.

Stimuli

To control the level of motor complexity, one vowel and one sentence were used for the experimental tasks. The vowel was used because it primarily requires respiratory and phonatory adjustments, with minimum articulatory modifications. In contrast, the sentence was used because it requires adjustments and coordination of the entire vocal tract system. In order to maintain a constant linguistic environment only one sentence was employed. To control the effect of syntactic boundaries, the sentence did not contain phrase or clause boundaries, and it was comprised of short words with a high frequency of occurrence in speech.

Experimental Tasks

The experimental tasks consisted of four categories of tasks: pitch, loudness, rate, and prosody. Each task was repeated three times, producing a sample of 78 utterances for each subject. The pitch tasks required the subjects to produce the vowel with their habitual, high, low, glide up, and glide down pitches. The sentence was produced with subjects' habitual, high, and low pitches. The loudness tasks required the subjects to produce both the vowel and the sentence with their habitual, loud, soft, and whisper. The rate tasks required the subjects to repeat the vowel and the sentence with their habitual, fast, and slow rates. Lastly, the prosody task required the subjects to repeat the sentence with a statement, question, happy, and angry intention.

RESULTS

The results of the present study were as follows:

1. Compared to the habitual pitch, the glide up and glide down tasks significantly increased the vocal tics.

2. Compared to the habitual vowel, the loud vowel significantly increased the vocal tics.

3. Compared to the habitual sentence, the soft and whispered sentences significantly decreased the vocal tics.

4. Compared to the habitual rate, fast and slow rates did not affect vocal tics at either vowel or sentence level.

5. Linguistic and emotional prosody did not have a differential effect on vocal tics.

6. Compared to the vowel productions, sentence productions significantly increased the vocal tics.

CONCLUSIONS

The following conclusions were derived based on the results of the present study:

1. It appears that as the speech motoric complexity increases, the vocal tics increase.

2. It appears that a relationship exists between the vocal tics and laryngeal adjustments for the rapid changes of pitch and for the laryngeal and respiratory adjustments for loudness.

3. The results of the present study appear to suggest that vocal tics result from an intermittent lack of inhibition of the cortical voluntary systems over the reflexive systems during speech.

4. These findings also suggest that the feedback system involved in the motor speech control is implicated in Tourette syndrome, indicated by the inappropriate temporal-spatial activation of the speech system.

5. Diagnostically, the experimental approach used in this study may provide important information about the relationship between the vocal tics and motor speech functions in a client with Tourette syndrome.

6. Therapeutically, methods which enhance the cortical activity during speech may be useful in reducing the vocal tics.

APPENDIX

APPENDIX

SUBJECT'S NUMBER OF TIC-FILLED UTTERANCES FOR ALL THE EXPERIMENTAL TASKS

SUBJECT'S NUMBER OF TIC-FILLED UTTERANCES ASSOCIATED WITH PITCH TASKS

TASK	S1	S2	S3	S4	S5	S6	S7	S8	S9	S10
VOWEL /a/										
Habitual	0	0	1	0	0	0	0	0	0	1
High	2	0	1	1	2	0	0	0	0	1
Low	0	0	0	2	1	1	0	0	1	0
Glide up	2	0	3	1	0	2	0	0	0	2
Glide down	2	0	1	2	0	0	0	0	1	2
SENTENCES										
Habitual	1	0	3	1	3	0	0	0	1	1
High	3	1	0	3	2	0	0	0	0	0
Low	3	0	2	3	2	0	1	0	0	1

SUBJECT'S NUMBER OF TIC-FILLED UTTERANCES ASSOCIATED WITH LOUDNESS TASKS

TASK	S1	S2	S3	S4	S5	S6	S7	S8	S9	S10
VOWEL /a/										
Habitual	0	0	2	0	1	0	0	0	0	0
loud	3	1	0	2	2	1	0	2	0	0
Soft	1	0	0	1	0	0	0	0	0	0
Whisper	0	0	1	2	0	1	0	0	0	0
SENTENCES										
Habitual	3	1	3	1	3	0	0	0	1	1
Loud	3	0	2	3	1	1	0	0	0	0
Soft	1	0	3	0	1	0	0	0	0	0
Whisper	0	0	0	2	0	0	0	0	0	0

SUBJECT'S NUMBER OF TIC-FILLED UTTERANCES ASSOCIATED WITH RATE TASKS

TASK	S1	S2	S3	S4	S5	S6	S7	S8	S9	S10
VOWEL /a/ *										
Habitual	1	0	3	2	2	0	0	0	0	0
Fast	2	0	2	3	1	1	0	0	1	1
Slow	0	0	3	2	1	0	0	0	2	0
SENTENCES										
Habitual	2	1	2	2	0	0	0	0	2	0
Fast	3	0	0	1	0	0	0	0	0	1
Slow	0	0	3	2	1	0	0	0	1	0

* Vowel /a/ was repeated for the rate tasks.

SUBJECT'S NUMBER OF TIC-FILLED UTTERANCES ASSOCIATED WITH PROSODY TASKS

TASK	S1	S2	S3	S4	S5	S6	S7	S8	S9	S10
LINGUISTIC										
Statement	2	0	3	2	1	1	0	0	1	0
Question	1	0	2	2	1	2	1	0	0	1
SENTENCES										
Happy	3	0	3	1	0	0	0	0	3	0
Angry	3	0	3	3	0	1	0	0	0	0

REFERENCES

Aronson, A.E., (1985). Clinical voice disorders, Thieme Inc., New York.

Abuzzahab. F.S., and Anderson, F.O. (1976). Gilles de la Tourette syndrome, Vol. 1. Mason, St. Paul, Minnesota.

Asam, U. (1982). A follow-up study of Tourette syndrome. In Advances in Neurology, 35, Gilles de la Tourette syndrome, (eds) A.j. Friedhoff and T.N. Chase, pp. 285-286. Raven Press, New York.

American Psychiatric Association (1980). Diagnostic and Statistical Manual of Mental Disorders (third edition), Author, Washington, DC.

American Psychiatric Association (1987). Diagnostic and Statistical Manual of Mental Disorders (third edition revised), Author, Washington, DC.

Azrin, N.H., Nunn, R.G., and Frants, S.E. (1980). Habit reversal vs. negative practice treatment of nervous tics. Behavior Therapy, 2; 160-178.

Balthasar, K. (1956). Uber das anatomishe substrat der generaliserten tic-krankeit (maladie des tics, Gilles de la Tourete), entwickleungshemmung des corpus striatum. Archives Psychiatry Nervenkr, 195; 531-549.

Barnes, G.J. (1983). Suprasegmental and prosodic considerations in motor speech disorders. In Clinical dysarthrias, (ed) Berry, W.R., College-Hill Press, California.

Beard, G.M. (1880). Experiments with the "jumpers" or jumping Frenchmen" of Main. Journal of Nervous and Mental Disease, 7, 487-490.

Bing, R. (1925). Uber lokale muskelspasmen und tics. Schweizerisch Medizinch Wochenschrift, 6, 993-997.

Billings, A. (1978). Self-monitoring in the treatment of tics, a single subject analysis. Journal of Behavior Therapy & Experimental Psychiatry, 9, 339-342.

Bliss, J. (1980). Sensory experiences of Gilles de la Tourette syndrome. Archives of General Psychiatry, 37, 1343-1347.

Brokshire, B., (1991). Fluency and language charactersitics of children with Tourette syndrome. ASHA.

Bruun, R. (1984). Treatment of Tourette syndrome with Pimozide (Orap). Tourettte Syndrome Association Newsletter.

Bruun, R. (1986). Treatment of Tourette syndrome with Pimozide (Orap). Tourette Syndrome Association Newsletter.

Bruun, R.D. (1988). The natural history of Tourette syndrome. In Tourette syndrome and tic disorders, clinical understanding and tratment, (eds) D.J. Cohen, Brunn, R.D., Leckman, J.F., pp. 21-40. Wiley & Sons. New York.

Burd, L. (1991). Education and TS Child. Paper presented at the Second Scientific International Symposium on Tourette Syndrome. Botston, MA.

Burd, L., and Kerbeshian, J. (1988). Nocturnal coprolalia and phonic tics. American Journal of Psychiatry, 145:1, 132.

Burns, K.L., and Beier, E.G. (1973). Significance of vocal and visual channels in the decoding of emotional meaning, Journal of Communication, 23, 118-130.

Caine, E.D. (1985). Gilles de la Tourette syndrome, a review of clinical and research studies and considerations of future directions for investigation. Archives of Neurology, 42, 392-397.

Caine, E.D., Polinsky, R.J., Ludlow, C.L., Ebert, M.H., and Nee, L.E. (1982). Heterogeniety and Variability in Tourette syndrome. In Advances in Neurology, 35, Gilles de la Tourette syndrome, (eds) A.j. Friedhoff and T.N. Chase, pp. 437-442. Raven Press, New York.

Canavan, A.G.M. and Powell, G.E. (1981). The efficacy of several treatments of Gilles de la Tourette syndrome as assessed in a single case. Behavior Research and Therapy, 19, 549-556.

Caprini, G., and Melotti, V. (1961). Un grave sindrome ticossa guarita con haloperidol. Rivivista Sperimentale di Freniatria e Medician Legale Delle Alienazioni Mentali, 85, 191-196.

Chase, T.N., Foster, N.L., Fedio, P., Brooks, R., Mansi, L., Kessler, R., and DiChiro, G. (1984). Gilles de la Tourette syndrome, studies with the fluorine-18-labeled fluorodeoxy-glucose positron emission tomographic method. Annals of Neurology, 15, 175.

Cohen, D.J., Leckman, J.F., and Shaywitz, B.A. (1984). The Tourette syndrome and other Tics. In The clinical guide to child psychiatry (Eds) Shaffer, D., Ehrhardt, A.A., and Greenhill, L.L., Free Press, New York.

Colsher, P.L., Cooper, W.E., and Graff-Radford, N. (1987) Intonational variability in the speech of right hemisphere damaged patients. Brain and Language, 32, 379-383.

Cooper, W.E., and Sorensen, J.M. (1981). Fundamental frequency in sentence production, Springer-Verlag, New York.

Comings, D.E., Comings, B.G., Devor, E.J., and Cloninger, C.R.(1984). Detection of major gene for Gilles de la Tourette syndrome. American Journal of Human Genetics, 36, 586-600.

Comings, D.E., and Comings, B.G., (1984). Tourette syndrome and attention deficit disorder with hyperactivity, are they genetically related? Journal of American Academy of Child Psychiatry, 23, 138-146.

Comings, D.E., & Comings, B.G. (1988).Tourette syndrome and attention deficit disorder. In Tourette syndrome and tic disorders, clinical understanding and tratment, (eds) D.J. Cohen, Brunn, R.D., Leckman, J.F., pp. 119-135. Wiley and Sons. New York.

Darley, F., Aronson, A., & Brown, J. (1975). Motor speech disorders, Saunders, Philadelphia.

Devinskey, O. (1983). Neuroanatomy of Gilles de la Tourette syndrome, possible midbrain involvement. Archives of Neurology, 40, 508-514.

Domino, E.F., Pigott, L., Demetrioue, S., and Culbert, J. (1982). Visually evoked responses in Tourette syndrome. In Advances in Neurology, 35, Gilles de la Tourette syndrome,(eds) A.J. Friedhoff and T.N. Chase, pp. 115-120. Raven Press, New York.

Enderby, P. (1983). The standardized assessment of dysarthria is possible. In Clinical Dysarthria, (ed) W.R. Berry, pp. 109-120. College-Hill Press, San Diego.

Fahn, S. (1982). A case of post-trumatic tic syndrome. In Advances in Neurology, 35, Gilles de la Tourette syndrome, (eds) A.J. Friedhoff and T.N. Chase, pp. 349-350. Raven Press, New York.

Fahn, S., and Erenberg, G. (1988). Differential diagnosis of tic phenomenoa, a neurologic perspective. In Tourette syndrome and tic disorders, clinical understanding and tratment, (eds) D.J. Cohen, Brunn, R.D., Leckman, J.F., pp. 41-54. Wiley & Sons. New York.

Fairbanks, G. (1960). Voice and articulation drillbook. Harper, New York.

Frankel, M., Cummings, J.L., Robertson, M.M., Trimble, M.R., Hill, M.A., & Benson, D.F. (1986). Obsessions and compulsions in Gilles de la Tourette syndrome. Neurology, 36, 378-382.

Goodglass, H. and Kaplan, E. (1972). The assessment of aphasia and related disorders, Lea and Febiger, Philadelphia.

Golden, G.S., (1982). Tourette syndrome in children, ethnic and genetic factors and response to stimulant drugs. In Advances in Neurology, 35, Gilles de la Tourette syndrome,(eds) A.J. Friedhoff and T.N. Chase, pp. 287-289. Raven Press, New York.

Goldman-Eisler, F. (1968). Psycholinguistics, experiments in spontaneous speech, Academic Press, New York.

Goetz, C.G., Klawans, H.L. (1982). In Advances in Neurology, 35, Gilles de la Tourette syndrome, (eds) A.J. Friedhoff and T.N. Chase, pp. 1-16. Raven Press, New York.

Graybiel, A. (1991). Functional neuroanatomy of basal ganglia. Paper presented at the Second International Symposium on Tourette syndrome. Boston.

Hammond, W.A., (1984). Myriachit, a newly described disease of the nervous system and its analaogues. British Medical Journal, 2, 758-759.

Hirano, M., Koiko, Y. and von Leden, H. (1968). Maximum phonation time and air usage during phonation. Folia Phoniatrica, 20, 185-201.

Huttar, G.L. (1968). Relations between prosodic variables and emotions in normal American English utterances. Journal of Hearing and Speech Research, 11, 481-487.

Itard, J.M.G. (1825): Memoire sur quelqes fonctions involontaires des appareils de la locomotion de la prehension et de la voix. Archives of General Medicine, 8, 384-407.

Kent, R.D., Rosenbeek, J.C. (1982). Prosodic disturbances and neurologic lesions. Brain and Language, 15, 259-291.

Kerbeshian, J. (1991). North Dakota Tourette syndrome peravlence study. Paper presented at the Second International Symposium on Tourette Syndrome. Boston.

Kertesz, A. (1982). Western Aphasia Battery, Grune and Stratton, New York.

Kirk, R.E. (1968). Experimental design: procedures for the behavioral sciences. Second Edition, Brooks/Cole, California.

Krichner, J. (1985). Laryngeal reflex systems. In (Eds) Baer, T, Sasaki, C., and Harris, K. Laryngeal function in phonation and respiration, College-Hill Press, Calafornia.

Kreul, E. (1972). Neuromuscular control examination for parkinsonism: vowel prolongation and diadochokinetic and reading rates. Journal of Speech and Hearing Research, 15, 72-83.

Krumholz, A., Singer, H.S., Niedermeyer, E., Burnite, R., Harris, K. (1983). Electrophysiological studies in Tourette's syndrome Annals of Neurology, 14, 638-641.

Lacey, D. (1986). Diagnosis of Tourette syndrome in childhood. Clinical Pediatrics, 25, 433-435.

Leckman, J.F., & Cohen, D.J. (1988). Descriptive and diagnostic classification of Tic disorders. In Tourette syndrome and tic disorders, clinical understanding and tratment, (eds) D.J. Cohen, Brunn, R.D., Leckman, J.F., pp. 3-21. Wiley and Sons, New York.

Lanier, B. (1985). Alternative approches to Tourette syndrome. Symposium on Tourette syndrome (presentation), Mt Sinai School of Medicine, New York.

Lees, A.J., Robertson, M., Trimble, M.R., & Muray, N.M.F. (1984). A clinical study of Gilles de la Tourette syndrome in the United Kingdom. Journal of Neurology, Neurosurgery, & Psychiatry, 47, 1-8.

Lieberman, P. (1967). Intonation, perception, and language, MIT Press, Massachusetts.

Lieh-mak, F., Chung, S.Y., & Chen, S. (1982). Tourette syndrome in Chinese follow-up of 15 cases. In Advances in Neurology, 35, Gilles de la Tourette syndrome, (eds) A.J. Friedhoff and T.N. Chase, pp. 281-283. Raven Press, New York.

Logemann, J. (1983). Evaluation and treatment of swallowing disorders, College-Hill Press, Calafornia.

Ludlow, C.L. and Bassich, C.J. (1884). Relationship between perceptual ratings and acoustic measures of hypokinetic speech. In (Eds) Mc Neil, M.R., Rosenbek, J.C., Aronson, A.E., The dysartrias: physiology, acoustics, perception, and management, College-Hill Press, Calafornia.

Ludlow, C.L., Bassich, C.J., and Connor, N.P. (1985). An objective system for assessment and analysis of dysarthric speech. Speech and language evaluation in neurology: adult disorders, (Ed) John, K. Darby, Grune and Stratton, Florida.

Ludlow, C.L., Polinsky, R.J., Caine, E.D., Bassich, C.J. and Ebert, M.H. (1982). Language and speech abnormalities in Tourette syndrome. In Advances in Neurology, 35, Gilles de la Tourette syndrome, (eds) A.j. Friedhoff and T.N. Chase, pp. 351-361. Raven Press, New York.

Martindale , C. (1977). Syntactic and semantic correlates of tics in Gilles de la Tourette syndrome, a quantitative case study. Brain and Language, 4, 231-247.

Meige, H., and Feindel, E. (1907): Tics and their treatment, translated and edited by S.A.K. Wilson. William Wood and Co., New York.

Murray, T.J. (1982). Doctor Samuel Johnson's abnormal movements. In Advances in Neurology, 35, Gilles de la Tourette syndrome, (eds) A.j. Friedhoff and T.N. Chase, pp. 25-30. Raven Press, New York.

Mutti, M., Sterling, H., Spalding, N. (1978). Quick Neurological Screening Test, Revised Edition. Academic Therapy Publications, California.

Netsell, R. (1973). Speech physiology. In Normal aspects of speech hearing and language, (eds) T. Hixon, and F. Williams. Prentice-Hall, New Jersy.

Nomura, Y. and Segawa, M. (1982). Tourette syndrome in Oriental children, clinical and pathophysiological considerations. pp. 227-280. In Advances in Neurology, 35, Gilles de la Tourette syndrome, (eds) A.J. Friedhoff and T.N. Chase, Raven Press, New York.

Nuwer, M.R. (1982). Coprolalia as an organic symptom pp. 363-368. In Advances in Neurology, 35, Gilles de la Tourette syndrome, (eds) A.j. Friedhoff and T.N. Chase, Raven Press, New York.

O'Brian, J.A. (1883). Latha, Journal of British Asiat Society, 1; 381.

Portnoy, R.A., and Aronson, A.E. (1982). Diadochpkinetic syllable rate and regularity in normal and in spastic and atxic disarthric speakers. Journal of Speech and Hearing Disorders, 47, 324-328.

Price, A.R., Kidd, K.K., Cohen, D.J., Pauls, D.L., and Leckman, J.F., (1985). A twin study of Tourette syndrome, Archives of General Psychiatry, 42, 815-820.

Riddle, M., Hardin, M.T., Towbin, K.E., Leckman, J.F., and Cohen D.J. (1987). Letter to the editor, Tardive diskinesia following haloperidol treatment in Tourette syndrome. Archives of General Psychiatry, 44, 98-99.

Robertson, M. (1991). Self injurious behavior in Tourette syndrome. Paper presented at the Second International Symposium on Tourette Syndrome. Boston, MA.

Ross, M., Duffy, R., Cooker, H.S., and Sargeant, R.L. (1973). Contribution of the lower audible frequencies to the recognitio f emotions. American Annals of The Deaf, 118, 37-42.

Sacks, O. (1987). The man who mistook his wife for a hat, pp. 92-101, Harper & Row, New York.

Sacks, O. (1982). Acquired Tourettism in adult life. In Advances in Neurology, 35, Gilles de la Tourette syndrome, (eds) A.j. Friedhoff and T.N. Chase, pp. 89-92. Raven Press, New York.

Sacks, O. (1992). A neurologist's notebook: a surgeon's life. New Yorker, March.

Sayadi, R. and Cooke, P.A. (1991). The effect of prosody on vocal tics in Tourette syndrome. Poster presented at the Second International Symposium on Tourette Syndrome. Boston.

Scherer, K.R. (1986). Vocal affect expression: a review and a model for future research. Psychological Bulletin, 99, 143 - 65.

Seignot, M.J.N. (1961). Un cas de maladie de Gilles de la Tourette gueri par le R-1625. Annals of Medical Psychology, 119, 578-579.

Shapiro, A.K., Shapiro, E. (1982). Tourette syndrome history and present status. In Advances in Neurology, 35, Gilles de la Tourette syndrome, (eds) A.j. Friedhoff and T.N. Chase, pp. 17-23, Raven Press, New York.

Shapiro, A.K., & Shapiro, E. (1988). Treatment of tic disorders with haloperidol. In Tourette syndrome and tic disorders: clinical understanding and treatment, (eds) J.Cohen, R.D., Bruun, J.F., Leckman. John Wiley and Sons, New York.

Shapiro, A.k., Shapiro, E. Brunn, R.D., & Sweet, R. (1978). Gilles de la Tourette Syndrome, Raven Press, New York.

Shapiro, A.K., Shapiro, E., Wayne, H.L., & Clark, J. (1972). The psychopathology of Gilles de la Tourette syndrome. American Journal of Psychiatry, 129, 427-423.

Shapiro, A.K., Shapiro, E., Young, J.G., and Feinberg, T.E. (1988). Gilles de la Tourette syndrome, Raven Press, New york.

Silver, A.A. (1988). Intrapsychic processes and adjustment in Tourette syndrome. In eds. Donald J. Cohen, Bruun, R.D., and Leckman, J.F. Tourette"s syndrome and tic disorders:clinical understanding and treatment. Wiely and Sons, New York.

Silverman, F.H., (1985). Research design and evaluation in speech-language pathology, 2nd edition, Prentice-Hall. New Jersey.

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

Stevens, H. (1971). Gilles de la Tourette syndrome and his syndrome by serindipity. American Journal of Psychiatry, 128, 489-492.

Storms, L. (1985). Massed negative practice as a behavioral treatment for Gilles de la Tourette's syndrome. American Journal of Psychotherapy, 39, 277-281.

Sutherland, R.J., Kolb, B., Schoel, W.M., Whishow, I.Q., Davies, D. (1982). Neuropsychological assessment of children and adults with Tourette syndrome, a comparison with learning disabilities and schizophrnia. In Advances in Neurology, 35, Gilles de la Tourette syndrome, (eds) A.j. Friedhoff and T.N. Chase, pp.311-322. Raven Press, New York.

Sweet, R.D., Solomon, G.E., Wayne, H.L., Shapiro, E., and Shapiro, A.K. (1973). Neurological features of Gilles de la Tourette syndrome. Journal of Neurology, Neurosurgery, & Psychiatry, 36, 1-9.

Turpin, G., and Powell, G.E. (1984). Effects of massed practice and cue-controlled relaxation on tic frequency in Gilles de la Tourette syndrome. Behavioral Reserch and Therapy, 22, 165-178.

Williams, C.E., and Stevens, K.N. (1971). Emotions and speech: some acoustical correlates. Journal of acoustical society of America, 52, 1238-1250.

Yorkston, K.M., & Beukelman, D.R. (1981). Assessment of intelligibility of dysarthric speech.Tigard,OR.

Zemlin, W.R. (1988). Speech and hearing science, anatomy and physiology, Prentice Hall, New Jersey.

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