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Influence of Musical Engagement on Symptoms of Tourette's Disorder

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Influence of Musical Engagement on Symptoms
of Tourette's Disorder

by

William Christopher Brown

A dissertation submitted in partial fulfillment
of the requirements for the degree of
Doctor of Philosophy
School of Music
College of the Arts
University of South Florida

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Dedication

This research is dedicated to all those affected by Tourette's and especially to my new friend, Jim Couchenour, whose musicianship, spirituality and encouraging speech about his dealings with this disorder inspired the study.

I am especially grateful to all of those people within my life who made an impact educationally in my development not only as a musician but as a researcher. I'm particularly thankful for my father, Bill Brown, who not only provided continued financial support through many difficult days but always exemplified character in spite of adverse conditions. He has also served as a role-model as a father, spiritual leader, friend and musician.

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I would also like to acknowledge my Savior who has provided the blessings and gifts I have in order to serve Him who is the author and perfecter of our faith.

Table of Contents

List of Tables.....	iii
List of Figures.....	iv
Abstract.....	v
Chapter One: Introduction	1
Historical Context.....	1
Rationales of this Study	3
Research Purpose and Questions	7
Chapter Two: Review of Literature	9
Etiology.....	9
Classification and Symptomology	10
Genetics	12
Neurological Research.....	14
Measurements and Assessments.....	17
Environmental Factors and Interventions	18
Current pharmacological therapies.	19
Surgical Intervention.....	21
Behavioral therapies.....	22
Music as therapy.	23
Developmental Implications	26
Biological Implications?.....	28
Other Considerations	29
Summary.....	31
Chapter Three: Method.....	32
Research Design	32
Participants.....	32
Survey Instrument.....	34
Procedures.....	36
Ethical Considerations	36
Chapter Four: Results	38
Demographic Data	38
Ethnicity.....	38
Age.....	39
Gender.....	42
Diagnosis and Engagement.....	43

Addressing Research Questions.....	45
Research Question 1.	45
Research Question 2.	47
Research Question 3.	52
Research Question 4.	55
Chapter Five: Discussion and Conclusions.....	59
Research Questions Discussed.....	59
Implications for Music Educators.....	64
Future Research Implications	65
Conclusion	66
References.....	68
Appendix A: On-line Consent Document.....	97
Appendix B: Survey Instrument	99
Appendix C: IRB Exemption Letter.....	101

List of Tables

Table 1	Ethnicities Reported by Participants.....	39
Table 2	Primary Musical Instrument Reported.....	44
Table 3	Percentages Totals for Each Scale Response.....	46
Table 4	Symptoms Reported and Total Percent of Cases per Category.....	50
Table 5	Correlations, Means and Standard Deviations for Regression of Criterion.....	51
Table 6	Stepwise Regression Analyses Results of Symptoms Influence on Predictor Variables.....	51
Table 7	Symptoms Influence and Musical Instrument Crosstabulations.....	54
Table 8	Physical Involvement with Musical Activity.....	57

List of Figures

Figure 4.1	Male respondent age frequencies, descriptives and distribution.....	40
Figure 4.2	Female respondent age frequencies, descriptives and distribution.....	41
Figure 4.3	Combined genders with age specific totals for frequencies and descriptors.....	42
Figure 4.4	Total response ratings of symptoms influence while engaged in a musical activity.....	45
Figure 4.5	Minute ranges per week of musical engagement activity.....	48

Abstract

Tourette's is currently considered a neurodevelopmental genetic disorder. Georges Gilles de la Tourette is given primary credit for the diagnoses of the disorder in the late 1800s. Clear answers have been elusive although modern research and improvements to neuroimaging have enabled the causal factors of Tourette's Disorder (TD) to be examined with greater scrutiny. Currently, there is no known cure or pharmaceutical treatment that has been proven 100% effective for all patients and symptoms of Tourette's. Anecdotally, there have been recent media and self-reports of people diagnosed with Tourette's finding relief from their symptoms through involvement in focus-based activities such as video games, athletic endeavors and musical engagement, albeit little empirical evidence exists on these subjects. The author is seeking empirical data showing the influence of musical engagement on the symptoms of Tourette's. This research does not focus on the receptive activity of listening to music, but *engagement*. This performance-based music making or engagement can be described as the body being physically involved in the creation and production of music which comes from such activities as playing a musical instrument or singing. This study seeks to answer the question, "does musical engagement influence the symptoms of Tourette's?" A self-reported survey instrument was generated to question those claiming to be musicians who have been diagnosed with TD to explore what effect engaging in a musical activity has on their symptoms. Participants' responses to ten questions were analyzed. The survey apex involved a Likert-type scale asking to what extent these musicians experienced changes in their symptoms. One hundred eighty-three ($N = 183$) respondents rated their perceptions from one to five where one equaled drastic symptoms

increase by engaging in a musical activity and five equaled drastic symptoms decrease. The mean response from the scale was 4.45, clearly showing that these musicians with TD experienced a great deal of relief when engaged in their activity. This study presents evidence and support for research into neurodevelopmental and musical training correlations and a strong case for childhood music education as a means to facilitate this training.

Chapter One: Introduction

Historical Context

La Maladie des tics (or Tics Disease) is the initial description given by early physicians in the late 17th century now known as Tourette's disorder. This disorder has previously been described (for nearly 175 years) as a syndrome. *The Diagnostic and Statistical Manual of Mental Disorders (DSM-5)* (American Psychiatric Association, 2013) has clarified the hierarchy given to symptoms, syndromes, disorders and diseases and therefore recategorized what was formerly known as Tourette's syndrome to Tourette's disorder. Like a syndrome, a *disorder* refers to a cluster of symptoms however, the concept includes the idea that the set of symptoms is not accounted for by a more pervasive condition with no implication of etiology.

Eighteen twenty-five marked the first medical diagnosis of two known cases by noted French physician, Jean Marc Gaspard Itard. The most famous case being that of the Marquise de Dampiere, a Parisian aristocrat who was reported exhibiting sudden involuntary movements along with sudden vocalizations and outbursts of obscenities. Itard was a student of now famed Philippe Pinel (known as the Father of Psychiatry) whose famous observation methods were noted in his attempts to civilize "the wild child of Aveyron." These written details set him apart as an empiricist based upon sensory observations and facts and not medication, surgery or both. Itard's methods would be the

foundation for future psychiatric and neurological studies that would influence others to follow (Newman, 2006).

In the mid-1800s another physician emerged as a leader in the field of neurology in his time, Jean-Martin Charcot. At age twenty-three Charcot had qualified for a hospital internship at Salpêtrière in Paris. After his dissertation defense he was appointed Chief Residence (*Chef de Clinique*) in 1853 and later promoted to *Professeur Agrégé* in 1860. It was during this time that Charcot would often assign duties to his assistants or residents whose responsibilities included nosological (disease study and classification) tagging. The practice of the day was for these assistants and interns to take lecture notes and publish the work (Teive, Chien, Munhoz, & Barbosa, 2008). This could explain why a famous Tourette's syndrome case involving the Marquise de Dampiere first reported by Charcot, would later surface in a work by his student, Georges Gilles de la Tourette who, because of his more detailed description of the symptoms of the disorder, bears his name (Díaz-Anzaldúa & Rouleau, 2008).

Before the medical accounts were recorded in the 1800s, there were however, earlier written records that described motor and vocal tics. Sprenger and Kraemer detail a priest with motor and vocal tics in their 1489 pamphlet, *Malleus Maleficarum* (Institoris, Sprenger, & Summers, 2000). Several hundred years later, in the 17th century French court, the Prince de Condé was said to stuff objects in his mouth to suppress involuntary noises (Goetz, Chmura, & Lanska, 2001). The historical accounts of convulsive tics by George Gilles de la Tourette (de la Tourette, 1899) and more recent records pointing to his discoveries and personal notes (Lajonchere, Nortz, & Finger, 1996) help detail the

metamorphosis in theories surrounding Tourette's disorder (which henceforth will be noted as TD).

At the time of initial diagnosis and discovery, the accounts of TD were thought to be very rare and considered a neurological disorder. With the development and blossoming of Freudian sciences in the early twentieth century there began a diagnostic shift to psychiatry (Goetz et al., 2001). Recent discoveries and modern advancements in neuroimaging have woven the formerly dichotomous branches into what is now referred to as neuropsychiatry. More recently, the parental science of neurology has also studied the influence of music on the brain with the help of famed neurologist Oliver Sacks.

Author of over a dozen books, Sacks has been a primary proponent for the use of music as therapy and the intrinsic power it has on the brain. His interest in music and the role it may play in neurological disorders has been documented in multiple media outlets, videos and personal interviews; moreover, Sacks believes the brain and music are inseparable. "We humans are a musical species no less than a linguistic one. Music seems hard-wired into our very being. It moves us, stirs us to action, sets us in motion, sticks in our memories and minds" (Sacks, 2010). Although these thoughts of Sacks seem to be culturally accepted, a great deal of research needs to be accomplished to produce empirical strength.

Rationales of this Study

Anecdotal references abound for music being an aid as therapy for various types of neurological disorders and diseases especially in the aged, yet little has been done in the area of Tourette's and for children. Music educators see the benefits of music on their students in the classroom but may not truly understand why their craft is so important or

valuable to a young developing mind. The author's initial research into these topics revealed a need for methods by which to bridge these gaps with empirical research that details why music and music education are crucial for a child's neurological development. Although there is a growing list of musicians who have found music helpful that can be accessed on the Tourette's Association of America website, no empirical data is presented to support what they perceive and why (K. McNaught & Scullin, 2016). Another hope for an understanding and cure to TD lies in the collaboration of *all* research groups in a synergistic approach using modern research and reporting techniques along with the World Wide Web. Thirty years ago such a collaboration would have been much more difficult to attain however, today's *DSM-5* contains many more internationally cooperative entries than its first edition of 1952 (Wilson, n.d.). With this global cooperation of the sciences and the inclusion of empirical studies of music as therapy, perhaps the symptoms of Tourette's can not only be understood but curtailed.

One might speculate why a twenty-year music educator would delve into the waters of Tourette's and music therapy? First, as a music educator the author is convinced that music should be taught to everyone and experienced by all and is a key component to the academic and neurological development of a child. Although students with special needs diagnosed with TD in the United States represent between 0.4% and 5% of the population (Bitsko et al., 2014), these students should have the opportunity to make music a fulfilling part of their lives; this includes music as a holistic therapeutic alternative that could potentially lessen their symptoms severity.

Second, public school music educators have or will be exposed to special needs students including those with TD at some point in their career. The knowledgeable professional educator should have an understanding of what it is and how to properly teach those students diagnosed with TD. In a recent conference presentation Adams and colleagues presented data showing that teachers receive little education about TD and the students with TD view their teachers as lacking knowledge about their disorder (Adams et al., 2015).

Third, research has shown that neurological development and specifically that of the trained musician occurs both structurally as well as functionally (Schlaug, 2015). The author seeks to explore these similarities and propose a partnership among the fields of music and neurological sciences to consider the possibility of symptoms abatement or cessation.

The author's desire to understand Tourette's disorder and the influence of music on its symptoms followed the hearing of a speech given by Jim Couchenour, who detailed the struggles in his life as a TD sufferer. Now in his late fifties, Jim spent the first half of his life not knowing why he was different, shunned, or alienated during his adolescence. The only solace he described was playing the piano at which times Jim said, "His symptoms stopped." For Jim, and others like him, an empirical attempt should be made at answering the question of why engaging in a musical activity has some apparent influence on TD symptoms and is it possible for music to make life more satisfying for TD sufferers? For Jim's children or grandchildren who may also have the gene associated with the disorder, is it possible to provide developmental therapies of music that would deter or stop symptoms from ever occurring?

Although there are ample studies into the pathogenesis, neurobiology, genetics, neurochemistry and neuropsychiatry of Tourette's there is very little empirical data for the use of music as a therapeutic device or behavioral modifier. There are two subsets of music research that have only been anecdotal to date. The first is the effect of rhythmic pulses within music that have been attributed to heighten the sensitivity of, or possibly instigate, tics. Sacks mentioned a gentleman he called John S. who stated his tics were brought on by "certain kinds of music heavy with rhythm" (Sacks, 2010, p. 247). This area of what is termed receptive music therapy (that of listening to music and the effects thereof) is not the focal point of this study.

The second musical effect, from the same chapter in his book, Sacks described another man Sydney, who suffered from multiple symptoms of TD and yet when he "took up his guitar and sang an old ballad there was no ticking at all" (Sacks, 2010, p. 249). Other studies have reported that focused attention can alleviate tics but do not specifically mention music as a behavioral modifier (Cohen, Leckman, & Bloch, 2013). Leckman (2002) too posed the development of music abilities as a possible protective factor against the symptoms of TD along with academic gifts or athletic skills, but only presented it within a boxed table in the article with no empirical data as support. His original comment continued to receive citations but with no further provisions for evidence (Leckman & Martino, 2013). A more recent study has pointed to the tic-reducing effects of music in TD patients however, the research covers both listening to and playing music. The results reported that practice-based musical experiences influence cortical functions in TD subjects and that the "practice of well-trained music leads to frequency reduction." The definition of "well-trained music" was unclear although those

who reported playing instruments did show symptom reduction the small sample size of eight casual musicians (non-professionals) elicited the need for further studies (Bodeck, Lappe, & Evers, 2015, p. 45). These items prompted the author to develop his statement of purpose and research questions for this research. Those diagnosed with TD might reveal data to provide insight into the influences of musical engagement on their symptoms.

Research Purpose and Questions

The primary question for this research study was, “What is the influence of musical engagement on the symptoms of Tourette’s?” By extension, *engagement* is defined as *active* physical involvement versus the *receptive* activity of listening to music. This question must reveal significant evidence between physical involvement in a musical activity and symptom responsivity in order to justify continued time and resources of future studies. The following four research questions emerged as essential to this study:

1. To what extent does musical engagement relate to symptoms change for those diagnosed with TD?
2. What are the correlations between symptom changes and time per week engaged in musical activity, age, and total number of symptoms reported?
3. To what extent do types of instruments and gender influence symptom activities reduction for musicians diagnosed with TD?
4. Are there physical factors such as breathing and manual operation of the instruments that could influence symptom severity while engaged in a musical activity?

In the following chapters that constitute this study the author has detailed the body of extant literature relevant to the topics discussed and formulated the methodology and design for an on-line survey instrument. The results of the Musical Engagement Influence on Tourette Symptoms survey have been reported in chapter four with discussion topics and items for future research outlined in the final chapter.

Chapter Two: Review of Literature

In this chapter the author seeks to detail the underlying research and treatment methodologies which currently explore the cause and effect (or etiology) of Tourette's disorder. Providing clarity from the observations of the past and directing efforts toward possible future solutions can be achieved by understanding the various areas that have become the focal points of more modern research and how these have evolved. The most recent classification and symptomology associated with TD have been summarized in the following paragraphs along with the current genetic propensities of the disorder. The neurological research that embodies the substance of this chapter illumines the various factors possibly associated with the causes and effects of TD. This includes the means by which symptoms and diagnoses are determined clinically through measurement and assessment tools and their effectiveness as well as environmental factors and current interventions. This literature review also mandates a broadening of ideas supporting developmental implications previously disconnected and seeks to explore possible biological influences and other considerations.

Etiology

Earliest accounts of "*tics disease*" (before it became known as Tourette's Syndrome and now Tourette's Disorder) can be traced back to ancient Rome where the Greek physician Aretaeus of Cappadocia provides documented accounts of tic-like symptoms now associated with TD such as twitching, barking and cursing (Díaz-

Anzaldúa & Rouleau, 2008; Sacks, 1995). Fifteenth and Seventeenth century examples were cited earlier but in the century to follow a now famed case involving the first writer of the English Language Dictionary emerged. Samuel Johnson, according to accounts from James Boswell in his book *The Life of Samuel Johnson* written in 1873, “held his head to one side towards his right shoulder, and shook it in a tremulous manner, moving his body backwards and forwards” (Boswell & Ross, 1945, p. 402). Johnson was also reported making unusual vocal sounds that are now attributed as TD symptoms.

Of the most famous and recent of debates is the case for W. A. Mozart. The unedited letters of Mozart were released in its latest form in 1985. The earlier versions first publicly seen in 1928 (1938 and 1950 respectively) were edited for content. The most recent letters detail obscenities associated with scatological disorders and were initially reported to be possible evidence of Tourette’s (Aterman, 1994; McBride et al., 1992; Simkin, 1992). This was especially tantalizing in wake of the movie release of *Amadeus*. Further investigation by Ashoori and Jankovic have since shed new light on the subject (Ashoori & Jankovic, 2008) prompting the release of later material that would reject those initial claims (Karhausen, 1998; Sacks, 1992). There is still a great deal of mystery after nearly 175 years of clinical study that seeks to reveal what *is* TD, what causes it, who is affected by it, and what are the best methods for treatment?

Classification and Symptomology

The most widely accepted classification and descriptors of TD can be seen in the *DSM-5* and the work of Díaz-Anzaldúa and Rouleau. These authors note that Georges Gilles de la Tourette’s initial diagnoses in 1885 in six of his initial nine patients were correctly identified by today’s standards. The other three are now classified as mild tic

disorders not encompassed in the TD classification due to the lack of phonic tics (Díaz-Anzaldúa & Rouleau, 2008). The current Tourette's diagnosis in the *DSM-5* concurs that TD is the presence of multiple motor and one or more vocal tics occurring longer than a year with symptoms onset before 18 years of age (American Psychiatric Association, 2013). The most common symptoms associated with this diagnosis are:

Tics of the face (eyes, nose or mouth)

Grunting

Tics of the arms/legs

Throat clearing

Trunk tics

Shouting

Kicking/Stamping

Barking

Touching

Echolalia (echo speech)

Coprolalia (involuntary utterance of obscene words)

The age of symptoms onset has been unclear possibly due to the comorbidity factors associated with the diagnoses of multiple disorders in children and the adolescent. Earlier research proposed onset as early as age three (Leckman, 2002; Leckman, 2003) although, more recent studies have suggested both 5-7 and 6-8 years as the mean ages for most cases (Cavanna & Rickards, 2013; Cohen et al., 2013; Felling & Singer, 2011; Tallur & Minns, 2010). The exact causes of the disorder have been the topic of much scholarly literature. Perhaps these causes can be traced even further within the building

blocks of our own DNA. This would provide the justification for medical research and the development of targeted treatment options.

Genetics

With advancements in genetics, there were modern approaches to aide in the determination of the gene(s) associated with the disorder. Were there genetic inferences to explain TD? Gilles himself reported some of the familial occurrences within his patients. In fact, one of his patients was said to have had a grandmother who was insane; therefore, the assumption became that it indeed was hereditary (Cavanna & Rickards, 2013). There were however, a number of empirically based genetic studies for the discovery of the gene associated with Tourette's (e.g., (Davis et al., 2013; Dietrich et al., 2015; Paschou, 2013)). This has been made possible due to the decreasing costs of gene sequencing. For example, to sequence the known genes in a human genome (estimates in the 20,000-25,000 range) in 1998 would have cost over \$3,000,000. Current cost estimates are at \$550 (State, 2015).

Within the genetics body of literature there was recent research that identified rare copy number variations (CNV) genes associated with TD. CNVs are the structural mutations contributing to certain inherited diseases such as those attributed to TD and similar disorders (Carter, N., & Hurles, M., 2015). Numerous studies since have confirmed that genetics play a significant role in the etiology of TD. Although there was no conclusive evidence of which genes are directly affected, nor which could be manipulated for a possible cure (Felling & Singer, 2011; Friel, 1973; Leckman, 2002; Tallur & Minns, 2010), recent reports have emerged claiming specific gene "candidates." Huertas-Fernandez and colleagues (2015) claimed that the polymorphism (which

involves one of two or more variants of a particular DNA sequence) of rs3096140 glial cell line-derived neurotrophic factor gene (GDNF) was significant and that a defect in the production of GDNF could compromise the survival of parvalbumin interneurons, therefore varying the excitatory/inhibitory balance in the corticostriatal circuitry. This gene has been associated with other disorders such as anxiety and mood by Kotyuk and collaborators which might align with common Tourette comorbid disorders but before this study had not been previously linked to TD (Kotyuk et al., 2013a; Kotyuk et al., 2013b). Thomas Fernandez from the Yale Child Study Center recently reported a narrowing of 36 genes as strong candidates. These genes have been linked through other studies to the striatum, thalamus and signaling pathways previously implicated in TD pathology (Fernandez, 2015). In an update released in the spring of 2015 by the Tourette Syndrome Association International Consortium for Genomics confirms strong evidence for heritability in TD but lists the possibility of hundreds of candidates (Davis et al., 2013). It should be noted however, that TD genetics has not reached the level of quantitative study and analysis as other disorders such as schizophrenia or autism and most of the related literature lists only “candidates,” not specific “risks.” Although the aforementioned areas of genetics may provide promise for aiding in the causes and study of TD, the complex nature of the symptoms that coincide with other disorders continue to cloud the pictures of analysis, diagnosis and therefore treatment. This may be the rationale for the majority of recent literature that has focused on the neurological underpinnings as a means for a more tangible description of the disorder.

Neurological Research

Most studies have centered upon the behavioral, motor and cognitive symptoms of TD and the overlap in the related fields of psychiatry, neurology and psychology. The psychopathology and proposed GTS spectrum by Cavanna and Rickards (2013) further to blur the lines of demarcation associated with the disorder. Positively, there were data which show an increase in the amount of research surrounding TD and tic related disorders. PubMed reports the numbers have increased almost ten times from 1980-2014. There were less than 500 studies in the early 1980s but approximately 5,000 by mid-2010s (Black et al., 2014).

A great deal of research within the first decade and a half in the 21st century was focused on the role of the basal ganglia (or basal nuclei). Situated in the base of the forebrain the basal ganglia was responsible for several functions including routine behaviors, procedural learning, voluntary motor movements and emotion (Weyhenmeyer & Gallman, 2007). The main components of the basal ganglia, as defined functionally, were the dorsal striatum (caudate nucleus and putamen), ventral striatum (nucleus accumbens and olfactory tubercle), globus pallidus (internal and external), ventral pallidum, substantia nigra, and subthalamic nucleus (Fix & Wilkins, 2008).

The bulk of TD neurological research pointed to the alterations to the basal ganglia functioning including reduced activation in the control sections of the cortico-striato-thalamo-cortical circuits (Bronfeld, Bebelovsky, & Bar-Gad, 2011; Ganos, Roessner, & Münchau, 2013; Mink, 2006; Peterson et al., 2003; Wang et al., 2014). These complex circuits included the somatosensory, motor and premotor cortices and

related portions of the basal ganglia and thalamus including various prefrontal regions that may have been involved in the suppression of tics (Plessen et al., 2004).

Other studies have postulated an inhibitory dysfunction inside the anterior striatum, which in a precursory manner affected different cortico-basal ganglia circuits that led to motor tics and have been linked to behavioral disorders (Mink, 2006; Tremblay, Worbe, Thobois, Sgambato-Faure, & Féger, 2015; Worbe et al., 2010). These dysfunctions could have been linked to the neuron transmitters of either the excitatory or inhibitory categories as previously mentioned in the section on genetics.

It has been reported that neurotransmitters were the chemicals responsible for sending information throughout your body. The chemicals from the inhibitory standpoint included serotonin (which helped control cravings, pain control and sleep cycles), GABA (gamma-aminobutyric acid) which attempted to regulate over-firing and dopamine. Dopamine was unique in that it fell into both categories; for example, it helped with both depression and focus. From the excitatory side; dopamine was used in conjunction with norepinephrine, which at high levels could produce anxiety whereas epinephrine was linked to stress, heart rate and blood pressure (Helmly, Henry, Roberts, Frias, & MacDonald, 2015).

Another key component to the neurotransmitter puzzle was the understanding of parvalbumin inhibitory neurons. These were found throughout the cerebral cortex and have been linked with impressive abilities to synchronize the electrical activities of other brain cells including the developmental interludes when the brain's plasticity undergoes rewiring (Bilimoria, 2013). Abnormalities in these pivotal cells were believed to be the groundworks of many neurological disorders including those of TD. Two post-mortem

studies supported this hypothesis in that they showed not only a loss of parvalbumin inhibitory interneurons but a decreased volume of the caudate and putamen (Kalanithi et al., 2005; Kataoka et al., 2010). These smaller volumes of the caudate and putamen in children have been linked as a predictor in symptom severity in adulthood (Bloch, Leckman, Zhu, & Peterson, 2005).

Another area of focus has centered upon the prefrontal cortex. Since this area was known for executive functioning, decision making, and moderating social behavior, it too has become an area of study especially within the context of pre-monitory urges (sensations similar to scratching an itch) and phenomenology studies such as copropraxia, coprolalia, echolalia, and echopraxia (inappropriate behaviors, gestures and speech or the repetition of words or gestures). Several studies have shown structural connectivity deficits and abnormalities in these regions (Makki, Govindan, Wilson, Behen, & Chugani, 2009; Muller-Vahl et al., 2009; Singer & Minzer, 2003). In fact, these studies have been associated with what has been termed functional or developmental immaturity resulting in a “developmental hypothesis” (Cheng et al., 2014; Church et al., 2009; Makki et al., 2009; Worbe et al., 2010; Worbe et al., 2012). This hypothesis suggested that the thinning or reduced volumes of these areas might shed light on the pathophysiology of TD and similar disorders. This thinning or reduced volumes was also reported by Ganos and colleagues in the prefrontal cortex but noted there were no correlations between tic severity and tic inhibitions in their patients (Ganos et al., 2014). Other volumetric changes and alterations have been reported in the interhemispheric regions of the brain; those near the corpus callosum.

The various studies into these interconnected regions of the brain remain as evidence of uncertainty for causation of the disorder. Cheng and cohorts have agreed that “Gilles de la Tourette syndrome can be viewed as a model of aberrant activity in neuronal networks involved in planning, executing and controlling motor actions” (Cheng et al., 2014, p. 174) and although there have been great strides in the narrowing of causes in the pathophysiology of TD, the clear neural mechanisms causing motor tics are still elusive (Bronfeld & Bar-Gad, 2013; Cheng et al., 2014).

Measurements and Assessments

Other topics in research and categorization of tic disorders, symptomatology and severity immersed through the development of various rating scales and measures. The most widely used assessment to measure tic severity was the Yale Global Tic Severity Scale (YGTSS) (Leckman et al., 1989). This was shown to be the most reliable and valid of instruments in the assessment of tic disorders (Storch et al., 2005; Walkup, Rosenberg, Brown, & Singer, 1992). Additional measures have also been developed to determine how TD affects the day to day functions of the patient or if it is debilitating in nature (also termed functional impairment). Functional impairment was defined as the inability to perform routine and age-appropriate tasks in the domains of school, home and social activities (Rizzo, Gulisano, Pellico, Cali, & Curatolo, 2014; Storch et al., 2007). One such scale was developed by Storch and colleagues, The Child Tourette’s Syndrome Impairment Scale Parent Report about Child (CTIM-P). This scale was a parent report about their observations of their child’s ability to function in their routine activities; however the report did not address issues such as broken homes, lower socio-economic matters, home-schooled children (which may lessen the anxiety and tic exacerbation

problem) and makes assumptive statements that may be difficult to mark accurately for some parents/guardians. In a similar measure developed by Cavanna et al, quality of life has also been described more specifically as health-related quality of life but admits that the reports done by children and by parents show disparity either in their communication, lack thereof, or parent knowledge of what actually occurs to their child during the normal course of a school day away from their supervision (Cavanna et al., 2013; Crossley & Cavanna, 2013). This lack of communication and knowledge between parents and children is not a new issue, however the severity of it may be revealed in a more recent study by Storch and colleagues (2015); the day to day struggles for sufferers of TD and tic related disorders has also been linked to higher suicidal thoughts, in fact, more than three times than that of the control group. Clearly there are more unseen problems than clinical assessments can measure. The systematic and quantitative measuring of a TD sufferer's perception of their routine battles with the symptoms of the disorder may be elusive, but nevertheless important and attempts should be made to produce reliable, valid measurement instruments. Further understanding of TD might surface through the studies of environmental triggers. If these factors can be identified they might provide a means and justification for proper intervention strategies.

Environmental Factors and Interventions

Although the pathophysiology remains ambiguous for TD, some studies have pointed to environmental non-genetic triggers as contributing to tics and tic severity. These factors included prenatal events such as drug or alcohol use including nicotine from smoking, medication exposure in utero both prescribed and other, hyperemesis gravidarum (severe morning sickness), bleeding, gestational diabetes, maternal

malnutrition and psychosocial stress. One of the earliest of such studies by Leckman and colleagues (1990) looked into prenatal causes and correlations of tic severity among mothers and their children with TD. This study reported severe life-stresses during pregnancy, severe nausea/vomiting during the first trimester as well as gender of the child as being significantly associated with their current tic symptoms severity. Other studies followed which included studying perinatal (the time right before and after birth) instances and their correlates such as jaundice, cesarean section, hypoxia (lack of oxygen at birth), forceps delivery and other forms of traumatic deliveries including low birth weights but conclusions have been mixed and inconsistent (Hoekstra, Dietrich, Edwards, Elamin, & Martino, 2013; Mathews et al., 2006; Motlagh et al., 2010; Silva, Munoz, Barickman, & Friedhoff, 1995). More recent systematic reviews of this body of literature have confirmed problems with reporting methodologies and changing of earlier reports (Chao, Hu, & Pringsheim, 2014; Mathews et al., 2014). Newer reports have questioned more thoroughly both cannabis and alcohol usage as contributors of non-genetic causes of TD (Mathews et al., 2014) but these too need further research rigor. Since the existence of TD and tic related disorders cannot be dismissed or ignored, comprehensive therapies have been, and continue to be, developed to aid those suffering with TD with the routine handling of their daily struggles with their symptoms. These could be categorized arguably as either non-invasive or invasive procedures; but what, if any are most effective? There are several pharmacological options which have been studied and reported although, as with most medications, complete success is elusive.

Current pharmacological therapies. With the pathogenesis unclear, pharmaceuticals are also elusive. The comorbidity issues also cloud which symptoms

should and can be targeted successfully. Typically, pharmacological therapies are treated as “less is best.”

Current prescribed medications for tic disorders from the Tourette Association of America website include:

Clonidine (Catapres)

Neuroleptics (including)

1. Haloperidol (Haldol)
2. Pimozide (Orap)
3. Fluphenazine (Prolixin)
4. Others

Other Drugs

Botulinum Toxin (injections)

However, of these listed only two are currently FDA approved - Haloperidol and Pimozide (Kurlan, 2015; McNaught & Mink, 2011). It is also very important to note that with each and every drug intended to lessen symptoms severity for the TD sufferer there always exists some adverse side effects; of which, some are as debilitating as the symptoms themselves. In fact, these adverse effects are typically the cause for cessation of the prescribed medication (Cuenca et al., 2015) and there are no current pharmaceutical treatments that have proven highly effective (Cavanna & Rickards, 2013; McNaught & Mink, 2011). Each case must be considered on a risk-benefits model for assessment and non-invasive treatment.

One of the least invasive procedures is the Botulinum Toxin injections (also known as BOTOX). Although this procedure has proven effective in other situations and for other medical conditions it has not been proven highly effective for TD. BOTOX

works by blocking the release of neurochemicals that leads to the inactivity of the nerves supplying movement to the injected muscles or glands; although this sounds as if it could be particularly helpful for tics, recent research has been inconclusive in small clinical samples (Persaud et al., 2013). These and similar findings with pharmacological studies have prompted the use and argument for more invasive procedures including deep brain stimulation.

Surgical Intervention. Deep brain stimulation (or DBS) involves the surgical placement of a thin wire (with four electrical contacts at its tip) into a very specific and carefully selected brain region. The main parts of the brain where the DBS lead can be placed currently target the globus pallidus internus, the thalamus, globus pallidus externus, and the internal capsule/nucleus accumbens. The DBS lead is connected to a pacemaker-like device that is implanted in the chest region below the collarbone. This device, called the neurostimulator or implantable pulse generator (IPG), contains the battery and computer source that generates the electrical pulses that were delivered via the lead to the brain. The clinician can select which one or more of the four electrodes on each brain lead that are to be activated to provide electrical stimulation. This process allows electrical stimulation to be delivered to a very precise part of the brain. There are reports of positive effects on tics although the sometimes associated comorbid behavior disorder results vary (Marjama-Lyons & Okun, 2014; Visser-Vandewalle et al., 2014). The most common question associated with DBS is the best location for the sensing targets. Recent research centered within the basal ganglia has shown promise in the ventral anterior and ventrolateral motor part of the thalamus as well as the pallidal areas. These results include long-term positive effects not only in movement but emotional

stabilization (Dong, Zhang, Li, & Li, 2014; Huys et al., 2014; Zhang et al., 2014). These reports however, consist of small samples with severe symptoms that have continued into adulthood and have been targeted because of their functional impairment. This procedure has yet to be attempted on young adults or children. Some of the most distinguished names in the field of TD in conjunction with the Tourette's Association of America organization released a clinical review for DBS which details inclusion criteria. Although this document was released some ten years ago, to the knowledge of the public, its adherence continues to be followed (Mink et al., 2006). Some of the issues surrounding DBS are the obvious accessibility of clinics/physicians qualified to perform the surgical implants and costs associated with the procedure. The FDA has approved DBS for Parkinson's disease and a few other disorders but Tourette's is not currently approved. This could make the procedure unattainable for most. In 2014, the estimated costs without insurance assistance was reported \$35,000-\$100,000. Risks associated with the procedure are reported as "low" with the likelihood of death or stroke as a result of the procedure below 3% (Marjama-Lyons & Okun, 2014). Although the benefits may outweigh the risks for some TD sufferers this procedure was designed for adults only who have extreme movement disorders and who have exhausted all other behavioral and medicinal therapies. This greatly reduces the number for which it can possibly benefit. What are the viable options for the mainstream TD patient?

Behavioral therapies. Behavioral therapies have centered on activities that require attention and focus and the redirection of pre-motory urges. These have been attributed to success in alleviating tics at least temporarily (Conelea & Woods, 2008; Misirlisoy et al., 2015). Congruent therapies were called habit reversal and were reported

as effective. These therapies seek to reduce the influences of urges and tics by inspiring patients to be more cognizant of their tics as they occur (Bate, Malouff, Thorsteinsson, & Bhullar, 2011; Deckersbach, Rauch, Buhlmann, & Wilhelm, 2006; Wilhelm et al., 2003; Wilhelm et al., 2012).

Douglas Woods has been at the forefront for behavior therapy for Tourette's patients and reports that his Comprehensive Behavioral Intervention for Tics (CBIT) were beneficial. Woods claims this non-drug treatment makes his patients more aware of their urge to tic, trains them to do a competing behavior when they feel that urge, and seeks to avoid those circumstances or environmental triggers in an effort to reduce tics. He has stated that this method is not a cure for TD but a behavioral management system (Woods, Piacentini, & Walkup, 2015). Although this is a fairly new program, and most of the data belongs to Woods and his colleagues, it has received supporting success reports from other sources (Rowe, Yuen, & Dure, 2013; Wile & Pringsheim, 2013). This form of cognitive behavioral therapy has recently been shown to be more beneficial than that of psychoanalytic psychotherapy (Peterson et al., 2016). Although the research is promising for CBIT, the limitations of a new behavioral treatment are exacerbated by a lack of intervention clinics as well as clinicians trained in the method. What other known therapies can provide help within a reasonable time frame at a reasonable cost?

Music as therapy. The notion of music as therapy is not a new one. "Music has charms to sooth a savage breast" is the famed quote by William Congreve (Congreve, 1697, p. 19). Plato also stated, "Rhythm and harmony find their way into the inward places of the soul" (Plato, 1955, p.399). These quotes may not be *just* literary or philosophical statements, however empirical research has more recently been sought.

Perhaps, Sparr said it best in his title “Music Therapy: The Poor Stepchild Whose Time has Come” (Sparr & Tomaino, 2015). Music therapy has been associated with the term entrainment. Entrainment was described as different amounts of energy transferred between two moving bodies. This asynchronous movement period causes negative feedback which forces an adjustment period. After time these bodies move in resonant frequency or synchrony (Thaut, 2015). The use of entrainment for therapeutic purposes was first researched by Thaut and colleagues. They showed through several studies how periodicity of auditory rhythmic patterns in conjunction with patients with movement disorders could entrain movement patterns in their patients with the use of Neurologic Music Therapy (NMT) (Hurt-Thaut & Johnson, 2015; Thaut, Kenyon, Schauer, & McIntosh, 1999). Most of the NMT movement research has occurred in the areas of Parkinson’s disease and gait influences (Ashoori, Eagleman, & Jankovic, 2015; Bella, Benoit, Farrugia, Schwartz, & Kotz, 2015; de Bruin et al., 2015; De Dreu, Van Der Wilk, Poppe, Kwakkel, & Van Wegen, 2012; Elefant, Baker, Lotan, Lagesen, & Skeie, 2012; Nombela, Hughes, Owen, & Grahn, 2013; Okuda et al., 2012). More recently studies have also occurred with patients with Epilepsy (Raglio, Farina, & Giovagnoli, 2014), stroke rehabilitation (Fogg-Rogers et al., 2015; Raglio, 2015; Rosin, Ericsson, & Larsson, 2015; Street, Magee, Odell-Miller, Bateman, & Fachner, 2015) and traumatic brain injury (Gardiner & Horwitz, 2015; Gentle, Barker, & Bower, 2015; Hegde, 2014; Tamplin, 2015).

Li and colleagues (2015) have recently released a comprehensive review of the possible effect of music therapy on older adults. Their research methods and meta-analysis consisted of reporting the results for two main types of clinically practiced music

therapies; that of receptive music therapy which involves listening to music provided by the therapist and that of active music therapy which engages the patient in physical participation of music making either with instruments or by singing and perhaps movement improvisations. The data and meta-analysis of these studies involving the influence of music as therapy on the cognitive functions of the elderly suggested that positive therapeutic outcomes were best expressed with active music therapy usually involving singing in conjunction with another activity (Li, Wang, Chou, & Chen, 2015). Clearly there are cognitive functioning connections with empirical research support for the impact of music on the brain; not only for the old but for the young alike. These and similar studies should encourage researchers to seek clearer answers with empirical rigor. Is there a neurological link between active music engagement (whether the participant/patient be a casual musician or professional) and cognitive functioning and development?

Altenmüller (2008) suggested for musicians that engage in active music making their ability to make music results from the combination of multimodal sensory and motor information with precise monitoring of the motor performance via auditory feedback. What has been termed performance-based music making relies primarily on this complex auditory-motor integration and represents the rationale in this document for studying the engagement activities of music making. Although the idea of training-induced structural changes are not specific to only musicians it is clear there are developmental inferences (Draganski & May, 2008).

Developmental Implications

Recent studies involving psychologists and music educators have begun to unfold the intricacies of music, the brain, and child development. Trehub suggests that children during infancy show a predisposition to music, which in turn implies a possible biological basis for music (Trehub, 2001). This has led others to explore evidence for what is now termed a sensitive period for musical training. Evidence has suggested that musicians that began training earlier in age show better task performance with noticeable changes in both motor regions of the brain as well as auditory areas (Draganski et al., 2004; Kraus & Chandrasekaran, 2010; Penhune, 2011). Musical training has also been shown to aid in linguistic development and abilities (Moreno et al., 2009; Patel, 2003; Patel & Iversen, 2007; Patel, 2010; Patel & Daniele, 2003) and for neuro-rehabilitation (François, Grau-Sánchez, Duarte, & Rodriguez-Fornells, 2015). Does music make you smarter? This initial media exaggerated report was repudiated by noted psychologist, Glenn Schellenberg and others. The Mozart Effect (Rauscher & Shaw, 1998) was shown to be false, however Schellenberg offered that music lessons (training) beyond just listening to music could offer small but measurable intellectual benefits for children (Schlaug, Norton, Overy, & Winner, 2005).

Günther and colleagues (1996) provided a ground-breaking study implementing electroencephalogram (EEG) analysis that lead the way for further studies facilitating imagery. Their study focused on the observation of the areas of the brain affected when tics occurred as well as introducing music perception tasks to correlate the different areas affected. Gottfried Schlaug was one of the earliest neuroscientists to pose the differences in musicians' brains versus those who do not engage in music performance two decades

ago suggesting that the corpus callosum is larger in musicians (Schlaug, Jäncke, Huang, Staiger, & Steinmetz, 1995). Others have followed with their reports of brain plasticity and structural changes due to musical training (Hyde et al., 2009; Tervaniemi, 2009) including white and grey matter (Groussard et al., 2014; Imfeld, Oechslin, Meyer, Loenneker, & Jancke, 2009) and that of the corpus callosum (Steele, Bailey, Zatorre, & Penhune, 2013). This is a significant statement for both fields of neurology and music education. The corpus callosum is identified as being the interhemispheric connector between the right and left brain. One of the earliest reports of the difference in the size of the corpus callosum in children was published by Plessen et al. (2004) and confirmed in similar studies including the area near the anterior corpus callosum (Bäumer et al., 2010; Muller-Vahl et al., 2009; Plessen et al., 2004; Plessen et al., 2006a).

Within the last decade significant improvements in neuroimaging have not only allowed this to be a topic of discussion but a visual representation of measured data. The images from the Gaser and Schlaug study clearly showed the differences in gray matter sizes between amateur, professional and non-musicians (Gaser & Schlaug, 2003). The implications of this research to this proposal topic are relevant due to the apparent nature and change that occurs in the brain due to musical influences. In fact, recent research by Tallur and Minns and others has suggested that the corpus callosum is *smaller* in children with Tourette's (Plessen et al., 2004; Plessen et al., 2006b; Steele et al., 2013; Tallur & Minns, 2010). Speculation is therefore amplified by the idea that a child diagnosed with Tourette's disorder might alter their neurological development (and specifically the plasticity in the corpus callosum) through a musical engagement activity. If this could be proven empirically, it would seem the most logical and natural path in attempting to

affect the symptoms of Tourette's. Could the symptoms of Tourette's be altered or alleviated prior to the usual age of onset? This would seem to be the logical course of action in that there are only two current options for treatment. Symptoms treatment is currently managed with either behavioral modification techniques or medication but again, the complex nature of comorbidity with congruent disorders compounds this issue. Are the physical symptoms more important than the possible social ramifications? Do sustained social and emotional problems associated with the symptoms mandate the need for further chemical intervention? Is musical training initiated at an early age a possible solution to the symptoms or severity thereof?

Biological Implications?

One of the most interesting theories surrounding the causation of TD is not neurological or environmental but biological. Two dentists have made claims that their mouth piece device called a Neurocranio Vertical Distractor (NCVD) initially created to deter pain caused by Temporomandibular Joint Disorders (TMJ) has also shown promising results in their patients with Tourette's and similar disorders. They propose that TD is a structural deformity that manifests itself as a neurological issue and by fitting the patient with the manufactured NCVD they are able to eliminate nearly all of the symptoms (Sims & Stack, 2009). These two ascribe to the theory that the influences of the neurons believed to be affected within the cortico-striatal-thalamo-cortical circuitry are affected much earlier as a "reflexive disorder in the spinal cord region within the spinal trigeminal nucleus, specifically the subnucleus caudalis." They also speculate that one of the most common tics in TD, facial tics or blinking, are associated with this reflexive disorder. The trigeminal area has been associated with reflex blinks in other

studies (Espinosa, Roig, Perez, & Mas, 2015; Ryan, Kaminer, Enmore, & Evinger, 2014) and may add merit to their claims. Their scientific knowledge and the manner in which they propose these ideas are overshadowed by what they call a case-study with a very small sample ($N = 6$). Their study however, has not gone unnoticed and recent funds have been approved by the Tourette's Association of America for additional clinical research into their NCVD and its effect on TD symptoms (McNaught & Scullin, 2016).

Other Considerations

As the scientific, genetic and medical questions continue to elude answers there are at least two other complications undermining the assessment and treatment of TD. The first challenge is the logistical task of addressing the dichotomous areas of research that TD spans and the global collaboration necessary to make uniform and cooperative decisions. Although the World Wide Web has provided the means to conduct unprecedented research in *any* area of interest, its growth and expansion have created an unmanageable system of accumulated information that cannot be easily monitored (Teixeira da Silva, Jaime A & Dobránszki, 2015). This vast publishing information from scholarly peer-reviewed journals and publication numbers have surfaced in a document called the STM Report which is produced from a marketing firm in association with the International Association of Scientific, Technical and Medical Publishers. This report claims there are approximately 28,100 active scholarly English-language journals at the end of calendar year 2014 which, coincidentally, was the same number reported in an earlier 2012 edition of the same report (Ware & Mabe, 2015). The prevailing issue here is the management of data and research from a wide spectrum of scientific and medical fields and the accurate reporting and assimilation of that data as well as its free use.

In response to these difficulties groups have formed in the areas of support and/or research internationally, including Tourette's Action-UK, the European Society for the Study of Tourette's Syndrome, and their North American counter-part, the Tourette Association of America. In the summer of 2015 great strides were made at the 1st World Congress of Tourette Syndrome and Tic Disorders in London, U.K. to unite the normally divergent groups of science, therapy, medicine, genetics and education to discuss current relevant topics that could provide hope and direction for a global solution to the disorder.

The second challenge in discovering the answers behind TD lie in its comorbid nature with other neurological disorders. Recent literature associates Tourette with four other disorders: ASD (Autism Spectrum Disorder), DCD (Developmental Coordination Disorder), OCD (Obsessive Compulsive Disorder) and ADHD (Attention Deficit Hyperactivity Disorder) (Freeman, 2015). These disorders are exacerbated by other psychopathologies such as anxiety, depression, aggression, self-injurious behavior (SIBs), sleep difficulties, coprophenomena, sexually inappropriate behavior, stuttering and social skills (Wanderer et al., 2012). Examples of conflicting research abound. In a recent study of OCD comorbidity, Lochner and colleagues reported that the highest rate of concurring activity occurred with tic disorders (Lochner et al., 2014). In a similar study, Malhany and cohorts reported that ADHD is the most common comorbid activity with TD (Malhany, Gulisano, Rizzo, & Curatolo, 2015). Lebowitz and company also studied the complexities of tic severity and how they coincided with comorbidity of OCD and ADHD and concluded that if the comorbid conditions existed, the symptoms severity also increased (Lebowitz et al., 2012).

These and similar findings began emerging with the creation of a multisite,

international database known as the Tourette Syndrome International Database Consortium (TIC) developed in the 1990s whose results were first published in 2000. This was the largest effort of its kind to date consisting of data collected from 3500 individuals in 22 collaborating countries from the leaders in their fields from Canada, the US and UK (Freeman et al., 2000). With collaborative efforts such as these, hopefully researchers will bridge the gap in all fields studying TD and tic related disorders.

Summary

The majority of the literature reviewed has provided fairly consistent findings in the areas of clinical definitions, age of onset, symptoms, genetic tendencies and the neurological foundations for primary areas of clinical focus. More recent studies have offered evidence for behavioral therapies which have been labeled effective but have not included music and early music education training as viable alternatives for treatment methods to this point. The body of literature which presents the neuro-developmental hypothesis of TD coupled with the results of musical training studies changing brain structure are the backbone of this study. This literature contains the foundation for music educators to promote and advocate music education and early training with musical activities as the basis for healthy neurological development.

Chapter Three: Method

Research Design

This study was designed to use a self-report on-line survey instrument to gather data about the perceived relationship between types of musical engagement and the possible influences on TD symptoms for musicians diagnosed with the disorder. A survey instrument is a logical step (and often used) to propose possible future actions. The goal of this research was to reveal the subset of population with TD who are self-proclaimed musicians and explore their perceptions of musical engagement as either a therapeutic device for, or instigator of, symptoms. The justification for future research, time, resources and funding could be revealed in the number of respondents and the answers to these survey questions.

Participants

The desire to study musicians diagnosed with TD was a reasonable outcome given the amount of research data on the effects of music on the brain coupled with the fact that it is currently considered a neurodevelopmental disorder. Although the subset of TD musicians may not be enough for generalizations to the broader population, it was the author's hope to explore this phenomenon reported by musicians.

Initial recruiting of participants was done through the aid of three of the largest organizations responsible for the study and advancement of the disorder: The Tourette Association of America, European Society for the Study of Tourette Syndrome and

Tourette's Action-UK. These three entities were responsible for research announcements and email dissemination for recruitment of survey personnel. Once the Institutional Review Board (IRB) protocol was approved, the researcher solicited the help of the organizations above who aided in the recruitment of participants. Additional tools were created via the creation of a research website and social media page. When the researcher was contacted and provided an email address, the IRB consent document (see Appendix A) was then forwarded to the prospective participant. After the creation of the research sites a direct consent link was added. For the sake of anonymity and the fulfillment of review board requirements no internet protocol addresses were requested or collected, however email addresses might have been used to answer further questions and offer encouragement for the completion of the instrument. Survey participants who voluntarily responded to announcements, email solicitation, social media or research website were instructed to select the *submit* text button within the consent document which served as an external link to the Google documents survey measure. Participants included any person responding to the survey announcements placed on the Tourette's Association of America research page, author's research page or through social media solicitation that engage in a musical activity and have been diagnosed with TD. Protocols mandated parent or guardian completion of the survey for minors. The number of self-report musicians with TD related symptoms who responded to the research web pages advertisements and social media outlets were 191. Timestamp analysis and non-informative demographic data revealed one response that was repeated (duplicate submission) and therefore eliminated. Seven other respondents were excluded that did not meet inclusion criteria which left a total $N = 183$ in the data for analysis.

Survey Instrument

The survey instrument (Musical Engagement Influence on Tourette's Symptoms, or MEITS) consists of ten questions (see Appendix B for the full survey). The selection of fewer items seemed plausible for successful response rates in light of other survey instruments which may be too long generating lower completion rates. The author proposed that this issue can be exacerbated by survey participants with comorbid disorders such as ADHD. In a large on-line survey Conelea and colleagues (2013) reported an approximate 55% completion rate. Over twelve hundred consented ($N = 1216$) to take the survey and 970 answered at least one question but the final data reported was from 672 participants. This could be attributed to the length of their survey instrument (which she reported took an average of sixty minutes to complete) and/or comorbid disorders making the task of completing a lengthy measure difficult. This served as the rationale for creating a brief instrument with higher completion percentage rates. There are also possible benefits to increased participation based upon on-line measures shorter in duration. These internet surveys may be more accessible for people in rural communities or of lower socioeconomic status that might have basic internet access but not access to a large town where most clinical studies might occur. Research should not be biased and based upon those individuals with the resources to make frequent clinical visits but should be accommodating to include all classes. This was the rationale to include a web-based survey with the hopes of reaching as many as possible. The current version of the MEITS is available in English only but may be translated and adapted for future studies.

The types of questions designed for the MEITS were categorized as:

- Six nominal questions attributed to diagnosis, symptoms, musical activity, musical instruments, gender and ethnicity
- Two ratio questions analyzing minutes per week of musical engagement activities, age and
- One ordinal question using a Likert-scale of one to five.

The aforementioned survey question number five was the primary outcome response item (or dependent variable). Question 5 represented the survey respondent's perception of symptom reaction to musical engagement activities. The respondent's answer to the survey apex should reveal a substantial total mean to merit further analysis and future studies.

Question one was designed to ascertain clinical diagnosis. If their response was "no" they were eliminated from the study. Although subsequent questions revealed they may have met most of the DSM criteria, the length of symptoms activity was not determined in this instrument. *If* they have been diagnosed with Tourette's then they proceeded with question two which targets the independent variable of "are you regularly engaged in a musical activity such as playing or practicing a musical instrument?" Further stratification of data is possible to analyze the correlations between other mediating variables such as types of symptoms (question 6) or types of instruments played (question 4) as well as gender and age (questions 7 & 8). Current research suggests that the onset of symptoms can appear around the age of six years and peak during adolescence (Cohen et al., 2013). Age could become a dramatic factor if future research could show that the engagement of a musical activity and especially musical training correlates with the severity of the symptoms experienced by a family genetically

prone to the disorder. Further analysis could also reveal the correlation of engagement in an instrument requiring diaphragmatic breath control with that of the vocal symptoms associated with TD (i.e., throat clearing, shouting, barking or grunting).

Procedures

Protocol guidelines suggested twenty minutes to complete the survey. Once finished and submitted, the responses were recorded on a password protected site and saved in spreadsheet format for analysis. Data collection date range for the survey was 431 days (approximately 14 months). It should also be noted that the initial survey announcement bulletins on organizational websites were painstakingly difficult to find. Typical organizational restrictions would not allow a direct link to the survey measure which made finding the announcements challenging. These circumstances led to very few initial responses to the research solicitations; it was only after the creation of a personal research page and subsequent social media outlet that responses and interest in the study increased dramatically. Careful wording on all postings and documentation was considered to avoid potential biases towards research outcomes.

Ethical Considerations

All ethical concerns were addressed via the IRB review process through the University of South Florida. The consent document addressed adults answering for their minor children as well as other potential risks, which are considered minimal, and not outside the realm of daily normal lives. Any potential issues related to privacy of the participants were controlled by a non-identifying voluntary survey. There was no screening for diversity or ethnicity. Internal Review Board protocols also required an

extra consent proponent should some participants be faculty members or students of the sponsoring University to ensure measures against any feelings of coercion were avoided.

Chapter Four: Results

The MEITS survey data were compiled from 183 respondents answering ten questions regarding demographics, diagnosis, musical engagement activities, symptomology and the effects on symptoms. Statistical results of the survey instrument was accomplished using IBM SPSS Version 23.

Demographic Data

The survey instrument included questions pertaining to ethnicity, age and gender but were void of identifiable information. The demographic data provided information about the characteristics of the sample, as expressed in descriptive statistics.

Ethnicity. Findings suggest that there was a large percentage of white/Caucasians among the study participants. Although this report is unique in its study sample of musicians with TD it is line with similar reports of Tourette populations (see Table 1).

Table 1

Ethnicities Reported by Participants

Ethnicity	Frequency	Percent
White/Caucasian	164	89.6
Hispanic/Latino	9	4.9
Multiracial	5	2.7
American Indian or Alaskan Native	3	1.6
Asian	1	0.5
Other	1	0.5

These percentages from these results resemble a large dataset ($N = 672$) gathered by Conelea and colleagues with very similar statistics (Conelea et al., 2013). This report is consistent with other prevalence and survey studies that describe ranges of 80-97% of those diagnosed with TD, Caucasian (Conelea et al., 2013; Lebowitz et al., 2012; Specht et al., 2013; Storch et al., 2007; Wilhelm et al., 2012).

Age. There was a wide range of ages reported from respondents and their average was relatively young ($M = 24.69$, $SD = 14.69$). The minimum age submitted was eight and the maximum was 65. The most commonly reported age was 13. Fifty-five percent (55%) of the group were twenty or younger. Concurrent research supports this data on age and the apparent waning of symptoms as age increases (Cohen et al., 2013; McNaught & Mink, 2011). The male average age of the participants in this study was twenty five ($M = 25.05$, $SD = 15$) with acceptable skewness of .87 ($SE = .21$) and kurtosis of -.32 ($SE = .43$). Female age average was 23 years 11 months ($M = 23.87$, $SD = 14.04$) with a slightly higher skewness of 1.16 ($SE = .32$) and an acceptable kurtosis of .55 ($SE = .63$). The histograms below detail the visual distribution, descriptives and

frequencies of ages for both male (Figure 4.1) and female (Figure 4.2) respondents.

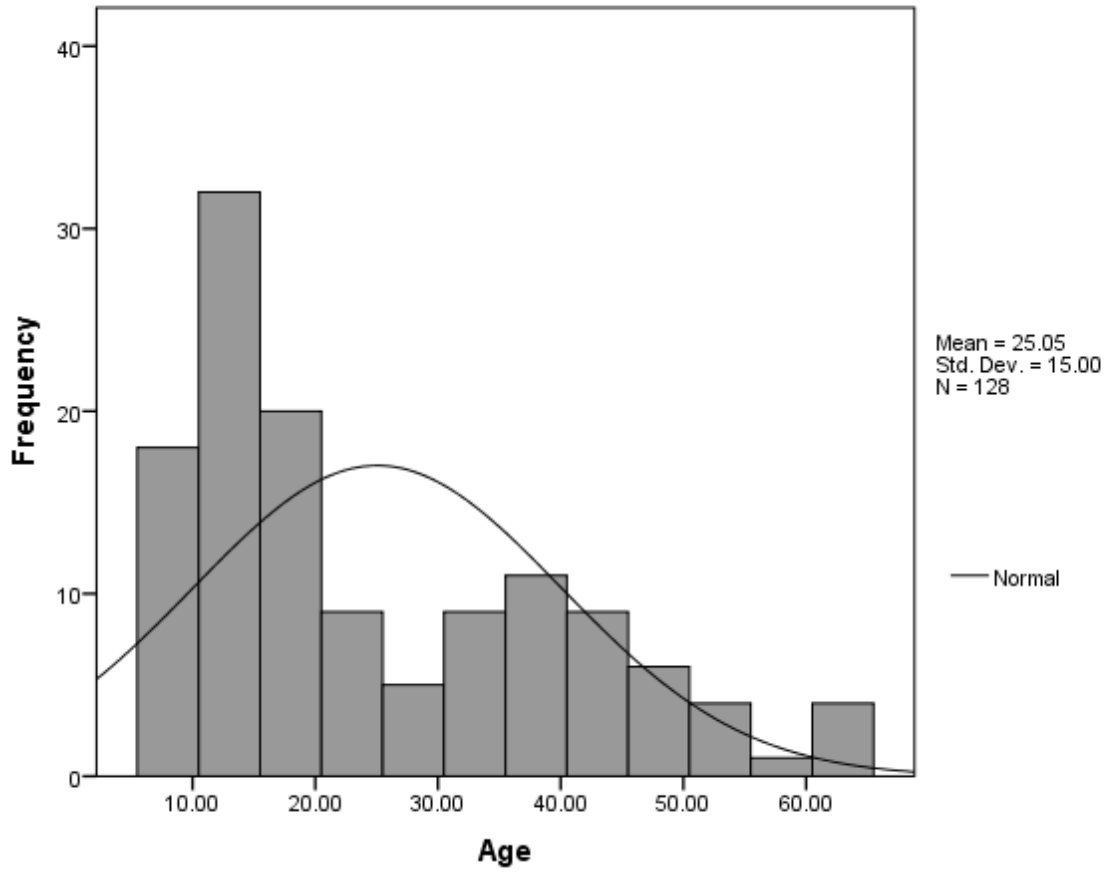


Figure 4.1. Male respondent age frequencies, descriptives and distribution.

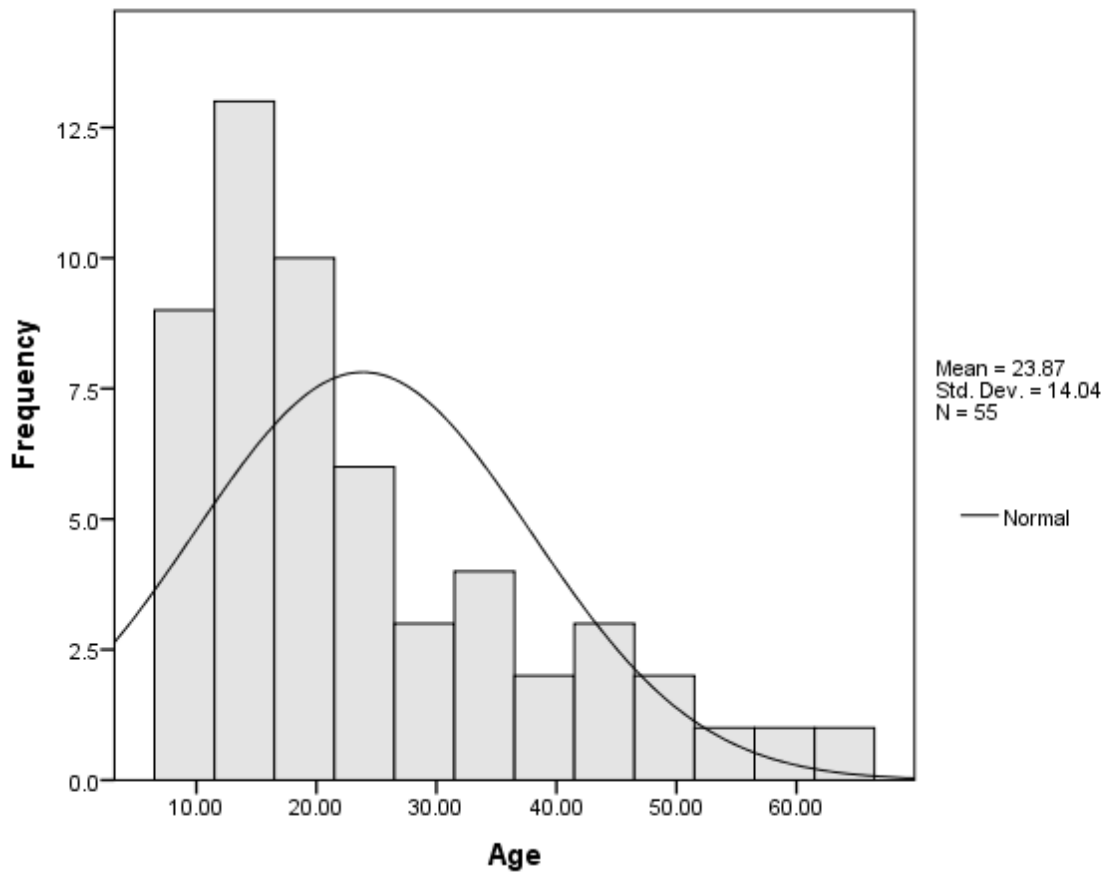


Figure 4.2. Female respondent age frequencies, descriptives and distribution.

Figure 4.3. details the combined gender data on the age of the participants. Results revealed a modest skewness of 0.94 ($SE = 0.18$) and kurtosis of -0.13 ($SE = 0.36$).

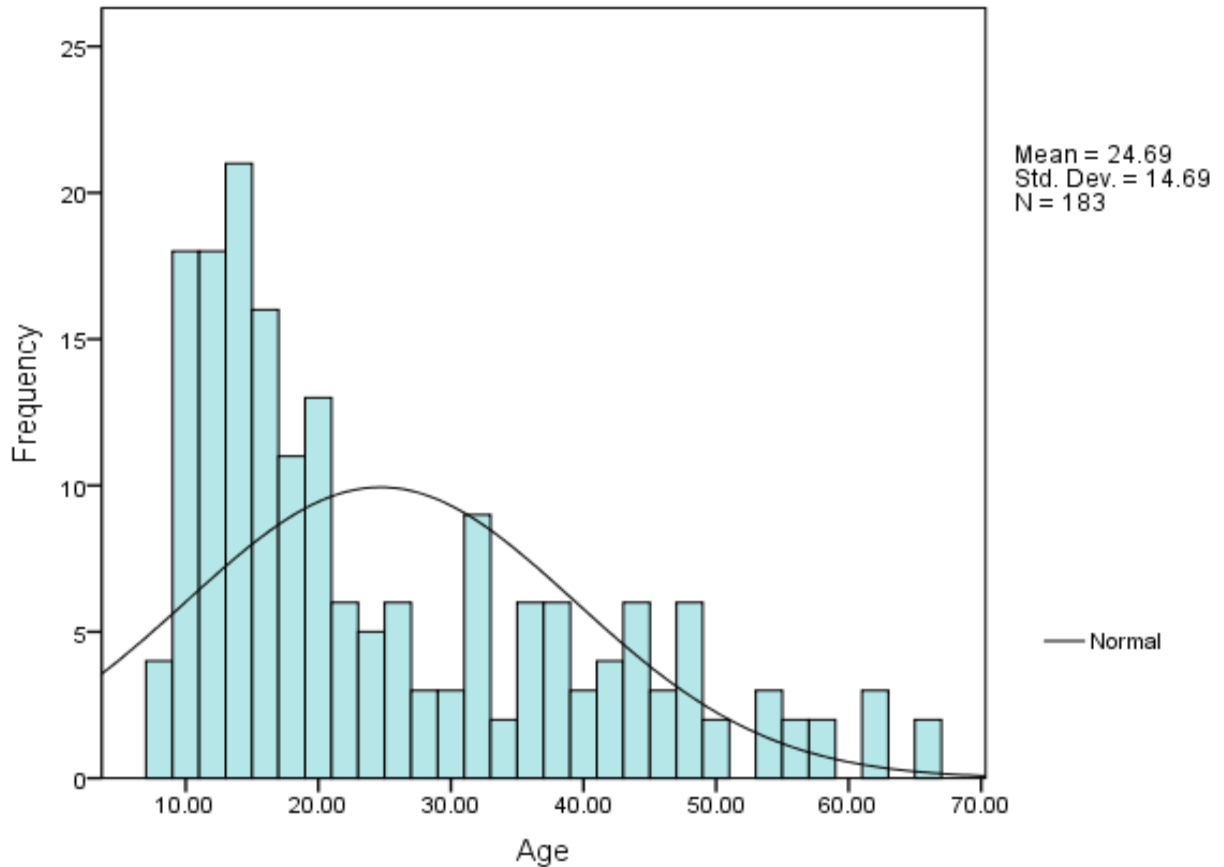


Figure 4.3. Combined genders with age specific totals for frequencies and descriptors.

Gender. In light of more recent studies the gender reports are not unusual by comparison. This study reported seventy percent (70%) males and thirty percent (30%) female respondents and is consistent with other recent studies with male-predominate TD diagnoses and its accompanying statistical data results within the sixty to eighty percent range (Conelea et al., 2013; Cuenca et al., 2015; Lebowitz et al., 2012; Plessen et al., 2004; Rizzo et al., 2014).

Diagnosis and Engagement

The dichotomous question one of the survey sought to address clinical assessment. In lieu of a clinical study there was an attempt made to deter self-diagnosed individuals from continuing the survey (see Appendix B for complete survey). Of the one hundred ninety-one initial responses to the survey five responded that they had not received a diagnosis for having Tourette. These were excluded from analysis. If they responded “yes” they were directed to proceed to question 2. Question two was also binary in nature, which was subordinate to the first, and required a response of “yes” to indicate that they were regularly engaged in a musical activity such as practicing or playing an instrument. Three respondents replied that they were not regularly engaged in a musical activity and were also eliminated from data analysis. The question two response led to determining how many minutes per week the respondent was regularly involved in their activity (question 3).

These activities included active engagement on their musical instrument(s) whether electronic, acoustic or biological (the voice). The 183 respondents reported playing over 250 instruments prompting the creation of a “multiple” category for purposes of analysis. If the types of instruments reported in the multiple category were of the same type (i.e., mandolin, banjo and guitar) and could be easily consolidated as one they were combined. All instruments reported (including the human voice) were then categorized by classification or construction. These constructed categories included stringed instruments (either bowed or strummed), woodwind instruments, brass instruments, keyboard-type instruments (either electronic or acoustic) or percussion instruments (anything struck to make a sound). These are the typical categories used in

music education to describe the various “families” of instruments. The category “Percussion-ME” was added to describe those musicians who reported playing percussion instruments using all appendages (i.e., drum sets) and therefore termed “multi-extremital.” It was possible that this type of playing represented more physical (and possibly cognitive) involvement than someone playing a percussion instrument that uses only two hands such as a bodhrán (hand drum) or tambourine. These data were stratified later within this document. The categorized totals from the survey responses can be seen in Table 2 below.

Table 2

Primary Musical Instrument Reported

Instrument	Frequency	Percent
Percussion	3	1.6
Brass	11	6.0
Woodwinds	11	6.0
Percussion-ME	15	8.2
Voice	16	8.7
Keyboard-type	40	21.9
Multiple*	43	23.5
Stringed	44	24.0
Total	183	100.0

Note. 23.5% of respondents reported playing multiple instruments and did not list a single primary preference.

The middle group of survey questions were designed to address the symptoms reported by the participants. To go deeper than the basic diagnosis, one subsidiary question was asked if the participant experienced “other” symptoms outside of the current accepted diagnoses. Forty percent of participants (40%) reported other symptoms not currently included in the DSM. These “other” symptoms were possibly due to comorbid

disorders such as OCD and ADHD but were not the focus of this study and therefore excluded from further analysis.

Addressing Research Questions

The following analyses are designed to address specific research questions outlined in this study.

Research Question 1. The apex question (survey question 5) was a Likert-type scale seeking to determine the influence of musical engagement perceived by these self-reported musicians with TD. This was developed to answer the research question: To what extent does musical engagement relate to symptoms change for those diagnosed with TD? Figure 4.4. presents the total number of participants per response rate.

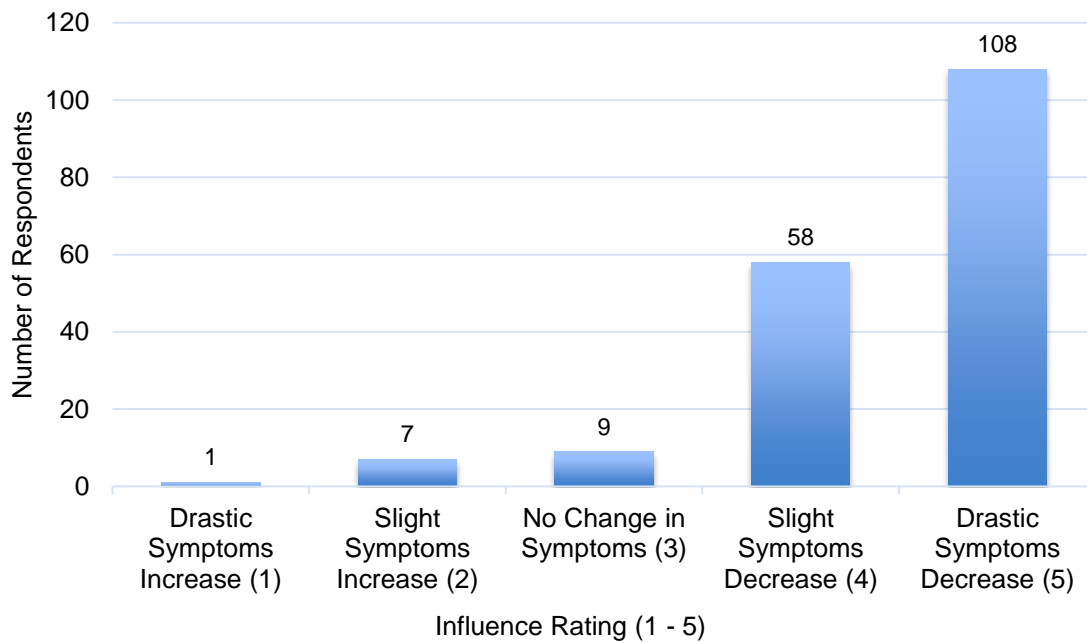


Figure 4.4. Total response ratings of symptoms influence while engaged in a musical activity.

The statistical mean of the response items for the scale suggests that there *is* a perception of symptoms decrease in musicians reported to have TD ($M = 4.45$, $SD = 0.80$) while they are engaged in a musical activity. The report of symptoms change has an abnormal distribution, with a skewness of -1.70 ($SE = 0.18$) and kurtosis of 3.03 ($SE = 0.36$). This deviance in distribution is attributed to the unusually high number of responses answering either a four or five on the rating scale and few reporting symptoms increase of any kind (less than 5%). Of interest to the author was the sole respondent reporting a drastic symptoms increase. This single case also reported an “other” symptom outside of the normal DSM categorization which might represent evidence of co-occurring movement disorders with comorbid activities. Without this single case, the skewness became -1.55 ($SE = .18$) and the kurtosis remained high at 2.22 ($SE = .36$).

Symptoms influence percentages can be seen in the table below. This data revealed that ninety percent (90%) of respondents affirmed perceptions of at least some symptoms decrease when engaged in a musical activity. The responses from individuals have been grouped by their answer frequencies to achieve a percentage total from each scale rating (see Table 3).

Table 3

Percentages Totals for Each Scale Response

Symptoms Influence Categories	Percent
Drastic symptoms increase	.5
Slight symptoms increase	3.8
No change	4.9
Slight symptoms decrease	31.7
Drastic symptoms decrease	59.0
Totals	100.0

Research Question 2. The second research question was “what are the correlations between symptom changes, time per week engaged in a musical activity, age and total number of symptoms reported”? The “average time per week” variable disclosed an open range with the weekly average time reported slightly less than four hundred minutes ($M = 397.45$, $SD = 855$). This data distribution was clearly abnormal with skewness of 5.82 ($SE = 0.19$) and kurtosis of 38.83 ($SE = 0.39$). These extreme results are the product of such a wide range of minutes reported where the mean, median and mode were not consistent. The most common time selected was two hours yet the vast majority of participants (75%) were engaged less than six hours per week (or less than an hour a day). There was an apparent contingent of professional musicians involved as well, which also contributed to the abnormal distribution, reporting practice times in excess of 15 hours per week.

The self-reported musicians in this study represented a wide range of competency levels based on their answers to this question. Terms such as “casual” or “professional” musicians were not included as a label for categorization. The data represents merely the range for which the respondents reported engaged in an activity. One hundred fifty-six responded with a number of hours per week to this survey question ($N = 156$). Twenty-seven were excluded from analysis because they did not provide a single numbered response but an engagement of activity range (i.e., “4-6 hours per week”). Figure 4.5 below represents the ranges of time spent per week on their musical activity.

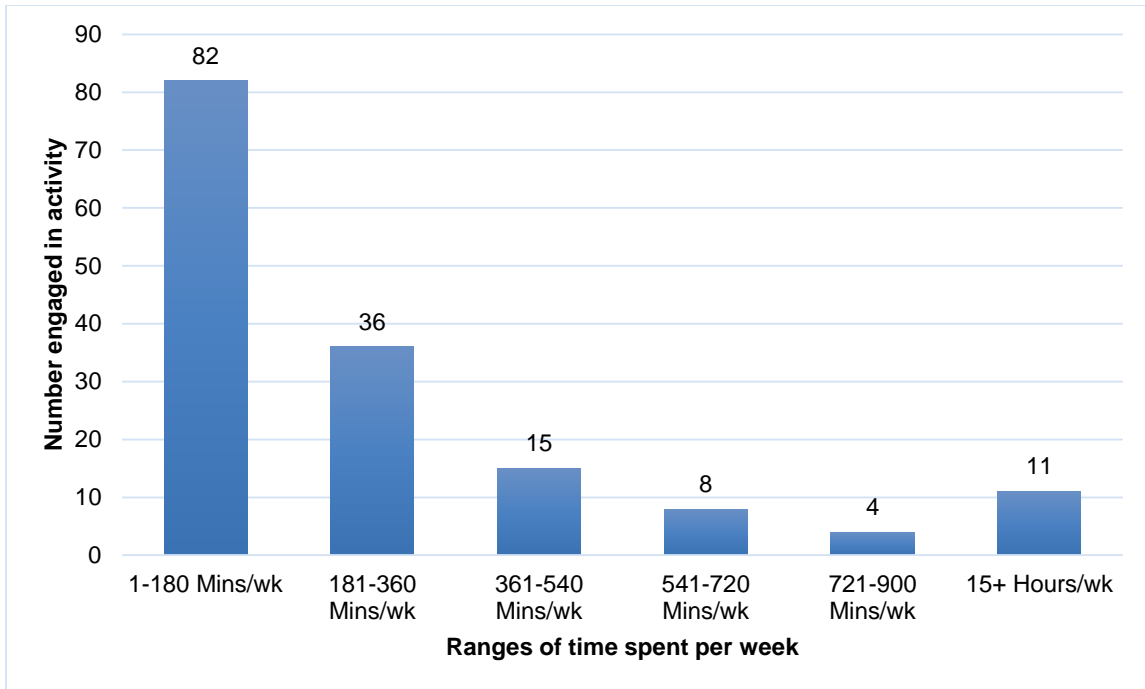


Figure 4.5. Minute ranges per week of musical engagement activity.

The symptoms reported by participants were chosen for the survey instrument using the current DSM Manual referencing the most common indicators for tics and vocalizations. Respondents were asked to check all boxes representing their symptoms. One extra checkbox was added to record any other symptoms that may currently fall outside of the DSM norm but is still experienced by the participant. The symptoms list is as follows:

1. Tics of the face (eyes, nose, mouth)
2. Grunting
3. Throat clearing
4. Tics of the arms and legs
5. Trunk tics
6. Shouting

7. Kicking/stamping
8. Barking
9. Touching
10. Coprolalia (involuntary utterance of obscene words)
11. Echolalia (echo speech)
12. Other

Symptoms were coded and separated to provide a total number, per participant, for further investigation. This dichotomous data shown below indicates the total number of occurrences for each symptom. The total number of each symptom which was recorded by the respondents ($N = 951$) equals the total number of symptoms reported from *all* participants. Percent of cases reveals the total number selected by category per participant group ($N = 183$). Nearly ninety-seven percent (97%) or 177 of the 183 respondents selected “Tics of the Face (Eyes, Nose or Mouth)” which was by far the most prevalent symptom. Participants averaged slightly over five symptoms each ($M = 5.20$, $SD = 2.41$) resulting in a minimal skewness of 0.87 ($SE = 0.18$) and kurtosis of 0.64 ($SE = 0.36$) (See Table 4).

Table 4

Symptoms Reported and Total Percent of Cases per Category

Symptoms Reported	Responses		% of Cases
	Frequency	% of total Frequency	
Tics of the Face (Eyes, Nose or Mouth)	177	18.6%	96.7%
Tics of the arms/legs	139	14.6%	76.0%
Throat Clearing	126	13.2%	68.9%
Grunting	92	9.7%	50.3%
Touching	77	8.1%	42.1%
Other Reported Symptoms	73	7.7%	39.9%
Trunk tics	69	7.3%	37.7%
Echolalia	52	5.5%	28.4%
Kicking/Stamping	46	4.8%	25.1%
Shouting	40	4.2%	21.9%
Coprolalia	35	3.7%	19.1%
Barking	25	2.6%	13.7%
Total	951	100.0%	

Correlation and multiple regression analyses were conducted to examine the relationship between symptoms influence and the predictors, minutes per week of musical engagement, age, and total number of symptoms reported. Table 5 summarizes the descriptive statistics, correlation and regression results. Age ($r = .212, p = .004$) and number of total symptoms ($r = .251, p = .001$) reported positively and significantly correlated with the criterion variable. Minutes per week of musical engagement activity showed no significance with symptoms influence ($p > .05$). This could indicate that *any* amount of activity is beneficial and does not require the construct of time as a predictor of the participant's reported symptom influence.

Table 5

Correlations, Means, and Standard Deviations for Regression of Criterion (N = 156)

	1	2	3	4	M	SD
1. Symptoms Influence		.12	.21**	.25**	4.44	0.81
2. Mins per week of Musical Engagement			.45**	.31**	399.05	856.36
3. Age				.38**	24.11	14.70
4. Number of Total Symptoms					5.25	2.43

Note. ** $p < .01$

A stepwise multiple regression was conducted to predict symptoms influence based on minutes per week of musical engagement, age, and number of total symptoms reported. The overall model was significant, $F(3, 152) = 6.71, p < .001$, and accounted for 12% of the variance. Results also indicated that age and number of total symptoms reported, but not minutes per week in musical engagement, were significant predictors of the influence on symptoms (see Table 6).

Table 6

Stepwise Regression Analyses Results of Symptoms Influence as Criterion Variable

Predictor Variables	B	SE	β	t	p	r
Number of Total Symptoms Reported	0.081	0.026	0.24	3.16	0.002	0.251
Age	0.011	0.004	0.21	2.72	0.007	0.212
Mins/week of Musical Engagement	0.000	0.000	0.11	1.43	0.156	0.116

Note. $F(3, 152) = 6.71, p < .001, R^2 = .118; R = .342$

Point-biserial analyses were conducted on the correlation between symptoms influence from musical engagement and each of the reported symptoms. Of the listed symptoms from the above table (Table 4), four showed significant results.

These included the symptoms of “grunting” $r = .243, p = .001, N = 183$, “shouting” $r = .166, p = .024, N = 183$, “tics of the arms/legs” $r = .299, p < .001, N = 183$, and “kicking/stamping”, $r = .195, p = .008, N = 183$. Participants with these symptoms tended to perceive more reduction of symptoms as influenced by musical engagement. It was postulated that these results might be related to the physical requirements entailed in musical activities and are explored further in Research Question 4.

Research Question 3. The next part of the analysis sought to reveal “to what extent do types of instruments used for participant engagement and gender influence symptoms change for those diagnosed musicians with TD?” Gender results showed no significance on symptoms influence ($r = .04, p = .64$) yet there were some inconsistencies with other studies that have reported male/female ratio in TD patient research. Earlier accounts have the ratio for male/female in the 3-4.3:1 range (Freeman et al., 2000; Robertson & Stern, 2000; Robertson, Eapen, & Cavanna, 2009) and subsequent studies accepted these for their reports (Woods, Walther, Bauer, Kemp, & Conelea, 2009; Woods, Conelea, & Himle, 2010). Some of the more recent studies are reporting more equal numbers in the 1.4-2.3:1 range (Caurín, Serrano, Fernández-Alvarez, Campistol, & Pérez-Dueñas, 2014; Conelea et al., 2013). Caurín and colleagues errantly reported their ratio data. They published a 3:1 ratio yet their numbers were 67 boys out of 92 subjects; this should equal a ratio of 1.37:1. In the only other musical study with Tourette’s and symptoms severity Bodeck and colleagues (2015) reported even closer ratios (.55:1). The author’s data results were 2.33:1, which details the need for more thorough analysis with current data. Future studies should include possible gender or response biases which may

be the case in this study if females play instruments more than males and therefore align more closely the dataset.

The respondents reported playing many different types of musical instruments: The categories included the brass and woodwind families, stringed instruments, keyboard-type instruments which encompassed acoustic and electronic, percussion instruments and the voice. The brass instruments reported were the tuba, baritone, trumpet, trombone and euphonium. Stringed instruments were grouped together for analysis based on their similarities in the method by which they are played as well as constructed. These included acoustic, electric and bass guitars, the violin family of instruments as well as a few used in folk music. Woodwind instruments are described as such because of the manner in which air is blown through a tube or reed constructed from wood (or in the case of a silver flute, formerly made from wood) but similarly to brass, the production of sound is controlled by air blown or forced through an aperture controlled and supported by diaphragmatic breathing. This breathing technique is essential to producing an audible tone and could also be compared to the only biological instrument in the survey, which coincidentally, also involves diaphragmatic breathing; the human voice. Percussion instruments can be described as any musical device which is struck to produce a tone. There are two types of percussion instruments; pitched and unpitched. These are usually separated based upon the acoustic quality of the sound produced and whether or not its tone can be detected among a diatonic (musical) scale. For example, a xylophone or marimba are said to be pitched percussion, while a bass drum, snare drum or cymbals are unpitched. For the purposes of this study the type of

percussion instrument was not differentiated (pitched/unpitched) although the methods by which they are played were.

The table below details the sub categories of brass (11), voice (16) and woodwinds (11) reporting either slight or drastic symptom decrease on the symptoms influence scale. None of the participants in these subcategories reported slight or drastic symptoms increase or no change in symptoms (ratings 1, 2 or 3 from the scale). Although the frequency counts in these subcategories were small the patterns seen in the results may warrant future investigation and are discussed in the next chapter.

Table 7

Symptoms Influence and Musical Instrument Crosstabulations

Symptoms Influence	Brass	Keyboard	Multiple	Percussion	Stringed	Voice	Woodwinds	Total
Drastic symptoms								
increase	0	0	0	0	1	0	0	1
Slight symptoms								
increase	0	3	1	2	1	0	0	7
No change in								
symptoms	0	2	2	1	4	0	0	9
Slight symptoms								
decrease	4	10	10	4	14	10	6	58
Drastic symptoms								
decrease	7	25	30	11	24	6	5	108
Total	11	40	43	18	44	16	11	183

A One-way ANOVA was conducted to ascertain if the seven groups of musical instruments played by the participants might also influence their symptoms. The results showed no significance, $F(6, 176) = .585, p = .742$; however the patterns (as seen above) led to additional investigation as a point of inquiry into possible correlations with diaphragmatic breathing (a concept very familiar to musicians) and the physical requirements needed to engage in their reported musical activity.

Research Question 4. The final research question sought to discover if there might be correlates with the body's physical involvement during a musical activity and symptoms responsiveness. The two sub categories that emerged from this data were the requirements upon the body either through action alone or the combination of diaphragmatic breathing with physical involvement. The first group of instrumental coding was done based upon the physical method used to engage the participant in music making. Empirical studies including neuroimaging and cognition research have labelled these physical methods as being unimanual or bimanual (Debaere, Wenderoth, Sunaert, Van Hecke, & Swinnen, 2004; Lang, Obrig, Lindinger, Cheyne, & Deecke, 1990; Marteniuk, MacKenzie, & Baba, 1984; Nair, Purcott, Fuchs, Steinberg, & Kelso, 2003). Each term implies the manner in which a person is engaged in their activity and to what extent the body (including cognitive processes) is involved. For example, a brass player's involvement could be described as unimanual; they position the instrument at the mouth and blow into the mouth piece but only one hand is used in operating the valves (or slide, in the case of a trombone) while the other hand is virtually inactive holding the instrument in place. Bimanual operation can be more easily seen in pianists or saxophone and clarinet players. The obvious involvement of both hands to produce pitches in order

to make music and the cognitive effects of this involvement has also been studied (Jäncke, Shah, & Peters, 2000; Sluming et al., 2002). These studies have typically been conducted on professional or expert musicians whereas casual music making of instruments such as the guitar or drums has not been vigorously explored in the cognitive domain. It was hypothesized that percussion players which participate in musical ensembles outside of the orchestral setting might require another category based upon their *multi-extremital* physical involvement. It was the author's assumption and theoretical view that from a cognitive and neurological standpoint that these musicians might experience greater cessation of symptoms based on their more rigorous physical and probable cognitive involvement. This can be exemplified in those reported playing brass (unimanual) and woodwind instruments (bimanual) as well as vocalists (biological). Descriptive analysis based upon the types of physical involvement required by the participants during their engagement activity showed that over half of the respondents (55.7%) were involved in bimanual operation of their instrument (See Table 8). This was determined based upon constructional categories and the method by which their instruments are played whether percussion, stringed, keyboard or wind. Brass instruments unvaryingly fall into the unimanual category by the nature in which they produce a tone. The groups analyzed by physical involvement included Uni-manual, Bi-manual, Multi-extremital, Vocal, Bi-manual requiring diaphragmatic breathing and those reporting "multiple" instruments. Although a One-way ANOVA analysis detailed no statistical significance, $F(5,177) = .435, p = .824$ a pattern emerged for instruments requiring breathing as the method of tone production.

Table 8

Physical Involvement with Musical Activity

Physical Involvement			
	Instrument(s)	Frequency	Percent
Unimanual	Brass	11	6.0
Bimanual	Piano, Strings	102	55.7
Multi-extremital	Drum set	15	8.2
Vocal	Voice	16	8.7
Bimanual w/ Diaphragmatic Breathing	Woodwinds	11	6.0
Multiple*		28	15.3
Total		183	100

Note. The “Multiple” category included those participants with more than one instrument reported that could not be placed in only one group; however some dual reports were placed within one segment such as guitar and keyboard (both bimanual not requiring diaphragmatic breathing).

Could these results indicate a greater symptoms influence based upon diaphragmatic breathing and not from physical involvement alone?

The methods by which the instrumental activity is physically executed with or without diaphragmatic breathing was divided into three groups. There were a random number of cases ($N = 129$) selected to create nearly equal sample sizes across these groups. Group one consisted of instruments requiring diaphragmatic breathing ($N = 39$). Group two were instruments that did not require diaphragmatic breathing ($N = 63$) and the final group contained non-specified combinations ($N = 27$). The gender split was 74 males and 55 females. A Levene's Test of Equality of Variances was conducted to insure

there were no violations of assumption for homogeneity; $F(5, 124) = 0.94, p = .46$. A Factorial 2 x 3 ANOVA was conducted to determine the main effects of gender and instrumental groups and their interaction effect on the symptoms influence scale. There was no significant effect for gender, $F(1, 128) = 0.48, p = .49$, or instrumental groups $F(2, 127) = 0.94, p = .39$; there were however, other items of note that will be discussed in the following chapter.

The Musical Engagement Influence on Tics Scale did report important results for musicians diagnosed with Tourette's. Although there was no differentiation in this study on the competency levels of the self-reported musicians, there is clearly a perceived influence change for the better when engaged in their musical activity regardless of classification. Conclusions can be drawn from the current data in this study and will be elaborated upon in the next chapter.

Chapter Five: Discussion and Conclusions

The purpose of this chapter is to explore the answers to the research questions that have been posed for this study and to interpret their findings based upon the extant body of literature. The author draws implications for music educators and future lines of research for this disorder and provides reasoning for the necessity of collaborative efforts into longitudinal research exploring the effects of musical training as a viable solution for possible developmental solutions to the disorder.

Research Questions Discussed

The Musical Engagement Influence on Tourette Symptoms questionnaire was designed to answer the research question “does a musical engagement activity influence the symptoms of Tourette’s”? The average response to this question was 4.45/5.0 clearly indicating a perception of symptoms decrease for the vast majority of the respondents when they are engaged in a musical activity such as playing an instrument or singing.

Regardless of empirical strength, the *perceptions* of these participants are as important as research data and should be treated as such. It would appear that future clinical studies and funds are justified by the research provided in this paper and should be targeted to include music as a behavioral modifier. The methods and best practices are yet to be discovered on an individual account however educational services should be encouraged to include music education at all levels to all students regardless of their

academic “label.” This should include the classroom musical environment or the small or large ensemble setting for the sake of those suffering with TD and similar disorders.

The second research question sought to address correlations between the symptoms rating scale and the following three segments including age, number of symptoms reported, and time per week engaged in their musical activity. The results for age and number of symptoms reported were significant ($p < .01$). The age factor might be explained by participants whose mean age is relatively young and at which time are experiencing more severe symptoms. The combination of age increasing, musical engagement and symptoms severity lessening might also play a role given the existing body of research suggesting neurodevelopmental changes occurring through musical training. Those who are engaged in a musical activity could be experiencing a type of “re-wiring” that lessens the degree and severity of symptoms over time.

Analysis was conducted into the symptomology associated with TD and specifically whether there was a correlation between the total number of symptoms per participant and their perceptions of change when engaged in their activity. The results revealed a significant correlation, $r = .24$, $p = .001$, $N = 183$, suggesting that the greater number of symptoms reported, the greater the sense of relief from their symptoms because of their musical activity. Only 6% of the variance are shared ($r^2 = .058$) between the number of reported symptoms and the sense of relief from the symptoms.

For the purposes of this study there was no measure of severity for the symptoms reported only whether the participant reported symptoms associated with diagnoses based upon the DSM (*The Diagnostic and Statistical Manual of Mental Disorders*) and The National Institute of Neurological Disorders and Stroke. The symptoms list for the survey

instrument was derived from the later but did not differentiate between simple or compound tics or vocalizations for those reporting. The latest version of the DSM merely defines the Disorder and does not offer guidance for the categorization of symptoms. The MEITS instrument included an additional checkbox for “other” symptoms which was reported by forty (40%) percent of the respondents. Perhaps this represents a need for revisions by which those diagnosed with TD categorize their symptomology and the methods by which clinicians classify them. The current diagnosis definition of two or more movement tics accompanied with one or more vocal tics seems merited, although by construct, vague in nature.

The results from analysis for the areas of symptoms significantly affected by musical engagement could possibly be related to the means by which these instruments are played and the tone is produced. For example, a drummer engaged in his multi-extremital activity might be more likely to have his/her arm or leg tics lessened by the natural activity of the playing motion. Whether these are the normal results of taking up an instrument requiring physical control and focus or symptoms controlled based on cognitive functioning is yet to be determined. It is interesting to pose the thought that instrumental engagement requiring diaphragmatic breathing could also be a factor and worthy of future exploration. Some research has been conducted on the effect of diaphragmatic breathing in Tourette’s patients as an aid in relaxation, behavioral therapy, habit reversal and occupational performance (Mansueto & Keuler, 2005; Rowe et al., 2013; Woods, Twohig, Flessner, & Roloff, 2003; Woods, 2008) however the field of musical engagement, diaphragmatic breathing and symptoms responsivity has not been explored.

The degrees by which the symptoms would change were hypothesized to be correlated with the minutes per week engaged in a musical activity and investigated as the second research question. The analysis of data for this study revealed this was not true. This data suggests that *any* musical activity regardless of time spent was beneficial to the participants. Altenmüller's research supports this by suggesting that increased neuronal coupling can be physically observed in fMRI studies after just twenty minutes of engagement in an activity in non-professional musicians (Altenmuller, 2008). This implicates that even short periods of musical activity are neurologically beneficial, but what are the long-term effects of intense musical training? A summary of this literature was discussed earlier (see pages 32-33) but the generalizable comment for "are there benefits to musical training?" is "yes." The body of literature is growing as well as improving. These improvements are due to focus on specific areas of neurological development and executive functioning tasks in musicians through longitudinal study and improved imaging techniques (Ellis, Bruijn, Norton, Winner, & Schlaug, 2013; Zuk, Benjamin, Kenyon, & Gaab, 2014).

The third research question sought to explore influences of gender and types of instruments on symptoms activity. There currently exists no rationale in the body of literature as to why there is a general gender discrepancy which is dominantly male. This too is a matter of future exploration for researchers. The types of instruments played during their musical activity did not show specific statistical significance although the response rates of several types showed signs worthy of exploration. These discernable patterns helped develop the final research question. The brass, woodwind, and vocal

instruments had no responses less than 4 on the rating scale. Was this the result of diaphragmatic breathing as an aid, musical training or a combination of the two?

Question four sought to reveal other possible factors including physical involvement. These were categorized into two groups for analysis. The first group analysis based on the physical method used for engagement suggests that although a singular instrument did not emerge with statistical significance for symptoms influence the overall response rates do suggest further studies are merited. Of interest to the author was the theory that the more appendages that were involved in playing of an instrument (especially the drummer with the multi-extremital activity) would produce greater cognitive activation and therefore greater cessation of symptoms this however, did prove statistically true. Of further interest and possible merit for additional study would be the correlation of those who reported playing multiple instruments and if this too is more beneficial. Another component for future studies might explore cognitive comparisons defining whether the consistent reading of musical notation during their engagement activity serves as an aid in the cognitive processing and influence rating. The second group consisting of mechanical construction of the produced tone revealed trends with the way in which the tone was produced; specifically the use of diaphragmatic breathing. Of the thirty nine cases, none reported less than 4 on the rating scale ($M = 4.46, N = 39$). The second subset of instruments being played that did not require diaphragmatic breathing had the lowest mean based on the rating scale but still in the 4-5 range ($M = 4.38, N = 117$). Interestingly, the final subset which included multiple instruments played with or without breathing had the highest mean ($M = 4.7, N = 27$). This might suggest

that multiple types of instruments played in various ways may be more beneficial but further research is merited.

These results were not statistically significant however, the small values of variance that emerge per category may suggest another piece of the larger puzzle to be considered. Additionally, of possible value for further study might be the demarcation between subcategories of diaphragmatic breathing. For example, although brass instruments are all unimanual they can be included with instruments that also require diaphragmatic breathing while parsing out the bimanual instruments that are stringed or keyboard-type that do not. Also included in the diaphragmatic breathing category would be vocalists. Although their instrument is biological in nature and produces tones in a different way than the human-made instruments their basic production of a healthy tone is predicated upon the correct use of the diaphragm.

Another consideration could be biological in nature. All three of these instruments use similar facial muscles coinciding with the shaping of the mouth to produce a tone. In musical terms music educators call this the embouchure (from the French root *bouche*, meaning “mouth”). Stack and Sims might support this idea that there is a correlation with the use of these instruments, the shape of the mouth and protrusion of the jaw that simulate their invention (the Neurocranio Vertical Distractor). A current clinical study is underway sponsored by the Tourette’s Association of America which should help shed light on this subject (McNaught & Scullin, 2016).

Implications for Music Educators

Given the overwhelming evidence for musical training activities and their influences on neuroplasticity development there need be no further argument about the

importance of music education for the developing child; especially those with this neurological disorder. Perhaps the more pertinent question is that whether starting at the typical age of five (when a student enters most primary educational systems) early enough.

Music educators could contribute to the knowledge base on TD and similar disorders by becoming more aware of the neurological benefits music has on the young and developing brain and advocating those effects. This is especially important as a basis for informed policy making as well as knowledge for parents and school administrators. Unfortunately, budget restraints and cuts in education have caused the less-informed to make inappropriate decisions to cut music programs claiming standardized testing priorities and remediation classes as more important (Dillon, 2006). A rationale errantly exists that school districts can't afford music while the author would propose due to this and concurrent data districts can't afford *not* to have music for the sake of the children and the enhanced neurological benefits, including the data from this research, which it provides. Ellis and colleagues (2013) state that "practicing a musical instrument has a profound impact on the structure and function of the human brain" (Ellis et al., 2013, p.1). For those who use words carefully in empirical research this is a bold claim. Bold claims only surface after years of study and data. This information and the hundreds of studies with similar content must be brought to the attention of those who can change the way we educate our children.

Future Research Implications

For the respondents of the MEITS Survey attempts were made to insure that self-diagnosis did not occur but cannot be verified without clinical assessment and analysis.

Perhaps future studies might include study of more video clips of musical engagement such as found on the author's [website](#) and proper ways to visually analyze them for statistical strength without the impediment of the time involved in travel and costs to medical clinics.

Additional growth and collaboration studies should be done across the boundary lines of the current disciplinary fields of neurology and music education. The goals for all fields should be the same as researchers seek to understand and possibly eliminate this disorder known as Tourette's.

Although empirical data exists suggesting waning of symptoms as age increases (Bloch, State, & Pittenger, 2011; Freeman et al., 2000; Leckman, Bloch, Sukhodolsky, Scahill, & King, 2013) there is no current body of literature for musicians with TD or longitudinal studies for the developmental implications of musical training on their diagnosed symptoms. Future research should explore these correlations more vigorously.

Conclusion

Although probably not through scientific reports or research, the musicians in this study with Tourette's are fully aware of the benefits of musical engagement on their symptoms. This research sought to ascertain their level of understanding as well as provide an empirical foundation for direction and future studies. Although perceptions are by nature difficult to quantify that does not make them less valid. Recent survey literature of two hundred ninety-five parents details their feelings that general health professionals have little knowledge of treatment options for TD sufferers and prefer a wider variety of said options (Cuenca et al., 2015). Adams and colleagues (2015) echo this sentiment among teachers who may also be ill advised or uninformed. These studies

would seem to exacerbate the feelings of frustration from parents or guardians seeking knowledge-based alternatives for their loved ones. It is the author's opinion that these issues *could* be addressed but would require a broad range of policy changes that can only occur from knowledgeable decisions based on continued empirical research and data such as this. Although the clear answers to "why" musical engagement activities affect the symptoms of such a disorder are currently elusive, this author would pose that enough evidence exists to include musical activities as a viable alternative, or at least supplement to, other treatment options.

The most tantalizing and rewarding research for those suffering with TD or their caregivers trying to manage it may lie in the musical training arena. Given the recent discoveries of the areas of the brain physically affected by TD and especially those correlates with musical training and neuro-plasticity; parents, music educators and policy makers would be wise to condone teaching as much music as possible to children as early as possible. Policy makers should be made aware of this and other pertinent research for the betterment of all children and the possible cessation of a neurological disorder.

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Appendix A: Online Consent Document

The Influence of Playing a Musical Instrument on Tourette Syndrome Symptoms

(Pro00017048)

You are invited to participate in a research study on the effects of musical engagement activities on the symptoms of those diagnosed with Tourette's Syndrome. This study is conducted by William C. Brown, at the Center for Research in Music Education from the University of South Florida, Tampa.

This study will take approximately 20 minutes of your time. You will be asked to complete an online survey about the effects of musical engagement on the Tourette symptoms for you or your minor child for whom you have legal custody or guardianship.

Your decision to participate or decline participation in this study is completely voluntary and you have the right to terminate your participation at any time without penalty. You may skip any questions you do not wish to answer. If you do not wish to complete this survey just close your browser.

Your participation in this research will be completely confidential and data will be averaged and reported in aggregate. Possible outlets of dissemination may be the TSA organization and affiliates and/or peer review journals of publication. Although your participation in this research may not benefit you personally, it will help us understand how music affects those with Tourette syndrome and provide possible avenues for future studies to aid those affected by these symptoms.

There are no known risks to individuals participating in this survey beyond those that exist in daily life.

If you have questions about this project, you may contact the Principal Investigator of this study, Wm. Chris Brown at wcbrown1@mail.usf.edu or by phone, 813-997-2137. If you have any questions about your rights as a participant in this study or any concerns or complaints, please contact the University of South Florida Institutional Review Board at 813-974-2880.

Please print a copy of this consent form for your records, if you so desire.

I have read and understand the above consent form, I certify that I am 18 years old or older with Tourette symptoms for whom this survey is about OR, I, as the parent or legal guardian have asked my child for approval to respond to this survey that will request information about my child on their behalf. By clicking the submit button to enter the survey, I indicate my willingness to voluntarily take part in the study. (By clicking on the word 'SUBMIT' below you will be directed to the survey instrument.)

SUBMIT

If you happen to be a student or adult affiliated with the University of South Florida, please read and respond below:

Your decision to participate, decline, or withdraw from participation will have no effect on your current status or future relations with the University of South Florida. By clicking submit, you have read and understand the above statement.

SUBMIT

Appendix B: Survey Instrument

The Influence of Musical Instrument Playing on Tourette Syndrome Symptoms

Wm. Chris Brown, University of South Florida

TS survey #1 for on-line anonymous survey distribution

- 1) Have you (or your child if you are the guardian/parent of a diagnosed minor) been diagnosed with Tourette syndrome? (yes/no) If yes, please continue to question 2.

- 2) Are you (or your diagnosed child) regularly engaged in a musical activity such as playing or practicing a musical instrument? (yes/no) If yes, please continue to question 3 and following.

- 3) How many minutes per week do you (or your child) engage in this activity?

- 4) What musical instrument do you (or your child) primarily play?

- 5) On a scale from one to five, please rate the level of change (if any) that occurs in your symptoms while playing your instrument.

1/drastic symptoms increase 2/slight symptoms increase 3/symptoms do not change 4/ slight symptoms decrease 5/ drastic symptoms decrease

6) Please use the following list and check some or all of the symptoms of TS you (or your child) have been diagnosed;

Tics of the face (eyes, nose or mouth)	Grunting
Tics of the arms/legs	Throat clearing
Trunk tics	Shouting
Kicking/Stamping	Barking
Touching	Echolalia (echo speech)
Other (_____)	Coprolalia (involuntary utterance of obscene words)

6a) If you checked "other" please specify. _____

7) What age are you (or your child) for which you're completing this questionnaire?

8) Gender? Male/Female

9) Ethnic group? White, African American, Hispanic, Asian American, Indian American, Middle Eastern, Native American

Thank you for your time. Please submit this survey only once.

Appendix C: IRB Exemption Letter



RESEARCH INTEGRITY AND COMPLIANCE
Institutional Review Boards, FWA No. 00001669
12901 Bruce B. Downs Blvd., MDC0353 • Tampa, FL 33612-4799
813.974-5638 • FAX 813.974-7091

6/19/2014

William Brown
USF School of Music
4202 E. Fowler Avenue MUS 101
Tampa, FL 33620

RE: **Exempt Certification**

IRB#: Pro00017048

Title: The Influence of Playing a Musical Instrument on Tourette Syndrome Symptoms

Study Approval Period: 6/19/2014 to 6/19/2019

Dear Mr. Brown:

On 6/19/2014, the Institutional Review Board (IRB) determined that your research meets USF requirements and Federal Exemption criteria as outlined in the federal regulations at 45CFR46.101(b):

(2) Research involving the use of educational tests (cognitive, diagnostic, aptitude, achievement), survey procedures, interview procedures or observation of public behavior, unless:
(i) information obtained is recorded in such a manner that human subjects can be identified, directly or through identifiers linked to the subjects; and (ii) any disclosure of the human subjects' responses outside the research could reasonably place the subjects at risk of criminal or civil liability or be damaging to the subjects' financial standing, employability, or reputation.

Your study qualifies for a waiver of the requirements for the documentation of informed consent as outlined in the federal regulations at 45CFR46.117(c) which states that an IRB may waive the requirement for the investigator to obtain a signed consent form for some or all subjects if it finds either: (1) That the only record linking the subject and the research would be the consent document and the principal risk would be potential harm resulting from a breach of confidentiality. Each subject will be asked whether the subject wants documentation linking the subject with the research, and the subject's wishes will govern; or (2) That the research presents no more than minimal risk of harm to subjects and involves no procedures for which written consent is normally required outside of the research context.

As the principal investigator for this study, it is your responsibility to ensure that this research is conducted as outlined in your application and consistent with the ethical principles outlined in the Belmont Report and with USF IRB policies and procedures. Please note that changes to this

protocol may disqualify it from exempt status. Please note that you are responsible for notifying the IRB prior to implementing any changes to the currently approved protocol.

The Institutional Review Board will maintain your exemption application for a period of five years from the date of approval or for three years after a Final Progress Report is received, whichever is longer. If you wish to continue this protocol beyond five years, you will need to submit a new application at least 60 days prior to the end of your exemption approval period. Should you complete this study prior to the end of the five-year period, you must submit a request to close the study.

We appreciate your dedication to the ethical conduct of human subject research at the University of South Florida and your continued commitment to human research protections. If you have any questions regarding this matter, please call 813-974-5638.

Sincerely,

A handwritten signature in cursive script that reads "John A. Schinka, Ph.D.".

John Schinka, Ph.D., Chairperson
USF Institutional Review Board